Report Information from ProQuest

09 November 2023 07:47

TABLE OF CONTENTS

Search Strategy..... v

1. Community-based health literacy focused intervention for cervical cancer control among Black women living with human immunodeficiency virus: A randomized pilot trial..... 1

2. The importance of personal documentation for patients living with long-term illness symptoms after pituitary surgery: A Constructivist Grounded Theory study..... 14

3. Becoming a secondary actor of one's own life: A qualitative study of the experiences of informal caregivers in the care of people with chronic pain..... 29

4. Alignment of patient-centredness definitions with real-life patient and clinician experiences: A qualitative study..... 42

5. Problem-based shared decision making: The role of canonical SDM steps..... 57

6. Exploring first-time mothers' experiences and knowledge about behavioural risk factors for stillbirth..... 67

7. A valued voice: A qualitative analysis of parental decision-making preferences in emergent paediatric surgery..... 84

8. Service-user experiences of an integrated psychological intervention for depression or anxiety and tobacco smoking in improving access to psychological therapies services: A qualitative investigation into mechanisms of change in quitting smoking..... 98

9. Children, young people and parent engagement in health intervention design and implementation: A scoping review..... 114

10. Use of co-design methodology in the development of cardiovascular disease secondary prevention interventions: A scoping review..... 136

11. Evaluating the role and effectiveness of co-produced community-based mental health interventions that aim to reduce suicide among adults: A systematic review..... 155

12. Co-designing an intervention to improve the process of deprescribing for older people living with frailty in the United Kingdom..... 197

13. Pathways to recovery model of youth substance misuse in Assam, India..... 213

14. ‘I don't know what to do or where to go’. Experiences of accessing healthcare support from the perspectives of people living with Long Covid and healthcare professionals: A qualitative study in Bradford, UK..... 228

15. COVID-19 community assessment hubs in Ireland: A study of staff and patient perceptions of their value 242

16. ‘It feels like my metabolism has shut down’. Negotiating interactional roles and epistemic positions in a primary care consultation..... 260

17. Bereaved parents involvement in maternity hospital perinatal death review processes: ‘Nobody even thought to ask us anything’..... 272

18. Experiences of antibiotic use among Brazilian healthcare users: An exploratory study..... 298

19. Compliant citizens, defiant rebels or neither? Exploring change and complexity in COVID-19 vaccine attitudes and decisions in Bradford, UK: Findings from a follow-up qualitative study..... 310

20. Influences on indoor environmental trigger remediation uptake for children and young people with asthma: A scoping review..... 324

TABLE OF CONTENTS

21. Developing a Health Literacy Scale for adults in Hong Kong: A modified e-Delphi study with healthcare consumers and providers..... 337

22. ‘What price do you put on your health?’: Medical cannabis, financial toxicity and patient perspectives on medication access in advanced cancer..... 351

23. ‘it was that … specialist … that finally listened to us … that's probably a weird answer to what you were expecting’: Clinician and carer perspectives on brilliant feeding care..... 366

24. Co-creation of information materials within the assent process: From theory to practice..... 379

25. How do clients with multiple problems and (in)formal caretakers coproduce integrated care and support? A longitudinal study on integrated care trajectories of clients with multiple problems..... 399

26. Integrating Patient-reported Experience (PRE) in a multistage approach to study access to health services for women with chronic illness and migration experience..... 415

27. A qualitative investigation of perceptions towards antibiotics by members of the public after choosing to pledge as an Antibiotic Guardian..... 426

28. Preferences of patients with chronic low back pain about nonsurgical treatments: Results of a discrete choice experiment..... 442

29. Incorporation of the emotional indicators of the patient journey into healthcare organization management 473

30. Patient views on asthma diagnosis and how a clinical decision support system could help: A qualitative study..... 478

31. What are the reasons for unfinished nursing care as perceived by hospitalized patients? Findings from a qualitative study..... 491

32. Acceptability of integrating smoking cessation treatment into routine care for people with mental illness: A qualitative study..... 507

33. Understanding the cultural environment of the outpatient care setting for patients with dementia receiving cancer treatment: A qualitative study..... 520

34. Implementation of a patient-reported experience measure in a Dutch disability care organization: A process evaluation of cocreated tailored strategies..... 533

35. For autistic persons by autistic persons: Acceptability of a structured peer support service according to key stakeholders..... 556

36. Public contributors' preferences for the organization of remote public involvement meetings in health and social care: A discrete choice experiment study..... 573

37. Barriers and enabling factors in weight management of patients with nonalcoholic fatty liver disease: A qualitative study using the COM-B model of behaviour..... 595

38. Parental COVID-19–related health information practises, sources, evaluations and needs: A qualitative interview study..... 619

39. Youth engagement in mental health research: A systematic review..... 634

40. Development of an inflammatory bowel disease (IBD) Patient-Reported Experience Measure (PREM): A patient-led consensus work and ‘think aloud’ study for a quality improvement programme..... 685

TABLE OF CONTENTS

41. Comparing an in-person workshop and a postal Delphi survey for involving health service users in health care and health research prioritization..... 705

42. Embedding health literacy research and best practice within a socioeconomically and culturally diverse health service: A narrative case study and revised model of co-creation..... 722

43. Trauma-informed patient and public-engaged research: Development and evaluation of an online training programme..... 736

44. Stakeholder outcome prioritization in the Biologic Abatement and Capturing Kids' Outcomes and Flare Frequency in Juvenile Spondyloarthritis (BACK-OFF JSpA) trial..... 750

45. Women's experiences along the ovarian cancer diagnostic pathway in Catalonia: A qualitative study..... 764

46. Conceptual and practical challenges associated with understanding patient safety within community-based mental health services..... 776

Bibliography..... 794

SEARCH STRATEGY

Set No.	Searched for	Databases	Results
S1	Health Expectations	Ebook Central, Public Health Database, Publicly Available Content Database	420337*

* Duplicates are removed from your search, but included in your result count.

Community-based health literacy focused intervention for cervical cancer control among Black women living with human immunodeficiency virus: A randomized pilot trial

Hae-Ra Han [1]
; Mendez, Kyra J W [2] ; Perrin, Nancy [3] ; Cudjoe, Joycelyn [4] ; Taylor, Gregory [5] ; Baker, Dorcas [6] ; Murphy-Stone, Jeanne [7] ; Sharps, Phyllis [3] [1] The Johns Hopkins University School of Nursing, Baltimore, Maryland, USA; The Johns Hopkins University Bloomberg School of Public Health, Baltimore, Maryland, USA; Center for Community Programs, Innovation, and Scholarship, Baltimore, Maryland, USA [2] National Cancer Institute, Bethesda, Maryland, USA [3] The Johns Hopkins University School of Nursing, Baltimore, Maryland, USA [4] United States Government Accountability Office, District of Columbia, Washington, USA [5] University of Maryland School of Medicine, Baltimore, Maryland, USA [6] Older Women Embracing Life (OWEL), Baltimore, Maryland, USA [7] School of Nursing, The George Washington University, Washington, DC, USA

ABSTRACT (ENGLISH)

Background
Health literacy plays an essential role in how individuals process health information to make decisions about health behaviours including cancer screening. Research is scarce to address health literacy as a strategy to improve cancer screening participation among women living with human immunodeficiency virus (HIV), particularly Black women who, despite the heavy burden of cervical cancer, report consistently low screening rates.
Aim
To assess the feasibility, acceptability and preliminary efficacy of a health literacy-focused intervention called CHECC-uP—Community-based, HEalth literacy focused intervention for Cervical Cancer control—among women living with HIV.
Methods
We conducted a community-based, single-blinded randomized pilot trial. A total of 123 eligible women were enrolled and randomized to one of two conditions, control (i.e., cervical cancer brochure) or intervention (cervical cancer brochure plus 30–60 min health literacy-focused education followed by monthly phone counselling and navigation assistance for 6 months). Study assessments were done at baseline, 3 and 6 months. The final analysis sample included 58 women who completed all data points and whose Papanicolaou (Pap) test status was confirmed by medical records.
Results
All intervention participants who completed the programme would recommend the CHECC-uP to other women living with HIV. However, adherence in the experimental conditions was low (49.6% attrition rate including 20 women who dropped out before the intervention began) due, in large part, to phone disconnection. Those who had received the intervention had a significantly higher Pap test rate compared to women in the control group at 6 months (50% vs. 21.9%, p=.025). Participation in the intervention programme was associated with improved health literacy and other psychosocial outcomes at 3 months but the trend was attenuated at 6 months.
Conclusions

The CHECC-uP was highly acceptable and led to improved Pap testing rates among Black women living with HIV. Future research should consider addressing social determinants of health such as phone connectivity as part of designing a retention plan targeting low-income Black women living with HIV.

Implications

The findings should be incorporated into a future intervention framework to fulfil the unmet needs of Black women living with HIV to facilitate their decision-making about Pap test screening.

Patient or Public Contribution

Nineteen community members including women living with HIV along with HIV advocates and care providers participated in four focus groups to develop cervical cancer screening decision-relevant information and the health literacy intervention. Additionally, a community advisory board was involved to provide guidance in the general design and conduct of the study.

FULL TEXT

INTRODUCTION

Despite considerable progress in US cancer control over the past decades, certain groups continue to experience significant health disparities. Women living with human immunodeficiency virus (HIV) (WLH) experience a disproportionate cervical cancer burden because of an impaired immune response to the human papillomavirus, the virus that causes cervical cancer.[1] In particular, Black women have the highest cervical cancer mortality.[2] Regular Papanicolaou (Pap) testing is accepted as a critical strategy in the early detection and timely treatment of cervical cancer and precancerous lesions.[3] Yet, a large cross-sectional study found that cervical cancer screening decreased in the United States between 2005 and 2019.[4] WLH, especially Black women have reported consistently lower Pap test rates compared to other groups.[5]

Health literacy—'the degree to which individuals have the capacity to obtain, process, and understand basic health information and services to make appropriate health decisions' (para. 1)—is a key social determinant of health and is recognized as an essential element of access to high-quality, patient-centred care.[6,7] Health literacy deficits are a significant barrier to obtaining Pap tests.[6] While research on health literacy among WLH is scarce,[7] studies involving women without HIV[8–10] have reported that women with limited health literacy are more likely to misunderstand health information provided and find it difficult to convert and interpret proportions of their cancer risk, which increases women's misperceptions and lowers their personalization of such risks. Consequently, low health literacy negatively affects knowledge, attitudes and self-efficacy with regard to cervical cancer screening.[8,11] Approximately 25%–38% of people living with HIV have limited health literacy,[12,13] compared to the national rate of 9% for the general US population.[14] The rate of low health literacy is even higher among Black WLH. For example, in a recent cross-sectional study, nearly half (49.6%) of the Black WLH had a reading level at or below sixth grade, suggesting that the women may struggle with most written health information.[15]

Systematic reviews and meta-analyses[16,17] of interventions designed to increase Pap screening participation among ethnic minority populations revealed that interventions have focused primarily on increasing knowledge (e.g., causes, risk factors or signs and symptoms of the disease) or accommodating women's needs and have produced small effect sizes of 5%–24%. None of the studies in these reviews has attempted to directly address study participants' health literacy deficits as a strategy to improve cancer screening participation rates. Examples of health literacy interventions may include training on how and when to access healthcare, medical terminology training or numeracy training by using visual aids.[18] Further, only one study addressed WLH, in which a randomized controlled trial was conducted to test an intervention where WLH collected their own human papillomavirus samples and then received counselling based on their results.[19] This intervention failed to improve Pap test screening among WLH.[19] There is a need for promising innovations that can address the health literacy needs of WLH, who suffer disproportionately from unequal cervical cancer burden.[1,2]

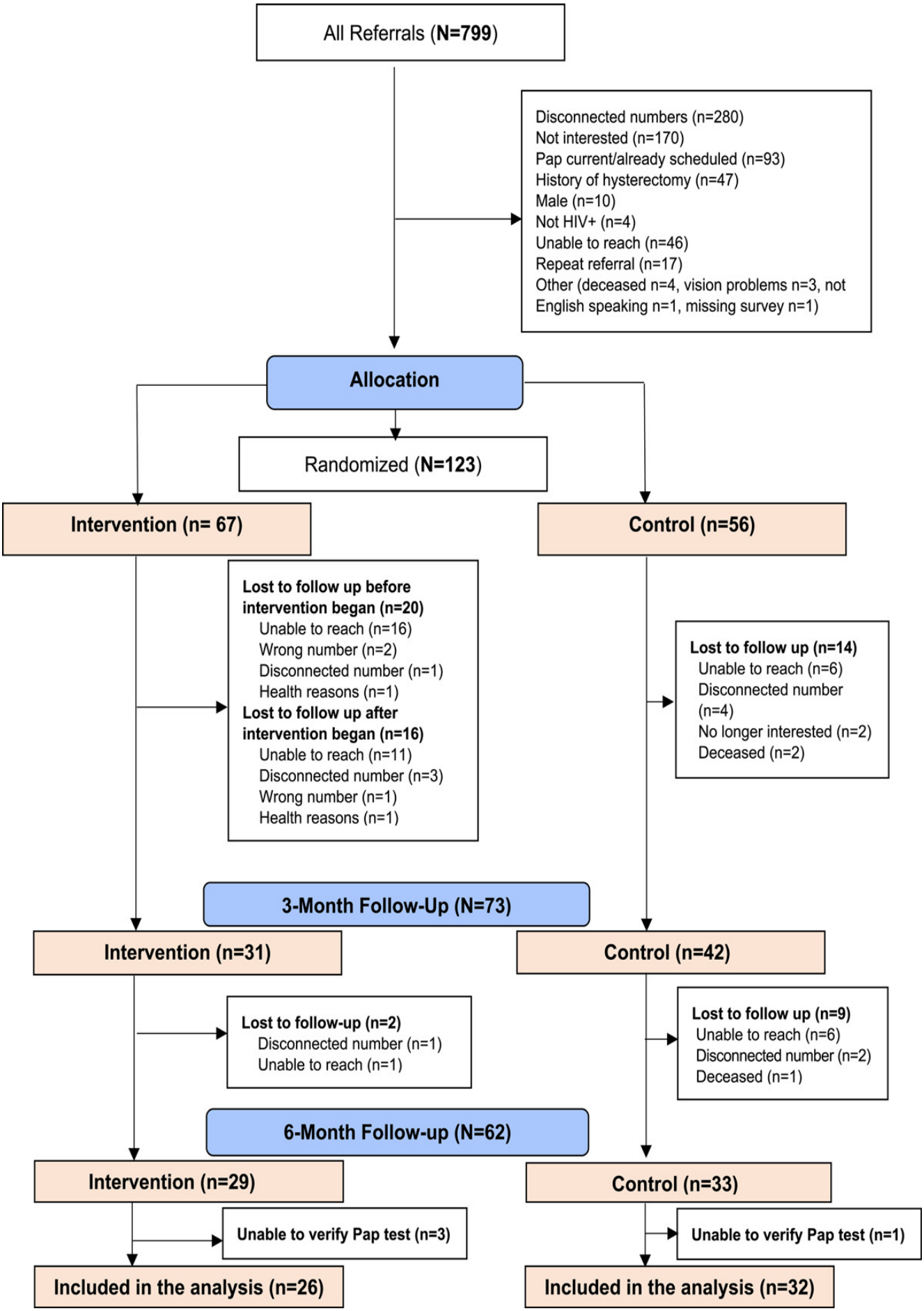
The current study was designed to address this gap by testing a health literacy-focused intervention programme called CHECC-uP—Community-based, HEalth literacy focused intervention for Cervical Cancer control among WLH. We conducted a pilot study with 3- and 6-month follow-ups to evaluate the feasibility, acceptability and

preliminary efficacy of this intervention. We hypothesized that participation in the CHECC-uP intervention would be associated with an increase in Pap testing and improvements in psychosocial outcomes.

METHODSDesign and sample

We used a community-based, randomized controlled trial design to pilot test the CHECC-uP intervention compared to an educational control (Clinical Trials Registry NCT03033888). Women were recruited from inner-city HIV clinics, community organizations serving people with HIV or a university-based HIV/AIDS research centre in Baltimore, MD, by posting study flyers in these organizations or advertising the study through social media (e.g., Facebook, Craigslist) and attending health fairs.[20] Additionally, the study team received names and contact information of potential study participants through a university-based HIV/AIDS research centre hotline; people living with HIV would call the HIV/AIDS research centre for potential research participation and hotline staff would provide their information to the study team if they met the study eligibility criteria.[20] Upon these self- or direct referrals, trained study staff screened potential study participants for eligibility over the phone and scheduled a study visit for informed consent and baseline data collection. Eligible participants were: (1) women aged 18 years or older; (2) diagnosed with HIV; (3) overdue for a Pap test (e.g., no Pap test within the last 12 months at the time of study enrollment) and (4) could speak and understand English. Women with a hysterectomy were excluded. The study was designed to detect an increase of 20% in the proportion of women in the intervention arm completing Pap testing at 6-month follow-up, compared to those in the control arm, with 80% power and α of .05.[21,22] Assuming a drop-out rate of 30%, we estimated that we would need to enroll a total of 122 women.

A total of 123 eligible women completed the study assessment at baseline and were randomized (intervention, n = 67; control, n = 56). Of those who completed the baseline assessment and were assigned to the intervention arm, 20 dropped out before the intervention began for several reasons including unable to reach after multiple attempts (n = 16), wrong or disconnected phone number (n = 3) and health reasons (n = 1). Those, whom we were unable to reach after attempting up to eight calls on different days and times (including weekends and evening hours), were considered dropouts. As a result, our intervention was delivered to 47 participants. Of those, 18 discontinued their participation before the final data collection assessments at 6 months were done, yielding 29 in the analysis sample for the intervention arm. As for the control arm, 23 dropped out over the course of the study, yielding 33 in the analysis sample (Figure 1).



Enlarge this image.

Randomization and intervention

We used computer-generated random numbers to randomize women to either the intervention or control arm. The

control arm received an educational brochure related to cervical cancer among WLH created for the purpose of the study. Women in the intervention arm received the educational brochure plus the study intervention, which consisted of health literacy education and phone counselling with navigation assistance. Specifically, a trained community health worker delivered health literacy education at a community centre conveniently located near a subway station in the central downtown area. The health literacy education (Table 1) was designed to promote WLH's understanding of basic medical terminology used in cervical cancer screening; relevant medical instructions, such as appointment slips or follow-up screening instructions and familiarity with how to navigate the healthcare system for Pap test screening.

Table 1 Main educational topics with examples of medical terminologies and role-play contents

Topic	Example medical terminology practised	Example content covered in role-play
HIV and cancer	Human papillomavirus, cancer, sexually transmitted disease	
What is Pap smear	Cervix, Pap smear	In the doctor's office: History taking
Why is it important to receive a Pap test	Cervical cancer, reproductive organs	
Cervical cancer symptoms	Hormones, genital warts	
How is Pap smear done	Laboratory (lab), pelvic exam, polyp, speculum	In the doctor's office: Pap smear
Things to remember before and after Pap smear	Abnormalities, history taking	

Abbreviations: HIV, human immunodeficiency virus; Pap, Papanicolaou.

Development of our intervention to include health literacy as its core component was guided by the Precede-Proceed model,[23] which identifies critical constructs as predisposing (e.g., individual characteristics), enabling (e.g., health literacy knowledge, self-efficacy) and reinforcing factors (e.g., cultural beliefs and attitudes). HIV community advocates, Black WLH and clinicians working closely with WLH engaged in the formative work to develop the health literacy intervention by sharing their experiences at OB/GYN clinics and identifying common scenarios and dialogues that occur between the patient and medical staff when navigating a Pap test screening. Based on this formative work, a picture guidebook was created as educational material for WLH.

At the end of the health literacy-focused education session, women in the intervention group received a copy of the picture guidebook to reinforce what they had learned and practised in class. The follow-up portion of the study intervention included monthly phone counselling for up to 6 months. Using a checklist addressing key talking points, the objectives of the follow-up were to (1) reinforce health literacy knowledge and skills learned and practised from the education session; (2) address any questions or concerns the participant might have and (3) provide tailored navigation assistance with individually identified barriers to Pap test screening over a 6-month period.

Procedures

The Johns Hopkins Institutional Review Board approved the study protocol. Once eligible women were identified, trained research assistants scheduled a visit to obtain written informed consent and collect baseline data at several

community sites (e.g., nurse-run community health centres or community organizations serving people with HIV). Upon completion of the baseline assessment, a trained community health worker delivered health literacy education to women assigned to the intervention arm.

Initially, education sessions were scheduled for groups of six to eight women. However, the group-based format presented substantial scheduling challenges to the study team with high rates of no-shows. This led to the study team's decision to adopt individual education delivery. The education sessions lasted about 30 min, when offered individually, and 45–60 min, when offered as a group due to discussion during the group session. Within 1–2 weeks after completing education, intervention participants received monthly phone counselling sessions for 6 months. During each phone call, a counsellor checked the participant's progress toward completing a Pap test and answered questions or concerns about Pap test screening.

For both intervention and control arms, we provided a copy of the Pap test brochure tailored to WLH, highlighting causes and symptoms of cervical cancer, risk factors for cervical cancer among WLH, the value of Pap screening and how to prepare for a Pap test. All of our educational materials were written at a sixth grade level or lower, as assessed by Flesch-Kincaid grade-level statistics in Microsoft Word. Additionally, all women in the study received a list of local community resources where a Pap test could be obtained free, or at a reduced cost, based on a sliding scale.

Trained study staff who were blinded to the group assignment collected data at baseline, 3 and 6 months from the start of the intervention. After 6 months, intervention women were invited to postintervention qualitative interviews to share their experiences with CHECC-uP. Every woman provided informed written consent. Enrolled participants received $20 at baseline and 3 months and $40 at 6 months for their time. Postintervention interview participants received an additional $30.

Measures

A study questionnaire was used to collect participants sociodemographic and medical characteristics. Data regarding Pap test status were assessed via medical record review. We used several study instruments to assess changes in WLH's psychosocial outcomes: Health literacy, cancer knowledge, self-efficacy, cultural beliefs/attitudes and depression. We include the internal consistency for each instrument, which was calculated using the full sample (n = 123) at baseline.

To assess health literacy, we used familiarity, navigation and numeracy subscales from the Assessment of Health Literacy in Cancer Screening (AHL-C), a validated comprehensive health literacy instrument with α coefficients ranging from .70 to .96.[24] Building on Baker's conceptual model of health literacy,[25] the AHL-C addresses multiple types of health literacy in cancer screening, such as reading ability, familiarity, navigation, comprehension and numeracy. We chose familiarity, navigation and numeracy because they have been associated with cancer knowledge,[26] risk perception,[27] intent to get cancer screening[26,28] and actual cervical cancer screening behaviour.[29] The familiarity subscale includes 12 items (5-point Likert scale; 1 = never heard before to 5 = can use fluently) with scores ranging from 12 to 60. The navigation and numeracy subscales include 12 and 7 items, respectively; each correct response to the items on the subscales is coded as 1, with possible scores ranging from 0 to 12 and 0 to 7, respectively. Example questions included: ‘How familiar are you with the following words’ or ‘Please read the passages below and select a word to fill in each blank’. α Coefficients ranged from .51 to .94 in the study sample.

Cancer knowledge was measured by the Cervical Cancer Knowledge (CCK) Test which consists of 10 items (α coefficient = .80–.89).[30] An example question is ‘If one smokes heavily, the risk for cervical cancer increases’. Given the direct link between HPV and cervical cancer, we added 12 items about HPV to the CCK Test (e.g., ‘A person who has HPV needs to have Pap smears more often than others’). Correct responses to each of the knowledge questions were scored 1, with possible total scores from 0 to 22. Higher scores indicated higher cancer knowledge. The modified CCK Test had an α coefficient of .77 in the study sample.

Self-efficacy related to cervical cancer screening was assessed by the Cervical Cancer Self-Efficacy scale.[31] The self-efficacy scale includes four items (4-point Likert scale; 1 = not at all confident to 4 = very confident) asking how confident a woman is in carrying out tasks related to Pap tests, with higher scores indicating higher self-efficacy. An

example question is ‘Do you feel confident that you can schedule a Pap test appointment and keep it?’ The scale was validated in Mexican and Korean American women with high internal consistency reliability coefficients ranging from 0.92 to 0.95.[32,33] The α coefficient was .89 in this sample.

Cultural beliefs and attitudes were assessed using a modified nine-item inventory (5-point Likert; 1 = strongly disagree to 5 = strongly agree), which was adopted from the cultural barriers to breast and cervical cancer screening questionnaire.[34,35] The original scale was validated on young Asian American women and older Chinese American women with α coefficients ranging from .61 to .72. Example questions include ‘I would feel embarrassed with a doctor examining my cervix as part of a medical exam’, and ‘I only see a doctor when I am having a health problem’. The α coefficient of the modified scale was .8 in the study sample.

Depressive symptoms were measured using the Patient Health Questionnaire-9 (PHQ-9). PHQ-9 is a well-validated and widely disseminated screener for depressive symptoms.[36] The score of each participant is calculated by summing the scores for nine questions (4-point Likert scale; 0 = not at all to 3 = nearly every day) asking about the presence of signs and symptoms of depression during the 2 weeks before the survey. Total scores range from 0 to 27, with scores of 5, 10, 15 and 20 represent mild, moderate, moderately severe and severe depression, respectively. The Cronbach's α of the PHQ-9 was .88 in the study sample.

We also collected data on the feasibility and acceptability of the CHECC-uP. The feasibility of the study was examined using multiple sources of data, such as study recruitment and retention, attendance at education sessions and follow-up phone counselling completion rates. Acceptability was assessed using a questionnaire developed for the purpose of this study. The survey included self-reported satisfaction with the intervention programme, as well as the receipt (e.g., reading the intervention material), helpfulness and application (e.g., applied contents from the material to get a Pap test) of intervention materials.

Statistical analyses

Analysis was performed using data from the 58 participants who completed all data points and whose Pap test status was confirmed objectively by medical records (Figure 1). We used descriptive statistics such as means, standard deviations (SDs) and frequencies to establish analysis sample characteristics and study variables. Intervention and control groups were compared at baseline using chi-squared tests or independent sample *t*-tests. The primary efficacy outcome was the completion of a Pap test, which was tested with a χ^2 test. Change over time in the psychosocial outcomes was tested with repeated measures analysis of variance with time, group and the group ×time interactions included in the model. We calculated effect sizes using the group difference in the mean change from baseline to 3-month follow-up, and the group difference in the mean change from baseline to 6 months follow-up, each divided by the baseline SD.[37]

RESULTSSample characteristics

The final sample size included 58 participants (Figure 1). There were significant differences in age and cultural beliefs and attitudes scores between the participants who completed the study and those who did not. Specifically, participants who completed the study were 4 years older (p = .008) and had a 2.3-point lower score on the cultural beliefs and attitudes scale (p = .048) at baseline. Among the intervention group women, there were no significant differences between participants who did not complete the intervention (n = 38) and those who did (n = 29).

The baseline characteristics of 58 participants included in the analysis are summarized in Table 2. The only significant difference between intervention and control groups at baseline was that participants in the intervention group were about 6 years younger, on average than participants in the control group (p = .003). Overall, the participants in the analysis sample were middle-aged (mean: 53.5 years, SD: 7.8) and all were Black or African American. Most women were never married (49.1%), separated, widowed or divorced (29.8%). More than 40% of women had less than a high school education. Nearly 9 out of 10 (89.5%) were unemployed, retired or disabled, and only 27.3% of WLH reported they could live comfortably or very comfortably with their income. The majority of our sample was renting their current residence (69%). Finally, most had a primary care physician (98.2%) and 96.4% of women reported having a pap test at some time in their life.

Table 2 Analysis sample characteristics at baseline (N = 58)

Variable	Total (*N* = 58), *n* (%) or mean ±SD	Control (*n* = 32), *n* (%) or mean ±SD	Intervention (*n* = 26), *n* (%) or mean ±SD	*p* Value
Age in years (range = 28–67)	53.5 ±7.8	56.3 ±5.6	49.8 ±8.9	.003
Black/African American	58 (100)	32 (100)	26 (100)	
Marital status				.053
Married or partnered	12 (21.3)	3 (9.4)	9 (36.0)	
Separated, widowed or divorced	17 (29.8)	13 (40.6)	4 (16.0)	
Never married	28 (49.1)	16 (50.0)	12 (48.0)	
Missing	1		1	
Education				.246
<High school	24 (42.1)	14 (45.2)	10 (38.5)	
High school	18 (31.6)	7 (22.6)	11 (42.3)	
Some college or more	15 (26.3)	10 (32.3)	5 (19.2)	
Missing	1	1		
Employment				.820
Working full- or part-time	6 (10.5)	3 (9.7)	3 (11.5)	
Unemployed, retired or disabled	51 (89.5)	28 (90.3)	23 (88.5)	
Missing	1	1		
Income level				.826
Very comfortable or comfortable	15 (27.3)	9 (29.0)	6 (25.0)	
Just OK	24 (43.6)	14 (45.2)	10 (41.7)	

Difficult/very difficult to manage	16 (29.1)	8 (25.8)	8 (33.3)	
Missing	3	1	2	
Type of residence				.957
Own	3 (5.2)	2 (6.3)	1 (3.8)	
Renting	40 (69.0)	22 (68.8)	18 (69.2)	
Public housing	10 (17.2)	5 (15.6)	5 (19.2)	
Other	5 (8.6)	3 (9.4)	2 (7.7)	
Have health insurance	58 (100)	32 (100)	26 (100)	
Have PCP	56 (98.2)	31 (96.9)	25 (96.2)	.373
Ever had a Pap test (Yes)	54 (96.4)	31 (96.9)	23 (96.9)	.109
Own a smartphone	37 (77.1)	18 (72.0)	19 (82.6)	.382

Abbreviations: Pap, Papanicolaou; PCP, primary care provider.

Feasibility and acceptability

We recruited and randomized the target sample size of 123 with a retention rate of 50.4% (or 60.2% after accounting for early dropouts in the intervention arm; e.g., those who completed the baseline assessment but left the study before receiving the study intervention, $n = 20$). The size of health literacy education classes ranged from 1 to 5 participants before we changed it into an individually based format. The intervention participants in the analysis sample completed on average about one phone counselling session (range = 0–4; median = 1).

The CHECC-uP intervention was highly acceptable. All intervention women who responded to the acceptability questionnaire ($n = 26$) would recommend the programme to other WLH. Nearly all respondents to the questionnaire were satisfied or very satisfied with the information they learned about Pap test screening (96.2%) and the way they learned (96.2%). More than two thirds (73.1%) of the responders indicated they read the picture guidebook on their own, partially or entirely. Slightly more than a quarter of them (26.3%) used the picture guidebook when getting a Pap test to better understand the process. Eighty percent of women who used the picture guidebook found it helpful and 20% somewhat helpful. Similarly, the majority of responders reported that they read the Pap test brochure and the community resource list partially or entirely (88.5% and 76.9%, respectively). Of those who read the brochure, 87% found it helpful or very helpful. Forty percent (40%) of women who read the community resource list indicated that they used the list to find a place for a Pap test; 75% noted the list as being very helpful or helpful and 25% somewhat helpful.

Changes in Pap test screening and psychosocial outcomes

At 6 months, 50% of WLH in the intervention group received a Pap test, compared to 21.9% of women in the control group (28.1% difference; $\chi^2 = 5.02$, $p = .025$). The effect sizes of the CHECC-uP intervention on psychosocial outcomes at 3 and 6 months are presented in Table 3. At baseline, the overall analysis sample had relatively high

levels of familiarity (mean = 39.5, SD = 13.2) and health navigational literacy (mean = 10.9, SD = 1.76) and low levels of numeracy (mean = 3.44, SD = 1.62). At 3 months, the mean increases in familiarity and numeracy were greater in WLH in the intervention arm compared to those in the control arm with effect sizes of 0.34 and 0.23, respectively. The mean increase in health navigational literacy was higher in WLH in the control arm at 3 months, but the trend reversed favouring the intervention arm at 6 months with a negligible effect size.

Table 3 Outcome changes over 6 months[a]

Note: Health literacy variables are italicized.

Abbreviation: Pap, Papanicolaou.

a

n = 58 with full data on both psychosocial outcomes and Pap test status based on medical record review.

b

Group difference in mean change scores from baseline to 3 months divided by the standard deviation at baseline.

c

Group difference in mean change scores from baseline to 6 months divided by the standard deviation at baseline.

For other psychosocial variables, the absolute value of effect sizes ranged from 0.25 to 0.58 at 3 months and from <0.01 to 0.54 at 6 months. For cervical cancer knowledge, the intervention arm had a greater increase at 3 months, but the difference was not sustained at 6 months, with relatively no difference between the intervention and control arms. For self-efficacy, the intervention arm had a greater increase, but with a reduced effect size at 6 months. Cultural beliefs addressing cultural barriers, such as modesty, declined for both groups at 3 months, with the intervention arm having a significantly greater reduction. At 6 months, the intervention arm maintained the declining trend while the control arm reversed back. Finally, depression scores declined for both groups at 3 months, but the intervention arm had a greater decrease with an effect size of 0.28; though the difference was not sustained at 6 months. For all psychosocial variables, the statistical test of change over time was significant only for cultural beliefs at 3 months (p = .024).

DISCUSSION

We found that a multifaceted, health literacy-focused intervention (CHECC-uP) can promote Pap testing among Black WLH. However, we experienced a high attrition rate in the study sample. The findings demonstrate the preliminary efficacy of CHECC-uP for Black WLH as a potential strategy to reduce cervical cancer disparities in this population. To the best of our knowledge, CHECC-uP is the first intervention to integrate health literacy education as an active component to promote Pap testing among WLH. The statistically significant difference in Pap test rates observed among WLH in the trial (28.1% difference) is higher than other reported rates for HIV-negative women, ranging from 5% to 24%.[16,17] The theory-driven intervention programme was well received by our sample, as evidenced by the acceptability measures including 100% of intervention women in the analysis sample who would recommend the CHECC-uP to other WLH. We believe the involvement of community stakeholders in developing the intervention approach may have helped to promote the credibility of CHECC-uP as relevant to the target community.[38]

Health literacy consists of multiple dimensions that go beyond one's reading ability.[29] Of the three dimensions measured in the study, the effect sizes for both familiarity and numeracy favoured the intervention and remained consistent at 3 and 6 months. In contrast, the effect size for navigational health literacy was either not in favour of the intervention arm, or negligible. Navigational health literacy addresses one's understanding of how to navigate the process of undergoing cancer screening (e.g., check-in and -out at an OB/GYN clinic, dialogue between a woman and a doctor about risk factors for cervical cancer).[24] Our finding may be a result of the study sample mostly being recruited from HIV clinics or an HIV/AIDS research centre (65.9%).[20] Different from prior research, in which women without HIV were recruited from nonclinical settings such as ethnic churches, the current study sample included women with prior exposure to the healthcare system. Nearly perfect baseline scores on the navigation subscale (possible ranges = 0–12) observed in both the intervention and control arms (about 11 points) indicate a high ceiling effect with the limited utility of the subscale as a health literacy outcome measure in our sample of WLH.

Our retention rate was not optimal. We had higher dropouts among younger women and women who scored higher on the cultural attitudes and beliefs scale. The role of age in cervical cancer screening participation is not at all consistent.[39–41] Cultural beliefs and attitudes in cancer screening address embarrassment about the body or sexuality and modesty.[34,35] A recent focus group study involving WLH noted feelings of shame and embarrassment when talking about cervical cancer and Pap smears as a barrier to screening for WLH.[38] These findings suggest the need for more tailored retention approaches to those at risk for dropout by showing empathy, active listening and open communication to allow expressing one's feeling, while also sharing acceptable strategies based on beliefs (e.g., community resources listing clinics with female doctors).[42] Additionally, a recent review of the literature for recruitment and retention of WLH in clinical studies reported attrition rates between 15% and 33%.[20] The published studies included in the review used on-site staff and/or multiple engagement methods to retain participants (e.g., sending holiday or birthday cards, sending newsletters or offering stipends for childcare or transportation to study sites).[20] Due to constraints in terms of resources, our study used trained study staff to recruit women from participating sites, upon referrals, with the main methods of engagement being reminder calls and nominal stipends for transportation to the data collection sites. The findings highlight the need for working with HIV clinical partners and the use of multiple, individually tailored engagement approaches to retain WLH in a clinical trial.

Another important lesson learned from this pilot trial is that at least one-third of the attrition observed in our study was early dropouts in the intervention arm, which led to a change in the education format from group sessions to individually based sessions. Benefits of group-based education have included cost saving[43] and peer support.[44] Despite our best efforts, the study team experienced logistical challenges in scheduling group education sessions (with delays of up to a month or longer) due to the different schedules and needs of WLH. The challenges were due, in large part, to phone disconnection. According to a national report,[45] adults living in poverty (69%) or in rented homes (76%) had a higher probability of being ‘wireless only’, with no landline telephone, compared to higher-income adults (59%) and adults living in a house owned by a household member (53%; p. 3). Nevertheless, physical access to cell phones may not be enough to ensure connectivity. Adults with low incomes often must purchase minutes because they do not have cell phone plans.[46] Without contract plans, users often must change numbers, or get disconnected, until more minutes are purchased, resulting in people experiencing periods of ‘phonelessness’ (p. 1428).[46] Future trials involving low-income WLH should consider addressing phone connectivity as part of their retention plan. For example, the federal Lifeline programme provides discounted or free phones and services for low-income families in the United States.[47]

Study limitations include an insufficient sample size to detect a statistically significant change in outcomes, which resulted from high attrition. Nevertheless, the effect sizes estimated for the study variables are encouraging and warrant further investigation to test the efficacy of the intervention, especially given the high acceptability and satisfaction of WLH with the study intervention. Additionally, given the multifaceted nature of the study intervention, we are unable to tease out active intervention components. The intervention acceptability indicators (e.g., satisfaction with the intervention and receipt, helpfulness and application of the intervention materials) seem to suggest overall synergy between health literacy education and follow-up components, which should be maintained in future implementations of CHECC-uP. Finally, the generalizability of study findings is limited by the inclusion of only Black, African American women in the study sample from a low-income, urban community. Nationally, 41% of WLH are Black, with 49% having less than a high school education and 44% having household incomes at or below federal poverty guidelines.[48] In Baltimore, 72% of WLH are Black, 35%–55% have less than a high school level of education, and about 64% have low-income status.[49,50] Future research should include more diverse groups of WLH from different cultural and racial/ethnic backgrounds.

CONCLUSION

Pilot testing of the CHECC-uP intervention resulted in promising effect sizes and high acceptability among low-income Black WLH. We incorporated health literacy education as a new approach to promote Pap test screening among WLH. The findings support integrating health literacy into a future intervention framework to transform the design of cervical cancer screening interventions for WLH. High attrition observed in our study sample highlights the

need for considering systematic strategies, such as the federal Lifeline programme for free phones and services, in future trials to successfully retain a study sample from underserved, low-income communities. It is possible that the positive effects of improved health literacy required for the uptake of cervical cancer screening may be more evident with a larger sample size than that of our pilot trial.

AUTHOR CONTRIBUTIONS

All authors approved the final version of the manuscript. Hae-Ra Han originated the study and led the writing. Jeanne Murphy-Stone and Phyllis Sharps contributed to the development of the study concept and design. Hae-Ra Han, Kyra J. W. Mendez, Nancy Perrin, Joycelyn Cudjoe, Gregory Taylor and Dorcas Baker contributed to the acquisition, analysis or interpretation of data. Hae-Ra Han drafted the manuscript, and all authors contributed to the critical revision of the manuscript. Hae-Ra Han also supervised the study.

ACKNOWLEDGEMENTS

This study was funded by Johns Hopkins University Center for AIDS Research (P30AI094189) and Dorothy Evans Lyne Fund.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Study data will be made available upon reasonable request to the corresponding author. De-identified data will be made available upon reasonable request.

ETHICS STATEMENT

The study was approved by the Johns Hopkins Medicine IRB. Informed consent was obtained from all individual participants included in the study.

DETAILS

Subject:	Intervention; Attrition; Literacy; Guidebooks; Efficacy; Human papillomavirus; Human immunodeficiency virus--HIV; Counseling; Health literacy; Data points; Disconnection; Cervical cancer; Feasibility; Health education; Black people; Psychosocial factors; Social factors; Health behavior; Information processing; Medical screening; Health information; Medical records; Jargon; Decision making; Cancer screening; Womens health; Cervix; Cross-sectional studies; Social networks; Women; Navigation; Brochures; Medical practices; Unmet needs; Viruses; Research centers; Cancer; Immune system; Clinical trials; Community health care; Data collection; Medical personnel; Pap smear; Acceptability
Business indexing term:	Subject: Social networks
Location:	United States--US
Identifier / keyword:	cervical cancer screening; health literacy; human immunodeficiency virus; pilot; randomized trial
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	172-182

Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Languageof publication:	English
Document type:	Evidence Based Healthcare, Journal Article
Publication history :	
Online publication date:	2022-11-28
Milestone dates:	2022-10-05 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-05-27 (manuscriptReceived); 2022-11-28 (publishedOnlineEarlyUnpaginated); 2022-10-19 (manuscriptAccepted)
Publication history :	
First posting date:	28 Nov 2022
DOI:	https://doi.org/10.1111/hex.13644
ProQuest document ID:	2767102298
Document URL:	https://www.proquest.com/scholarly-journals/community-based-health-literacy-focused/docview/2767102298/se-2?accountid=211160

Last updated:	2023-04-26
Database:	Public Health Database,Publicly Available Content Database

The importance of personal documentation for patients living with long-term illness symptoms after pituitary surgery: A Constructivist Grounded Theory study

Heckemann, Birgit [1]
; Graf, Tatjana [2] ; Eva Jakobsson Ung [3]
; Jakobsson, Sofie [4]
; Ragnarsson, Oskar [5]
; Olsson, Daniel S [5]
; Blomdahl, Christina [6]
[1] Section for Care in Long-term Conditions, Institute of Health and Care Sciences, Sahlgrenska Academy, University of Gothenburg, Gothenburg, Sweden; Department of Anesthetics, Surgery and Intensive Care, Sahlgrenska University Hospital, Gothenburg, Sweden [2] Department of Sociology, University of Lucerne, Lucerne, Switzerland [3] Section for Learning and Leadership for Health Care Professionals, Institute of Health and Care Sciences, Sahlgrenska Academy, University of Gothenburg, Gothenburg, Sweden [4] Section for Care in Long-term Conditions, Institute of Health and Care Sciences, Sahlgrenska Academy, University of Gothenburg, Gothenburg, Sweden [5] Department of Medicine, Sahlgrenska University Hospital, Gothenburg, Sweden; Department of Internal Medicine and Clinical Nutrition, Sahlgrenska Academy, Institute of Medicine, University of Gothenburg, Gothenburg, Sweden [6] Research and Development Centre Södra Älvsborg, Research and Development Primary Healthcare, Region Västra Götaland, Borås, Sweden

ABSTRACT (ENGLISH)

Introduction
Despite surgical treatment, pituitary adenomas often cause long-term illness symptoms, that profoundly impact patients' quality of life physically, psychologically and socially. Healthcare professionals often fail to recognize and discuss the ensuing problems. Personal documentation, such as symptom monitoring, reflective writing or even posts on social media, may help this patient group to manage their daily life and support communication of their care needs. Documentation strategies and the role of documentation for people with long-term symptoms after pituitary adenoma surgery are currently unknown.
Aim
To examine the effects and strategies of documenting symptoms, activities and physical and emotional well-being among people living with long-term pituitary adenoma.
Methods
In this Constructivist Grounded Theory study, 12 individuals living with long-term illness symptoms after pituitary adenoma surgery described their documentation strategies in in-depth interviews using teleconferencing and photo-elicitation between August and October 2020.
Results
Strategies for documentation included analogue and digital media. One core category (Exercising autonomy) and

three categories describing processes (Gaining insight, Striving for control and Sharing) emerged from the analysis. These three interrelated processes become an expression of autonomy to manage life and make sense of chronic illness. Personal documentation is a flexible tool that is used more extensively in times of ill health and less in times of relative well-being. Sharing documentation with healthcare professionals facilitated care planning and sharing with friends and family fostered emotional well-being.

Conclusion

Personal documentation is a valuable resource for managing life after pituitary adenoma surgery. The current findings may be relevant to other chronic illnesses. Further research exploring potential tools for personal documentation is needed.

Patient or Public Contribution

We deliberately chose a Constructivist Grounded Theory approach for this interview study. Using Constructivist Grounded Theory, we gave people living with long-term symptoms a voice, allowing them to freely speak about managing their illness in connection with personal documentation. The theoretical sampling approach enabled us to invite participants that could provide a broad overview of the landscape of personal documentation.

FULL TEXT

INTRODUCTION

Pituitary adenomas are rare[1,2] but can entail a heavy symptom burden and a chronic illness course despite surgical treatment.[3,4] Long-term illness symptoms can vary significantly between patients depending on the type of adenoma.[3] Patients with acromegaly typically develop progressive changes in facial appearance and growth of the hands, feet and internal organs.[5] Cushing's syndrome often causes proximal muscle weakness, fatigue and sleeping problems, central obesity, a round face and dorsocervical fat accumulation. Comorbidities with Cushing's syndrome include hypertension, diabetes, infections and hypogonadism.[6] Prolactinoma typically causes infertility.[7] Long-term health-related overall quality of life is often reduced in patients with pituitary adenoma.[8] Pituitary adenomas are typically treated with surgery, medical therapy and potentially radiotherapy, except for prolactinomas, which are usually treated with medication.[9]

There is a robust body of quantitative research exploring the quality of life of this patient group.[3,5] While patients' health-related quality of life may increase initially after surgery, it often fails to normalize in the long term.[8] Depending on the underlying condition, long-term symptoms can include joint pain or headaches, hypertension, memory deficiencies or fatigue.[8] Qualitative studies have reported that patients often live with unpredictable symptoms, cognitive problems such as attention, concentration and memory problems, altered physique, sexual dysfunction, fatigue, pain and psychological complaints such as depressive symptoms, anxiety, stress and fear.[10,11] Personality-related issues may occur, including personality changes and altered emotional functioning. Patients report work and social problems, including changes in social functioning, adverse effects of the illness on personal relationships, reduced social networks and a general lack of sympathy and support.[4,10]

Despite the heavy symptom burden associated with the condition, qualitative research examining the experiences of living with long-term illness symptoms after pituitary adenoma surgery is scarce. Several recent studies have highlighted that patients' care needs are often not adequately met. Healthcare professionals may lack specialist knowledge or fail to recognize and discuss issues regarding sexuality, fatigue and other psychological or personal problems that impinge on patients' lives[4,10,11] Patients require structured, continued support that addresses their physical, cognitive and existential challenges.[4]

Personal documentation may play an important role in improving care and self-care in this patient group. The benefits of various types of personal documentation in managing chronic illness are well known. For example, monitoring improves the control and day-to-day management of physical symptoms, such as blood sugar levels or blood pressure.[12–14] Recently, there has been increased interest in Patient-Generated Health Data (PGHD) to improve chronic illness treatment. PGHD are any health-related data generated by a patient, such as ‘biometric data, symptoms, lifestyle choices, and treatment history’.[15] With PGHD, patients are responsible for collecting and choosing with whom they share their data.[15] However, although the approach is patient-oriented, this documentation

focuses on biomedical information. Other forms of documentation, such as expressive writing, can be more therapeutic, as they support physical and psychological health.[16] Narration and storytelling facilitate making sense of traumatic or stressful experiences.[17,18] Emotional catharsis, narration and storytelling can reduce psychological stress and contribute to developing a coherent life narrative.[16]

For the current study, we drew on Plummer's[19] seminal work to define personal documentation from a sociological angle. Plummer[19],p.17 writes: 'People keep diaries, send letters, make quilts, take photos, dash off memos, compose auto/biographies, construct websites, scrawl graffiti, publish their memoirs, write letters, compose CVs, leave suicide notes, film video diaries, inscribe memorials on tombstones, shoot films, paint pictures, make tapes and try to record their personal dreams. All of these expressions of personal life are […] in the broadest sense' documents of life'. Based on these deliberations, we defined personal documentation to include narrative writing and journaling but also the recording of symptoms or activities or creating images. In line with this definition, we regarded documentation not in a generic sense but as an expression of different processes, for example, individual reflection and social network engagement.

Although previous evidence suggests that various documentation techniques may be valuable resources, the roles of documentation and the documentation strategies employed by patients in managing a condition with diverse chronic symptoms, such as pituitary adenoma, are largely unknown. This study explored patients' strategies for documenting information about symptoms and physical and emotional health in a life affected by chronic symptoms following pituitary adenoma surgery.

METHODDesign

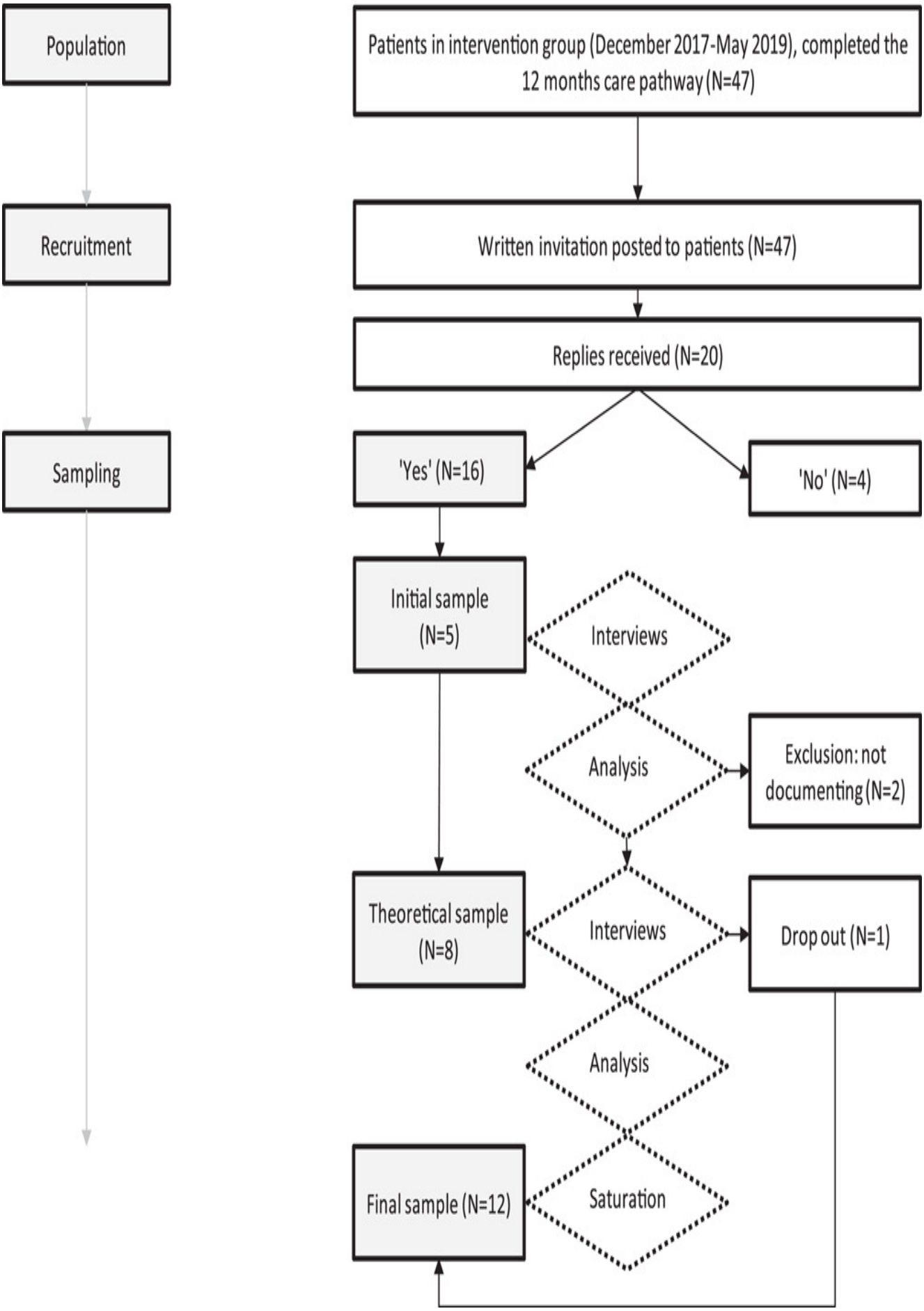
This qualitative interview study used a Constructivist Grounded Theory approach.[20] We chose this approach because it enabled us to understand the meaning of personal documentation in people living with a pituitary adenoma. In contrast to other qualitative research approaches, the Constructivist Grounded Theory approach permitted us to study personal documentation in great depth, explore its role concerning individual and social life and generate a theory about this scantly investigated phenomenon.[21]

Study context

This interview study was conducted in the context of a larger quasi-experimental study designed to evaluate the implementation of a person-centred care pathway for patients receiving treatment for pituitary adenoma.[22] The interview study population comprised 47 patients who participated in the trial's intervention group between December 2017 and May 2019.

Sample and sampling

We invited 47 patients that had undergone pituitary adenoma surgery to participate in the study. Twenty of the 47 patients replied, and 16 were interested in participating in the study. In line with the Constructivist Grounded Theory approach,[20] we included five participants during the initial sampling phase. These were the patients that had replied first. We selected eight additional participants during the theoretical sampling phase and excluded two further participants because they did not perform documentation. One participant dropped out of the study, resulting in a final sample of 12 participants (six women; six men) (Figure 1). The age range was 35–58 years (mean 54 years). Table 1 shows the characteristics and demographics of the study participants.



Enlarge this image.

Table 1 Participant characteristics and demographics

Characteristics and demographics (*N* = 12)	Number of participants
Gender	
Female	6
Male	6
Age	
30–50	5
51–80	7
Diagnosis	
Nonfunctioning pituitary adenoma	5
Acromegaly	4
Cushing	1
Prolactinome	1
Rathke's cyst	1
Operation	
First	7
Reoperation	5
Country of birth	
Sweden	11
Other European country	1
Educational level	
High school/vocational training	6
Middle school	2
University	4

Occupation	
Working	8
Other	4
Living situation	
Cohabiting	9
Living alone	3

Except for the first two interviewees, all participants either kept personal documentation and/or read their official personal online health records, which are accessible to patients in Sweden. Keeping personal documentation or reading their online health record became one of our theoretical sampling criteria because it became apparent during the analysis process that personal documentation played a complex and multifaceted role in managing life with a chronic illness.

Data collection

The interviews took place between August and October 2020, using teleconferencing and photo-elicitation,[23] which enabled safe data collection during the coronavirus-2019 pandemic. One co-author (C. B.), with substantial experience in arts-inspired face-to-face interviewing, conducted the interviews. All interviews were conducted in Swedish. We used ‘Mitt vårdmöte’ (My Care Meeting), an application that enables safe healthcare online consultations, to protect our participants' anonymity. The app is a CE-marked (CE marking indicates conformity with health, safety, and environmental protection standards for medical technology products sold within the European Economic Area. https://eurlex.europa.eu/LexUriServ/LexUriServ.do?uri=CELEX:31993L0068:en:HTML). The app complies with the Patient Data Act, the General Data Protection Regulations (GDPR) and the Swedish Data Protection Authority's rules.[24] The videoconferencing technology enabled participants to freely choose a place from which to join C. B. for the interview. A set of introductory questions such as ‘Do you document in daily life?’, ‘Can you tell me about how you document?’ or ‘Which information about your symptoms or health in your daily life is important?’ were used in the opening phase of the interviews. Further questions were generated from the concurrent analysis and the participants' narratives. In line with our definition,[19] we aimed to encourage the participants to reflect broadly on their documentation activities. We invited participants to look at 20 photos of objects related to writing (e.g., a typewriter, a computer, a bookshelf, a mobile phone and headphones) and scenes that conveyed different moods that could be linked to the illness experience. All images were from B. H.'s private stock. Photo-elicitation is an established method for stimulating interview participants' thinking, encouraging storytelling, fostering reflection and facilitating free association.[25–28] Photo-elicitation is particularly suited to exploring activities, interactions and processes.[26]

The interviews lasted for 18–78 min (mean 49 min), and were digitally recorded and transcribed verbatim.

Data analysis

According to the Constructivist Grounded Theory approach, the interview transcripts were analysed concurrently during the data collection phase.[20] We used a constant comparative method to analyse the data; B. H. and T. G. were the main coders. B. H. and T. G. coded independently, but they had 17 weekly meetings, each lasting 60–90 min during the analysis, to discuss and compare the codes and their meanings.

The coding process for each interview included initial reading and rereading of the transcript and open coding of the texts. We did not code line-by-line but instead moved straight to coding meaning units, which provided deep insights. We labelled text excerpts with codes or phrases that were as close to the original meaning as possible, thus maintaining a low level of abstraction. During the coding process, we constantly compared meanings and identified

similarities and differences within and between the interviews, continuously moving to more abstract and more specific codes, categories and concepts.[23,29] We also recorded reflections, questions and ideas in memos. B. H. and T. G. used the memos to inspire the exploration of additional literature to broaden their horizons and maintain a questioning mind to better understand the possible meaning of the data. The authors B. H., S. J., E. J. U., C. B. and T. G. met four times to discuss the analysis. This collaborative approach enabled critical discussion, spawned ideas, identified gaps in data collection and clarified the relationships between the codes and categories. As a team, we decided that we had reached saturation after 12 interviews.[20] We used Nvivo software,[29] Word documents and Excel spreadsheets to manage, analyse and visualize the data.

Quality criteria

Credibility, originality, resonance and usefulness have been proposed as quality criteria for Constructivist Grounded Theory research.[20,30]

We ensured credibility with a theoretical sampling process guided by concurrent data analysis. After 12 interviews, we closed the data collection because we had reached a saturation of categories (instead of data),[20] not due to lack of participants or resources. Our study is original in that it explores documentation from a purely patient-driven angle. This angle extends the current ideas about patient documentation, which is often symptom oriented instead of capturing the lived experience. Resonance pertains to how the study relates to larger collectives and individual lives. The study's usefulness relates to its contribution to knowledge, further research and generic processes. We discuss the resonance and usefulness in the discussion section of this paper (transferability).

Reflexivity statement

A reflexive stance is essential in Constructivist Grounded Theory.[20,30] In the following paragraphs, we briefly outline our reflections drawing on Gentles et al.[31]

Researchers' influence on research design and decisions

B. H., E. J. U. and S. J. initiated this project because of their professional and personal interest in documentation, person-centred care and chronic illness. Sharing a nursing background, we felt compelled to highlight this often-overlooked patient group's experiences and challenges in the research community and clinical practice. The initial decisions regarding the research topic, research questions and methodology were taken jointly during meetings and ongoing discussions. The remaining authors joined at the later stages. All authors contributed to the data analysis, interpretation and manuscript writing using their different professional lenses from occupational therapy (C. B.), sociology (T. G.) and medicine (D. S. O. and O. R.). We considered this input from different angles to be important because we did not want to produce research that was biased towards a nursing agenda.

Researcher–participant interactional influences during data collection

C. B. collected the interview data and was in email contact with the participants before the interviews. Having a background in occupational therapy and mental health and no prior knowledge about this patient group, C. B. interviewed participants from a place of curiosity, using empathy to establish rapport. C. B. evolved as a researcher during the data collection. The first few interviews taught her about the experience of life after pituitary adenoma surgery. During the later interviews, she immediately related to participants' experiences, which enabled deeper discussion and co-construction of meaning. This evolution affected the power balance during the interviews. Because C. B. was initially inexperienced regarding the condition, the power balance tipped towards the participants, who held specialist knowledge. With her increasing experience, the power balance shifted towards equality, which opened space for co-constructing meaning and interpretation in the interviews.

Researchers' influence on the analysis

The data analysis was a team effort. We discussed and interpreted the data through the lenses of nursing, occupational therapy, mental health and sociology. We ensured theoretical sensitivity through several measures: C. B. and B. H. discussed their thoughts about each interview. B. H. and T. G. kept memos on their thoughts, feelings and insights. B. H. and T. G. frequently met to discuss coding and data visualizations. While we explored participants' experiences in-depth, we also continuously reflected on the meaning and application of the data within the broader context of health care and society.

Researchers' influence on the writing

We designed this study to improve visibility and care for this patient group. We conducted this study from a place of empathy, which might have biased our writing. However, we feel that this stance was also essential to do justice to the participants' experiences.

Influence of the research on the researcher

This research gave the research team a heightened understanding of living with long-term symptoms after pituitary adenoma surgery and patients' resourcefulness in managing their challenges with the aid of documentation.

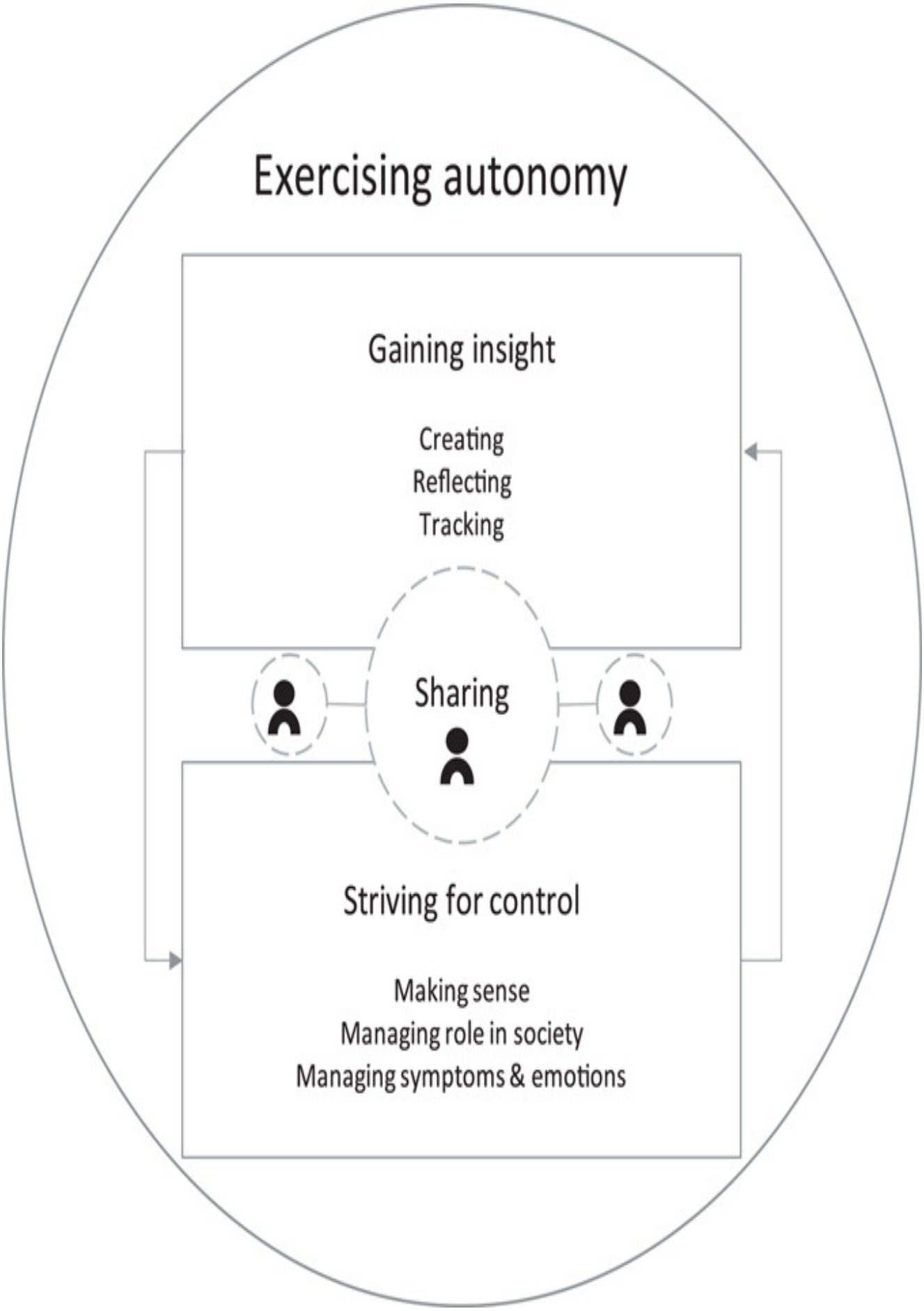
Ethical considerations

Approval of the study was obtained from the Swedish Ethical Review Board (DNR 2019-06485 and 2020 03025). The data collection complied with the Declaration of Helsinki principles.[32] All data were de-identified. Data were stored on a secure university server and only accessible to members of the research team. Participants received written and verbal information about the study and provided written informed consent before the interviews.

RESULTSStrategies for documentation

Many participants in our study used analogue media, including health diaries (as part of a care plan), diaries, calendars, notebooks or scraps of paper to document. Others used digital media, such as mobile phones or digital applications. Some posted on social media such as Instagram or Facebook, and some used a combination of analogue and digital media. Younger participants (35–40 years old), were more inclined to use digital media compared with older participants. The documentation content oscillated between mundane scribbles and quick, spontaneous note-taking about symptoms or activities in calendars, to more elaborate, reflective writing to preserve memories or release emotions. Some participants used images such as emojis to express feelings. Others took photographs to document their well-being or physical changes over time. Participants who struggled with the emotional burden of the illness tended to write more reflectively and elaborately.

In the following paragraph, we describe the theory that emerged from our analysis. The theory explains how three interrelated processes (described in categories) enable *Exercising autonomy* in managing life with a chronic illness (Figure 2).



Enlarge this image.

Core category: Exercising autonomy

Healthcare services are essential for the adequate medical management of chronic illnesses. However, services

such as clinic appointments are often scheduled at spread-out intervals of weeks or months, leaving patients to manage the effects of a chronic illness on their daily lives on their own in the interim. Our analysis showed that personal documentation plays an important role in the autonomous management of chronic illness, as it has diverse functions. The written texts or images mirror thoughts, feelings, experiences and situations, thus enabling new understanding or insights and an opportunity for self-reflection.

Oddly enough, I think that's what feeling bad is all about: That is why I felt bad and that's what made me feel bad. And things like that, and what happened, or how I experienced it. I think that's quite interesting, because you learn from your mistakes […]. (PT 8)

Self-reflection is an opportunity to ponder and understand oneself and one's situation in a novel way. Enhanced knowledge of the self provides access to strategies to deal with the illness, make health decisions or cope with life's challenges. The results revealed that patients use documentation creatively. Additionally, patients were mostly self-motivated but sometimes inspired by others, such as family or healthcare professionals.

[…] I got a great little book from my daughter. She had written nice things in it, some of her own things, a little here and there. And I'd read it a number of times, but never added anything. But about two weeks ago […], I started writing in the book […]. (PT6)

The core category comprised three interconnected categories describing processes in personal documentation. One category concerns *Striving for control* in one's social role, the second relates to *Gaining insight* about life with chronic symptoms, and the third regards *Sharing* personal documentation. The activities associated with *Gaining insight* provide the basic information to enable *Striving for control*. The third category, *Sharing*, lies at the intersection of *Gaining insight* and *Striving for control*. *Sharing* documentation with healthcare professionals can support care planning; sharing documentation with loved ones can foster feelings of trust, belonging and connection. However, our interview analysis did not enable us to determine whether these processes were sequential or of equal importance.

Categories

Gaining insight is a process that comprises actions including (1) Creating, (2) Reflecting and (3) Tracking. These actions capture current information about different aspects of life, such as symptoms, emotional states or events. In *Striving for control*, these pieces of information are aggregated to enable (1) Making sense, (2) Managing Symptoms and Emotions, as well as (3) Managing (one's) role in society. The third category, *Sharing*, lies at the intersection of the two categories. *Sharing* selected pieces of documentation with specific, trusted persons (family, friends, healthcare professionals) can increase the benefit of personal documentation: *Sharing* with loved ones can foster feelings of proximity, whereas sharing symptom diaries with healthcare professionals can enable diagnosis or care and treatment planning.

Gaining insight

Three actions supported the process of *Gaining insight*.

- (1)

  Creating

  Expressing oneself through creating images or text can preserve personal memories or a legacy, or simply give a sense of achievement:

  […] I love photography, I take a lot of pictures, […]; it's my way of expressing myself as well. And I sit and write. (PT11)

- (2)

  Reflecting

  Living with a chronic illness entailed a loss of abilities and capabilities that were previously taken for granted, such as abundant physical and mental energy, memorizing, pursuing a successful career or having a slim, well-trained body. Writing can break endless ruminating cycles and release emotions, at least temporarily.

Yes, I think [writing can give release from ruminating], on some things at least. And even with this [illness], if you feel bad or if you get annoyed with someone, or you are pissed off about something, then you write it down. And [you think] yes, to handle this is oh so hard. And then [you] kind of let go, it's released. (PT9)

•(3)

Tracking

Tracking comprised the mundane activity of recording symptoms such as blood pressure, headaches or body temperature and daily activities, such as going to work or working out, or future tasks or appointments. Tracking was undertaken in apps, analogue calendars or notebooks.

Striving for control

Information gathered through creating, reflecting and tracking actions is contextualized to gain control. Because phases of relative well-being alternate with times of poorly controlled symptoms, personal documentation enables a more comprehensive overview of how symptoms and emotions fluctuate over extended periods, and how daily activities might influence health and well-being. Tracking can also help a person to situate their illness in the larger context of their biography.

•(1)

Making sense

Making sense pertains to understanding and coming to terms with what has happened in the past. Personal documentation allows a person to compare, understand or even become more compassionate with themselves and the current situation. Revisiting documentation can foster sense-making and learning for the future:

I have to take pictures every day because it's like my diary or my way of remembering, to record things, this is for real, or this has happened. This is what I looked like. So that I can understand it. Otherwise, in a year, I do not remember at all what I looked like. Or how it was […]. (PT11)

•(2)

Managing symptoms and emotions

Connecting activities such as physical, social or work activities and symptoms can provide an overview of the illness trajectory and may reveal causal relationships between actions and their effect over time:

I write, for example, if I had a very severe headache, I have had problems with that quite a lot. And then I can write down when I have a headache or if I'm tired one day, or if I'm feeling good one day and so on. So I can go back. It's also like that, and you feel that, oh my God, now I've had a headache every single day, [at least] that's what it feels like. And then you look at [your notes and realize] no; I was actually feeling good, I did this or that. Or I see a pattern, well, but now I have done quite a few things, maybe that's why I'm tired, and I should take it a little easier. (PT11)

In times of relative well-being, documentation may become less important:

So you do not write on these good days, because you are busy feeling good and having a good time. And that's a good sign […] I have not written so much lately. (PT9)

•(3)

Managing (one's) role in society

Personal documentation can be vital to managing one's role as a member of society. Being able to refer to written documentation about past activities becomes vital when living with severe memory loss. Personal documentation becomes ‘proof’ of productivity:

If I have not written things down, then I have no idea what to do. […] I check the diary in the kitchen; I check my little book. I check to see what the day looks like for me. Then I have an idea of how the day will be, and I eat (breakfast) in peace. (PT6)

Sharing

Sharing occurs at the intersection of *Gaining insight* and *Striving for control*. Whether, what, to what extent, and with whom to share personal documentation is an individual decision as it incurs the risk of making oneself vulnerable through disclosure. However, mindful sharing of personal documentation with selected persons can be beneficial. Sharing personal documentation with healthcare professionals can support care planning or getting a diagnosis in the first place:

But before, before, [the diagnosis] it was mainly to understand what was happening and also then be able to get help when I was not really believed [by physicians] that I was actually feeling bad. Therefore, I documented, to be able to show, this is my everyday life right now, it is something that is not right, with these symptoms. (PT12)

Sharing diaries or memories with family and friends fosters feelings of connection. Digital media such as Instagram or instant messaging services even enables 'real-time sharing' of messages about their experiences with friends or family:

Yes, but this was probably when I felt the worst, and then when I lived in Y. and my family lived in Z. It [writing instant chat messages] is an easy way to keep in touch. And I talked a lot with my brother when I felt so bad, and with my best friends. If you could not see each other, then you could always sort of write with them. (PT12)

In turn, reactions to posts on digital media can be rewarding:

Yes, but for now [a post about good blood results and an indication that the medication worked] there was a bit of cheering, like 'oh how good', 'nice to hear', 'good results' or like, positive answers. And then, as before I was to have surgery and after the operation, I also posted and then I also got comments like this, hugs, hearts and a few positive words for strength. (PT11)

DISCUSSION

This study sheds light on the role of documentation in managing life after surgery for pituitary adenoma. Based on our current findings, we propose a theory grounded in the analysis of 12 in-depth interviews. The theory explains how three interrelated processes described as *Gaining insight, Striving for control* and *Sharing* enable *Exercising autonomy* in managing life after pituitary adenoma surgery.

The three processes did not emerge as sequential, or even of equal importance from our interviews. Rather, they emerged as individual processes that, in combination, enabled the exercising of autonomy. Because this study offers the first insights into personal documentation, we cannot make claims about how and to what extent each process may be relevant for self-management in general. However, the current results complement previous research findings on managing life with a chronic illness.[33] Patients have been reported to mobilize resources such as attitude, willpower and creativity to manage the challenges associated with a chronic illness.[33] Personal documentation may be considered as one particular resource amongst several other creative strategies (working at health, participating in life, connecting with other people and developing new coping strategies) that enable patients to re-engage in a meaningful life despite a chronic illness.[33]

The core category that emerged from this analysis was 'exercising autonomy'. We assumed an extended concept of autonomy that encompasses individual and social-relational autonomy. In accordance with this assumption, the analysis revealed that personal documentation was not a purely private act but a resource that can support patients' social and relational contexts or functioning. Note-taking, for example, allows patients with memory loss to keep

track of their daily activities. Moreover, our findings indicated the benefits of sharing documentation for personal relationships and communication with healthcare professionals. This finding is novel. While sharing clinical notes with patients is currently widely practised and advocated,[34,35] there is, to the best of our knowledge, no published research exploring the benefits of sharing personal documentation. Further research is needed to explore the benefits of this approach and how personal documentation may be applied to improve self-management of long-term illness symptoms following pituitary adenoma surgery and possibly other chronic illnesses.

Some types of personal documentation are widely encouraged in the context of more patient-oriented chronic illness management. These documentation types often include digital applications for monitoring individual parameters such as weight or blood pressure.[12–14,36,37] PGHD are focused on collecting biomedical data and patient-defined information, such as observations of daily living, for example, feelings, thoughts or behaviours.[38,39] This combination provides a more holistic overview that enables planning, treatment and management beyond biomedical symptom and sign control. However, the patients in our study freely chose the data and documentation mode, making the documentation a genuinely personal and flexible process. Personal documentation was a tool used on an as-needed basis (as opposed to following a prescribed format). It was adapted to optimally serve patients' changing needs during periods of better or worse health. Patients who choose to keep and maintain personal documentation produce an invaluable wealth of knowledge about their illness, which, as our findings showed, is not necessarily included in care planning. The study participants were selective about what to share with whom, and how they shared information. Considering the benefits of personal documentation, whether shared or not, our findings indicate that documentation is beneficial for managing life with the chronic symptoms following pituitary adenoma surgery.

Transferability

This study included 12 participants living with chronic symptoms following pituitary adenoma surgery (Table 1). Depending on the type, the long-term symptoms that occur after pituitary adenoma surgery can vary widely.[8] In addition to comorbidities, patients must cope with unpredictable symptoms, cognitive or psychological problems, an altered physique or personality, sexual dysfunction, fatigue, pain and difficulties regulating emotions.[10,11] The range of long-term symptoms associated with the illness may make our findings apply to other patient groups living with a chronic illness. However, further studies are needed to confirm the theory derived from this study.

Limitations

This study has some limitations. First, we explored our research questions by recruiting a theoretical sample of participants who used personal documentation. Excluding participants who did not use personal documentation prevented an exploration of the potential benefits of ‘non-documentation’. However, this type of exploration exceeded the scope of our research question. Second, we recruited our participants from a study cohort of patients living with long-term symptoms following a pituitary adenoma. The patients' age range was 35–58 years (mean 54 years). Including younger participants (20–30 years old) may have provided more information regarding social or digital media use. Instead, we obtained an overview of the use and benefits of various media (pen and paper, digital media).

Our theoretical sampling strategy included some participants who had started documenting of their own accord, and some were inspired or encouraged by family members of healthcare professionals. Some participants had kept diaries before their diagnosis, while others started documenting after their diagnosis. Thus, our findings entail a risk of bias that is inherent in the theoretical sampling process. Our results came from participants with the motivation, skills and creativity to develop an individualized approach to their illness management. Finally, the theory was generated from a small theoretical sample and may therefore not be applicable to patients with fewer resources to self-manage their illness.

CONCLUSION

This study sheds light on the role of personal documentation in managing the chronic symptoms following pituitary adenoma surgery. We propose a theory that explains how three interrelated processes (*Gaining insight, Striving for control* and *Sharing)* enable *Exercising autonomy* in managing daily life with a potentially serious symptom burden. Future research should elucidate whether our theory translates to other patient groups and develop tools adapted to different needs, preferences and abilities to promote personal documentation.

AUTHOR CONTRIBUTIONS

Birgit Heckemann: Conceptualization; data curation; investigation; formal analysis; project administration; supervision; visualization; writing –original draft preparation; writing –review and editing (all lead). **Tatjana Graf**: Data curation; formal analysis; validation; investigation; writing –original draft preparation; writing –review and editing. **Sofie Jakobsson**: Conceptualization; formal analysis; supervision; validation; writing –original draft preparation; writing –review and editing. **Eva Jakobsson Ung**: Conceptualization; formal analysis; supervision; validation; funding acquisition; writing –original draft preparation; writing –review and editing. **Oskar Ragnarsson**: Writing –original draft preparation; writing –review and editing. **Daniel S. Olsson**: Writing –original draft preparation; writing –review and editing. **Christina Blomdahl**: Conceptualization; investigation; data curation; validation; writing –original draft preparation; writing –review and editing.

ACKNOWLEDGEMENTS

We are extremely grateful to all participants for generously giving us their time and sharing their thoughts, experiences and insights. Their creativity and spirit in managing illness through documentation are inspirational to healthcare professionals and people living with a chronic illnesses.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

DETAILS

Subject:	Documentation; Demographics; Health care; Storytelling; Teleconferencing; Surgery; Physical symptoms; Emotions; Quality of life; Emotional well being; Illnesses; Insight; Long term symptoms; Care plans; Tumors; Patients; Medical records; Grounded theory; Pituitary; Adenoma; Social networks; Chronic illnesses; Mass media; Digital media; Autonomy; Elicitation; Voice communication; Interviews; Symptoms; Social media; Hypertension; Activities of daily living; Health professional-Patient communication; Data collection; Medical personnel; Qualitative research; Friendship
Business indexing term:	Subject: Social networks
Location:	Sweden
Identifier / keyword:	chronic disease; chronic illness; Constructivist Grounded Theory; patient documentation; personal documentation; person-centred care; pituitary adenoma
Publication title:	Health Expectations; Oxford

Volume:	26
Issue:	1
Pages:	226-236
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-06
Milestone dates:	2022-09-06 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-01-25 (manuscriptReceived); 2022-11-06 (publishedOnlineEarlyUnpaginated); 2022-10-22 (manuscriptAccepted)
Publication history :	
First posting date:	06 Nov 2022
DOI:	https://doi.org/10.1111/hex.13648
ProQuest document ID:	2767102272
Document URL:	https://www.proquest.com/scholarly-journals/importance-personal-documentation-patients-living/docview/2767102272/se-2?accountid=211160

Lastupdated:	2023-10-04
Database:	Public Health Database,Publicly Available Content Database

Becoming a secondary actor of one's own life: A qualitative study of the experiences of informal caregivers in the care of people with chronic pain

De Sola, Helena [1] ; Failde, Inmaculada [1] ; Estalella, Itziar [2] ; Maquibar, Amaia [2 1] The Observatory of Pain, University of Cádiz, Cádiz, Spain; Biomedical Research and Innovation Institute of Cádiz (INiBICA) Research Unit, Puerta del Mar University Hospital, University of Cádiz, Cádiz, Spain; Preventive Medicine and Public Health Area, University of Cádiz, Cádiz, Spain [2] Department of Nursing I, Faculty of Medicine and Nursing, University of the Basque Country UPV/EHU, Bizkaia, Leioa, Spain

ABSTRACT (ENGLISH)

Introduction

The physical limitations experienced by people with chronic pain (CP) produce a greater need for care and assistance, most of which is provided by an informal caregiver (IC). Despite the key role ICs play in the everyday lives of individuals living with CP, knowledge about their experiences and needs is limited. We aimed to address this limitation by exploring the experiences of IC of people with CP.

Methods

This is a qualitative descriptive study using semistructured interviews. Participants were 12 ICs purposively chosen from the Unit of Pain at the University Hospital in Cádiz. Individual interviews were recorded, transcribed verbatim and analysed following thematic analysis.

Results

We developed one overarching theme ‘Becoming a secondary actor of one's own life’ and three themes: 1. Key elements that shape a caregiver's experiences; 2. It's the hand that life dealt me; 3. The burden of being a caregiver and coping strategies.

Conclusions

This study's findings highlight how the CP impacts IC lives. Being an IC for a relative with CP became the most important role in the IC's life, to the point of casting a shadow over their own needs. Besides, participants felt not having other options but to keep going with that role. Yet, the context was essential in shaping the experiences as caregivers and the burden derived from caregiving. In this line, differences related to gender roles were found in the narratives of participant women and men.

Patient or Public Contribution

Participants were purposively chosen from the Unit of Pain at the University Hospital ‘Puerta del Mar’ who attended the consultation accompanying their relatives. All the eligible participants were approached by the clinician. After this initial approach by the clinician, one of the researchers met the potential participant and they went to a quieter place in a clinical setting for the interview, before which the participant was shown a letter with more comprehensive information about the study and its aim. The participants were left alone to read and think carefully before giving

their written informed consent. Participation was voluntary and the subjects received no financial contribution for their time.

FULL TEXT

INTRODUCTION

Chronic pain (CP) is an important public health problem that affects between 10% and 30% of the adult population in Europe, and 17% of the general Spanish population.[1] Additionally, this prevalence is expected to increase in coming years, due to the ageing of the global population and prolonged exposure to risk factors such as obesity or occupational factors.[2–4] The mean duration of CP in Spain is 10 years,[5] with the majority of the sufferers reporting moderate or severe pain.[5–7] The physical limitations experienced by people with CP produce a greater need for care and assistance, most of which is provided by an informal caregiver (IC), that is, an unpaid family member or friend who provides assistance with everyday activities.[8] In this vein, the IC makes an important contribution to the formal health system. The mean length of CP mentioned above, suggests that the ICs have to accompany their relative for a prolonged period of time.

The role played by ICs is not unique to CP patients, as they play an essential role in many diseases. Therefore, the needs, experiences and consequences of being an IC have been widely studied, especially in the case of IC of the elderly, cancer patients, mental health and in palliative care. In the case of ICs for cancer patients, a study found that the new role and tasks performed can be emotionally, physically, socially and financially demanding.[9] Similarly, ICs of individuals with mental illness were found to have limited social and recreational activities as a result of their dedication to the patient.[10] Additionally, financial stress, derived from being responsible for continuous care and maintaining the family income, was identified as one of the most crucial challenges in daily living, contributing to mental illnesses among ICs.[11,12]

More limited knowledge is available about the experiences of ICs of individuals living with CP patients.[13] Studies have found there are some commonalities with IC of other diseases and some specificities related to the characteristics of CP. Similarly to what happens in other diseases, the IC has to perform new tasks related to the role, such as administering medications, dealing with the possible side effects of treatment and managing medical appointments. Some studies have shown[14,15] how ICs share the emotional experiences of people in pain, including stress, distress and insomnia which significantly reduces their own well-being. Being the everyday ‘witness to pain’ obliges IC to act as the reporter and defender of the pain to those who may question the sincerity of the pain[13] due to its ‘invisibility’,[16] for example by explaining to the physician the characteristics of the pain of their family member.[13]

The heterogeneity in individual's pain perceptions, and the biological, psychological and social nature of pain, requires not only a clinical approach but also fulfilling the social expectancies and responsibilities to offer informal care.[13] Additionally, CP is frequently associated with other pathologies, making the role of the IC more challenging and complex.

Experiences of IC have been found, as with other diseases, to vary in relation to sex and gender. Due to traditional gender roles, women, who are the vast majority of IC,[17] frequently assume the greatest share of responsibilities for maintaining the family's organization and providing nurturance to family members.[17] This gender gap is especially visible in mature-age caregivers, that is, those between 40 and 64 years,[18] who have been described in previous studies as the ‘sandwich generation’.[19] These women find themselves in a situation of caring for elderly parents, having dependent children and remaining active in the labour market.[8] Consequently, women may experience losses of identity, privacy and time for themselves and are at greater risk of poorer health than their male counterparts.[20]

Taking into account that CP prevalence is already significant and is expected to increase in coming years,[4] leading to an increase in the number of ICs for individuals living with CP, and that previous studies have found some specificities related to the characteristics of CP, this study aimed to explore the experiences of ICs of people with CP to better understand and respond to their needs. Likewise, these experiences were analysed from a gender perspective.

METHODSStudy design

This is a qualitative descriptive study[21] in which data were collected through semistructured interviews to explore the experiences of ICs of CP patients.

Setting and participants

The participants were the main IC of a patient with CP, understanding IC as the person responsible for the help needed by the patient to perform basic daily activities during the main part of the day without receiving a salary for providing this help.

The inclusion criteria for the participants were adults who accompanied their relative with CP to the consultation of the Pain Clinic of the Puerta del Mar University Hospital and consider themselves to be him/her main caregiver. After an analysis of the medical record and a physical evaluation of the CP patient, the clinician asked the person who accompanied him or her if they were the main IC of the CP patient. In case him/her responded affirmatively, the clinician explained to them the aim of the study. All the potential participants answered affirmatively to the question and agreed to participate. After this initial approach by the clinician, the interviewer met the participant and they went to a quieter place in a clinical setting for the interview.

Data collection

Data collection took place between May and October 2021. Twelve ICs agreed to participate in the interviews (Table 1), which were conducted by HDS. Individual, semistructured, qualitative interviews following a guide were conducted in Spanish. The guide was based on open-ended questions developed with guidance from the literature regarding CP and IC experiences (Table 2). Interviews were audio-recorded, transcribed verbatim and anonymized. All names used here are pseudonyms. We conducted interviews until very similar experiences were described in the last interviews as the previous interviews.

Table 1 Characteristics of the sample

	Gender	Age	Income/occupation	Relationship	Time dedicated to care	Task performed
Natalia	Female	44	Total permanent disability	Wife	Since 2016	24 h assistance
Juan	Male	72	Retired (bricklayer)	Husband	Since 2012	24 h assistance
Marco	Male	77	Total permanent disability (carpenter)	Husband	Since 2017	24 h assistance
Pablo	Male	73	Retired (Waiter)	Husband	-	Shopping and accompany to doctor
Rocío	Female	34	Teacher	Wife	Since 2019	Housework and tasks involving carrying weight
Celia	Female	46	Administrative assistant	Daughter	Since 2020	Company and supervision of all mother's activities
Marta	Female	47	Unemployed	Daughter	Since 2012	Housework and company

Elena	Female	51	Teacher	Daughter	Since 2016	Company and supervision
Ana	Female	70	Unemployed	Wife	‘a lot of years’	Housework, company and supervision
Javier	Male	68	Retired (Sailor)	Partner	Since 2015	Some housework, company and supervision
Miriam	Female	66	Businesswoman	Wife	Since 2010	24 h assistance
Milagros	Female	45	Medical assistant	Daughter	Since 2013	Company and Supervision

Table 2 Interview guide used for the semistructured interviews

B1. Sociodemographic data
Age/Education level/Employment status/Marital status
B2. Origin of the situation
How are you related to the person that you look after? (mother/father, son/daughter, brother/sister, husband/wife…)
What illness/es does your relative suffer from? How does the illness affect their physical and/or mental capacities?
How long have you been looking after them? How did this situation come about?
B3. Information received
Have you received any information about how to take care of your relative? How were you given this information?
In the event of having to make decisions about healthcare, treatment, looking after your relative, who makes the decisions? How do you decide?
B4. Daily life, care experience and perception of your health
How many hours per day do you dedicate to providing care? What kind of care does your relative need?
What is a normal day like for you?
How do you think taking care of your relative affects your health? What about your emotional/psychological well-being? What changes have you noticed in your emotional/psychological well-being?

B5. Family, social and working life
How has taking care of your relative affected your social life?
How do you disconnect from your obligations?
How is your relationship with the rest of your family? Has this relationship changed since you started taking care of your relative? Do other members of your family help you?
How has taking care of your relative affected your working life?
B6. Final questions
How would you describe your experience as a caregiver?
Is there anything else you would like to add?

Data analysis

We analysed all the interview transcripts following thematic analysis as described by Braun and Clarke in their six-step methodological guide.[22] The data analysis was inductive, thus thematic construction was data-driven; no initial hypothesis guided the preliminary coding and subsequent development of themes.

Three investigators performed an initial line-by-line coding of the interview transcripts, ensuring each interview had been coded by at least two of them independently to develop a robust and consistent code set. All the codes were then discussed and refined between the same three researchers. The resulting codes were then sorted into potential themes.

The elaborated themes were refined using the three stages proposed by Braun and Clarke for this part of the analysis, with the participation of all the authors. First, all the coded extracts for each theme were read thoroughly to check coherence in the pattern that led to that theme definition. Once necessary adjustments had been made, the preliminary themes were contrasted with the whole data set to refine them. Finally, a detailed analysis of each theme, including the meaning and scope, as well as its relationship with the other themes, was conducted and written based on the data extracts coded in each one.

RESULTS

Twelve people aged from 34 to 77 were interviewed (8 women and 4 men). Two of them had a declaration of total disability to work, and five were retired or unemployed. The majority of them (n = 8) were the intimate partner, and four participants were daughters of the CP patient (Table 2).

During data analysis, one overarching theme—‘Becoming a secondary actor of one's own life’—and three themes were elaborated: ‘Key elements that shape this experience’; ‘It's the hand that life dealt me’ and ‘The burden of being a caregiver’.

The overarching theme captures an idea that underpins the other three themes, while the combination of the themes ‘Key elements that shape this experience’ and ‘It's the hand that life dealt me’ lead to the third theme named ‘The burden of being a caregiver’.

Becoming a secondary actor of one's own life

This overarching theme is about how caring for their relative with CP, regardless of the relationship, was a central part of the lives of all the participants in the study, to the point of casting a shadow over their own lives and experiences. This shadow was expressed on the one hand by explicit statements about the full-time dedication to the care, and by narratives about how they have minimized the importance of their own health issues and needs on behalf of those of their relatives, as the following quote exemplifies:

I have been raging with pain for 10 years, but as my husband was in pain I did not pay attention to mine and when I went to the doctor, he told me I no longer had a hip, it had completely worn out. I didn't pay attention to my pain because I was taking care of him. (Miriam, 66 years old, caregiver for her husband 24 hours per day)

On the other, this loss of relevance resulting from caring for the other person was reflected in the way participants answered the open questions. Although most of the questions were focused on their experiences as caregivers, all the interviews were full of detailed and rich information about the health issues, mental health status, fears, hopes, difficulties and so on of the persons they were taking care of, while theirs were described more succinctly or minimized (Table 3). In this same vein, they used ‘we’ to describe problems or issues that were in fact their relative's, as it is shown in the Ana quote (Table 3). Additionally, the IC has to defend the credibility of their relative in relation to the pain experienced against those who question the severity or even the existence of pain, as Natalia exemplified (Table 3).

Table 3 Quotations illustrating categories and theme.

Overarching theme: ‘Becoming a secondary actor of one's own life’
Natalia, 44 years old, caregiver for her husband: ‘I've already come to terms with what my daily life is like and it doesn't bother me and if it should bother me, I don't mind …because he's here for me and I'm here for him. I'm not going to put having no lymph nodes in my breast before saying to him, “give me your hand, I'll help you get up,” I don't mind. It's no trouble’.
Ana, 70 years old, caregiver for her husband: ‘I was recently admitted to the hospital here for my prostate. And now on the 25th we'll be hospitalized again. •- You will be hospitalized? •- Him, him. I'll be admitted with him. Who takes care of him? Me, so I have to be in hospital with him 24 hours a day’.
Ana, 70 years old, caregiver for her husband: ‘He had an operation on his knee and I always keep an eye on him and make sure he takes his medicine. His illness doesn't hinder me at all. The thing is there's just the two of us and it's normal, I cook for both of us, I go shopping for him, go to the chemist's, go to the doctor for him …Very often he doesn't even go to see the family doctor, I tell the doctor where it hurts, “Look, he's not getting any better, his leg hurts today” (as if speaking to the doctor)’.
Natalia, 44 years old, caregiver for her husband: ‘We found people who think you're faking it. Let's see, I'm telling you that this, this and this happens to him, they say: “it's that you …don't go every time with the crutches.” When you see him with the crutches, it's because he can't stand it more’.
Miriam 66 years old, caregiver for her husband: ‘If he calls me 20 times, I go 20 times …Having to look after him doesn't bother me at all, thank God. Tiredness is the only thing and when the night finishes you end up really tired because of the “bring me this, give me that” …you know. For his shower he sits in a chair and I shower him because he can't stay standing up …anyway’.
Marco 77 years old, caregiver for his wife: ‘I have my pains too. For example, I sometimes get sharp pains from my hip to my calf and I have to stop and squeeze my leg and that …I have problems with my neck and shoulders and I get dizzy, but no problem, I try to ignore it’.

Category: ‘Key elements that shape this experience’
Pablo, 74 years old, caregiver for his wife: ‘I am retired, I have a fairly decent pension. The thing is, I live in a rented house. So, half of my pension goes on rent, electricity, water …For example, this month I'm having problems making ends meet’.
Juan, 72 years old, caregiver for his wife: ‘Well, I could take her out for breakfast every day, for example, which she likes. We'd go by car, I'd take her to the shopping centre, take her into town to have a coffee, but I can't do that every day. I can't spend 4 or 5 euros every day. I do it from time to time’.
Marco, 77 years old, caregiver for his wife: ‘We requested the help of a professional caregiver from the Junta de Andalucía (regional government), and after a while they replied that I'm a Grade 1 dependency care case. We think Grade 1 is too little, so we appealed to be considered Grade 2. That was two years ago, it'll be two years soon, and still they haven't replied’.
Natalia, 44 years old, caregiver for her husband: ‘My husband is a builder. He can't work and he has a 55% disability. When the medical board called him and saw him, they said, “you can't work as a builder, but you can do other work”—“What other work? I can't stand up, I can't sit down and I can't spend a long time in the same position”’.
Rocío, 34 years old, caregiver for her husband: ‘The doctors advised my husband not to go up and down stairs and we live on the fourth floor with no lift. So, it's a nightmare every time he goes up or down the stairs’.
Pablo, 73 years old, caregiver for his wife: ‘My children only help if I'm a bit overwhelmed. I do tell them that they have to take their mother to certain places, but not very often, very rarely because it's not necessary if I'm alright …there's no need for me to give them work to do’.
Category: ‘It's the hand that life dealt me’
Milagros, 45 years old, caregiver for her mother: ‘I want her to be happy for the time she is with …while she is alive. And that she is as comfortable as possible, as happy as possible and that she feels loved and close, know what I mean? That's my aim, mothing else’.
Miriam 66 years old, caregiver for her husband: ‘I'm alright for now. I hope I can take care of him like this for a long time. And that God gives me good health. That's what I ask for. And that God gives him good health so I can take care of him for a long time. Yes. Because he's such a good man and he's been so good to me. Seeing him like this…’.
Marta, 47 years old, caregiver for her mother: ‘They are all my family. I like the mother hen and the others are my chicks. You know what I mean? So, I have never thought about living in any other way’.
Celia, 46 years old, caregiver for her mother: ‘My brother is not as competent as me, do you understand? I'm much better at these things. When my mother was in hospital for her hip, I was the one that help her have a shower, I was the one … you know? I'm more competent. For coming with her to the hospital, he's no use, and well I'm a bit better at it (she laughs)’.

Rocío, 34 years old, caregiver for her husband: ‘I have experience as a caregiver for my mother, who passed away 7 years ago, after a long cancer. So, at home we are experts at handling and managing situations that are a bit difficult, and carrying on with life despite illness. But it's purely down to experience and to the slaps in the face that life brings. Not because anyone has told us how to do it’.
Category: ‘The burden of being a caregiver’
Rocío, 34 years old, caregiver for her husband: ‘I much more tired than I should be. That's simple and obvious. My back hurts because I sometimes do more than I should. It affects me, of course it does. A lot. As far as care goes, there are no limits’.
Elena, 51 years old, caregiver for her mother: ‘If her illness gets more complicated in the future, we'll see what I do. Do you understand? So, now is when I start getting worried, but it's alright for now. Like I say, I want to be positive and anything I can do for her now …so that she gets better …well. But it affects me, of course it does, it's normal, isn't it?’
Celia, 46 years old, caregiver for her mother: ‘I haven't been out for a long time, and haven't wanted to either because I won't feel at ease knowing that my mother is alone’.
Natalia, 44 years old, caregiver for her husband: ‘Well, we hardly have a social life, we don't have a social life. I might see one of my friends, but we don't usually go out’.

Key elements that shape this experience

This theme englobes the codes that describe all the contextual elements beyond the individual characteristics of the caregiver and the person with CP that were essential in shaping the experiences as caregivers. The elements mentioned by the participants in this study included: the advanced age of both the caregiver and their patient, the caregiver's health status, the socioeconomic status, the COVID outbreak and the (mainly) lack of social/family support. In relation to age and the health status of the caregiver, some of the participants were themselves facing health issues related to ageing while simultaneously providing care for their relatives.

The relevance of economic status was repeatedly mentioned by participants in their discourse, especially when they were facing economic difficulties. Difficult financial circumstances were often related to a basic educational level and a life of unskilled work in precarious conditions. Living on a limited budget was described by interviewees as a source of worry and a limitation to the care they could provide, from the food they could buy to making the home accessible, and the possibility of employing someone to help them with the housework or the leisure activities they could plan. As Juan said: ‘My problems …the real problem I have is financial, more than her health. Well, her health is very important, but if we didn't have money problems, we might live much better’.

All the caregiver tasks were performed with no specific support from the healthcare services and with little support from social services, which were described as insufficient and slow to respond, as the Marco and Natalia quotes show (Table 3).

As this study was conducted in 2021, the COVID outbreak was mentioned by participants as a relevant event in their experiences. The most extreme impact was the loss of family members due to COVID, this being the case for two of the participants. In the remaining cases, the impact was related to the lockdown measures and the restrictions imposed by the government regarding leisure and public spaces, and whether they were afraid of going out due to the risk of infection. As Juan stated: ‘My wife isn't very inclined to go out and with this pandemic even less so; she's very scared’. In addition, the lockdown also had an impact on the physical and mental sphere, as Ana stated: ‘The lockdown has been absolutely awful for my husband because he has hardly moved for 3 months …but at least we had the treadmill’.

Yet the cornerstone of all these elements was the wider family/social support or the lack of it. The participants' experiences varied in this sense and ranged from very good emotional support to the feeling of being totally alone. In any case, the support received was strongly intertwined with all the elements described above, as the support the other family members could offer depended to a large extent on family members' age, socioeconomic status or employment status. Thus, in some of the narratives, participants explained or justified the lack of support from their relatives by explaining the economic or health struggles those relatives were dealing with.

My husband's sisters' have settled lives, so they don't have much contact. If we won the lottery, they'd all come round, but nobody comes to deal with the pain. (Natalia, 44 years old, caregiver to her husband)

It's the hand that life dealt me

Although it was not a question included in the interview guide, participants explained or somehow justified why they were in that role. The diverse reasons given were underpinned to different extents by statements about having no other option but to remain in the role with a sense of resignation. This lack of alternatives was strongly linked to the previous category, where many participants described the lack of wider support. All these different positions were sustained by feelings of responsibility and obligation because of love or in the case of daughters with parents, the feeling of having to repay the care their parents had given them, as Milagros explained:

I took on the responsibility myself because, in truth, it's what my parents have always done for me. I'm the eldest, I was given the responsibility and that's how it has stayed. I've never wanted to stop …not because I wouldn't like to but because I'd feel bad, like selfish, if I did, you know what I mean? (Milagros, 45 years old, caregiver to her mother)

Their caregiver role implied a long list of tasks. Participants described how they were in charge of medical appointments, dealing with medication, managing medical information, making decisions, keeping their relative company, doing the housework, trying to cheer up their relatives or even assuming the cost of moving house, while at the same time having to cope with their own worries and anxiety about the future.

Well, you see, my husband needs me to put his socks on because he can't due to the pain. I have to shower him …he walks with the walking frame, with a walking stick, but when I come to the hospital I bring him with the wheelchair because he can't walk for more than 10 minutes. So, except for feeding him, I have to do nearly everything for him. (Miriam, 66 years old, caregiver to her husband)

In the description of what taking care involved, it is of note how the narratives of the women differed from those of the men. When describing the activities, they did as part of the care, men mentioned explicitly doing the housework while women did not. Moreover, men referred to the tasks they did as providing ‘help’ to their partner and not one of their responsibilities in the house. As Juan said:

I help her, she practically …I even help her make the lunch, something I didn't know how to do before, but there's no alternative but to learn. I clean for her, make the beds for her, go shopping for her…. (Juan 72 years old, caregiver to his wife)

In some cases, they described how despite living with CP their wives still did most of the housework. Pablo stated that: ‘my wife is a housewife and, no matter how much I want to, she says she prefers to do the cleaning herself. She does everything to be honest; she's a traditional woman and that's alright. She doesn't stop from the moment she gets up. Our daughters have left home so the only responsibility she has, you could say, is me’.

In this sense, women participants felt better equipped to assume the caregiver role as most of them had previous experience of taking care of someone else (Table 3), while for the men who took part in this study, this was their first experience in this role.

I've always been someone that …I've taken care of my father, I was with my brother when he was ill, when my sister had a breast removed too …So, I'm someone that can adapt to any situation and it doesn't affect me, thank God. I mean, losing a loved one affects me. That gets you down, but then you say ‘Well, if that's the way it is…’ If God meant it to be like this …it's the hand that life dealt me. (Miriam, 66 years old, caregiver to her husband)

In fact, for some women participants taking care of their relative with CP was simultaneous to taking care of their children or grandchildren, despite the extra responsibility and time it implies. As Elena, who takes care of her

mother, said:

I'm also at an age where, it's not that I'm very old, but you know, I do the housework, take the girls to school, come back to look after my mother; I go with her, up, down, running around …stress, lots of stress. And then my back and neck feel the effects and it affects me. I have contractures, I get dizzy and I have to take Enantium and Diazepam all the time. It affects me a lot. (Elena, 51 years old, caregiver to her mother)

The burden of being a caregiver and coping strategies

The combination of the different components described in the previous two themes leads to a wide range of perceived burdens resulting from the caregiver's role. The narratives of those participants who were wealthier and had good family or social support expressed fewer consequences for their physical and mental health than other participants who were struggling with other issues such as limited economic resources.

However, it is important to note that under this theme and within each interview there were inconsistencies, in the sense of participants saying there were no consequences and later describing some, such as a very limited social life, abandoning hobbies to take care of their relative, physical issues, anxiety, fear for the future, emotional exhaustion or physical consequences to cite some examples (Table 3).

I take pills and I have been to many psychologists and they all say the same: 'it's your mother …You are your mother's mother and, in the end, that muddles everything'. (Marta, 47 years old, caregiver to her mother)

At the same time, statements showing resilience, hope and a positive attitude were commonplace. In line with them, participants described their coping strategies and how they were making an effort to have their own space or, in some cases, after realizing they were somehow losing their life, making a conscious effort to regain it.

Uff …if you don't want to bang your head against the wall along the way, it isn't a physical journey, it's a mental one. It's mental. How you are influenced …by certain emotions and you don't understand …you have to understand that, despite the pain, life goes on. (Rocío, 34 years old, caregiver for her husband)

DISCUSSION

The findings of this study show that being an IC for a relative with CP became the most important role in the IC's life, to the point of casting a shadow over the priorities of their own lives. The experiences also varied depending on the contexts and key elements such as socioeconomic level or family support. Likewise, independently of the context, the ICs had the feeling of being the only person responsible and able to perform the care, regardless of the consequences.

A significant result of this study was that the ICs interviewed neglected to live their own lives to care for their relative with CP. It is remarkable how the ICs referred to their relative's illness as if it were their own and to the pain process as a shared experience. These findings are in line with studies conducted to assess the needs of ICs of individuals with other pathologies, where participants lacked time to care for themselves and address their own health concerns,[23] in some cases with severe detrimental consequences for their health. It has been shown how CP becomes the focus of patients' lives, and they have to both redefine their identity and adjust to the new constraints and physical limitations of their bodies.[24] In this line, and in the context of our findings, ICs change their daily lives according to treatment regimes, helping to manage pain and side effects, attending medical appointments and resolving everyday problems.[9,25] Thus, the length of time spent on care is not the only important predictor of the overall burden on ICs.[8,26] There is also the time lost for themselves, the lack of privacy and the development of a new identity as a caregiver.[27] The enormous impact the CP had on the lives of the IC found in this study suggests the concept of 'we-disease' can be applied to the described experiences. This concept, developed initially by the participants of a study about stress and coping strategies among breast cancer patients and their partners, reflects how breast cancer impacts not only the patient but their intimate partners as well and, furthermore, how the coping strategies with the stress of each of the members of the couple are interrelated.[28,29] Future research on the experiences of IC could further explore the applicability of the 'we-disease' concept to IC and enrich knowledge about their experiences by adding this interrelational dimension.

Social and structural determinants of health related to the burden of people suffering from CP have been widely studied.[30,31] Our study enhances this knowledge by highlighting how social determinants of health are also a key

element in the burden of being an IC. The limited budget was described by interviewees as a source of worry and a limitation to the care they could provide, and thus contributed to the perceived burden. Moreover, economic difficulties aggravated the decrease in the time that both the IC and the CP patient had for leisure and social activities. This finding is relevant in light of the results from Miller et al.,[11] who found the inability or difficulty to pay for basic needs and not having a social life were related to a high prevalence of depression among ICs. It has been argued that no other health problem causes as much disability as CP.[32] In this line, the lack of economic resources to make the patient's home accessible prevented both the patient and their family member from spending more time outside of their home. Prior studies[33] have shown housing as an important factor driving health inequalities, with long-term isolation producing adverse effects on mental well-being. Various studies suggest that among the variables that have contributed to the current crisis in CP care are policies that influence the socioeconomic climate of the healthcare system.[34] In this line, our results indicate ICs experience a feeling of helplessness due to a lack of resources provided by public health or social services, such as financial support or formal caregivers. Therefore, addressing social inequalities associated with CP is an essential initial step in improving this health problem, using collaborative approaches based on the chronic care model, which would optimize not only the patients' quality of life but also reduce the burden on caregivers.[35]

This study was conducted in the context of the COVID-19 pandemic. The fear of contracting COVID-19, along with public health measures such as home confinement, increased exponentially the time spent at home. Recent literature[36] has emphasized that one of the most important consequences of the lockdown was its impact on mental health, particularly fear, anxiety and negative thoughts about oneself and the future.[37,38] In this respect, the lockdown together with a combination of the different factors resulting in a limited social life—taking care of their relative, physical issues, emotional exhaustion or physical consequences—lead to an increase in the burden perceived by the caregiver. Future public health measures like those implemented during this pandemic should take into consideration the impact they have on vulnerable populations to minimize health inequities instead of increasing them.

However, the caregiver burden is not a universal experience.[27] Some individuals are able to adapt easily to the responsibility and demands of caregiving, whereas others report significant strain and distress.[39] In line with previous research, the study showed intergroup differences in gender, depending on the relationship with the family, sources of support, duration of care and stage of the disease. The findings suggest that gender and the type of relationship are important concepts in understanding the caregiving process and that they are often interwoven. In the case of the women, there were differences in recognizing them as a caregiver since they assume the role motivated by love and the desire to return the love received by their relatives. In fact, some of them had played or were playing this role with various members of their families simultaneously, accepting that they had no choice. This finding is in consonance with previous research on the ‘sandwich generation’.[19] Men and women have been shown to cope differently with caregiving situations.[40] Women are more concerned with the enhancement of others' emotional well-being and with the provision of emotional support. They are more emotionally involved in the caregiving, while at the same time being largely responsible for doing the housework. Men have a more task-oriented approach to caregiving. This suggests that both the level and impact of the burden develop differently over time for men and women, as other articles have shown.[41]

STRENGTHS AND LIMITATIONS

As previously described, several steps were taken to strengthen the trustworthiness of the findings. They do, however, need to be interpreted with some limitations in mind. Concerning transferability, it is important to consider the context in which this study was conducted: a group of individuals who accompanied their relatives to a pain clinic in the Spanish healthcare system. With this in mind, the results from this study could be relevant for understanding the experiences of ICs for a relative diagnosed with CP in other countries with a similar sociocultural background and healthcare systems since the consequences they face and concerns they have may be similar.

Regarding credibility, participants with different sex/gender, ages and experiences were chosen to increase the likelihood of shedding light on the research question. However, the vast majority of people interviewed were women.

Nonetheless, as discussed in this study, this is in line with the percentage of women who tend to be caregivers since they assume the role motivated by love, while also accepting that they have no choice.
Another limitation of this study is that, although to take part in this study the participants had to consider themselves to be the main caregiver, the results suggested gender differences in the way the idea of care is conceived and understood. However, the data were not rich enough to support a deep analysis and the elaboration of conclusions about the social construction of the term. Further research with this aim is required.

CONCLUSION

This study's findings highlight how the CP impacts IC lives. Being an IC for a relative with CP became the most important role in the IC's life, to the point of casting a shadow over their own needs. Besides, participants felt not having other options but to keep going with that role. Yet, the context was essential in shaping the experiences as caregivers and the burden derived from caregiving. In this line, differences related to gender roles were found in the narratives of participant women and men.

Practice implication

The number of people suffering from CP is expected to continue rising, and consequently so will the number of IC. This study shows that ICs of individuals with CP have specific needs similar to the IC of individuals with other chronic conditions but with certain specificities. The lack of a formal caregiver provided by the state and delays in financial help lead to the family member feeling more and more isolated with greater responsibility and a bigger burden, leading to physical and mental problems. This should be taken into account in the implementation of policies and healthcare programs aimed at the attention of individuals living with CP.
Finally, IC should be considered an integrated part of the CP illness process by healthcare providers. In this enhanced person-centred care, to meet both patient and IC's needs, social determinants of health and social support should be assessed in each individual case from a gender perspective to implement evidence-based measures that prevent negative consequences for IC.

ACKNOWLEDGEMENTS

We would like to thank the participants of this study for generously sharing their experiences. We would also want to thank the physicians and nurses from the Unit of Pain in the University Hospital ‘Puerta del Mar’ for letting us be part of their team for a period of time, especially Dr. Cristina Delgado Naveiro. Helena De Sola has disclosed that she has received a grant for research in the field of Pain from Fundación Española del Dolor.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

The study protocol was approved by the Clinical Research Ethics Committee (Reference Number of the study: EXPCUI2020), ensuring compliance with the standards of good clinical practice. All informants gave their consent to participate after they had received individualized and sufficient information. This also included the possibility of taking back the consent to participate.

DETAILS

Subject:	Informal care; Womens health; Relatives; Coping strategies; Sex roles; Gender; Mental disorders; Gender differences; Families &family life; Data analysis; Caregivers; Chronic pain; Pain; Interviews; Patients; Caregiver burden; Informed consent; Supervision; Data collection; Housework; Qualitative research

Business indexing term:	Subject: Supervision
Identifier / keyword:	burden; chronic pain; experiences; informal caregivers
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	409-418
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-12-08
Milestone dates:	2022-10-24 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-05-26 (manuscriptReceived); 2022-12-08 (publishedOnlineEarlyUnpaginated); 2022-11-08 (manuscriptAccepted)
Publication history :	
First posting date:	08 Dec 2022
DOI:	https://doi.org/10.1111/hex.13671
ProQuest document ID:	2767102269

Alignment of patient-centredness definitions with real-life patient and clinician experiences: A qualitative study

Babione, Julie [1] ; Panjwani, Dilshaan [1] ; Murphy, Sydney [1] ; Kelly, Jenny [1] ; Jessica Van Dyke [1] ; Santana, Maria [2] ; Kaufman, Jaime [1] ; Sargious, Peter [3] ; Rabi, Doreen [3] [1] Department of Medicine, Cumming School of Medicine, O'Brien Institute for Public Health, University of Calgary, Calgary, Alberta, Canada [2] Departments of Paediatrics and Community Health Sciences, Cumming School of Medicine, O'Brien Institute for Public Health, University of Calgary, Calgary, Alberta, Canada [3] Department of Medicine, Cumming School of Medicine, W21C Research and Innovation Centre, O'Brien Institute for Public Health, University of Calgary, Calgary, Alberta, Canada

ABSTRACT (ENGLISH)

Introduction

Patient-centred care (PCC) has come to the forefront for many institutions, funding agencies and clinicians, and is integrated into care. Does a disconnect in understanding still exist between patients, healthcare organizations and clinicians in what PCC means and how outstanding issues might be addressed?

Methods

We conducted interviews and focus groups with self-reported chronic care patients and clinicians providing care to these patients exploring PCC experiences, expectations and practices. These data were initially analysed using inductive thematic analysis. This paper reports on the findings of a secondary analysis examining the alignment between patients and clinicians on five key predetermined dimensions of PCC.

Results

Eighteen patients participated, representing a range of chronic conditions. Thirty-eight clinicians participated. One thousand and three hundred patient and 1800 clinician codes were identified and grouped into 5 main topics with 140 unique themes (patients) and 9 main topics with 54 unique themes (clinicians). A total of 166 quotes (patient = 93, clinician = 73) were identified for this PCC definition alignment analysis. Partial or complete alignment of patient and clinician perspectives was seen on most dimensions. Key disconnects were observed in patient involvement, patient empowerment and clinician–patient communication. Only 18% of patients reported experiencing patient-

centred communication, whereas 57% of clinicians reported using patient-focused communication approaches.
Conclusion
Overall, study patients and clinicians endorse that many PCC elements occur. This study highlights key differences between patients and clinicians, suggesting persistent challenges. Clinician participants relayed their PCC approaches of informing and educating patients; however, patients often perceive these approaches as didactic, role-diminishing and noncollaborative. Collaborative approaches, such as shared decision-making, hold promise to bridge persistent PCC gaps and should be integrated into medical education programmes.
Patient or Public Contribution
This project was conceived and executed with a co-design approach wherein patients with chronic conditions who are trained in research (i.e., see descriptions of Patient and Community Engagement Research in the text) were involved in all stages of the research project alongside other researchers on the project team. Healthcare providers were involved as participants and as principal investigators in the project.

FULL TEXT

INTRODUCTION

The Institute of Medicine defines ‘patient-centred care’ (PCC) as ‘care that is respectful of and responsive to the preferences, needs, and values of the patient’. The Wagner Chronic Care Model[1,2] has been the template for care provision for over two decades, with significant investment in PCC at policy and mandate levels,[3–5] through patient advocacy[6] and in performance measures.[7,8] In Canada, there is a proliferation of primary care teams and medical homes that provide PCC to diverse patients. While evidence suggests these care models (with particular emphasis on interprofessional collaboration and care integration) effect positive clinical outcomes,[9–12] it is unclear whether the patient's care experience is significantly improved, or tangibly different, than with traditional models.

The importance of the patient and their lived experience in informing care is recognized by healthcare organizations and research funding agencies that prioritize patient inclusion and partnership.[13–16] Clinicians may see PCC as a service delivery structure that better supports patients' needs. Concurrently, patients may reasonably expect PCC to centre care on their experience of a condition, and formally include them in care processes and decisions that relate to them. A shared understanding of PCC's meaning is fundamental to achieving health care centred on patients, but the degree to which the delivery of PCC at the clinician level and the experience of PCC at the individual level are not extensively characterized.

Recognizing this potential disconnect between the agents and the objects of PCC, we conducted a secondary content analysis of patient and clinician narratives using principles of natural language processing[17] to determine (1) whether contemporary care experiences are patient-centred and (2) how patient and clinician perceptions of PCC align.

METHODS

Our team collected patient and clinician narratives as part of focus groups and interviews in the context of developing a patient-centred planning tool for adults with multiple chronic health conditions. We conducted a secondary content analysis exploring PCC experiences from two distinct perspectives: patients and clinicians. Using a conceptual model of PCC[18–20] (detailed below) we identified keywords and themes to capture experiences of PCC in patient and clinician narratives.

Focus groups and interviews with patients

Patients were recruited through the Patient and Community Engagement Research (PaCER) programme at the University of Calgary.[21] PaCER researchers are patients trained in research methodologies (i.e., interviews, focus groups and surveys). PaCER's research is iterative, with three distinct data collection and analysis phases: Set, Collect and Reflect.[22] We recruited study participants through outpatient speciality clinics and through existing networks.

Within the PaCER patient ‘Collect’ phase, data were captured through audio recordings, flip charts and process recording notes. Audio recordings were transcribed and analysed, deducing themes that informed the last (Reflect) phase, which explored participants' experiences of self-management. Supplementary (Collect phase) interviews

were added until no new themes emerged.

Focus groups and interviews with clinicians

This research team conducted semistructured interviews and focus groups with clinicians, following a guideline script that explored the understanding of patient-centredness, how they involve patients in care planning, potential digital platform information elements and perceived general barriers and facilitators to digital platform uptake. With the script as a guideline, researchers were able to expand and elaborate, exploring topics, concepts, examples and responses to patient content as they emerged in the same and in subsequent sessions. Local primary care networks facilitated recruitment through targeted invitations supplemented by existing networks and sessions were audio-recorded.

Participant data analysis

All sessions were anonymized, transcribed and verified. Four researchers (J. B., J. K., S. M., J. V. D.) including a social sciences expert in qualitative research methods (J. K.) conducted the inductive thematic analysis.[23,24] Researchers independently reviewed and coded transcripts in pairs, each team delegated half the transcripts. Codes were then reconciled through consensus discussions.

Alignment analysis

In parallel, another researcher (D. P.) used the analysis to identify specific quotes that aligned with the top five dimensions of PCC. Table 1 shows these dimensions that were derived following systematic review, consensus approach and selected as representative of a rigorous PCC concept synthesis.[19]

Table 1 Dimensions of PCC[18]

Dimension	Definition
Patient as a unique person	Each patient's individual needs, preferences, values, feelings, concerns, ideas and expectations as well as exploring both the patient's disease and illness experience, the impact on functions (e.g., the patient's idea of how the illness affects his or her daily life; effects of the illness on the patient and his or her family), and his or her individual explanatory model. This also entails providing care that is tailored to each specific patient.
Clinician–patient communication	Many aspects of how we communicate in a patient-centred manner are included in the definitions of patient-centredness. They include general communication skills, (e.g., setting the stage, setting an agenda, prioritizing the patient's problems). A broad range of verbal and nonverbal behaviour can be used to engage in patient-centred communication (e.g., using open-ended questions, summarizing important information, asking the patient to repeat, making eye contact, nodding).
Patient information	This dimension highlights the importance of sharing knowledge and information reciprocally between the clinician and the patient. The clinician should give tailored information (regarding all aspects of care from prevention to treatment, as well as information on how to access medical, psychosocial, physical and financial support) while eliciting and respecting the patient's information needs and preferences. Some definitions also described the provision of informational resources and tools (e.g., audio records of consultations, multimedia resources, information brochures). Furthermore, the patient should be encouraged to share information (e.g., regarding symptoms and concerns).

Patient involvement in care	A prominent dimension often described in the literature on patient-centeredness is the patient's active involvement in care. While older publications use terms like ‘informed consent’ or ‘sharing power and responsibility,’ more recent publications define in more detail the importance of encouraging the patient to participate actively in the consultation and of engaging the patient in the decision making regarding his or her own health (shared decision making). The importance of helping the patient in making informed choices is highlighted in many definitions. This includes respecting the patient's preferences for involvement as well as encouraging the patient's feedback on care (e.g., using patient surveys).
Patient empowerment	‘…by acknowledging the patients' perceived ability to self-manage important aspects of his or her illness, activating and encouraging the patient to take responsibility to solve health related problems and to take actions to improve his or her health and becoming an expert regarding the management of his or her health condition. This also entails supporting the patient's autonomy by offering educational programs, patient activation and health promotion interventions’.

Note: Content reproduced with permission under the Creative Commons Attribution License.
Abbreviation: PCC, patient-centred care.

Two researchers (D. P. and J. B.) independently coded quotes within dimensions according to *concept presence* and *concept concordance*. For concept presence, quotes were evaluated whether they *referred* to a specific dimension. For concept concordance, quotes were examined for *alignment with* the dimension.

For example, for the dimension ‘patient as a unique person’, concept presence was (‘recognition of each patient's uniqueness—individual needs, preferences, values, feelings, beliefs, concerns, ideas, expectations’) and concordance ‘the patient *is* a unique person’. If a quote spoke about patient uniqueness *and* that the patient *is* unique, both criteria are met. Quotes were colour-coded as:

- *Green* (both concept presence and concordance match), illustrating the selected quote was present and in complete alignment with the dimension.
- *Yellow* (concept presence *or* concordance match), illustrating that either the quote was relevant for the dimension *or* the viewpoint was in alignment.
- Red (neither concept presence nor concordance match). This might occur if the first researcher (D. P.) reconsidered the quote, or the second researcher (J. B.) disagreed that the quote was sufficiently applied.

Coding consensus was established on all included quotes and explicit comparisons between patient and clinician alignment quantified and visualized.

RESULTSFocus groups and interviews

Eighteen patients participated, representing a range of chronic conditions, including cancers, diabetes, liver failure, leukaemia, bone marrow transplant, heart problems, scleroderma, chronic obstructive pulmonary disease, Hashimoto's thyroiditis, arthritis, depression, bipolar disorder and anxiety. Table 2 summarizes these demographics. Thirty-eight clinicians participated in interviews or focus groups with representation as shown in Table 3.

Table 2 Demographic descriptors of patient participants

	N (%)

Basic demographic	
Sex (female)	11 (64.7)
Age	60.94 (12.3)
Education level	
High school diploma	3 (17.6)
College or university	14 (82.4)
Working status	
Full time/part time	4 (23.5)
Retired	7 (41.2)
Disability/sick Leave	5 (29.4)
Unemployed	1 (5.9)
Ethnicity	
Asian	1 (5.9)
East Indian	1 (5.9)
Caucasian	15 (88.2)
Family physician	
Yes	16 (94.1)
Self-reported health status	
Very good	3 (17.6)
Good	7 (41.2)
Fair	5 (29.4)
Poor	2 (11.8)
Other descriptive demographics	

Number of medical conditions	3.59 (1.5)
Average number of healthcare providers on patient's team	3.82 (2.5)
Time managing health conditions (hours/week)	25.45 (46.97)

Table 3 Clinician participant demographics

Clinician type	# Participants
Physician	3
Specialist (Geriatric Medicine, Internal Medicine (2), Respirology, Neurology, Infectious Disease, Nephrology)	7
Pharmacist	5
Nurse	11
Social work	3
Dietician	1
Other (Kinesiologist, Medical Office Assistant, Behavioural Health Consultant (2), Patient Flow Coordinator, Clinic Manager, Unit Manager, Allied Health Manager)	8
Urban/rural	30/8

Primary participant data analysis

One thousand and three hundred patient and 1800 clinician codes were identified. Once both data sets were coded, two researchers (S. H. and J. K.) grouped codes into 5 main topics with 140 unique themes (patients) and 9 main topics with 54 unique themes (clinicians).

PCC definition alignment analysis

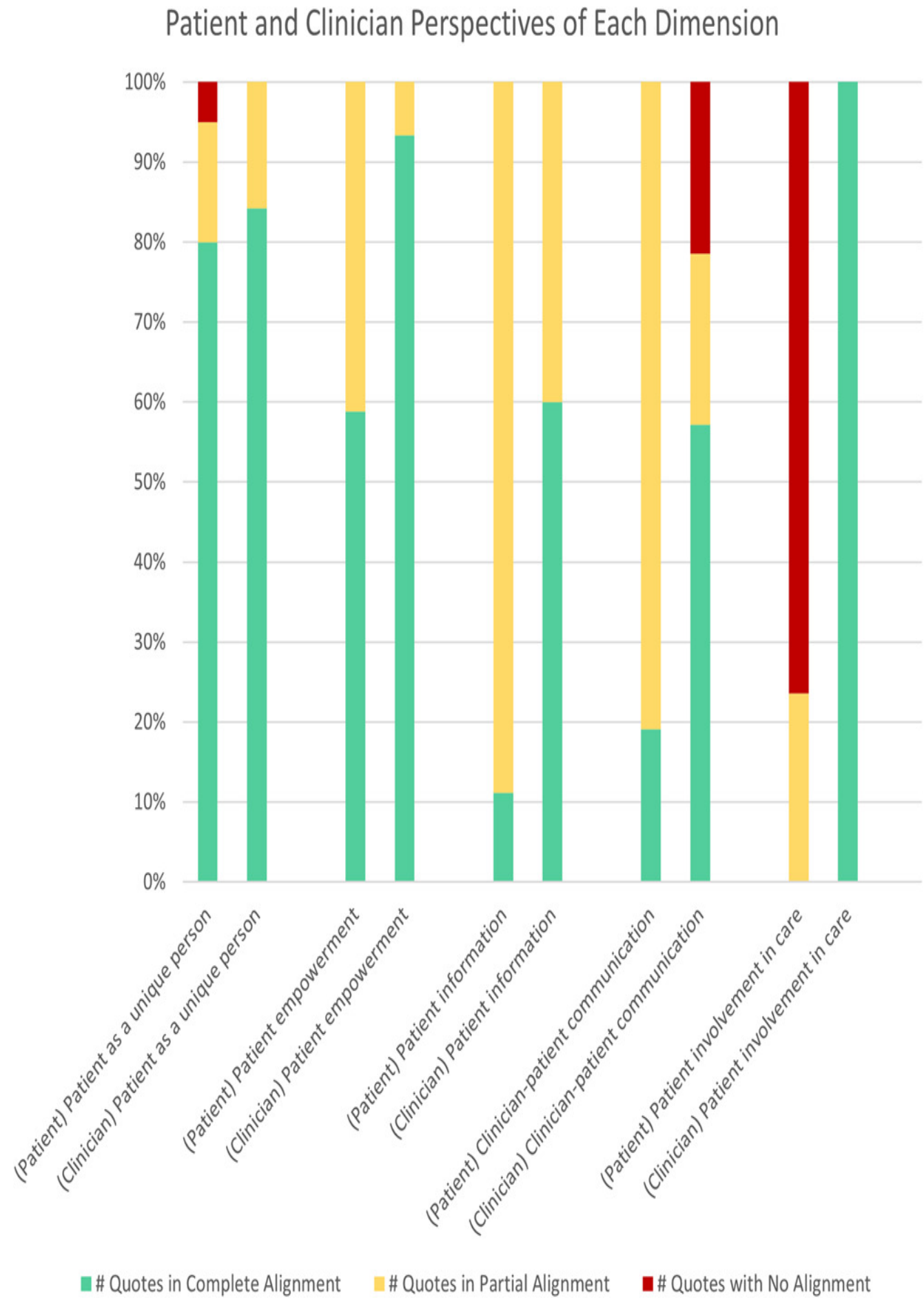
A total of 166 quotes (patient = 93, clinician = 73) were identified for this PCC definition alignment analysis. The distribution of quotes across themes is summarized in Table 4.

Table 4 Number of patient, clinician and total quotes as included in alignment analysis

Dimension	Patient quotes	Clinician quotes	Total quotes
Patient as a unique person	20	19	39
Clinician–patient communication	21	14	35
Patient information	18	15	33

Patient involvement in care	17	10	27
Patient empowerment	17	15	32
Total	**93**	**73**	**166**

As Figure 1 shows, patient and clinician narratives suggested that many elements of PCC are being achieved across most dimensions. Table 5 captures key example quotes for each dimension referenced in the text.



Enlarge this image.

Table 5 Sample patient and clinician quotes included in this secondary alignment analysis

Dimension	Patient quotes	Clinician quotes
Patient as a unique person	[IMAGE OMITTED. SEE PDF.] ‘Having been depressed before and having been worsely depressed, being terrified about where that spiral went, I actually did a lot of self-controlling and saying, “I'm not going there again” and how do you not go there again? You continue to fight. So, from a previous experience had developed a whole array of tools’.	[IMAGE OMITTED. SEE PDF.] ‘Well, I'm seeing someone, we made some really good diet and exercise goals. Her goals, they're doable, we checked you know, confidence, importance, readiness and from those found some more barriers and then kind of did a plan B…’.
	[IMAGE OMITTED. SEE PDF.] ‘And when I got back to my family a couple of weeks ago, one of my cousins said to me “You look just like five years ago. You haven't changed, or you don't act different. You're not marked,” she said’.	[IMAGE OMITTED. SEE PDF.] ‘The dynamic has switched here. It's easy for me to sit here in my seat and say you need to do a, b, c, d, but you need to tell me what you are able to do and willing to do’.
Patient empowerment	[IMAGE OMITTED. SEE PDF.] ‘…you have to be an advocate for yourself as much as possible because if they say, “Don't call us, we'll call you,” I never take that advice. I call and make sure I'm told’.	[IMAGE OMITTED. SEE PDF.] ‘I try to encourage them to like be involved as actively as they can, so whether that is voicing their concerns, or just kind of updating people as things go along so we know, and to provide us enough information so we know where we're coming from’.
	[IMAGE OMITTED. SEE PDF.] ‘Of course, the first thing he said to me, when he got my results back—**he eventually did agree to do them because I said “well if you're not going to run these tests for me, I will find someone who will”** because I knew a lot of the things were happening to me and I didn't like what was happening to me. So eventually he did agree to run them and the first thing he said was “Are you ok?” I didn't even register on the scale on this one type of test he did’.	[IMAGE OMITTED. SEE PDF.] ‘And yes, we do involve our patients […] we do the congestive heart failure teaching, the diabetes teaching, the COPD teaching. If I have the opportunity to do that. […] Very basic, but putting it back to them, that they need to do that for themselves’.

Patient information	[IMAGE OMITTED. SEE PDF.] ‘I had my huge meltdown and I actually had been told the plan. And it was just around the very beginning and the plan was that one induction and two consolidation chemos. But I didn't hear that I had two consolidation chemos. I thought I only had one. So, when I went in there, I found out that I had two and that was meltdown. **And I actually had been told that, I was just so sick that I hadn't got it**’.	[IMAGE OMITTED. SEE PDF.] ‘I'm imagining something they can interpret versus like our actual care plan that we always see where you need to understand the medical jargon and understand what everything means’.
	[IMAGE OMITTED. SEE PDF.] ‘You have reached a point where you are **not quite sure what was just said**, you don't remember what drugs you were supposed to take etc. **And there are a number of breakdowns in that interaction**’.	[IMAGE OMITTED. SEE PDF.] ‘I do a lot of discharge instructions sometimes for the patient because you can see that sometimes they don't understand or even though they are given a discharge summary with everything in there, they are still overwhelmed by everything. So, I'll just give the 1, 2, 3, 4, and that's after talking to the residents if they're going to do discharge instructions’.
Patient–clinician communication	[IMAGE OMITTED. SEE PDF.] ‘I think it's incredibly important. I would say […] so this is what is going to happen now. And then after that somebody is going to discuss with you what the next step is, and this will take this many days. And then you are going to talk to this person and this person and then they will determine what the next time is that you're looking at’.	[IMAGE OMITTED. SEE PDF.] ‘I think one of the things that I have seen is the knowledge, the actual ability to understand the implications of the medical perspective. A lot of the information that a team will talk about is very technical […] you have to simplify it quite a bit so even if you were to attempt to …I don't know if patients and families would ever fully understand the whole picture’.
	[IMAGE OMITTED. SEE PDF.] ‘It's one thing to go to the doctor and he knows and does everything, and the nurses do everything, **but they don't really communicate with you and tell you…**’.	[IMAGE OMITTED. SEE PDF.] ‘Or taking that with them to go home and how they can further educate themselves. So, I educate them why I get them to do deep breathing to prevent pneumonia, to move fluids around, and I call it chest physio. And I explain to them exactly why I get them to do that’.
Patient involvement in care	[IMAGE OMITTED. SEE PDF.] ‘Now I've got other health issues […] and I tell a doctor, a new system of doctors something, and they say, “oh no, that's nothing” and **they don't believe me**. You know all that credibility I had built up at the [other] clinic, in [this] suddenly I'm stupid, and you know that's not true right. **I find that really insulting**’.	[IMAGE OMITTED. SEE PDF.] ‘I think […] to keep putting it back to the patients so what can you do, and how are you going to make this work, and just keep working with them on it until they start to take it on and start to get the hang of it’.

	[IMAGE OMITTED. SEE PDF.] 'Given what I have figured out from the literature and stuff, **it feels like the doctors I was dealing with were very—this is the way I have done it for twenty years, why should I change what I'm doing**?'	[IMAGE OMITTED. SEE PDF.] '…this only applies to some of our patients, sometimes some of the stuff about patient accountability and patient responsibility for their own health. And how do we draw them in as a partner rather than a victim of health care'.

Note: Quotes deemed to be in perfect alignment (concordance) with the dimension are precluded with a green [IMAGE OMITTED. SEE PDF.], and those in partial alignment (partial discordance) with light orange [IMAGE OMITTED. SEE PDF.]. Selective text bolding within quotes highlights those elements that signalled the partial classification of those quotes.

Abbreviation: COPD, chronic obstructive pulmonary disease.

Patient as a unique person

Patients and clinicians aligned well on the 'patient as a unique person' dimension, with over 80% of extracted quotes explicitly highlighting the importance of patients being recognized as individuals.

Patient empowerment

There was also similar alignment on patient empowerment with 58% of patient quotes expressing feelings of empowerment, and 94% of clinician quotes supporting patient empowerment. Despite no overt disagreement on this dimension, patient and clinician quotes suggest interpretational differences and thus discordance in understanding between groups. Often, patient–participants likened empowerment to self-advocacy and the ability to self-determine care and outcomes, as in:

I've become very resourceful so I think for me, managing it if I know where my resources are, I know what the treatments are; I'm very good at asking the questions and knowing what is the best way to manage it. (Patient) [IMAGE OMITTED. SEE PDF.]

Some patients often viewed empowerment as a desire for emancipation from conventional practices that limit their role in accessing health information or prevent autonomous decision-making. Conversely, clinicians often viewed patient empowerment as engagement in self-care, being 'educated', a patient's ability to self-manage or monitor or relaying information between clinicians. One example quote that speaks to this is:

You get them to go to the kiosk to look up information and follow up with it. They come in with diabetes but they also have hypertension so we get them to watch a video on why you want to monitor at home and why it's important to bring those readings to the doctor…. (Clinician) [IMAGE OMITTED. SEE PDF.]

Patient information

On this dimension, patient and clinician quotes suggest partial or complete alignment. Patient quotes were often partially aligned, expressing a desire for more reciprocal or situation-appropriate information sharing. They also expressed a desire for better access to their health information.

Clinician–patient communication

Only 18% of patients reported experiencing patient-centred communication, whereas 57% of clinicians reported using patient-focused communication approaches. Notably, 22% of extracted clinician quotes suggested that a patient-centred communication dimension was *not* being achieved. Patients often focussed on needing information about 'next steps' and having their concerns/questions addressed, as in:

It's one thing to go to the doctor and he knows and does everything, and the nurses do everything, but they don't really communicate with you. (Patient) [IMAGE OMITTED. SEE PDF.]

One patient expressed frustration and stress in recounting experiences awaiting direction. In contrast, clinicians

expressed feeling successful at communicating with their patients. Most quotes spoke of clinicians simplifying medical explanations or 'educating' their patients, as in:

Or taking that with them to go home and how they can further educate themselves. So I educate them why I get them to do deep breathing to prevent pneumonia, to move fluids around, and I call it chest physio. (Clinician) [IMAGE OMITTED. SEE PDF.]

Patient involvement in care

Patients and clinicians had strikingly different perceptions within this dimension. Most patients felt uninvolved, while interviewed clinicians expressed confidence in patient involvement. Clinicians spoke of attempts to engage patients in self-care; however, patients felt their involvement was often prescribed or lacked autonomy. Patients also felt an undesirable off-loading of care duties, making their 'involvement' burdensome with care workload concerns often going unheard. One patient participant recounted trying to engage in care decision-making felt their efforts to self-educate and inform care discussions were dismissed. Meanwhile, clinician participants saw patient involvement very differently—as executing prescribed self-management plans, as in:

…they are the care giver for their husband or wife at home and its really difficult for them to get out. Really all I need to know is […] what have you done, what are your blood sugar numbers, how have you been adjusting your insulin and we just need to do some tweaking…. (Clinician) [IMAGE OMITTED. SEE PDF.]

Sometimes, clinicians indicated trying to encourage patients to actively take responsibility for their own care. In other cases, clinicians indicated a desire to partner with their patients.

DISCUSSION

This study examined how patients' and clinicians' PCC experiences aligned across five dimensions considered integral to this care model and revealed important differences between patient and clinician perspectives and understanding of the dimensions, This suggests that there are several ongoing challenges in patient-clinician care collaboration. For example, the dimensions of patient empowerment and patient involvement in their care *are* difficult to distinguish and can easily be misinterpreted. Clinician participants relayed their PCC approaches of informing and educating patients; however, patients often perceive these approaches as didactic, role-diminishing and noncollaborative. If PCC is defined as being respectful and responsive to patients' preferences, values and needs, then reciprocal information sharing is integral for clinicians to fully understand each individual patient.

Our findings around unaligned PCC dimensions are consistent with prior research that patients feel insufficiently engaged or that the approach misses the target.[25] Practices such as shared decision-making (SDM) have been proposed to address these concerns. SDM is an approach where patients and clinicians collaboratively create personalized care decisions for each patient based on a shared review of relevant clinical and experiential data to the care decision and define a path that makes intellectual, emotional and practical sense to the patient.[26–28] When SDM occurs, patients report higher satisfaction with care decisions and more positive care experiences.[29–31] When done well, SDM epitomizes information exchange and patient involvement. However, despite being evidence-supported and highly recommended, SDM remains underutilized, with only 42% of Canadian patients reporting an SDM experience and only 21% reporting engagement matching their preference[25]—a finding that is corroborated by our work from multiple patient and clinician perspectives.

Primary care has seen remarkable investment and reorganization ensuring patients can access a multidisciplinary care team and expanded medical services provided within their 'medical home'. This organizational change has created more PCC delivery,[32] but as our study illustrates, this care structure does not guarantee collaborative care experiences. Unfortunately, PCC appears to conceptualize the patient as someone who needs to be educated, who must do more work to self-manage their conditions, and so forth, which effectively operationalizes a degree of

paternalism in care delivery and diminishes the patient. While attitudes towards SDM are generally favourable,[33] across medical disciplines SDM knowledge of the impact on decision outcomes, the patient's role in decision-making, and a misunderstanding of SDM's time intensity are identified barriers to application in practice. Our study illustrates this knowledge-practice gap in, for example, the nonalignment of ‘patient information’ and ‘patient involvement’ dimensions. Patients want more equitable care, which we might find in relationship-centred and strength-based approaches that honour the lived experiences of the patients and respect them as equal partners in their care, and approaches beyond PCC may facilitate this.

Clinicians also need better training to improve the implementation of collaborative care practices like SDM. Medical students were surveyed in four countries on SDM knowledge, ability to communicate risk and SDM attitudes, revealing SDM as a highly-trainable skill, however not routinely provided (i.e., the proportion of students receiving SDM training varies from 2% to 74%).[34] The literature also suggests that patient characteristics may drive clinician SDM engagement decisions. Older, racialized or female patients are less likely to experience SDM despite often strongly desiring engagement in their care decisions.[35]

Limitations

The research team blinded themselves to clinician professions when selecting quotes, in an effort to prevent any bias. However, we acknowledge that this blinding to clinician status (as, e.g., a physician, specialist or nurse) makes the assumption that individuals from these different professions would speak to these issues similarly, which may not have been the case. There may well be differences in perspective by professional groups, but this was not explored in this analysis. This work was conducted within a single provincial health region and may not reflect PCC experiences elsewhere. Nonetheless, clinicians trained outside this region practice here, suggesting SDM training can be universally improved. We endeavoured for diversity, with representation from a range of urban and rural clinical settings and chronic conditions, however, most participants identified as Caucasian with access to study participation opportunities. It seems to be a paradox of not hearing from the people who need equity-focused approaches the most, as these tend to be the people with the least trust in the establishments and the least capacity to participate in co-design activities—something that the research team is currently exploring. Finally, participants were asked about care experiences (patients) or how clinicians involve patients in their care without reference to SDM. Thus, conclusions drawn about the patient involvement variability may be biased. Concurrently, without priming study participants to SDM concepts allowed for unbiased and naturally shared experiences without assumptions or reference to specific care models—experienced or not. This work was also conducted in a prepandemic context, so we may not have captured new attitudes resulting from the patient and provider care or SDM experiences that may have shifted with the pandemic. Still, our work helps draw attention to challenging elements of PCC that may persist with the wider adoption of virtual care as a more durable or patient-centred way of interacting that might create the best opportunity to revisit and embed principles of SDM and PCC.

CONCLUSIONS

Patient care has recently been restructured to be patient-centred, however, care collaboration where patients feel respectfully engaged in their health is lacking. This study, which is part one part of a larger research, design and development project that uses a Human-Centred Design approach to create digital supports for patients with (multiple) chronic conditions in ways that support both PCC and SDM, examined PCC dimension alignment between patient and clinician perspectives. Clinicians may lack insight into the patient perspective on this. Recognizing and responding to patient needs should be foundational in PCC with continued work required to ensure patients are meaningfully engaged. While SDM has the potential to provide a patient–clinician collaboration framework, it remains underutilized despite interventions to support integration into standard care practices. As researchers, we

have the opportunity to intentionally push for more collaborative care practices and better education for our clinicians.

AUTHOR CONTRIBUTIONS

Julie Babione coordinated and conducted most components of the research project including data collection, conducting clinician interviews and focus groups, coordinating transcription and preliminary data analysis, co-conducting the alignment analysis, manuscript writing and coordinating internal revisions. Dilshaan Panjwani coordinated and co-conducted clinician interview and focus group sessions, co-conducted the alignment analysis and participated in manuscript writing and revisions. Sydney Murphy conducted some transcription, co-conducted the preliminary data analysis and contributed to manuscript writing and revisions. Jessica Van Dyke co-conducted the preliminary data analysis and contributed to manuscript writing and revisions. Maria Santana contributed to the project's conception and early guidance and contributed to manuscript writing and revisions. Jaime Kaufman oversaw the project and contributed to all stages of the project, as well as manuscript revisions. Peter Sargious contributed to the project's conception, guided the project and contributed to manuscript revisions. Doreen Rabi provided regular project guidance, including data analysis and alignment analysis guidance. She also guided the manuscript's conception, outline and wrote key components of the manuscript discussion. All authors reviewed and approved the final version of the manuscript before submission.

ACKNOWLEDGEMENTS

We wish to thank the following individuals and organizations for their contributions and support to this work: Marlyn Gill, Susan Nguyen and Colin Penman and other patient researchers from Pacer Innovates (pacerinnovates.ca), Dr. Nancy Marlett, Dr. Sheelagh Carpendale and her graduate students, Dr. Fateme Rajabiyazdi, Megan Crosby, Joseph Tropiano, Brad Haws and our patient and healthcare provider project participants. We could not have done this without you. This study has been supported by the Alberta Innovates Health Solutions (AIHS) Collaborative Research and Innovation Opportunities (CRIO) team grant (number 20130152).

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings as reported here are available upon request from the corresponding author, Julie Babione and Doreen Rabi. The data are not publicly available due to privacy restrictions, as the data may contain information that could compromise research participant privacy.

ETHICS STATEMENT

Ethical approval was obtained from the University of Calgary's Conjoint Health Research Ethics Board (REB#13-1081 and REB#14-0747).

DETAILS

Subject:	Patient-centered care; Communication; Chronic conditions; Patients; Demographics; Collaborative approach; Health care; Chronic illnesses; Content analysis; Audio recordings; Secondary analysis; Teams; Decision making; Health care industry; Medical education; Educational programs; Research methodology; Alignment; Co-design; Empowerment; Community involvement; Focus groups; Medical research; Primary care; Community participation; Health education; Patient participation; Group decision making; Qualitative research; Research projects; Patient communication

Identifier / keyword:	patient and public involvement; patient empowerment; patient experience; patient-centred care
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	419-428
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-12-03
Milestone dates:	2022-10-31 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-08-24 (manuscriptReceived); 2022-12-03 (publishedOnlineEarlyUnpaginated); 2022-11-08 (manuscriptAccepted)
Publication history :	
First posting date:	03 Dec 2022
DOI:	https://doi.org/10.1111/hex.13674
ProQuest document ID:	2767102268

Problem-based shared decision making: The role of canonical SDM steps

Montori, Victor M [1]
; Ruissen, Merel M [2] ; Branda, Megan E [3] ; Hargraves, Ian G [1] ; Kunneman, Marleen [4]
[1] Knowledge and Evaluation Research Unit, Mayo Clinic Rochester, Rochester, Minnesota, USA [2] Knowledge and Evaluation Research Unit, Mayo Clinic Rochester, Rochester, Minnesota, USA; Department of Medicine, Division of Endocrinology, Leiden University Medical Center, Leiden, Zuid-Holland, The Netherlands; Department of Biomedical Data Sciences, Section of Medical Decision Making, Leiden University Medical Center, Leiden, The Netherlands [3] Knowledge and Evaluation Research Unit, Mayo Clinic Rochester, Rochester, Minnesota, USA; Department of Quantitative Health Sciences, Division of Clinical Trials and Biostatistics, Mayo Clinic, Rochester, Minnesota, USA [4] Knowledge and Evaluation Research Unit, Mayo Clinic Rochester, Rochester, Minnesota, USA; Department of Biomedical Data Sciences, Section of Medical Decision Making, Leiden University Medical Center, Leiden, The Netherlands

ABSTRACT (ENGLISH)

Objective
To evaluate the extent to which the canonical steps of shared decision making (SDM) take place in clinical encounters in practice and across SDM forms.
Methods
We assessed 100 randomly selected video-recorded primary care encounters, obtained as part of a randomized trial of an SDM intervention in patients with type 2 diabetes. Two coders, working independently, noted each instance of SDM, classified it as one of four problem-based forms to SDM (weighing alternatives, negotiating conflicting issues, solving problems, or developing existential insight), and noted the occurrence and timing of each of the four canonical SDM steps: fostering choice awareness, providing information, stating preferences, and deciding. Descriptive analyses sought to determine the relative frequency of these steps across each of the four SDM forms within each encounter.
Results
There were 485 SDM steps noted (mean 4.85 steps per encounter), of which providing information and stating

preferences were the most common. There were 2.7 (38 steps in 14 encounters) steps per encounter observed in encounters with no discernible SDM form, 3.4 (105 steps in 31 encounters) with one SDM form, 5.2 (129 steps in 25 encounters) with two SDM forms, and 7.1 (213 steps in 30 encounters) when ≥3 SDM forms were observed within the encounter. The prescribed order of the four SDM steps was observed in, at best, 16 of the 100 encounters. Stating preferences was a common step when weighing alternatives (38%) or negotiating conflicts (59.3%) but less common when solving problems (29.2%). The distribution of SDM steps was similar to usual care with or without the SDM intervention.

Conclusion

The normative steps of SDM are infrequently observed in their prescribed order regardless of whether an SDM intervention was used. Some steps are more likely in some SDM forms but no pattern of steps appears to distinguish among SDM forms.

Clinical Trial Registration

ClinicalTrial.gov: NCT01293578.

FULL TEXT

INTRODUCTION

Clinical care requires noticing the problematic human situation of patients and responding with plans of care that fit. This has been defined as the work patients and clinicians do to iteratively develop a plan of care that is maximally responsive to this problematic situation, maximally supportive of patient goals, and minimally disruptive of each person's life and loves.[1] One process by which patients and clinicians work together to figure out what to do is called shared decision making (SDM). Guidelines and other policy instruments increasingly recommend and promote the use of SDM in clinical practice.[2,3]

Conventionally, SDM is framed as a decision-making process involving patients choosing between multiple acceptable treatment options.[4] Experts describe SDM as consisting of four consecutive steps: (1) fostering choice awareness, (2) providing information about the available options and their pros and cons, (3) deliberating about these options based on patient preferences, and (4) making a final decision.[5,6] This form of SDM is considered relatively rare in practice, its use is hampered by lack of time and other supportive resources (e.g., SDM tools), clinician's lack of ability or willingness, and other barriers.[7]

This canonical form of SDM, however, seems inappropriate as a tactic to address problems that require a method of making collaborative decisions other than weighing alternative options based on patient preferences. Recently, Hargraves and colleagues have proposed that the appropriate SDM method must purposefully match the kind of problematic situation patients and clinicians are facing.[8]

Recognizing a range of situations for which SDM is appropriate, purposeful SDM proposes four SDM forms, one for each kind of problematic situation: (1) weighing treatment alternatives, (2) negotiating intra-, or interpersonal conflicting issues, (3) problem solving and (4) developing existential insight.[8] After re-analysing a database of video recordings of clinical encounters between patients with diabetes and their clinician, Ruissen et al.[9] found that clinicians and patients frequently used SDM in practice, in 86 of 100 encounters, with the canonical SDM form of weighing treatment alternatives comprising only 33% of all purposeful SDM forms used.

After recognizing that SDM is common in the care of chronic patients and that a range of forms is used in practice, we sought to determine how often are the canonical steps of SDM seen in practice, appear in their normative order or at all within each of the forms of SDM observed. We hypothesized that the steps of SDM appear in the order prescribed when the canonical form of SDM is used (weighing treatment alternatives) but are less appropriate to describe other forms to SDM.

METHODS

We used the same data set developed for the study by Ruissen et al.[9] for this analysis. Briefly, M.M.R. used a random-number generator to randomly select 100 video-recorded encounters of the 350 encounters from both arms (without stratification by arm) of a multicenter clinical trial assessing the effect of a within-encounter SDM conversation aid (intervention) versus usual primary diabetes care for patients with type 2 diabetes in the United

States (ClinicalTrial.gov: NCT01293578).[10] The trial database was the source of patient and clinician characteristics and trial arm (usual care with or without SDM intervention) allocation.

The Mayo Clinic Institutional Review Board approved this secondary analysis before coding. Patients and clinicians provided written informed consent about the use of trial data and video recordings for research before the encounter.

Purposeful SDM provided the underpinning of the coding scheme to determine the form or forms of SDM used in an encounter.[8] When a form of SDM was identified, a distinction was made between SDM concerning (1) weighing treatment alternatives (canonical SDM), (2) negotiating intra-, or interpersonal issues, (3) problem solving or (4) developing existential insight. Only the start of the SDM process was coded, given the fact that a clear end of SDM can often not be distinguished. We then noted when the following conventional SDM steps appeared during the consultation: (1) fostering choice awareness, (2) providing information (including the pros/cons of available options), (3) expression of patient preference or desire, and (4) making a final decision.

We developed and refined a coding scheme based on 14 video-recorded encounters not included in our sample. Of the 100 included videos, 20 were used to train, and test the self-developed coding scheme. These videos and the other 80 recordings were coded using the final version of the coding scheme. All encounters were coded in duplicate by two investigators from different backgrounds (M.M.R., a medical doctor, and M.K., a clinical linguist and decision scientist). Disagreements were resolved by discussion and consensus.

Statistical analyses

We tested associations using the Kruskal–Wallis test for continuous variables and the χ^2 test statistic for categorical variables.

To visualize the distribution of purposeful forms and canonical steps within the encounters, we created a swimmer plot. Encounters were grouped into the plot by the number of forms present in each encounter (None, one, two, or three or more forms). The relative occurrence in time of each form noted or of each step identified is presented as the fraction of the encounter duration (i.e., from greeting to end of the visit indicated by the clinician and/or patient leaving the room or end of the recording) at which time the form or step started, expressed as a percentage of the encounter duration. Study data were collected and managed using REDCap electronic data capture tools, hosted at Mayo Clinic thanks to its Center for Clinical and Translational Science (funded by the National Institutes of Health—NCATS UL1TR002377).[11,12] Analyses were completed in SAS v9.4 (SAS, Inc.).

RESULTSParticipants

Table 1 describes the 100 patients (41% women, average age 60, 85% white) and 52 clinicians (28% women, average age 47) involved in the encounters included and coded. The average length of the clinical encounter was 17.0 min (range: 4.0–43.6 min).

Table 1 Participant characteristics

Patient characteristics	Patients (*n* = 100)
Encounter, usual care without/with SDM tool, *n*	31/69
Age (years), mean (SD)	60.0 (9.7)
Women, *n*	41
Body mass index, mean (SD)	36.7 (9.1)
Race, Black/White/other, *n*	9/85/6
Insurance, private/government/other, *n*	52/29/7

Education, high school or less, *n*	29
HbA1c, mean (SD)	8.9% (1.3)
Years in relationship with clinician, *n*	
<5	43
5 to <10	22
>10	25
Adequate health literacy, *n*a	81
Clinician characteristics	Clinicians (*n* = 52)
Age, years, mean (SD)	46.9 (11.2)
Women, *n* (%)	25 (48%)
Years in practice, mean (SD)	13.6 (10.5)
Number of encounters, mean (SD)	1.9 (1.3)
Median (IQR)	1 (1, 3)

Abbreviations: IQR, interquartile range; SDM, shared decision making. a

Based on 'never' or 'rarely' answers to the Single Item Literacy Screener ('How often do you need to have someone help you when you read instructions, pamphlets, or other written material from your doctor or pharmacy?').[18]

Purposeful forms and canonical steps of SDM

One or more SDM forms could be identified in 86 of 100 encounters. A single SDM form was evident in 31 encounters, 2 forms in 25, and 3 or more SDM forms in 30 encounters. Situations in which treatment alternatives were weighed accounted for 33% of the SDM forms used during the consultation, compared with 30% in which negotiating intra- or interpersonal conflicting issues was used, and 36% in which a problem-solving form was used. Developing existential insight accounted for 1% of the observed SDM forms.

Table 2 describes the distribution of SDM steps within the encounters. In these 100 encounters, we observed 485 steps or an average of 4.85 steps per encounter. In encounters with no discernible purposeful SDM form, we observed 2.7 (38 steps in 14 encounters) steps per encounter. In encounters with one SDM form, we observed 3.4 (105 steps in 31 encounters) steps per encounter. We observed 5.2 (129 steps in 25 encounters) steps per encounter in encounters with two SDM forms and 7.1 (213 steps in 30 encounters) steps per encounter in encounters with ≥3 SDM forms observed within the encounter. The most common steps were 'giving statements of preference or desire' during deliberations and 'providing information'; both steps were present in about a third of encounters with one or more purposeful SDM forms. 'Choice awareness' and 'deciding' were evident in a fifth of purposeful SDM forms.

Table 2 Distribution of shared decision-making (SDM) steps and forms within encounters

	Encounters by number of SDM forms observed				
	None (*n* = 14)	One (*n* = 31)	Two (*n* = 25)	≥3 (*n* = 30)	**All encounters (*n* = 100)**
Steps, *n* (%)					
SDM steps observeda	38b	105	129	213	485
Choice awareness	8 (21.1)	19 (18.1)	22 (17.1)	40 (18.8)	89 (18.4)
Providing information	13 (34.2)	32 (30.5)	39 (30.2)	53 (24.9)	137 (28.2)
Deliberating with statement of preferences	6 (15.8)	32 (30.5)	46 (35.7)	93 (43.7)	177 (36.5)
Deciding	11 (28.9)	22 (21.0)	22 (17.1)	27 (12.7)	82 (16.9)
Encounters with SDM steps in order, *n* (%)c	0 (0)	3 (9.7)	5 (20.0)	8 (26.7)	16 (16.0)

a

χ^2 test, $p = .048$.

b

Although no purposeful SDM was observed in these encounters, SDM steps were seen but without contributing to collaborative decision making to address the patient's problematic situation.

c

Fisher's exact test, $p < .001$.

When purposeful SDM was not evident, ‘giving statements of patient preference or desire’ during deliberation was less common (15.8% vs. 30.5%–43.7% when a form of purposeful SDM was observed) and ‘deciding’ (28.9% vs. 12.7%–21% when a form of purposeful SDM was observed) was more common.

SDM steps appeared in the canonical order (i.e., starting with fostering choice awareness and finishing with making a final decision) in 18 encounters. In 16 of these encounters, these sets of ordered steps were preceded or followed by other steps (Table 2). The distribution of steps within forms was similar whether the encounter was allocated to usual care with or without the SDM intervention (Appendix A).

Table 3 shows the distribution of SDM steps within each of the four forms to purposeful SDM. ‘Stating preferences’ was a common step when participants engaged in SDM by weighing treatment alternatives (38%) or negotiating intra-interpersonal conflicts (59.3%), but less common when they worked on solving problems (29.2%) or developing an existential insight (27.3%). Appendix B shows that allocation to the SDM intervention did not affect the frequency of steps observed in total or within each SDM form. Similarly, our post hoc exploration of the duration of the care relationship (<5 vs. ≥5 years) did not affect the results (data not shown).

Table 3 Distribution of shared decision-making (SDM) steps by the form of SDM in which they were observed

	SDM forma				
	Weighing alternatives	Negotiating conflict	Solving problems	Developing insight	Totalb
Steps, *n* (%)					
SDM steps observed	137	108	120	11	376
Choice awareness	24 (17.5)	10 (9.3)	22 (18.3)	4 (36.4)	60 (16)
Providing information	34 (24.8)	18 (16.7)	39 (32.5)	3 (27.3)	94 (25)
Deliberating with statement of preferences	52 (38)	64 (59.3)	35 (29.2)	3 (27.3)	154 (41)
Deciding	27 (19.7)	16 (14.8)	24 (20)	1 (9.1)	68 (18.1)

a

χ^2 *p* value = .0011.

b

Data limited to encounters in which a step followed the onset of an SDM form (i.e., 83 of the 86 encounters in which an SDM form was observed).

Figure 1 describes the steps observed within SDM forms presented by whether purposeful SDM was either not observed or when 1, 2 or 3 or more forms were observed.

Figure 1. Occurrence of shared decision-making steps and forms within encounters grouped by the number of SDM forms observed per encounter. (A) Encounters in which no shared decision-making form was observed (n = 14). (B) Encounters in which one form was observed (n = 31). (C) Encounters in which two forms were observed (n = 25). (D) Encounters in which three or more forms were observed (n = 30). Each row represents an encounter, with its duration represented on a 100% scale.

DISCUSSION

In this set of 100 clinical encounters obtained from a practice-based randomized trial of usual diabetes care with or without an SDM tool, in which two-thirds of patients with diabetes and their primary care clinicians used an SDM tool, we found that patients and clinicians engaged in SDM without necessarily completing the canonical SDM steps or following them in their prescribed order. We found that the canonical steps of SDM were present when no specific purposeful SDM form was identified. These steps also were commonly present when one or more purposeful SDM forms were used (of which the canonical form of SDM represented about a third), were similarly present regardless of which SDM form was used, and were present in the normative order in, at best, 16% of encounters. In 70% of encounters, clinicians and patients took different SDM steps as they entered and switched across different forms to SDM. These results suggest that, even under stimulated conditions of adding an SDM intervention, clinicians and patients infrequently follow the normative order of SDM steps to make decisions with patients in practice.

Along with the report by Ruissen et al.,[9] which found that almost 90% of these encounters demonstrated some form of SDM (with the canonical form representing about a third of the observed instances), this report documents the relative frequency of SDM steps in these encounters and the timing of their appearance within each encounter. The results are not directly comparable to other studies in which the frequency of steps has been analysed as if each encounter had only one form of SDM. Kunneman et al., for example, documented that choice awareness appeared

in 53% of clinical encounters drawn from a similar sample of video-recorded encounters within clinical trials of SDM tools.[13]

The results call into question SDM measurement forms that rely on the presence of SDM steps to determine the occurrence or quality of SDM.[14–16] SDM steps occurred, in one instance in the normative order, even when no purposeful SDM form was evident. The most assessed step of SDM, providing information,[15] appears in less than a third of instances of SDM.

These results, while novel, have limitations. Video recordings were randomly drawn from a set of encounters produced during the experimental evaluation of the use of an SDM intervention. The presence of the conversation aid, the video recorder, or of the randomized trial procedures may have affected the observations reported herein. We intuit that the direction of effect of these factors would have been to normalize the encounters to what is expected (i.e., a higher prevalence of the canonical form of SDM with the steps in the expected order). That, despite these factors, we found high variability in the range of purposeful SDM forms and canonical steps may thus represent a best-case scenario. These findings must be evaluated in independent data sets by other research groups. On the other hand, the carefully developed yet ad-hoc coding scheme based in part on purposeful SDM and its use by a clinician and an expert in SDM on actual clinical encounters across multiple primary care practices represent the strengths of this investigation.

These results, particularly the patterns observed in Figure 1, suggest a highly variable approach to SDM in primary care practice. This variability could be an indication of poor participant skill, or that the SDM intervention, present in two-thirds of visits, provided insufficient support in structuring the encounter. Alternatively, this variability could represent the natural process of trial-and-error, of uncovering how might a problem be addressed, that patients and clinicians use during consultations.

The most common depiction of SDM, by Charles et al.,[6] refers to stages (information exchange, deliberation, decision making) in which each one leads to the next. The Three Talk Model by Elwyn et al.[17] suggests, instead, a cyclical process by which patient and clinician move along the steps of SDM, a process that may very well describe the observations here, particularly those within the canonical form of SDM (weighing alternatives). Both models assume that a problem is defined at the start of the process and that the exchange focuses on how to solve it. Conversely, a major advantage of the purposeful SDM framework is the recognition that the nature of the problem and of how to respond to it can emerge from the joint effort of clinician and patient.[8] This view matches better with the observations reported here of multiple forms to SDM and multiple steps taken as the patient and clinician talk, think, and feel their way through the uncertain and problematic human situation of the patient. The variability observed may in fact suggest flexibility in the use of clinical skills within a participatory and empathic collaboration. This possibility may need to be explored using content analysis of the encounters.

These findings, if confirmed, would give credence to the purposeful SDM model and challenge ways of training, measuring, and assessing for SDM that rely on (a) a single canonical form of SDM, and (b) a set order of steps to do SDM well. This challenge may lead to new SDM tools designed to create the conditions for flexible collaboration, supporting whichever form appears more conducive to addressing the problematic situation of the patient.

Our findings may also challenge the notion that the key problem SDM addresses is patient participation when it seems as if both patient and clinician must take part in determining together what the problem is and how to address it in an iterative and, to the outside observer, somewhat chaotic process of exploration, discovery, and experimentation.

Finally, our findings challenge existing measures of the occurrence and quality of SDM that rely on detecting only one form of SDM and one set of steps.[14] Indeed, when clinicians say ‘but I do SDM already’ they may be referring to the processes depicted here, which depart in important ways from what has counted as SDM hitherto.

In conclusion, we found that the canonical steps of SDM are infrequently observed in their normative order in usual clinical practice (as observed in a practice-based randomized trial of adding or not an SDM intervention), regardless of whether an SDM tool was used. These steps do not appear more likely to follow a particular order when one or more SDM forms are used within a clinical encounter. The most common steps are for patients to state their

preferences or desires during deliberation and for clinicians to share information. These observations should be considered when developing new measures of SDM and interventions—for example, training and tools—to promote its optimal and purposeful use as a method of care in practice.

AUTHOR CONTRIBUTIONS

Victor M. Montori, is the principal investigator at the KER Unit, the research entity within Mayo Clinic that conducted the original trial from which the videos for this secondary analysis came and that maintains the database with such videos. He wrote the first draft of the manuscript with Merel M. Ruissen and coordinated revisions and final submission. Merel M. Ruissen worked with Marleen Kunneman under her supervision to develop the study protocol and the video coding scheme. She conducted with Marleen Kunneman the data collection, and video analysis, and contributed to the data analyses reported here. She worked with Victor M. Montori on the first draft of this manuscript, provided critical revisions and approved the final manuscript. Megan E. Branda is the study statistician and contributes to maintaining the database of video recordings and clinical data from KER Unit trials. She worked with the co-authors in designing and conducting the data analyses and produced the figure and tables that accompany this manuscript. She also provided critical revisions and approved the final manuscript. Ian G. Hargraves is the proponent of the purposeful shared decision-making framework that guides this work. He worked with Merel M. Ruissen and Marleen Kunneman providing feedback on the video coding scheme and on analysing the results. Ian G. Hargraves provided critical revisions to the manuscript and approved the final submission. Marleen Kunneman worked with and provided direct supervision to Merel M. Ruissen in the development of the study protocol, coding scheme, video analyses, and data analyses and interpretation. She provided critical revisions to the manuscript and approved the final version. She is the corresponding author and data warrantor.

ACKNOWLEDGEMENTS

M.K. has received funding from Nederlandse Organisatie voor Wetenschappelijk Onderzoek (Personal Veni award) and ZonMw (Award #016.196.138). No other funding was received by the authors for this project.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data extracted from video recordings that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

The Mayo Clinic Institutional Review Board approved this secondary analysis before coding. Patients and clinicians provided written informed consent about the use of trial data and video recordings for research before the encounter.

AAPPENDIX

See Table A1

Table A1 Table of shared decision-making (SDM) steps observed classified by (a) whether the encounter was allocated to usual care with or without the use of an SDM intervention, and (b) number of SDM forms observed.

	SDM forms observed per encounter					
Steps, *n* (%)	No SDM form	One form	Two forms	Three or more forms	All	*p* Value
SDM intervention	*N* = 23	*N* = 64	*N* = 89	*N* = 172	*N* = 348	.040 a

Choice awareness	4 (17.4)	10 (15.6)	14 (15.7)	33 (19.2)	61 (17.5)	
Providing information	8 (34.8)	20 (31.3)	28 (31.5)	42 (24.4)	98 (28.2)	
Deliberating with statement of preferences	3 (13.0)	19 (29.7)	30 (33.7)	76 (44.2)	128 (36.8)	
Deciding	8 (34.8)	15 (23.4)	17 (19.1)	21 (12.2)	61 (17.5)	
Usual care	*N* = 15	*N* = 41	*N* = 40	*N* = 41	*N* = 137	.96a
Choice awareness	4 (26.7)	9 (22.0)	8 (20.0)	7 (17.1)	28 (20.4)	
Providing information	5 (33.3)	12 (29.3)	11 (27.5)	11 (26.8)	39 (28.5)	
Deliberating with statement of preferences	3 (20.0)	13 (31.7)	16 (40.0)	17 (41.5)	49 (35.8)	
Deciding	3 (20.0)	7 (17.1)	5 (12.5)	6 (14.6)	21 (15.3)	

a
χ^2 *p* value.

B**APPENDIX**

See Table B1

Table B1 of shared decision-making (SDM) steps observed classified by (a) whether the encounter was allocated to usual care with or without the use of an SDM intervention, and (b) by the SDM form within which the step was observed.

	SDM forma				
SDM steps within a form, *n* (%)	Weighing alternatives	Negotiating conflict	Solving problems	Developing insight	Total
SDM interventiona	*N* = 106	*N* = 69	*N* = 88	*N* = 8	*N* = 271
Choice awareness	18 (17.0)	5 (7.2)	14 (15.9)	2 (25.0)	39 (14.4)
Providing information	23 (21.7)	11 (15.9)	28 (31.8)	2 (25.0)	64 (23.6)

Deliberating with statement of preferences	43 (40.6)	42 (60.9)	28 (31.8)	3 (37.5)	116 (42.8)
Deciding	22 (20.8)	11 (15.9)	18 (20.5)	1 (12.5)	52 (19.2)
*Usual care*b	*N* = 31	*N* = 39	*N* = 32	*N* = 3	*N* = 105
Choice awareness	6 (19.4)	5 (12.8)	8 (25.0)	2 (66.7)	21 (20.0)
Providing information	11 (35.5)	7 (17.9)	11 (34.4)	1 (33.3)	30 (28.6)
Deliberating with statement of preferences	9 (29.0)	22 (56.4)	7 (21.9)	0 (0.0)	38 (36.2)
Deciding	5 (16.1)	5 (12.8)	6 (18.8)	0 (0.0)	16 (15.2)

a
χ^2 *p* value = .052.
b
χ^2 *p* value = .072.

DETAILS

Subject:	Problem solving; Diabetes mellitus (non-insulin dependent); Intervention; Bargaining; Diabetes; Video recordings; Clinical research; Type 2 diabetes mellitus; Weighing; Primary care; Decision making; Clinical trials; Coders; Patients; Health care; Diabetes mellitus; Alternatives; Preferences; Timing; Group decision making; Clinical medicine
Company / organization:	Name: Mayo Clinic; NAICS: 622110
Identifier / keyword:	problem-based; purposeful; SDM; shared decision making; steps
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	282-289
Publication year:	2023
Publication date:	Feb 2023

Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-29
Milestone dates:	2022-10-07 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-08-05 (manuscriptReceived); 2022-11-29 (publishedOnlineEarlyUnpaginated); 2022-10-25 (manuscriptAccepted)
Publication history :	
First posting date:	29 Nov 2022
DOI:	https://doi.org/10.1111/hex.13654
ProQuest document ID:	2767102267
Document URL:	https://www.proquest.com/scholarly-journals/problem-based-shared-decision-making-role/docview/2767102267/se-2?accountid=211160
Copyright:	
Last updated:	2023-06-21
Database:	Public Health Database,Publicly Available Content Database

Exploring first-time mothers' experiences and knowledge about behavioural risk factors for stillbirth

Sánchez, Tamara Escañuela [1]
; Matvienko-Sikar, Karen [2] ; Meaney, Sarah [3] ; O'Donoghue, Keelin [1] [1] Pregnancy Loss Research Group, Department of Obstetrics and Gynaecology, Cork University Maternity Hospital, University College Cork, Cork, Ireland; Infant Centre, University College Cork, Cork, Ireland [2] School of Public Health, University College Cork, Cork, Ireland [3] Pregnancy Loss Research Group, Department of Obstetrics and Gynaecology, Cork University Maternity Hospital, University College Cork, Cork, Ireland; National Perinatal Epidemiology Centre (NPEC), University College Cork, Cork, Ireland

ABSTRACT (ENGLISH)

Background
Modifiable factors such as substance use, lack of attendance at antenatal care, overweight or obesity and sleeping position are associated with a higher risk of stillbirth. This qualitative study aimed to explore women's experiences of modifiable factors during pregnancy and their awareness of stillbirth.
Methods
Purposive sampling was implemented by hospital staff in a large tertiary maternity hospital in Ireland between November 2020 and March 2021. Women were approached during their stay in the hospital and were invited to participate in a semistructured interview 3–5 months later. Eligible women were primiparous, >18 years of age and had an uncomplicated pregnancy and delivery. Eighteen women who consented to be followed up were interviewed at 3–5 months postpartum. Thematic analysis was used to analyse the data.
Results
Four themes were identified: attitudes towards behaviour change, awareness regarding stillbirth and risk factors, the silence around stillbirth and risks, and attitudes towards receiving information about stillbirth. Women spoke about behaviour change in terms of outcomes, and most changes (e.g., ceasing alcohol consumption) were perceived as easy to manage. Awareness of stillbirth was limited among the women interviewed, and the association between risk behaviours and stillbirth was not known by any woman. Results suggest that there is a silence around stillbirth, including in antenatal care, which hinders information provision. However, most women highlighted the value of receiving information and extra education about modifiable risk factors and stillbirth.
Conclusion
There is a general lack of understanding of the link between behavioural risk factors and potential pregnancy outcomes such as stillbirth. Providing further information to women about stillbirth and providing additional support with behaviour change might contribute to enhancing preventive efforts.
Patient or Public Contribution
Patients were involved in this study by providing their experiences of antenatal care which were used as primary data.

FULL TEXT

INTRODUCTION

Stillbirth is one of the worst outcomes pregnant women and their families can experience.[1,2] Worldwide, the estimated average rate of stillbirth (defined as a baby being born with no signs of life at 28 weeks gestation or more) in 49 high-income countries is 3.5 per 1000 total births, with rates varying from 1.3 (Iceland) to 8.8 (Ukraine)

depending on the country.[3] In Ireland, according to the latest report published by the National Perinatal Epidemiology Centre, the stillbirth rate was 4.06 per 1000 births in 2019, which reflected a slight increase compared with 2018 data.[4] The overall perinatal mortality rate has remained flat in Ireland for several years, as opposed to the decrease observed in the decade before 2012.[4] Although the rates of stillbirth in high-income countries have reduced in the last 20 years, the variation between rates in high-income countries shows that a further reduction of stillbirth incidence is possible and needed.[5]

Previous research has associated different types of risk factors with an increased risk of stillbirth, including medical,[6–8] behavioural factors[9–11] and sociodemographic factors.[12–15] Some of these risk factors are potentially modifiable and addressing them could contribute to reducing the rates of stillbirth.[16,17] Previous research has associated substance use including smoking, alcohol use and illicit drug use with an increased risk of stillbirth. Flenady et al.[18] and Marufu et al.[19] conducted two separate meta-analyses examining the influence of smoking on the risk of stillbirth, and both concluded that smoking was associated with a 36% and 47% increase, respectively, in the risk of stillbirth.[18,19] Regarding alcohol consumption, Aliyu et al.[10] concluded that mothers who consumed alcohol while pregnant were 40% more likely to experience stillbirth as compared with nondrinking mothers. Additionally, a recently published study exploring the risk of stillbirth in women who consumed alcohol and smoked in the antenatal period concluded that the adjusted relative risk for all stillbirths was 1.75 (98.3% confidence interval [CI], 0.96–3.18) for dual exposure, 1.26 (98.3% CI, 0.58–2.74) for drinking only and 1.27 (98.3% CI, 0.69–2.35) for smoking only compared with the reference group.[20] Being overweight and obese has also been associated with an increased risk of stillbirth. A recent study exploring prognostic variables for stillbirth found that the most frequently reported maternal characteristic associated with stillbirth was high BMI and other measures of obesity, and these findings were supported by highly convincing evidence in most of the studies included in their review.[21] Engagement with and attendance at antenatal care has also been linked to the risk of stillbirth. Stacey et al.[22] concluded in their study that attending less than 50% of the recommended visits was associated with an almost three times increased risk of stillbirth, and this risk increased as the number of visits attended decreased. Last, women's sleeping habits have also been studied in association with stillbirth risk and research has specifically focussed on the maternal sleep position. Cronin et al.[23] conducted a meta-analysis which concluded that the supine going-to-sleep position is independently associated with late stillbirth.

Even though there is evidence in the literature associating these modifiable risk factors with stillbirth,[16] information about the role of these behaviours in stillbirth is not widely available amongst the public in Ireland, as demonstrated by Nuzum et al.[24] in their survey study. These authors, after surveying 999 members of the Irish population, reported that there is a lack of public awareness of the incidence, causes and risk factors for stillbirth, although over 50% of respondents personally knew someone who had experienced a stillbirth.[24] These findings are similar to those obtained in a survey study conducted among members of the American College of Obstetrics and Gynaecology.[25] The authors concluded that knowledge regarding epidemiology, risk factors and effective interventions to reduce stillbirth amongst participants was only fair. In the same study, only 30% of respondents were aware that preeclampsia, advanced maternal age, elevated α-fetoprotein, multiple gestations, cigarette smoking, illicit drug use and being postterm increased risk.[25] Regarding knowledge of behavioural risk factors, previous studies have also demonstrated that some women held serious misconceptions about the risks of substance use or engaging in weight management behaviours, as well as having limited reproductive knowledge, which interferes with their care-seeking behaviours and increases their risk of stillbirth.[26–28]

Previous research has also shown that stillbirth is still a taboo subject in society.[29] Hence, further efforts to increase women's awareness about stillbirth and its risk factors through routine antenatal care and public health campaigns are necessary to support stillbirth prevention efforts.

Involving patients' experiences in designing and developing public health campaigns and behaviour change interventions is vital to ensure that they are tailored to users' needs. Previous research has established that women engage in behaviour change during pregnancy due to different motivations, but the volume of expectations placed on them and the complexities of those changes are rarely acknowledged.[30] Understanding women's experiences of

behaviour change and information provision during antenatal care through their perspective is essential to inform the development of such interventions and public health campaigns. The main objective of this study was to explore women's experiences of modifiable factors during pregnancy and knowledge and beliefs regarding behavioural risk factors related to stillbirth. Additionally, we aimed to examine women's experiences, if any, of being informed about such risk factors during their antenatal care.

METHODS

To enhance the reporting of this study, the Standards for Reporting Qualitative Research (SRQR) checklist has been used[31] (see Supporting Information: File 1).

Design

A qualitative semistructured interview study was conducted using a Reflexive Thematic Analysis approach (see Supporting Information: File 2). Qualitative research allows the researcher to explore and understand social phenomena and psychological concepts such as experiences, beliefs, motivations and attitudes.[32] For this study, a constructivist paradigm was utilized.

Constructivism is based on the idea that knowledge is built from an interaction between the subject and reality, and hence, the same situation experienced by different people might result in different interpretations, depending on each individual's perception of such a situation.[32] This epistemological approach refuses the idea that there is only one truth but instead supports the importance of taking into consideration the subjective meaning given to a phenomenon and the understanding of the context of the same.[32] In constructivism, the researcher and the study participants are equally involved in the process to generate knowledge, hence, the researcher has an active role in the research processes by creating—rather than revealing—something which has to make sense within existing frameworks of meaning.[32]

This paradigm allowed us to understand the complexity of our participants' lived experiences from their point of view.

Recruitment

Women were recruited using a purposive sampling approach implemented by hospital staff in Cork University Maternity Hospital (CUMH), Ireland. Inclusion criteria included primiparous women, with a low-risk pregnancy and uncomplicated delivery, who had a healthy baby and were 18 years old or older and consented to participate in a one-to-one interview 3–5 months postpartum. The follow-up timeframe of 3–5 months postpartum for interviews was chosen to facilitate women adapting to their new routine with their babies before taking up their time to participate in the interviews.

Exclusion criteria included multiparous women, women currently pregnant, women with a history of pregnancy loss, women attending high-risk antenatal clinics, congenital anomalies, cases of maternal morbidity and cases of major complications during pregnancy or birth for both woman and baby. Given the aims of our study, it was decided that obtaining the experiences of primiparous women with uncomplicated pregnancies and deliveries would reflect the common practices regarding information provision and knowledge acquisition during antenatal care. It was assumed that including women with additional care needs during their pregnancy or delivery would have resulted in a biased picture of the normal day-to-day practices of the antenatal care services, as these women require additional education and are exposed to different types of healthcare professionals and levels of care.

Women were invited to participate in the study during their stay in the maternity hospital after delivering their babies from November 2020 to March 2021. If eligible, an obstetrician affiliated with our research group explained the study to the woman and provided them with the patient information leaflet and consent form. After providing women time to read the information and ask questions, their written consent was obtained by the obstetrician. The woman's contact details were obtained at this first encounter, and women were advised that a researcher would contact them 3–5 months later to participate in a one-to-one interview.

Setting and sample

CUMH is 1 of 19 maternity units in the Republic of Ireland and is situated within the south/southwest hospital group. In 2020, 7040 babies were delivered in the hospital of which 42.2% were born to first-time mothers.[33] Forty-four women consented to be followed up 3–5 months postpartum in the current study. One researcher (T. E. S.)

contacted all women who agreed to participate in the study between 3 and 5 months postpartum by phone or e-mail. Of the 44 women, 4 subsequently declined to participate and 22 were unreachable or did not attend the scheduled online interviews. Eighteen women subsequently participated in the individual semistructured online interviews between February and July 2021.

In Ireland, even though access to maternity services is free, the option of choosing private or semiprivate care also exists. All women participating in this study gave birth in CUMH. Some participants used the service as public patients, meaning all their antenatal care was provided by CUMH staff; other women used the service as private patients, meaning that their antenatal care was mostly provided by a specific clinician's private practice staff.

Data collection

Women were interviewed about their experiences of behaviour change on modifiable factors during pregnancy and information provision regarding stillbirth and modifiable risk factors for stillbirth during their antenatal care between February and July 2021. Online one-to-one interviews using Google Meet were used as the data collection method due to the restrictions imposed on in-person data collection during the COVID-19 pandemic. The women participating in the study were sent a link to a scheduled online Google Meet at the date and time of their choice, with instructions on how to access the online meeting. The researcher's contact details were also provided to be used in case of technical difficulties.

All online one-to-one interviews were conducted by T. E. S. The semistructured interviews were guided by a pre-agreed topic guide developed by the multidisciplinary team and informed by previous work of the research group.[26–28] The topic guide included the following areas: history and health habits, awareness of risk factors for stillbirth, feelings and opinions about receiving education on stillbirth, information sources and interventions (see Supporting Information: File 2). Some of the questions in the topic guide were designed to elicit information about specific topics such as knowledge about stillbirth and risk factors associated with stillbirth. The use of semistructured interviews allowed the women to introduce or discuss topics that were not strictly predefined in the interview topic guide. As the interviewing process progressed reaching the 14th and 15th interviews, it was clear that the depth and quality of the interviews indicated that it was not necessary to continue actively pursuing the nonresponders to the invitation following their initial consent. It was then considered that enough information power was achieved. The concept of information power is proposed by Braun and Clarke as an alternative to the concept of data saturation which they deem incompatible with Reflexive Thematic Analysis. The concept of information power relies on the fact that rich, relevant data requires fewer participants,[34,35] hence the more relevant information a sample holds, the fewer participants are needed.[35] However, all 18 women for whom an interview was scheduled were interviewed to respect the commitment that had been made with them.

Interviews lasted between 30 and 50 min, were recorded, transcribed verbatim and imported into NVIVO 12 for analysis.

Data analysis

The data analysis conducted in this study is based on the principles of Reflexive Thematic Analysis as described by Braun and Clarke.[32,36] Reflexive Thematic Analysis is a flexible method that is suited to experiential and critical framings of language, data and meaning, and it can be used in either a deductive or inductive way. Reflexive Thematic Analysis involves an interpretative reflexive process, coding does not follow a framework and themes are the outcome of the analytical process.[37] We conceptualized reflexive thematic analysis for this paper within a constructivist approach (see Section 2.1).

The different phases of thematic analysis described by Braun and Clarke are as follows: (1) data familiarization and writing familiarization notes; (2) systematic data coding; (3) generating initial themes from coded and collated data; (4) developing and reviewing themes; (5) refining, defining and naming themes and (6) writing the report.[32] The analytical process in the current study began by transcribing the audio of the recorded interviews (data familiarization), to facilitate this process, Tactiq was used during the interviews, which is an automatic transcription tool. One researcher (T. E. S) read and reread all of the interview transcripts. Inductive open coding was then initiated which facilitated the identification of units of meaning that related to the research aims. Subsequently, those

codes were categorized and grouped into themes and relabelled where appropriate. A record of the evolvement of the themes and the category names was always kept. Further analysis allowed the researchers (T. E. S. and K. M.-S.) to group the different categories into themes, by refining their meaning to portray the story the data tells. A second author (K. M.-S.) reviewed and followed the coding process at all stages, and discussions were held as necessary.

Ethical considerations

The clinical team who obtained consent from women to participate in the study ensured that the sensitive nature of the interview topic was discussed to anticipate potential distressing factors for the women. Women were informed about their rights to withdraw from the study at any stage without any potential impact on their care or any other type of consequences. We considered the potential for women to experience distress while participating in the interviews and so planned accordingly to provide support to women who experienced distress.

This involved ensuring that the researcher would remain in the online session with the woman after the interview and until she was happy to end the session. The researcher would then make a follow-up contact if deemed necessary or contact the appropriate required support (e.g., referral to specialist bereavement midwives or perinatal mental health services). However, none of the participating women reported distress or were observed to experience distress and so this support was not provided to any participants.

Reflexivity statement

The research team that conducted this study includes experts in health psychology, epidemiology, behavioural science, public health and obstetrics and maternal–fetal medicine. All interviews were conducted by T. E. S., who is a female PhD student with a background in health psychology. T. E. S. received training in qualitative methods and interview techniques before conducting this study.

All three remaining authors are highly experienced researchers who have been involved in multiple qualitative studies.

None of the authors had a previous relationship with any of the participants, and the participant only had contact with the researcher conducting the interviews (T. E. S.).

The participants were informed that the interviewer was a PhD student and the overall objectives of the PhD project were exposed to them before commencing the interviews.

All members of the research team interacted throughout all phases of this study, which enhanced the process by providing points of view from different disciplines.

RESULTSSample characteristics

The final sample of 18 women included 16 White Irish women, 1 White Eastern European woman and 1 White North American woman. Woman's ages ranged from 28 to 37 years old. All women were married or cohabiting with their partners (see Table 1).

Table 1 Women's characteristics

Women number	Age	Insurance status	Occupation	Relationship status
W1	31–35	Public	Management	Married
W2	25–30	Private	Healthcare	Married
W3	>35	Public	Education	Cohabiting
W4	>35	Private	Social Care	Married
W5	>35	Public	Retail	Married

W6	31–35	Private	Healthcare	Married
W7	31–35	Public	Social Care	Cohabiting
W8	31–35	Public	Management	Married
W9	31–35	Public	Management	Married
W10	31–35	Private	Management	Married
W11	>35	Private	Education	Married
W12	>35	Public	Hospitality	Cohabiting
W13	25–30	Public	Education	Cohabiting
W14	25–30	Public	Healthcare	Cohabiting
W15	31–35	Public	Healthcare	Married
W16	31–35	Public	Healthcare	Married
W17	25–30	Private	Engineering	Married
W18	31–35	Public	Management	Married

Findings

Several themes and subthemes were identified through the analytic process (see Table 2).

Table 2 Themes overview

Themes	Subthemes
Attitudes towards behaviour change	
Awareness regarding stillbirth and risk factors	Awareness regarding health advice
	Limited awareness about stillbirth
Silence around stillbirth and risks	Lack of discussion regarding stillbirth and risk factors
	Reliance on own information-seeking behaviours
Attitudes towards receiving information about stillbirth	‘Knowledge is key’
	Stillbirth perceived as a difficult topic

	Importance of language and preference for information provision

Theme 1: Attitudes towards behaviour change

All women expressed having made changes in their behaviours because of their pregnancies. In some instances, these changes started preconceptually in preparation for pregnancy and were maintained throughout the pregnancy. The preconception preparatory behaviours adopted by women involved attending their general practitioner (GP) for advice, adopting healthier eating behaviours and having a more active life, monitoring menstrual cycles, preconceptual alcohol abstinence, preconceptual intake of folic acid and antenatal vitamins, as well as prepregnancy weight loss. Women spoke about the changes they made as soon as they learnt they were pregnant, the most common behaviours being ceasing alcohol consumption, avoiding foods not recommended during pregnancy, taking vitamins and folic acid, staying active, moderating the intensity of physical activity, having a nutritious diet and/or increasing rest.

Women discussed newly adopting some of these behaviours to achieve their best health status (e.g., being as fit and eating as healthily as possible, taking folic acid), and also abandoning some previous behaviours to prevent illness and adverse outcomes (e.g., avoiding consumption of alcohol, avoid stress, quitting smoking, etc.). These results show that women perceive behaviour change during pregnancy to have a dual nature in that it can help to improve positive outcomes and reduce the likelihood of adverse outcomes.

I just took vitamins and I did go to the doctor, alright? For advice, because I was on the pill for so long and I was a bit worried (W11)

I used to drink, alcohol, at weekends, but I stopped. And I obviously stayed off as well while I was pregnant. (W6)

I was exercising intermittently and I was kind of prioritizing work and I just said ‘no I'm going to prioritize myself a little bit more’ so I made it my purpose to just do swims and run more often, that was it. (W15)

Women discussed their different experiences when engaging in behaviour change during pregnancy. All women in this study expressed a strong and clear opinion about the need to stop consuming alcohol during pregnancy. However, some women decided to stop consuming alcohol as soon as they started planning the pregnancy, while others waited until they had confirmation of pregnancy. Regarding smoking and illicit substance use, women also expressed negative attitudes. Only one of the women was a smoker before her pregnancy, and she quit as soon as she learnt about her pregnancy. Physical activity was perceived as beneficial to achieve an appropriate fitness level and general health status, rather than a tool to manage weight gain. Some women expressed difficulties keeping up with their levels of exercise through their pregnancy, which led them to slowly disengage from these behaviours in response to perceived challenges associated with pregnancy (e.g., lack of energy, nausea). On the other hand, the adopted changes in diet were mostly focused on avoiding foodborne diseases, with some women starting to relax their attitude towards their food consumption as the pregnancy progressed.

I think it was less about weight management and more about just fitness, or like wellbeing or health or whatever, more cardio and all that more so than the weight management thing. (W17)

Women had the perception that they had been able to continue with their normal life during their pregnancy with little interference from their pregnancies. Most of the women perceived their behaviour changes (e.g., stopping alcohol consumption, avoiding dangerous foods, engaging in low-demand physical activity) as easy to manage and natural because they were doing these changes for their babies. However, one woman expressed after the interview that actively thinking about all of the changes made during her pregnancy helped her realize that she had made more changes than she had previously perceived. Hence, it seems that the fact that women perceived all their behaviour changes as natural might have contributed to the fact that they were less consciously aware of the range of different changes they did engage in. Women also discussed behaviour change during pregnancy in terms of consequences for the health of their babies or their own health which acted as their drive to change their behaviour. As an example, several women spoke about exercise as useful to facilitate labour in general.

I thought that the having no drinks would be harder than it was because I'd never been one to be able to go months

and months without drinking. But it was actually easier than I thought because I was doing it for my baby. (W18)
Yes, I had the day sometimes that I really wanted to smoke but, you know, because I was waiting so long for that baby. I was like, ‘no, I'm not gonna put her in any risk no matter what’. (W12)
it's all actually just coming back now talking. I'm like, ‘no, I didn't really change much’. Yes, I actually did. (W5)

Theme 2: Awareness regarding stillbirth and risk factorsAwareness regarding health advice

Most women in this study were aware of the importance of maintaining a healthy diet and adequate levels of physical activity during their pregnancies, with little concern about the safety of the physical activity. Women were also aware of the relevance of antenatal vitamins and prepregnancy supplements, especially folic acid.
I just always knew that if I did want to fall pregnant that I should be on folic acid. I really started ensuring that I took it for the six months beforehand, but I've kind of really been taking it for years sometimes. (W10)
Risks associated with substance use were also discussed by many of the women. For instance, women were aware of the recommendations regarding alcohol consumption during pregnancy, and all women in this study decided to abstain from alcohol. All of the women also discussed smoking as a behaviour with risks for the baby. Although alcohol consumption and smoking were issues mentioned by almost every woman, some women also commented on the risks associated with illicit drug use.
I was at a party and I was said, like, ‘you know, one drink is okay for the baby’ but I was reluctant. (W15)
Um, obviously smoking is a big no-no or any drugs of any description. (W9)
Some women were also aware of the risks associated with sleep position and were aware of the importance of monitoring their baby's movements, which for one woman was a source of distress.

Limited awareness about stillbirth

Women were explicitly asked about their knowledge of stillbirth and the risk factors associated with stillbirth. Our findings suggest limited awareness about stillbirth among the women included in this study. Some women discussed an understanding that stillbirth is a pregnancy loss that can occur later during pregnancy. Further, most women openly expressed that their knowledge about stillbirth and related risk factors was very limited. When explicitly asked, none of the women reported that information about stillbirth was received from a healthcare professional during their antenatal care.
No, I think I have a very limited view of it. You know that baby is born and unfortunately, baby is not born alive but I wouldn't say that I know the reasons why are …you know, obviously it was something that crossed my mind […] but no, I can't say that I know a whole deal about it. (W3)
I suppose what I know about stillbirth is when a baby is born and they're dead. I know that there are stillbirths were you can go through full pregnancy and you can give birth and you don't know that your baby's not going to breathe when they come out, and that's just so sad. Then I do know that there's others where a baby may …their hearts might stop or they might stop breathing during pregnancy and you know that and then you have to give birth to the baby, you know? And they're …that's kind of as much as I know really. (W10)
Additionally, some women confused the term stillbirth with other adverse outcomes given their lack of knowledge of the definitions of pregnancy/infant loss. Women were not aware of the differences between the concepts of stillbirth and miscarriage regarding the gestational cut-off point that differentiates them, and some women conceptualized stillbirth as an event that can only occur during labour. Further, two women confused the term stillbirth with Sudden Infant Death Syndrome.
I'm not sure, can it happen at any stage during pregnancy? can the term stillbirth and miscarriage, you know, can they be used in the same? (W8)
I thought it was kind of random…, that it was most risky before kind of 20 weeks was the highest probability [of experiencing stillbirth] or whatever. Um, that's all I really know. (W17)
I don't know a huge amount about it, but it's if a baby unexplainably passes away and usually, when sleeping before they're six months and the child can seem to be perfectly healthy and there doesn't seem to be an explanation for it. (W4)
Most of the women in this study expressed that they were ‘guessing’ or ‘supposing’ when asked what their

knowledge was about stillbirth and risk factors. Some women stated their knowledge and awareness were drawn from other people's experiences of pregnancy loss in their social circles (e.g., friends of friends, distant relatives and neighbours).

The behaviours that women thought were most likely related to stillbirth were substance use, sleeping on their back or doing exercise lying down on their back, or ‘knocks on the stomach’. Women also spoke about and identified behaviours such as substance use or consumption of certain foods associated with other adverse pregnancy outcomes (e.g., long-term developmental delay, physical disabilities or extreme prematurity); however, the link between such risk behaviours and stillbirth was not present in the majority of women's discourses. These other potential adverse outcomes were perceived as very relevant, concerning and more present in women's minds during pregnancy than the possibility of stillbirth.

I think most women are more concerned with developmental issues with like the drinking and smoking and things like that. They are more thinking. ‘okay, when my baby comes out is it going to have learning issues?’ No one actually thinks about stillbirth. (W18)

I didn't actually worry about stillbirth, I worried about early miscarriage, I worried about falling like that kind of stillbirth, maybe an early delivery and extreme prematurity and neonatal units. (W6)

Theme 3: Silence around stillbirth and risksLack of discussion regarding stillbirth and risk factors

Many women felt that they had not received information about stillbirth, health habits or risk factors for stillbirth during their antenatal care. When asked, none of the women could remember a moment during their interactions with healthcare professionals (e.g., midwives, consultants) in which they had received information about stillbirths.

It is kind of common knowledge, you know, ‘don't drink. Don't…’ you know, like a lot of it, I think people kind of know already. (W13)

Regarding specific health habits, such as physical activity, women perceived that healthcare professionals adopted a very conservative approach towards offering advice, and the general message given to women was to ‘not overdo it’. Regarding weight gain, women also expressed a lack of advice in their interactions with healthcare professionals, and most women reported only being weighed at the time of antenatal booking.

No, no, not really [got any advice regarding physical activity]. Of course, that's if you did some like, very hard Exercise, you have to stop a bit so they warned me about it. (W12)

On the other hand, some women reported receiving some information regarding health habits and risk factors (e.g., keeping physically active, avoiding substance use, sleeping on the left side or monitoring the baby's movements), even though this information was never associated with the risk of stillbirth. In these cases, the information would normally be provided by the women's GP, the hospital midwives or the midwife at their consultant's private clinic.

They were both lovely [Consultant and midwife] and so Midwife would have done a lot of the antenatal education when I went in to see her before I went to see Doctor. (W2)

That's something I came across in my public health nurse, she said everything. And she did say to me, ‘sorry, if I'm being kind of condescending or telling you things, you know, but I need to tell you in case you don't know’. (W17)

The information that women discussed being provided by healthcare professionals regarding risk factors and health habits was mostly focused on sleep position, monitoring fetal movements and preventing food-borne diseases. However, even though some women were provided with this information, in most cases women reported that they were not informed about the reasons to adopt these behaviours. Most women received written information during their care, and even though they valued it and used it to prompt discussions with their providers, they did not feel it could replace a conversation with a healthcare professional.

It was just a brief conversation that was had in the earlier stages [about left-side sleeping]. And I don't even know why. (W15)

I was advised that if I felt any reduced fetal movement […] I presumed that was down to …chances of stillbirth or being at risk of something happened in the baby. This was all after 24 weeks and I know that stillbirth is after 24 weeks. So, I presume, I just put that down to being that without ever mentioning this is a risk of stillbirth. Nobody ever mentioned those words to me in that time. (W15)

According to women's accounts, it seemed that the limited information provided by their healthcare providers might relate to the fact that the participant women were healthy and were not experiencing complicated pregnancies.

I don't feel like they felt the need to have that conversation with me because I wasn't part of the criteria for it perhaps. (W15)

I suppose the fact that I was quite young, the fact that I was low risk and I had no other health concerns …It wasn't the thing that they really discussed with me. [.] So we were happy out and I didn't have any signs or symptoms to suggest that there might be something wrong. (W2)

Reliance on own information-seeking behaviours

Most of the women who participated in our study reported feeling able and confident when trying to find information about their pregnancies using information sources of their choice (e.g., websites and books). Our findings show that most of the awareness that women showed about stillbirth, risk factors and health habits during pregnancy was a consequence of the women's autonomous information-seeking behaviours. Information-seeking behaviours sometimes translated into decision-making modulating women's behaviour. However, in other instances, finding conflicting information (e.g., in online forums) acted as a source of concern.

I was happy with that because I knew myself that I got my information from a variety of sources […] I was kind of happy with my own research. (W16)

I was actually looking and I found like different information because there are some pages were saying that you should sleep on the left side, some people on the right side, so I was wondering, should I sleep on the left side or the right side? (W12)

Proactive information-seeking behaviour led some women to feel that they already had enough information through their research, and they did not require additional professional advice. However, some women acknowledged that only relying on the information provided by their healthcare professionals would have made them feel uninformed.

Like it was fine, I'm well able to kind of Google things and research myself. (W9)

I don't think so [that information provision would have been enough without her own research]. Because I have a friend who refused to do any research. She was just totally caught off guard. With lots of different things that happened, yeah, I don't think there was enough information there but I mean maybe they would have given me more information if they got the impression that I didn't know, you know. (W16)

Women also spoke about some of the characteristics of the sources of information that they used during their pregnancies. Women expressed a strong reliance on official sources, like websites or hospital-provided books, and had a critical attitude towards certain sources of information, especially those found online.

I did take on board what was in the HSE websites […] I did find the HSE website quite trustful as well. (W2)

Most of the information I just got from the HSE book, I found the HSE book actually to be excellent. That was probably the best, along with the nurses and, and the consultants, that was one of the best sources of information. I just kind of stuck to that because I it's accurate what was in it. (W4)

Some of the sources of information named by the women were: hospital-provided books, commercial mobile applications, websites, family and friends, conversations with healthcare professionals, social media, online antenatal classes, formal education, peers and podcasts. Most women engaged with commercial mobile phone applications (apps) to obtain information, and expressed that one of the features that they appreciated the most about apps in particular was the weekly updates and notifications; these features provided women with timely relevant information for their stage in pregnancy.

[weekly updates] it helped me along the journey. Kind of nearly makes it easier, you're better with something that you can see, now you know you're getting bigger, but like you can kind of just see the journey and it helps you along, but it was just really simple. (W10)

Theme 4: Attitudes towards receiving information about stillbirth'Knowledge is key'

Eight women in our study had a positive and open disposition towards receiving information about risk factors and stillbirth during their antenatal care from their healthcare professionals. For these women, information was perceived as a tool that might have an influence on preventive efforts, and also facilitate the grieving process of parents who

experience stillbirth.

I think it is good to know about stillbirth because I think it's a very Irish thing maybe, that we don't talk about things that we don't want to talk about. It should be spoken about. I think we should be told about it. We're told about everything else. Then, you know, if there's risk factors for stillbirth and it's also risk factor, we should be made wherever of it. A lot of people don't talk about it. There's a stigma around it, I understand why. It's, you know, it's sad, it's heart-breaking. (W10)

I think to have the knowledge would be way more beneficial than negative. Even if I did go away with a little bit of concern about it, I think that's very natural because you fear for your baby and you want to do everything that you can. But I think overall it would have been extremely positive. (W15)

It's better to be prepared basically for the unexpected. (W5)

Some comments made by the women indicate that women's concerns regarding pregnancy loss are focused on the first trimester and that the probability of loss is almost nil afterwards. This is a popular misconception that might have been supported by the women's family or social context.

I suppose with stillbirth it's the same kind of thing, preparing women that this is not a 'You've now reached 12 weeks it's not a 100% guarantee' and I suppose if you have no awareness of it and it happens to you would be so shocking. (W18)

Some women felt that receiving information about stillbirth and its modifiable risk factors would also work to facilitate women's decision-making, by providing them with enough information to facilitate making informed judgements about their risks.

I think information is power. Just to give them as much information as possible and then they eventually choose what they want to do with it. But at least they have the information then. (W9)

Stillbirth perceived as a difficult topic

Most of the women participating in this study engaged in a process of balancing the pros and cons of receiving this information, which was evident in their discourses. Some of the women in the study expressed that they would have found stillbirth a difficult topic to discuss, especially during pregnancy. However, some women with this opinion also recognized that stillbirth was a latent concern of theirs during their pregnancies.

It was something I didn't want to talk about and I wouldn't have brought up myself, but if they had approached me with this, I would have been absolutely fine to talk about it. (W3)

When you're pregnant, it's something you tend to not want to overly research just because […] you're hypersensitive at that time also. (W8)

Nine of the women expressed that discussing the topic of stillbirth might have the potential to increase their levels of anxiety during pregnancy. The word stillbirth was perceived as a societal taboo which, for some women, acted as a barrier to both information seeking and information provision. However, all of these women felt that the balance between being empowered with the information, despite having certain negative feelings, was still positive. Women rationalized their decision by considering the information as a positive resource, but they highlighted the importance of tailoring the information based on each woman.

I think it would have been counterproductive, but you know …saying that I'm a person who likes to know all the facts. So, you know, I would like to think that if I did ask that they would have been very forthcoming and very honest with the information that it wouldn't be a case of 'oh you don't need to worry about that'. (W8)

I think knowledge is key so I would have absorbed the information and taken on board for sure. However, being and discovering that I'm a bit of a worrier, there's no doubt it would have played in my mind. (W15)

I think it would be beneficial, but I'm not sure everyone would want to hear it. Like if you're quite emotional and it is an emotional time, but I do think it is good to kind of maybe touch on it and maybe not put too much emphasis on it. (W9)

One woman had a negative attitude towards receiving information about stillbirth and risk factors, for this woman, the benefits of obtaining the information did not justify the potential harm.

If someone just says 'don't drink alcohol' fine. You don't do it. If someone says 'don't drink alcohol because you

might have a miscarriage' and then you're worrying about it …Does worrying increase the risk? I don't know. No, it's probably better that it's not pointed out maybe, for fear that you do worry more about it and you worry unnecessarily. (W17)

Importance of language and preferences for information provision

Some of the women that participated in our study insisted on the importance of the language used when providing information about stillbirth and risk factors. Women felt that the best way to provide education about stillbirth and risk factors was to do so in a very sensitive way and avoid the use of 'blaming' vocabulary when talking about the behaviours that might increase the risk of stillbirth.

So while it might invoke some degree of upset. I think if it's done in a sensitive way and in a kind of a positive slant of, you know, 'we know there are some things that we can do to reduce this risk', that I would think that most women would be receptive to that. (W6)

Maybe do it without using the word stillbirth: 'avoid these behaviours during pregnancy' or something. Yeah. (W13)

Maybe it's just the people don't like to say the words, you know? (W15)

Regarding women's preferences for information provision, women highlighted the importance of having reliable sources, and providing specific advice or information, with the option for more information on demand. Women also expressed a preference for group educational sessions on behavioural risk factors for stillbirth and best health habits for pregnancy. According to these women's opinions, receiving information about stillbirth and risk factors for stillbirth on a one-to-one basis would have made them feel as if there was something wrong with their pregnancy, whereas receiving the information in a group format would be considered as 'just another topic to discuss'.

I think maybe groups, because I think if it's one on one, the person might feel it's being directed at them. Whereas if it's in a group, it's just, you're informing them. (W5)

When it comes to the best timing to provide women with information about stillbirth and risk factors, women felt that this information should be provided early in pregnancy. These women were aware of the importance of not engaging in risky behaviours as soon as possible, and so they considered that receiving the information early would help prevent adverse outcomes in pregnancy. Some women expressed that they had a heightened cautious attitude at the beginning of their pregnancies and that this could be taken advantage of in terms of increasing awareness about risk behaviours. Additionally, some women expressed the potential for conversations about stillbirth between women and healthcare providers to become more difficult as the pregnancy progresses, when women's concerns start focusing on labour and birth.

There's a lot of information at the start, but it's really that's when you need to be doing it [changing behaviour], […] I think giving the information earlier is better. (W15)

I think earlier in the pregnancy so they have time to set up for a healthier pregnancy. (W14)

However, other women considered that the information about stillbirth and risk factors should be provided in the second trimester since the first trimester represents a vulnerable period, in which discussion of stillbirth would constitute additional stress and pressure for the woman. Whereas other women, especially those with a background in healthcare, highlighted the importance of the preconceptual period as the best time to provide education to women.

Maybe in between 12 week and the anomaly can. I think a lot of people are kind of stressed about getting to the 12 weeks anyway […] I think maybe we should leave people get to that stage rather than stressing them out even more. (W1)

At the beginning I think there's enough stress anyway …after the half, if you know that everything is fine, that kind of information won't actually scare you so much like at the beginning. (W12)

DISCUSSION

Our findings have shown that information provision and awareness about stillbirth and associated behavioural risk factors are mostly poor amongst most women with uncomplicated pregnancies and births participating in our study. Women showed good awareness regarding health recommendations and behaviours to avoid during pregnancy, however, regardless of the level of knowledge that women expressed about stillbirth or potential behaviours

associated with an increased risk of stillbirth, it seemed that there was not a clear link establishing the association between risk behaviours and stillbirth in women's discourses. Further, the awareness that women expressed regarding risk factors and stillbirth was predominantly a result of their information-seeking behaviour. Women expressed that information provision during their antenatal care regarding health behaviours was poor, and information regarding stillbirth was nonexistent. Women in this study showed varying attitudes regarding being provided with information about stillbirth and behavioural risk factors for stillbirth. Whereas some women perceived information as a tool to improve prevention efforts, other women considered that this information was not necessary in all cases and it would only increase anxiety levels for them.

Women's reasons for engaging in behaviour change were commonly associated with obtaining the best possible outcomes for their babies and themselves. This motivation to make healthy choices and a sense of responsibility driven by the desire to improve the baby's health and reduce risk has been described in the extant literature.[26,27,28,38] Further, women's perception of risk, which can only be accurate with appropriate communication and awareness, will also influence their decision-making.[38,39] This highlights the importance of providing accurate information to women about stillbirth related to different behaviours during pregnancy. Providing women with this information might increase their level of motivation to tackle modifiable behaviours and reduce their risk of stillbirth.

Women in our sample were able and willing to conduct their research and find answers to their concerns, and one of the most mentioned information resources was the internet. However, we know from previous research that the information available online regarding stillbirth and behavioural risk factors for stillbirth in Ireland and the United Kingdom is scarce and difficult to access.[40] Further, women in our study were a small number of predominantly highly educated White Irish women. Previous research has demonstrated that sociodemographic characteristics are associated with the choice of a particular source of information, and also with the number of sources used,[41] with being female, educated and young the best predictors to engage in health information-seeking behaviours.[41] Therefore, it is possible that the positive attitude towards information seeking in our sample is not representative of the general population, and further research would be necessary to explore the needs of people from different sociodemographic backgrounds.

Previous research has demonstrated that the levels of knowledge and understanding that women have about advice received during pregnancy will have an impact on their psychological capabilities towards behaviour change.[28] Women who are less aware, or who held misconceptions about the consequences of their behaviours will be less likely to engage in behaviour change.[28,42] The women in our sample had limited knowledge about stillbirth and its associated risk factors; however, when prompted to speak about the advice received during their antenatal care, monitoring of fetal movements and the importance of sleep position was often mentioned. These findings are very similar to those obtained by Stacey et al.[43] in a recently published study, where they explored migrant women's awareness of health messages to reduce stillbirth risk. Our findings also highlight the importance of promoting health education during pregnancy from the healthcare professionals involved in women's care, as well as the value of generating additional written or online resources for women.

Additionally, our results show that women prefer antenatal groups to receive information regarding their health and their pregnancies, and most women found that antenatal classes represent appropriate spaces to share information about stillbirth and risk factors for stillbirth. This is positive considering that previous research has demonstrated that passive transfer of information is not sufficient to prepare women and their partners for birth and parenthood, and hence facilitating educational groups using different types of experiential methods should be recommended.[44]

Women in our study were also very reliant on the written information provided to them by their healthcare providers. Hence, health services should produce up-to-date user-friendly online and paper-based resources or materials for women and encourage engagement with such materials from the healthcare professionals' perspective to support discussion with patients. However, given the diversity in which women prefer to receive information, it is important to tailor and utilize diverse types of intervention approaches to maximize engagement and effectiveness of antenatal interventions.

Healthcare professionals should engage in active discussions with women, regarding risk factors for stillbirth.

Antenatal education standards and healthcare professionals' training programmes should guide to support healthcare professionals in promoting health with their patients, and also be able to discuss risk factors for stillbirth or other potential adverse outcomes. The National Women and Infant Health Programme developed the National Standards for Antenatal Education in Ireland in 2020.[44] Although this document highlights the importance of including an element of better health and well-being (Theme 4) in the optimal standards of antenatal education, this theme only refers to supporting healthy habits and addressing mental health concerns generally, without specifying the need to address issues such as the risk factors for stillbirth with women.[44]

Previous research has also demonstrated that stillbirth is a taboo topic in society.[29,45,46] However, as seen from our results, most women would welcome information about stillbirth and risk factors as a tool to improve their health and prevent it from happening to them, or even as emotional preparation after experiencing a stillbirth. However, a high number of women also acknowledged that receiving this information would have the potential to increase their levels of anxiety and concerns, which is an issue that should be taken into consideration. Literature in areas such as sexual health or weight management has shown that healthcare professionals are reluctant to discuss topics perceived as taboo or difficult with their patients,[47] and stillbirth is presumably a similarly unmentionable topic.[45] We believe that it is important that official bodies regulating antenatal education standards in different high-income countries include specific guidance regarding advising women about health habits, risk factors and potential adverse outcomes such as stillbirth to support healthcare professionals in providing antenatal education with confidence. Furthermore, as mentioned by multiple women, receiving this information has the potential to increase their anxiety levels or their concerns, and hence, healthcare professionals are prepared to address women's concerns and resolve their doubts, trying to alleviate their worries by providing accurate and sensitive information. The different types of pregnancy loss have different implications regarding care, and hence, breaking the silence around stillbirth is essential to ensure that women who experience stillbirth received the appropriate support.[29]

Our study has some strengths and limitations. The study's first limitation is related to the sample of interviewed women. The study participants were predominantly White, educated, heterosexual women from developed countries, and, likely, their experiences with behaviour change during pregnancy and challenges identified are not generalizable to people from different sociodemographic backgrounds. Additionally, the rates of smoking and alcohol consumption were also lower in our sample than in the general pregnant population, thus findings related to this modifiable risk factor for stillbirth are likely influenced by this. Further, another limitation that we encountered when conducting the interviews is that in some instances women struggled to recall some parts of their experiences, given the timeframe in which they were interviewed. This might also be explained by the changes in memory that occur during pregnancy and have been described in the literature before, where women reported memory deficits categorized as a general sense of ‘fogginess’.[30] The number of women approached to consent to this study was high for a qualitative design. This decision was made to ensure that we were able to obtain an appropriate amount of rich data and anticipate that most women might decline to participate a few months after leaving the hospital. Of the 44 women recruited, only 18 participated in the interviews, indicating that possibly our timeframes did not suit the majority of women and highlighting the challenges of recruiting women during the busy postnatal period. Furthermore, given that only the clinical team has access to the patient's medical record, we are unaware of how many women were approached by the clinical recruiting team and declined to participate as this number was not recorded. The researchers only obtained the contact information of the women who consented to participate in the study. This hinders the assessment of the efficacy of our recruitment method, and it also limits our knowledge about potential differences between women who agreed to participate and women who declined. It is possible that women who chose to take the time to participate in this study were adapting better to the postpartum period than those who could not participate. This might be related to their support networks, their baby's sleep patterns or temperament, their baby's health needs or their own health needs. Hence, it is important to acknowledge that women participating in this study might have been having a more positive experience regarding their pregnancy, birth and the postpartum period, and so these factors also need to be considered when trying to generalize these findings.

Despite these limitations, our study also has several strengths. Our inclusion and exclusion criteria were designed to

recruit women with uncomplicated pregnancies and births, enabling the exploration of experiences of women receiving the most ‘generalised’ type of care in our maternity services; this increases the generalizability and transferability of our findings to other low risk/uncomplicated pregnancies in similar maternity systems. Furthermore, the use of online interviews was praised by some of the women as a very comfortable way to engage in research, without causing much interference in their normal lives. To date, there is only one other study exploring women's perception of public health messages to reduce stillbirth, in this case, exploring the views of migrant women in the United Kingdom.[43] The findings of this study are similar to our findings in that they highlight the complexities of discussing stillbirth with women during their pregnancies and the importance of developing culturally appropriate resources to secure efficient communication.

CONCLUSION

This study aimed to explore women's experiences of modifiable risk factors during pregnancy, awareness of stillbirth and its risk factors, as well as their experience with information provision during their antenatal care. The findings of this study have shown that women with uncomplicated pregnancies receive very poor information about health behaviours, behavioural risk factors or stillbirth during pregnancy. Women have high levels of understanding of how to have a healthy pregnancy, but the link between behavioural risk factors and potential outcomes such as stillbirth is not considered. Information provision during antenatal care was not sufficient, and women had to rely upon their information-seeking behaviour. Most women perceived receiving information about stillbirth during antenatal care to be useful to help preventive efforts, although others acknowledge the potential for this information to raise some concerns highlighting the importance of using sensitive nonjudgemental language.

Information provision alone is not sufficient to support behaviour change, however, it might act as a first step to engage in discussions and facilitate women seeking adequate care for their specific needs. Healthcare professionals should break the silence around stillbirth and incorporate risk factors, health habits and stillbirth in their routine discussions with women, especially in terms of outcomes for their babies, to motivate women to engage in behaviour change. Tackling the modifiable maternal risk factors for stillbirth by providing information and supporting women with behaviour change during pregnancy might contribute to reducing the stillbirth rates in Ireland.

ACKNOWLEDGEMENTS

This study is part of a PhD project funded by Science Foundation Ireland (grant number 12/RC/2272). Open access funding provided by IReL.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

DETAILS

Subject:	Pregnancy; Behavior; Womens health; Risk behavior; Constructivism; Risk factors; Epidemiology; Intervention; Hospitals; Stillbirth; Information sharing; Risk analysis; Interviews; Body weight; Postpartum women; Sleeping position; Risk taking; Substance use; Risk perception; Smoking; Alcohol use; Attitudes; Substance abuse; Change agents; Knowledge; Obesity; Drug use; Public health; Alcohol; High income; Antenatal care; Qualitative research; Mothers; Silence; Babies; Prenatal care; Behavior change; Overweight
Business indexing term:	Subject: High income
Location:	Ireland

Identifier / keyword:	antenatal education; risk factors; stillbirth awareness; stillbirth prevention
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	329-342
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-23
Milestone dates:	2022-10-27 (manuscriptRevised); 2023-0 1-20 (publishedOnlineFinalForm); 2022-06-23 (manuscriptReceived); 2022-11-23 (publishedOnlineEarlyUnpaginated); 2022-10-31 (manuscriptAccepted)
Publication history :	
First posting date:	23 Nov 2022
DOI:	https://doi.org/10.1111/hex.13662
ProQuest document ID:	2767102251
Document URL:	https://www.proquest.com/scholarly-journals/exploring-first-time-mothers-experiences/docview/2767102251/se-2?accountid=211160

A valued voice: A qualitative analysis of parental decision-making preferences in emergent paediatric surgery

Carlisle, Erica M [1] ; Shinkunas, Laura A [2] ; Ruba, Emily [3] ; Klipowicz, Caleb J [4] ; Lieberman, Maxwell T [4] ; Hoffman, Richard M [5] ; Reisinger, Heather S [6] [1] Department of Surgery, Division of Pediatric Surgery, University of Iowa Hospitals and Clinics, Iowa City, Iowa, USA; College of Medicine, Program in Bioethics and Humanities, University of Iowa Carver College of Medicine, Iowa City, Iowa, USA [2] College of Medicine, Program in Bioethics and Humanities, University of Iowa Carver College of Medicine, Iowa City, Iowa, USA [3] College of Medicine, University of Iowa Carver College of Medicine, Iowa City, Iowa, USA [4] Department of Anthropology, University of Iowa, Iowa City, Iowa, USA [5] Department of Internal Medicine, University of Iowa Hospitals and Clinics, Iowa City, Iowa, USA [6] College of Medicine, Program in Bioethics and Humanities, University of Iowa Carver College of Medicine, Iowa City, Iowa, USA; Department of Internal Medicine, University of Iowa Hospitals and Clinics, Iowa City, Iowa, USA; Institute for Clinical and Translational Science, University of Iowa, Iowa City, Iowa, USA

ProQuest document link

ABSTRACT (ENGLISH)

Introduction

Shared decision-making, with an emphasis on patient autonomy, is often advised in healthcare decision-making. However, this may be difficult to implement in emergent settings. We have previously demonstrated that when considering emergent operations for their children, parents prefer surgeon guidance as opposed to shared decision-making. Here, we interviewed parents of paediatric patients who had undergone emergent operations to better understand parental decision-making preferences.

Methods

Parents of paediatric patients who underwent surgery over the past 5 years at a University-based, tertiary children's hospital for cancer, an emergent operation while in the neonatal intensive care unit (NICU) or extracorporeal membrane oxygenation (ECMO) were invited to complete a 60-min semi-structured interview. Interviews were digitally recorded and transcribed verbatim. Thematic content analysis was performed via deductive and inductive analysis. An iterative approach to thematic sampling/data analysis was used.

Results

Thematic saturation was achieved after 12 interviews (4 cancer, 5 NICU and 3 ECMO). Five common themes were

identified: (1) recommendations from surgeons are valuable; (2) ‘lifesaving mode’: parents felt there were no decisions to be made; (3) effective ways of obtaining information about treatment; (4) shared decision-making as a ‘dialogue’ or ‘discussion’ and (5) parents as a ‘valued voice’ to advocate for their children.

Conclusions

When engaging in decision-making regarding emergent surgical procedures for their children, parents value a surgeon's recommendation. Parents felt that discussion or dialogue with surgeons defined shared decision-making, and they believed that the opportunity to ask questions gave them a ‘valued voice’, even when they felt there were no decisions to be made.

Patient or Public Contribution

For this study, we interviewed parents of paediatric patients who had undergone emergent operations to better understand parental decision-making preferences. Parents thus provided all the data for the study.

FULL TEXT

INTRODUCTION

Thoughtful, effective engagement of patients in decision-making has been shown to decrease decisional conflict (i.e., level of discomfort with the decision) and reduce overutilization of healthcare resources.[1] Decision quality, the extent to which decisions are both informed and congruent with the patient's values, is also increased when patients are engaged in the decision-making process.[1] Given the elevated risks of morbidity and mortality as well as the potential emotional burden associated with discussions surrounding complex, high-risk operations, meaningful engagement of patients in the decision-making process may be especially important for patients considering such operations. Shared decision-making (SDM), a process in which physicians and patients develop mutually agreed upon care plans that incorporate clinical evidence as well as patients' values and preferences, is generally considered to be a favourable approach to clinical counselling.[2] However, relatively little data exists regarding the effectiveness of SDM for surgical counselling.[3]

Within the field of paediatric surgery, even fewer data are available regarding attitudes towards SDM or overall parental decision-making preferences.[4] Most available data focus on parental decision-making preferences for elective paediatric surgical procedures.[4] For example, Hong et al. evaluated the decision-making process of parents considering elective otoplasty, a surgical procedure to correct prominent ears, for their children. The authors found that overall, parents experienced significant decisional conflict when considering this operation. However, those parents who considered themselves to have greater involvement in the decision-making process reported less decisional conflict and regret than did parents who considered themselves to be less involved in the decision-making process.[5] Similar findings were echoed in a study of parents considering an elective tonsillectomy, adenoidectomy or tympanostomy tube insertion for their children. Parents who considered themselves to be more involved in the decision-making process reported less decisional conflict when deciding whether their children should undergo surgery.[6] While studies such as these suggest that parental involvement in the elective surgical decision-making process is important, they offer limited insight into the specific components of the decision-making process that promote parental involvement.

Parental decision-making preferences for elective surgery may be different than those for emergent surgery. Parental preferences for emergent surgical decision-making have not been extensively studied.[4] In a survey of parents of paediatric surgery patients, we found that parental decision-making preferences for paediatric surgery are context-dependent.[7] Specifically, we identified that parents prefer more guidance from surgeons when making decisions about operations they consider emergent as opposed to operations that they consider nonemergent.[7] Marsh et al.[8] had similar findings when they interviewed parents of children who sustained a life-threatening, traumatic brain injury on their preference for who should make the decision to proceed with an operation to place an intracranial pressure monitor. While parents desired information and communication from physicians regarding the procedure, they overwhelmingly preferred that physicians decide whether to perform the procedure.[8] These studies suggest that parents increasingly prioritize guidance from their child's surgeon when making emergent decisions; however, further investigation of parental decision-making preferences is needed to fully understand this issue.

Enhancing our understanding of parental decision-making preferences in emergent paediatric surgical settings will help surgeons communicate with parents in a way that appropriately promotes parental involvement, facilitates trust and minimizes decisional conflict and regret. We conducted semistructured interviews with parents of paediatric patients who had undergone emergent operations to identify common themes regarding their decision-making preferences.

METHODSStudy design and recruitment

We conducted semi-structured interviews to broadly explore parents' experiences and preferences towards decision-making in three emergent paediatric surgical settings at a university-based children's hospital.[9,10] The study was approved by the University of Iowa Institutional Review Board (#201905811) and is reported according to the Consolidated Criteria for Reporting Qualitative Research (COREQ) checklist (Appendix 1).[11] All methods were carried out in accordance with relevant guidelines and regulations. As approved by the University of Iowa Institutional Review Board, informed consent was obtained verbally at the commencement of each interview. All participants were over the age of 18.

We used a purposive sampling technique to identify parents of paediatric patients (<18 years old) who had undergone surgery by a paediatric surgeon for major resection of a solid tumour, an urgent operation while in the neonatal intensive care unit (NICU) or extracorporeal membrane oxygenation (ECMO) between 2015 and 2020. ECMO was included, as this is a procedure performed only when patients are in immediately life-threatening clinical circumstances. Solid tumour resection operations and operations on neonates in the NICU were selected based upon a prior survey conducted by our research team where parents of paediatric surgical patients were provided six hypothetical paediatric surgical scenarios and asked to identify the operations they considered emergent.[7] Parents identified operations for cancer and operations on babies to be emergent operations.

Eligible parent participants were sent an initial email or letter, which was followed with a phone call if there was no response after 2 weeks. Parent participants were also recruited through our hospital's Facebook page and Twitter feed. To maintain our purposive sampling technique, interested parents were asked to complete a brief online survey to ensure that their child met the eligibility criteria. Participants received a $20 Amazon gift card after the completion of the interview.

Interview procedures

Semistructured interviews were conducted by phone between June 2019 and April 2020. All interviews were conducted by a research specialist (L. A. S.) or a medical anthropologist (C. J. K. or M. T. L.) trained in the process of semistructured interviews. We designed a semistructured interview guide of 20 open-ended questions based on the literature review and our prior survey data (Appendix 2). During the interview, parents were asked to create an illness narrative about their child's surgery. The questions sought to explore ‘personal experience narratives’ by asking parents about the events leading up to their child's surgery, the process of making surgical decisions, and the consequences of the decisions that were made.[12] The illness narrative specifically emphasized the amount of guidance and interaction parents desired and experienced from the surgeon, the amount of involvement in the decision-making process parents preferred and experienced, how much procedural detail or information regarding procedural risks was desired and received, preferred approaches for receiving information and feelings regarding the outcome of the surgery and the surgeon. At the end of the interview, parents were asked what the term ‘shared decision-making’ meant to them. There were no question prompts. The main questions in the interview guide were intended to direct the conversation. However, the interview was not limited to the questions in the guide but rather shaped by each parent's responses to the main questions. Throughout the interview process, questions were refined based on the consensus of the research group as well as concurrent iterative analysis.

Data analysis

Interviews were digitally recorded on a secure device. Audiotapes were transcribed verbatim by a member of the research team and de-identified. Thematic content analysis was performed using deductive codes from the interview guide and the literature and inductive codes that were identified in the data.[10] An iterative approach to thematic sampling and data analysis was used. Three members of our research team (L. A. S., M. T. L. and E. M. C.) met

weekly to discuss the overall progress of the project. This team coded all transcripts using a deductive and inductive approach, which involved constant comparative analysis and multiple iterations of coding as the research team oscillated between gathering and analysing the data. A codebook was developed and repeatedly refined based on group discussion of the data. Each research team member independently coded the transcripts before all discussions or refinement of the codebook. Constant comparative analysis continued until data saturation, the stage where 'there are mounting instances of the same codes, but no new ones' was reached.[13] Coding was reviewed and saturation was assessed after completion of every two to three interviews. In total, we performed 12 interviews (4 cancer, 5 NICU and 3 ECMO). Transcripts were uploaded into MAXQDA 2020 (Verbi Software, 2019) for data management and analysis of themes across interviews.

RESULTSParticipants

Twelve parents of 12 paediatric surgical patients were interviewed. All participants were mothers of children who had undergone ECMO, an emergent operation while in the NICU or major resection of a solid tumour. Patient demographics are presented in Table 1. Parental race, ethnicity and socioeconomic status were not collected. Interview length ranged from 25 to 65 min.

Table 1 Patient demographics

Participant ID	Surgical category	Patient sex	Patient status	Patient's diagnosis	Patient age at first surgery
P1	Cancer	F	Alive	Wilms tumour	2 years old
P2	Cancer	F	Alive	Wilms tumour	3 years old
P3	Cancer	F	Alive	Wilms tumour	5 years old
P4	Cancer	F	Alive	Neuroblastoma	2 years old
P5	ECMO	M	Alive	Persistent pulmonary hypertension	1 day old
P6	ECMO	F	Alive	Congenital diaphragmatic hernia	9 days old
P7	ECMO	M	Alive	Sepsis	13 years old
P8	NICU	F	Alive	Bowel perforation	19 days old
P9	NICU	M	Deceased	Bowel perforation	10 days old
P10	NICU	M	Alive	Necrotizing enterocolitis	12 days old
P11	NICU	M	Alive	Persistent pulmonary hypertension	1 month old
P12	NICU	F	Alive	Gastric perforation	3 days old

Abbreviations: ECMO, extracorporeal membrane oxygenation; F, female; M, male; NICU, neonatal intensive care unit.

Common themes

Five common themes were identified: (1) recommendations from surgeons are valuable; (2) 'lifesaving mode': parents felt there were no decisions to be made; (3) effective ways of obtaining information about treatment; (4) shared decision-making as a 'dialogue' or 'discussion' and (5) parents as a 'valued voice' to advocate for their children. Themes did not vary based on the type of surgery the patient underwent or the age of the patient.

Theme 1: Recommendations from surgeons are valuable

The majority of parents (8 out of 12) specifically noted that receiving a recommendation about surgical intervention from a surgeon was valuable. Surgeons were described as 'the experts [who] knew what needed to be done' (P11). As one parent articulated, 'I mean alright if we do A, B and C, then her survival rate is XYZ. For me, I found that helpful' (P3). Parents explained that recommendations provided a sense of confidence in the care plan and made them feel involved in the process of caring for their children. As one parent pointed out, 'ultimately the surgeon needs to share what they're recommending and how they want to proceed' (P4). Another parent explained that the recommendation 'kind of gave me something to look up online to try to figure out [things]' (P12). Not all surgeon recommendations were perceived as helpful. One parent expressed that the recommendation provided was not helpful due predominantly to the blunt manner in which it was delivered by the surgeon, suggesting that a surgeon's approach to communication may influence parent's perceptions of recommendations as well.

Theme 2: 'Lifesaving Mode': Parents felt there were no decisions to be made

All parents independently expressed that they felt as if there were no surgical decisions to be made. No specific question was asked by interviewers to assess this sentiment, rather this feeling was organically identified by parents during every interview. As one cancer parent stated, 'When she first was diagnosed, they made all the decisions. They told me what was gonna happen' (P1). An ECMO parent acknowledged, 'No, there was no decision. We were all just in just lifesaving mode' (P5). Parents tended to express the belief that consenting to further care was seen not as a decision but as the best option. As one parent articulated, 'I don't feel like we made that many decisions. It was just you know being guided through by the doctors and nurses. Like doing the next best thing. This is the next right thing to choose' (P7). Death of the patient or transition to comfort measures were not seen as reasonable choices by any of the parents interviewed. For instance, one NICU parent indicated, 'That was one of those things where there were really no options. You either had to do surgery or death' (P10). Another parent shared this sentiment by stating, 'It was this or a funeral. There wasn't any other option' (P5).

Theme 3: Effective ways of obtaining information about treatment

All parents acknowledged the importance of direct communication with surgeons and other providers.

I know that it's a very involved process having a sick child. But I really appreciated the fact that the surgeon sat down with us, and discussed everything they were going to do. They told us about the surgery in detail. They told us exactly what medications he was going to be on. And they were completely honest with us as far as what his chances of survival were. (P9)

Three parents emphasized the importance of a slow conversational pace with significant repetition. As one parent expressed, 'It's easy to trust [the surgeon] when you feel like you're not being rushed out the door or rushed with any questions or concerns that you have and you know that they want to help you' (P12). Another parent verbalized, 'it was reassuring to hear a story three times so that we could actually gather the facts' (P7). Most parents also highlighted their appreciation of the surgeons' efforts to use drawings and other nonverbal, physical means of communication to clarify explanations. Specifically, four parents emphasized the favorability of a protocol or guidebook, four parents highlighted the use of images and diagrams and two parents emphasized the use of brochures or other printed materials. In reference to the helpfulness of images and diagrams, one parent stated, 'She drew pictures, and it made so much sense to me' (P7). Thinking about the helpfulness of a protocol or guidebook, one parent said:

Back in the beginning when she first was diagnosed I was really glad that they gave me this big old notebook with all

this information and it had that you know that protocol in there telling me what was gonna happen and when she was gonna have chemo and how many weeks apart they were and everything. And I think that was comforting …because you never know what's gonna happen but at least you have that timeline and you can kind of try to figure out your life with the timeline of chemo and surgery and whatever. (P1)

Two parents specifically emphasized the importance of being provided with as much information as possible '…regarding risks, outcomes, benefits, possible complications, and then what the plan would be if something were to happen. Just so that if something does happen, you don't feel like you're in the dark' (P8). Only two parents highlighted the use of statistics or descriptions of the surgical procedure in their assessment of useful information. Several parents expressed that poor surgeon demeanour (*n* = 1), repeated questions from multiple teams (*n* = 1) and acronyms (*n* = 1) were unhelpful forms of communication.

To supplement direct communication with surgeons, eight of the 12 parents discussed searching for information online. According to one parent, 'everybody says don't go to Google or don't, you know, live on the internet. But sometimes that's the only recourse you have when it comes to not having the medical staff around you' (P5). While one parent expressed that parents are 'not supposed' to do online searches, another parent was comforted by finding online evidence of international research on her child's diagnosis and another parent suggested, '…it made what the doctors said more understandable' (P3).Three parents used social media to connect with other parents. Additionally, several parents (*n* = 4) suggested that virtual communication opportunities may enhance communication with the healthcare team. For instance, one parent noted that,

If there was a way that, instead of having to call in and see how rounds went when we couldn't be there, if there was a way that we could have a monitoring system, almost, in their rooms. And we could check in …it would be nice if it was something that we could log in knowing that rounds were gonna' be done between like 9 and 11 or whatever. And then they could like teleconference rounds. (P5)

Theme 4: Shared decision-making as a 'dialogue' or 'discussion'

All parents were asked what the phrase 'shared decision making' meant to them. Overall, shared decision-making was seen as a 'dialogue' or 'open discussion between the surgeon and the family' (P2). Nine parents highlighted the importance of participating in the decision-making process. One parent defined shared decision-making as 'the doctor and the patient or the guardian you know agreeing upon what you're going to do. Not just the doctor coming in and saying this is what we're doing. Making the guardian or the parent or the patient feel involved in the decision-making process' (P4).

Five parents emphasized the importance of the surgeon's recommendation in the discussion.

If someone told me shared decision making, I would think that they were asking me to help make the decision on my child's treatment. I mean they're the experts. I mean so shared decision making, if you're saying so here's the recommendation and this is why I'm sharing with you my opinion why this is the best thing for your child, that's great. And I do want to know that. But at the end of the day, whatever they recommend, I'm gonna do it. (P3)

Three parents mentioned the importance of explaining options. For instance, one parent defined shared decision-making as '…explaining the options if there are options, I guess. I think I would always prefer a recommendation and then want to know the reasons behind that recommendation' (P2). Three parents said that the importance of shared decision-making might depend on the person or the situation.

You could say every parent needs to have input, but some parents don't have anything to input because they don't understand what's going on. So, I don't know, it's not, it just varies on the situation with the family. It's not like a cut and dry thing. It's kind of complicated. I guess it all depends on the person. (P1)

Only one parent mentioned the importance of parent responsibility for making the final decision.

Theme 5: Parents as a 'valued voice' to advocate for their children

All 12 parents relied on surgeons for information. Eight parents used internet searches and social media to supplement their knowledge. All parents expressed that even when they felt there was no specific decision to be made, the opportunity to use the information they had gathered to be involved in their child's care and included in discussions regarding their child was important. This engagement gave them a 'valued voice', which was described

by one parent as:

I felt like a valued voice in her care plan…. [Interviewer asks what makes parent feel valued]…. Just that they actually listen to us. Like if we're asked a question about how she's doing or different things that were going on that they actually wanted to hear the answer instead of just asking to ask. And that they actually kind of took it to heart when making decisions. (P12)

All parents also identified the need to advocate for their children; however, the approach parents chose varied based upon self-described personality characteristics. Three parents identified advocacy as speaking up when they perceived their child was being cared for incorrectly (including instances of wrong medication, wrong time to give medication or delayed stitch removal).

…like a couple times medications were given at the wrong times or given twice. Like if one nurse was busy, another nurse would come in and I would say, I think that was just given. You know and there were a couple times like that that I'm kind of glad that I got overly invested in it. (P7)

Four parents emphasized the importance of extending this advocacy to other patients or families. One parent shared that their family has '…stayed super involved with the NICU. We volunteer. We fundraise for them. We mentor some other parents now' (P11). Another parent revealed, 'I feel like we speak out more about stuff that we don't agree with like anti-vaxxers because [she] was exposed to whooping cough from an unvaccinated child while she was on chemo' (P3).

All parents emphasized the importance of gaining the knowledge that they needed to effectively advocate for their children. As one parent expressed, 'Once we figured out what rounds were, about a week later, we were there every day taking notes, and I would say we were very interested in learning the science and medicine behind what was going on in case we had to make decisions' (P11). Another parent conveyed, '…we wanted to go for the best option for her and we did a lot of talking and going back and forth with a couple different surgeons on the team to see, to get their different points of view' (P12).

DISCUSSION

Our qualitative analysis of parental decision-making preferences in emergent paediatric surgical settings has identified several key themes that can help paediatric surgeons promote parental involvement in ways that may facilitate trust and minimize decisional conflict and regret. Specifically, our work offers unique insight into parental perspectives on the relative value of autonomy, the perceived meaning of shared decision-making and the ways in which parents prefer to communicate with surgeons. Interpretation of these themes guides specific suggestions for how surgeons may assure meaningful parental engagement in surgical decision-making.

Much has been written about the prioritization of patient autonomy in healthcare decision-making.[2,14] Some ethicists argue that physician efforts to guide patients towards particular clinical decisions violate respect for patient autonomy.[15] Physicians have expressed concern that this seemingly rigid emphasis on patient autonomy creates encounters in which clinicians are expected to outline a spectrum of options that the patient may choose among as opposed to providing a recommendation.[16] Our findings suggest that parents of paediatric surgical patients value recommendations from the surgeon when considering emergent surgery for their children. The parents we interviewed describe that the recommendations make them feel more confident and involved in caring for their children. They emphasize that the recommendation provides a concrete scaffolding to guide the further pursuit of the knowledge they need to participate in the care of their children in an informed way—without the burden of decision-making. This finding supports an emerging body of literature that suggests that patients may prefer more guidance from physicians during decision-making as opposed to a greater emphasis on autonomy.[8,17,18]

Over the past several decades, SDM has been highlighted as a process that can prioritize patient autonomy by encouraging patients and physicians to work together to develop care plans that account for patient goals and values while inviting patients to participate in decision-making at a level they deem most appropriate.[2] Studies using the Degner control preference scale, which evaluates a patient's desired level of involvement along a continuum of autonomous patient decision-making to provider-based decision-making, suggest that most respondents favour some degree of collaboration with providers as opposed to a highly active or highly passive role in decision-making.

[19–21] While this concept of shared decision-making seems relatively straightforward to apply to clinical encounters, few studies have investigated what it means to patients to truly share in the surgical decision-making process.[22] Clarifying our understanding of how parents interpret the goals of SDM can help refine our approach to surgical counselling. The parents we interviewed emphasize that it is engagement in open discussion with the surgeon, as opposed to final decision-making autonomy, that defines shared decision-making. The parents in our study felt that they had engaged in SDM even though they unanimously felt that there were no decisions to be made. This apparent disconnect reinforces the notion that when considering emergent operations for their children, engagement and inclusion in the decision-making process are what parents desire—not final decision-making autonomy. Shared decision-making frameworks that overly prioritize parent autonomy thus seem inappropriate in the context of emergent paediatric surgery. The emphasis on involvement in the decision-making process has been highlighted in elective paediatric surgery decision-making[5,6]; however, the finding that this decision-making preference exists for parents considering emergent operations for their children has not been widely described.

Identification of the specific behaviours that help parents feel most involved in the decision-making process is a critical first step in assuring that surgeons promote the inclusion of these behaviours. The parents we interviewed expressed that the opportunity to ask questions about their child's care gave them a ‘valued voice’ even when they felt there was no specific decision to be made. This emphasizes the importance of providing ample opportunity for parents to ask questions throughout the decision-making process. Work by Pecanac et al.[23] suggests that patients may benefit from targeted guidance regarding how to assure pertinent, meaningful questions are asked during the surgical consultation. In their analysis of the types of questions patients asked before high-risk surgical procedures, this group found that patients focused upon logistical or technical questions such as whether the incision would be closed with sutures or staples, how to wash their hair postoperatively and vehicle parking options during their hospital stay.[23] While it is certainly important to consider all patient concerns, one may argue that time spent in consultation with the surgeon is best utilized to facilitate discussions of more critical issues such as complications of the procedure, risk of disease recurrence or risk of morbidity or mortality. Future work to develop a decision-support tool that prepares parents to effectively ask questions, and thus exercises their ‘valued voice’, during surgical consultation may improve the decision-making process for parents of children undergoing emergent operations.

A Question Prompt List (QPL) may be a well-suited decision-support tool for parents. QPLs are structured lists of questions that patients can use to identify meaningful questions to ask during discussions with clinicians.[24] QPL use has been shown to improve communication across many healthcare disciplines, but QPLs have not been evaluated in paediatric surgery.[25–27] Further work to identify the specific details parents and surgeons deem most critical to include during the decision-making process will help guide the development of a QPL that parents can use when discussing emergent surgery for their children. The QPL may need to be modified to address specific operations and pathologies; however, many of the fundamental questions (What are the risks of surgery? Are there alternatives to this surgery? etc.) would likely be relevant regardless of the operation being performed. Consideration of the distinction that the parents we interviewed made between seeking information to promote informed involvement in the care of their children as opposed to seeking information to promote decision-making preparedness will also help guide the intentional construction of a QPL. Analysis of surgeon preferences on the decision-making process for emergent paediatric surgery will further aid in the meaningful construction of such a decision-support tool and facilitate its integration into surgical practice.

Our work also demonstrates that while parents emphasized the importance of direct conversation with their child's surgeon to learn about the treatment, they also valued other information sources. Specifically, the parents we interviewed highlighted that reviewing written and graphical information (drawings, brochures, figures, etc.) helped promote their involvement in the decision-making process. Additionally, all parents in our study relied on online resources to supplement their understanding of their child's illness. The parents we interviewed expressed mixed emotions about whether surgeons approved of their online exploration. This suggests that an opportunity exists for surgeons to guide online searches by providing reliable websites or preferred social media groups to parents. Such guidance may both quell parents' concerns that they are ‘not supposed to’ seek information online as well as help

direct them to the most helpful and accurate online resources.

The parents we interviewed also suggested that increased opportunity for virtual communication adjuncts would facilitate their engagement in the decision-making process. Specifically, parents suggested that having a virtual presence during surgical rounds (when they were not able to be physically present) would promote enhanced communication with surgeons. The COVID-19 pandemic has brought with it increased focus on opportunities for telemedicine and virtual communication with patients and families.[28] Further exploration of how virtual communication platforms can be integrated into surgical counselling may yield insight into how to most effectively integrate these platforms into the care of paediatric surgical patients.

Parents in our study also emphasized the importance of advocacy—both for their own children as well as for families of other children with similar surgical issues. Parents' advocacy for their children was often subtle, such as being present for rounds and asking questions. However, some parents described engagement in more direct advocacy to prevent incorrect doses of medication from being given to their children or medicines from being given at incorrect times. Multiple parents highlighted the importance of advocating for other families by means of social media or hospital-sponsored programmes. Given the importance that parents place on these opportunities, surgeons should consider more directly providing parents with opportunities to advocate for their children as well as for families with children with similar surgical problems.

Our study has several limitations. Parent perspectives were solicited from a single, Midwestern, academic centre thereby limiting the generalizability of our results. Additionally, while the opportunity to interview was offered to both parents, only mothers volunteered to be interviewed. The inclusion of fathers or other guardians would increase the depth and generalizability of our analysis. Our work is also limited in that we did not solicit parent race, ethnicity, familial characteristics or socioeconomic status. This study was not designed to investigate thematic differences based on these variables, but subsequent studies should address these important factors. The voluntary nature of parental participation in our study may also have skewed results, in that those parents with less favourable attitudes towards the healthcare team may have been less likely to participate. Parents who chose to allocate their time to this study may also not be representative of all parents whose children are cared for in our institution and given that the researchers were from the same institution at which the children received surgical care, parents may have felt social pressure to respond positively during the interviews. Additionally, 11 of 12 parents in our study had living children at the time of the interview. The positive outcome for these children may have prompted parents to speak more highly of clinicians regardless of the quality of communication. Further, although grounded in prior work that evaluated parental interpretations of the urgency of given paediatric surgical operations,[7] our classification of the selected clinical settings (ECMO, NICU and cancer) as emergent is relatively subjective. One must also consider that identification of Theme 2 (‘Lifesaving mode’: parents felt there were no decisions to be made) may have occurred due to incomplete informed consent discussions in which clinically reasonable options to transition to palliative care or forego surgical intervention were not offered. Given the diversity of the clinical encounters as well as the fact that patients of all paediatric surgeons at the institution were included in the analysis, it seems unlikely that these options would have been omitted during every informed consent discussion. It seems more likely that parents may have considered options resulting in the likely death of their children to be intolerable even if the options may have been clinically and ethically reasonable (i.e., choosing not to undergo ECMO cannulation).

Parental recall bias may also have impacted our data in that we conducted interviews with parents at variable times following their child's surgery. Standardizing the length of time between surgery and the interview may limit this bias. However, this approach may impair our ability to recruit a sufficient number of parents for interviews. Our patient cohort also had significant clinical heterogeneity. A more specific focus on patients with a single diagnosis who underwent a particular procedure may have yielded more precise data. Additionally, in our attempt to focus specifically on the interaction between parents and surgeons, we failed to explore the impact of other care providers in the decision-making process. Although beyond the scope of the current study, future work should address how parents' interactions with oncologists, neonatologists and other care providers impact surgical decision-making.

Finally, we did strive to ensure internal validity through the inclusion of three different interviewers with varying

nonclinical backgrounds, interviewing multiple parents, inductive reasoning during analysis, independent coding of each interview transcript by three research team members and use of verbatim quotes to justify identified themes; we did not discuss the identified themes with participants or seek participant feedback on the accuracy of our themes. The inclusion of such participant checking should be considered in subsequent studies.

CONCLUSIONS

Our qualitative analysis of parental decision-making preferences during emergent surgical decision-making demonstrates that parents value guidance from surgeons by way of a recommendation. The parents we interviewed believe that inclusion in their children's care, as opposed to decision-making autonomy, is what defines shared decision-making. They feel included when given the opportunity to ask questions and use the information that they gather to serve as a ‘valued voice’ to advocate for their children as well as for families of children with similar surgical conditions. The parents in our study appreciate physical supplements to verbal communication, and they value the opportunity for telemedicine or virtual engagement when direct, in-person communication is not possible. Integration of these findings into surgical counselling will allow paediatric surgeons to modify their counselling for emergent procedures to optimize parental preferences. Such changes will promote parental involvement and are likely to facilitate trust and minimize decisional conflict and regret.

AUTHOR CONTRIBUTIONS

Conception and design of the study: Erica M. Carlisle, Laura A. Shinkunas, Heather S. Reisinger. *Acquisition of data* : Erica M. Carlisle, Laura A. Shinkunas, Emily Ruba, Caleb J. Klipowicz, Maxwell T. Lieberman. *Analysis and interpretation of data*: Erica M. Carlisle, Laura A. Shinkunas, Emily Ruba, Maxwell T. Lieberman, Richard M. Hoffman, Heather S. Reisinger. *Drafting and revision of the article*: Erica M. Carlisle, Laura A. Shinkunas, Emily Ruba, Caleb J. Klipowicz, Maxwell T. Lieberman, Richard M. Hoffman, Heather S. Reisinger. *Final approval of the version to be submitted*: Erica M. Carlisle, Laura A. Shinkunas, Emily Ruba, Caleb J. Klipowicz, Maxwell T. Lieberman, Richard M. Hoffman, Heather S. Reisinger. All authors read and approved the final manuscript.

ACKNOWLEDGEMENT

Dr. H. S. Reisinger would like to acknowledge funding from the National Center for Advancing Translational Sciences of the National Institutes of Health under Award Number UL1TR002537; she received to support her contribution to this research. The funding body had no role in the design of the study or collection, analysis, interpretation of the data or writing of the manuscript. This research did not receive any specific grant from funding agencies in the public, commercial or not-for-profit sectors.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

The study was approved by the University of Iowa Institutional Review Board (#201905811).

1APPENDIXCONSOLIDATED CRITERIA FOR REPORTING QUALITATIVE RESEARCH (COREQ) CHECKLIST

No.	Item	Description
Domain 1: Research team and reflexivity		
Personal characteristics		
1.	Interviewers(s)	Laura A. Shinkunas (L. A. S.), Caleb J. Klipowicz (C. J. K.), Maxwell T. Lieberman (M. T. L.)

2.	Credentials	C. J. K. (PhD candidate in anthropology); M. T. L. (PhD candidate in anthropology); L. A. S. (MS)
3.	Occupation	C. J. K. and M. T. L.: graduate research assistants; L. A. S: research specialist
4.	Gender	C. J. K. and M. T. L.: male; L. A. S.: female
5.	Experience and training	C. J. K.: Educational background in medical anthropology. Practical experience in qualitative research and teaching; M. T. L.: Educational background in cultural anthropology. Practical experience in qualitative research and teaching. L. A. S.: Educational background in psychology and bioethics. Practical experience in qualitative research.
Relationship with participants		
6.	Relationship established	No
7.	Participant knowledge of the interviewer	Participants knew that C. J. K. and M. T. L. were working as graduate research assistants with Erica M. Carlisle, a paediatric surgeon in the Department of Surgery.
8.	Facilitator characteristics	Participants knew that C. J. K. and M. T. L. had social science backgrounds.
Domain 2: Study design		
Theoretical framework		
9.	Methodological orientation and theory	Thematic content analysis (Bernard, 2016)
Participant selection		
10.	Sampling	Purposive sampling
11.	Method of approach	Email
12.	Sample size	12 participants
13.	Nonparticipation	Not applicable
Setting		

14.	Setting of data collection	Interviews were conducted by phone
15.	Presence of nonparticipants	NA
16.	Description of sample	Not collected
Data collection		
17.	Interview guide	Provided as supplemental material
18.	Repeat interviews	No
19.	Audio/visual recording	Audio recording
20.	Field notes	No
21.	Duration	Mean = 28.7 (range:25–65 min)
22.	Data saturation	Yes
23.	Transcripts returned	No
Domain 3: Analysis and findings		
Data analysis		
24.	Number of data coders	Three
25.	Description of the coding tree	Provided as supplemental material
26.	Derivation of the themes	Inductive and deductive (themes were derived from both previous literature and the interview data)
27.	Software	MAXQDA 2022 (Verbi Software, 2021)
28.	Participant checking	No
Reporting		
29.	Quotations presented	Yes
30.	Data and findings consistent	Yes
31.	Clarity of major themes	Yes

32.	Clarity of minor themes	Not applicable

2APPENDIXINTERVIEW GUIDE. SEMISTRUCTURED INTERVIEW GUIDE

Interviewer:

Date:

Participant Code:

Illness Narrative Overview

Could you describe for me the events that led up to your child's surgery? (Diagnosis, series of events, age of the child, etc.)

Could you tell me about your child's operation? (e.g., What did the doctors do? What did you do during the procedure?)

What happened after the surgery?

How has your child been since then?

Decision-Making Process

What did the doctor(s) tell you about the condition/surgery when you met with them? (e.g., did they offer you information about the surgery or condition, etc.?)

What were some of the decisions you had to make about your child's surgery?

What did you end up deciding to do? How did this make you feel?

How did you reach this decision? Who did you talk to help you make these decisions?

Do you think this process of reaching a decision would have been different if the treatment was for you and not your child?

Surgeon–Parent Interactions

How would you describe your interactions with the surgeon during the decision-making process?

Did the surgeon make a recommendation to you about surgery?

If Yes—Was it important/helpful to you for your surgeon to offer you a recommendation?

How do you feel about the guidance the surgeons/doctors offered you? (e.g., Do you have strong feelings about it? Did they meet your expectations?)

How might your feelings be different about your surgeon's guidance if your child's surgery was less of an emergency?

Reflection and Recommendations

Overall, how do you feel about this experience? (e.g., satisfied? Regret?)

How do you feel about the outcome of the surgery?

Looking back, do you wish anything had been done differently?

After this experience, how would you recommend surgeons work with parents that are in similar situations to the one you faced?

Is there anything that you would change or improve about the process of making decisions about surgery for your child?

Other/Wrap-up

Finally, in medicine, there is currently a push for doctors and surgeons to share in the decision-making process with their patients. Could you tell me, what ‘shared decision making’ mean to you?

Is there anything else you think is important that I did not ask you about that you would like to share?

Do you have any questions for me?

DETAILS

Subject:	Neonates; Review boards; Data analysis; Surgery; Pediatrics; Qualitative analysis; Decision analysis; Intensive care; Cancer; Decision making; Neonatal units; Children &youth; Group decision making; Patients; Oxygenation; Surgeons; Comparative analysis; Children; Parents &parenting; Sampling techniques; Content analysis; Physicians; Saturation; Interviews; Qualitative research; Parents; Health care; Extracorporeal membrane oxygenation; Voice; Autonomy
Location:	United States--US; Iowa
Company / organization:	Name: University of Iowa; NAICS: 611310
Identifier / keyword:	decision-making; emergent; paediatric surgery
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	531-541
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-12-08

Milestone dates:	2022-10-21 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-07-01 (manuscriptReceived); 2022-12-08 (publishedOnlineEarlyUnpaginated); 2022-11-23 (manuscriptAccepted)
Publication history :	
First posting date:	08 Dec 2022
DOI:	https://doi.org/10.1111/hex.13686
ProQuest document ID:	2767102232
Document URL:	https://www.proquest.com/scholarly-journals/valued-voice-qualitative-analysis-parental/docview/2767102232/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the "License"). Notwi thstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-04-24
Database:	Public Health Database,Publicly Available Content Database

Service-user experiences of an integrated psychological intervention for depression or anxiety and tobacco smoking in improving access to psychological therapies services: A qualitative investigation into mechanisms of change in quitting smoking

Kim Fredman Stein [1] ; Sawyer, Katherine [2]
; Daryan, Shadi [2]
; Allen, Jennifer [3] ; Taylor, Gemma [2]

[1] DClinPsy—Department of Psychology, University of Bath, Bath, UK [2] Addiction and Mental Health Group (AIM), Department of Psychology, University of Bath, Bath, UK [3] Department of Psychology, University of Bath, Bath, UK

ABSTRACT (ENGLISH)

Introduction

High smoking prevalence leads to increased morbidity and mortality in individuals with depression/anxiety.

Integrated interventions targeting both smoking and mood have been found to be more effective than those targeting smoking alone, but the mechanisms of change of these interventions have not been investigated. This qualitative study aimed to understand participants' experiences of the mechanisms underlying change in smoking behaviour following an integrated cognitive behavioural technique-based intervention for smoking cessation and depression/anxiety.

Methods

This study was embedded within an ongoing randomized-controlled acceptability and feasibility trial (http://www.isrctn.com/ISRCTN99531779). Semistructured interviews were conducted with 15 IAPT service users. Data were analysed using thematic analysis. During the interviews, participants were asked open-ended questions about their quitting experience and perception of how the intervention aided their behaviour change.

Results

Five themes were identified. *Acquiring an increased awareness of smoking patterns*: participants described an increased understanding of how smoking was contributing towards their mental health difficulty. *Developing individualized strategies*: participants described acquiring ‘a couple of tricks up your sleeve’ that were helpful in making smoking cessation feel more ‘manageable’. *Practitioner style as ‘supportive but not lecture-y’*: participants expressed how important the therapeutic alliance was in helping change their smoking behaviour. *Importance of regular sessions*: participants expressed the importance of ‘having someone that's checking in on you’. *Having the opportunity to access the intervention at ‘the right time’*: participants described the intervention as the ‘push’ that they ‘needed’.

Conclusions

Participants identified key factors towards smoking behaviour change. Perceived increased awareness of how smoking negatively impacted participants' mental health, and the opportunity to be offered smoking cessation treatment in a ‘non-judgemental’, ‘supportive’ environment, with regular sessions and individualized strategies contributed to successful smoking cessation outcomes. If similar results are found in more diverse samples, these aspects should be embedded within integrated interventions for smoking cessation and depression/anxiety.

Patient or Public Contribution

Persons with lived experience of depression, anxiety and tobacco addiction contributed towards the design of the interview schedule, participant information sheets and the debriefing process. This was to ensure that interview questions were relevant, nonjudgemental and acceptable for those who did not manage to quit smoking.

FULL TEXT

INTRODUCTION

Individuals with depression/anxiety are twice as likely to smoke than those without depression/anxiety[1]; this disparity increases mortality in people with depression/anxiety compared to the general population (mortality rate ratio, 1.92 [95% confidence interval (CI): 1.91–1.94]).[2] Despite being equally as motivated to quit, this population smokes more heavily, are more addicted and are less likely to successfully quit than the general population.[3–5] There are many reasons for the high smoking rates in this population; for example, they are less likely to be prescribed smoking medicines,[6] and it is widely believed that smoking cessation can worsen mental health.[7,8] However, a recent Cochrane review found evidence that smoking cessation can improve anxiety/depression compared to continuing smoking (standardized mean difference, −0.31 [95% CI: −0.40 to −0.22]).[9]

Another Cochrane review found that smoking cessation interventions offered alongside mood management support led to higher cessation rates compared to smoking cessation interventions alone for people with depression (risk ratio 1.47, 95% CI: 1.13–1.92),[10] indicating the importance of integrated interventions to improve smoking and mood outcomes. However, this review did not shed light on the mechanisms that led to behavioural change.

Integrated interventions may be more effective for quitting smoking for various reasons. For example, cognitive behavioural techniques (CBT) could alter unhelpful beliefs about the relationship between smoking and depression/anxiety (e.g., ‘smoking helps my mood’),[11] which could promote cessation and prevent relapse. Such an intervention could also promote alternative strategies for managing depression/anxiety to smoking.[12] For example, behavioural activation aims to increase pleasurable activities.[13] It is also possible that the therapeutic alliance in

psychological interventions could help facilitate behaviour change.[14,15]

There are some evidence-based models that we can use to investigate mechanisms of behavioural change. The Capability, Opportunity, Motivation-Behaviour model (COM-B model)[16] suggests that a person's capability (i.e., a person's physical and psychological capacity), opportunity (i.e., external factors that facilitate behaviour) and motivation are involved in behavioural change. The Smoking, Not smoking, Attempting to quit, Planning to quit model (SNAP)[17] suggests that smoking cessation involves moving through the four stages of (1) smoking, (2) attempting to quit, (3) planning to quit and (4) not smoking. The misattribution hypothesis suggests that smokers misattribute nicotine withdrawal symptoms of stress or anxiety/depression and believe that smoking alleviates symptoms of mental health difficulties.[18,19]

Understanding mechanisms of behavioural change and how they fit into evidence-based frameworks could improve our understanding of the active intervention components and help identify therapist characteristics that optimize therapeutic benefits, potentially leading to more effective and streamlined interventions.

Addiction research has been criticized for excluding the patient's view and focussing on intervention techniques rather than intervention mechanisms.[15] Therefore, as part of a wider trial,[20] the current study used qualitative interviews following an integrated intervention to investigate individuals' subjective experiences of the mechanisms underlying change in their smoking behaviour.

In this qualitative investigation, we aimed to use evidence-based models of behaviour change and behavioural intervention development[16–19] to explore participants' subjective experience of mechanisms of change in smoking cessation.

METHODSDesign

Our study was preregistered (https://osf.io/nfgu4/) and was part of an ongoing RCT (ESCAPE, http://www.isrctn.com/ISRCTN99531779). The preprint is available via medRxiv (https://doi.org/10.1101/2022.03.23.22272703). We followed COREQ reporting guidelines in writing this manuscript.[21] We used qualitative in-depth interviews to explore participants' perceptions of change.[22]

Participants

We approached 19 patients and conducted interviews with 15 who took part in the ESCAPE trial intervention arm and had attended three or more intervention sessions. Reasons for nonparticipation were not recorded. Participants in the intervention arm received a CBT-based smoking cessation intervention that was integrated into routine IAPT care for depression/anxiety[9,20] (Supporting Information: A). Improving Access to Psychological Therapies (IAPT) is a primary care service in the UK National Health Service providing evidence-based psychological therapies for depression/anxiety. Trial inclusion criteria were self-reported daily smokers of at least 1 year, aged ≥18 years, met thresholds for depression and/or anxiety (clinician-administered PHQ-9 score ≥10 and/or GAD-7 ≥8 scores) and were about to start IAPT treatment. Individuals were excluded if they did not have the capacity to give informed consent, or if they were pregnant or breastfeeding at trial entry.

Procedure and recruitment

Recruitment procedures for the ESCAPE trial can be found in the trial protocol (https://osf.io/nfgu4/). Purposive sampling was used to recruit participants for follow-up interviews about the intervention. During 3- and 6-month telephone ESCAPE trial follow-ups, participants were asked if they would like to be interviewed about their experiences in the study and attempting to quit. We recruited participants until information power was reached.[23] Information power is more suitable for pragmatic applied health research than data saturation. ‘Data saturation’ was originally developed for grounded theory analysis.[23,24] Participants gave oral consent before the interview, and again as an audio-recorded consent statement.

Interviews

The interview schedule (Supporting Information: B) aimed to explore participants' experiences of mechanisms of change. Interviews lasted 30–45 min and were embedded within a longer interview schedule, lasting no more than 60 min, which also investigated the acceptability of the intervention and trial procedures (the additional findings will be presented elsewhere). Interviews were conducted by K. F. S. and K. S.

Analysis

Data were transcribed using a University-approved service. Fifty percent of the audio data were checked against the transcripts to ensure fidelity. The data were analysed using a reflexive thematic analysis following the steps outlined by Braun and Clarke.[25,26] Reflexive thematic analysis was used as it is not tied to theoretical or epistemological approaches and can be used both inductively and deductively. A critical realist approach was adopted; meaning was viewed as both socially constructed and relating to individuals' experiential reality.[27] Braun and Clarke's[25] guidance for the six phases of thematic analysis was followed: (1) familiarization with the data, (2) coding, (3) generating themes, (4) reviewing themes, (5) defining and naming themes and (6) writing up. A combined inductive and deductive approach to coding was adopted, whereby codes and themes were developed both from the existing theory and the data. The framework of deductive codes was constructed based on the COM-B model (Supporting Information: C and Table S1), which has shown good reliability for categorizing components of behaviour change interventions.[28] Coding was conducted manually. Participant IDs were used throughout, and any potentially identifying information was removed.

Once the data were coded, all relevant coded data extracts were collated and organized into potential themes and subthemes. A series of tables were developed to explore possible relationships between codes, themes and subthemes. Potential themes and subthemes were then reviewed, refined and assessed according to the criteria of internal homogeneity and external heterogeneity.[29] Analysis was viewed as an iterative process; the researcher at times returned to previous stages rather than following a rigidly linear process. A self-reflexive stance was used throughout data collection and analysis to increase awareness of and limit the impact of the researcher's potential biases and assumptions.

Patient and public involvement

Persons with lived experience of depression, anxiety and tobacco addiction contributed to the study design, the design of study materials and the debriefing process. The research aims and design were reviewed by the UK Centre for Tobacco and Alcohol Studies Smokers' Panel. In general, the study's concept was well received, understood and thought to be an important area of research. We consulted with the UK Centre for Tobacco and Alcohol Studies Smokers' Panel and the Elizabeth Blackwell Institute's Patient and Public Involvement Panel to develop the interview schedules.

RESULTS

Fifteen participants were recruited (Table 1; Supporting Information: D and Table S2).

Table 1 Summary of participant characteristics

Participant characteristics	*N*
Gender	
Men	6
Women	9
Race/ethnicity	
White British	15
Highest level of education	
Higher degree	4

Degree	6
A-Level equivalent	1
GCSEO-grade/equivalent	1
Apprenticeship	1
Other vocational	2
Age (mean)	42.6
Age (range)	26–67

Note: *N* = 15 total.

We identified 5 themes and 12 subthemes (Table 2) and presented them with illustrative quotes (Table 3).

Table 2 Themes and subthemes

Themes	Subthemes
1. Increased awareness of smoking patterns	Increased awareness of triggers
	Increased awareness of smoking as a maintaining factor in vicious cycles
	Increased awareness of the need for alternatives
2. Developing individualized strategies: ‘What's in your toolkit?’	Finding alternative ways to fulfil the function of smoking
	Strategies enhance sense of capability
3. Practitioner style as ‘supportive but not like lecture-y’	Empathy and a nonjudgemental stance enabled the disclosure of setbacks
	Confronting avoidance
	Guided discovery rather than directive
4. ‘Having someone that's checking on you’: Importance of regularity of sessions	Opportunity for regular support and troubleshooting
	Feeling ‘answerable’
5. Having the opportunity at ‘the right time’	‘Sometimes good things come out of bad places’

	Life felt too difficult

Table 3 Illustrative quotes from participants

Theme	Subtheme	Quote(s)
Theme 1: Increased awareness of smoking patterns	Increased awareness of triggers	Quote 1: ‘Once I started thinking a bit more about why I needed to do that, or why I was doing it, and if you're a bit more conscious of that element you can almost pre-empt it and divert it’ (Participant 4).
		Quote 2: ‘Being aware of how you're spending your time and what is causing you to go, Oh, I need a cigarette (…). It was mapping out your day and working out when the trigger events were’ (Participant 14).
	Increased awareness of the need for alternatives	
	Increased awareness of smoking as a maintaining factor in vicious cycles	Quote 3: ‘It was like a coping mechanism that, in the short term, seemed to help, but in the long- term then I'd worry about it (…), it was making me feel worse’ (Participant 3).
		Quote 4: ‘You have the cigarette, you feel a bit better but when the nicotine is less effective (…) you feel worse again and you have the cravings and you feel more stressed and more anxious again after it's worn off, which I never really thought about before’ (Participant 5).
		Quote 5: ‘I was conscious of my weight and thought, “If I smoke, I'm going to have less of an appetite” (…) I'm a healthy weight and the way I was thinking was, and acting when I was younger was making me underweight and unhealthy, and I think it was just like having a realisation of that’ (Participant 13).

Theme 2: Developing individualized strategies: ‘What's in your toolkit?’	Finding alternative ways to fulfil the function of smoking	Quote 6: ‘I'd be anxious when I came home from school and I still had a lot of work to do, and I'd have a wine and sit in the courtyard and smoke because that helped, but in the short term. So, we looked at what could you do differently, so could you go for a run, could you go for a swim, could you play the piano, and things like that. So, then it gave me a bit more, changed the behaviour, rather than just stopped the smoking’ (Participant 3).
	Strategies enhance sense of capability	Quote 7: ‘The strategies are really helpful- rather than just going, “OK, I just will stop this habit I've had since I've been, 21 years,” that's a bit impossible, well, I found it a bit impossible before’ (Participant 3).
		Quote 8: ‘You could see that you were doing better, when they did the carbon monoxide test’ (Participant 1).
		Quote 9: ‘It was like, “wow, look how much money I've saved but also how much time I've saved”’ (Participant 3).
		Quote 10: ‘When I had the Champix, because it made the actual smoking make me feel sick, you couldn't smoke, it completely put you off it’ (Participant 2).
		Quote 11: ‘Because I wasn't really craving it because I sort of had the nicotine, but yeah, it was a lot easier and easier to manage at work. I was calmer, not trying to give up and work and get stressed, so that was really helpful’ (Participant 1).

		Quote 12: ‘I would not smoke for a couple of days but then I'd just find myself getting drawn back into doing it, through having colleagues that smoked or friends that smoked when we were out’ (Participant 13).
		Quote 13: ‘Then they're going to be aware of me doing it, and not be likely to offer me a cigarette (…) it did work and I think it did help having that’ (Participant 13).
Theme 3: Practitioner style as ‘supportive but not like lecture-y’	Empathy and a nonjudgemental stance enabled the disclosure of setbacks	Quote 14: ‘If you've got somebody that you're not connecting with (…) And that doesn't seem to be understanding of you, you're not going to take their advice because it's going to be: you don't understand where I'm coming from, how can you expect me to do something when you don't understand? Showing no empathy for what's going on. It makes you put a wall up, I suppose, and you get quite defensive. Whereas if you've got somebody that you get on with, you can open up to, that seems to understand what you're going through, the wall comes down so you're more likely to take on-board what's said’ (Participant 2).
		Quote 15: ‘She was very understanding of what I was going through as a whole anyway but it just meant that when it came to the smoking bit, it helped make that a more comfortable environment to discuss it’ (Participant 13).

		Quote 16: ‘The temptation is to be very self-critical when something you try doesn't work, and actually having somebody say, “It doesn't matter, these things happen, it's absolutely fine for it not to have worked this time, let's just have a reboot,” something about what it was that caused the hiccup last time, “See if we can try and avoid that next time” (…) There was no judgement or condemnation’ (Participant 4).
		Quote 17: ‘As soon as I turned around and said, “Yeah, I haven't had one” she said, “Oh that's fantastic!” Even just that little bit of encouragement was really good’ (Participant 5).
	Confronting avoidance	Quote 18: ‘She wasn't avoiding talking to me about, which was brilliant, because, “Let's talk about the smoking,” so I knew I had to confront it, it wasn't, “You said you'd stop smoking last week and you didn't so we're just going to ignore it,” it was, “What happened this time that made you feel…?,” so it was examining it, which is great’ (Participant 6).
	Guided discovery rather than directive	Quote 19: ‘I feel it was subtly getting me to make the decisions and getting me to make the choices (…) it was very much guiding rather than leading’ (Participant 4).

		Quote 20: ‘I guess the only thing I would suggest is if they had a bit more time for calls so they didn't feel …because you know that they're getting stressed too. The person who's supposed to be counselling you is getting stressed because they've got to get onto the next thing, and you can sense that they're trying to interpret what you're saying so they can get onto the next because they have six more questions and there's only four minutes left’ (Participant 10).
Theme 4: ‘Having someone that's checking on you’: Importance of regularity of sessions	Opportunity for regular and frequent support and troubleshooting	Quote 21: ‘If there was a blip, which there were, they didn't fester for too long before I was able to talk about it or get some more advice’ (Participant 4).
		Quote 22: ‘Knowing you're going to phone and speak to someone every week about it made it that bit easier for me to carry on and do’ (Participant 5)
		Quote 23: ‘It was the fact that I had to report into someone and look into why, that was what helped me rather than, so I think I didn't do it before because I just felt like it was too big a job to do on my own’ (Participant 3).
	Feeling ‘answerable’	Quote 24: ‘Having somebody checking on you, you've got to tell them that you've smoked, and you feel like you've let somebody else down rather than just yourself’ (Participant 7).
Theme 5: Having the opportunity at ‘the right time’	‘Sometimes good things come out of bad places’	Quote 25: ‘If I'm going to change to my whole mental state, I might as well try and change everything at the same time, and it just seemed a perfect opportunity to give it a go’ (Participant 8).

	Life felt too difficult	Quote 26: ‘It just seemed to be going from one trauma to the next, and I was just running out of energy to deal with everything, and quitting smoking just became very low on my list of priorities (…) I don't think anything would have helped, because I had to deal with the issues I was dealing with, and till I'd dealt with those I couldn't contemplate anything else’ (Participant 2).
		Quote 27: ‘I've still got the patches because I didn't use them all, so I have got some backup almost if say tomorrow I go, “That's it,” at least I've got that (…), it's almost like I'm in constant preparation because they're there, so that's good’ (Participant 6).

Increased awareness of smoking patterns

Participants described an increased awareness of smoking patterns as a key step in facilitating quitting smoking. They described a shift in their awareness, from smoking being automatic, where they would ‘smoke and not necessarily know I was smoking’ (Participant 2), to being more ‘analytical’ about their smoking patterns (Participant 4). This ‘understanding why you smoke’ (Participant 3) seemed to facilitate participants' sense of psychological capability to not smoke (Table 3, Quote 1).

One aspect of increased awareness was an improved awareness of smoking triggers (Table 3, Quote 2). One of the most common triggers for smoking was strong negative emotions. Participants expressed that through the intervention, they gained increased awareness of the link between strong negative emotions and smoking: ‘when I talk about it, I do smoke because I'm unhappy and because it's a distraction’ (Participant 11). Some expressed the realization that smoking was often an attempt to cope with negative emotions: ‘before, I felt that if I was stressed, I could step away and have a fag and that would help’ (Participant 5). This response reflects the misattribution hypothesis.[18] The increased awareness of this misattribution seemed to result in a change in thinking about the relationship between smoking and stress, as well as providing alternative options for managing stress: ‘I recognised that and that's more just stepping out of the situation and just chilling for a bit’ (Participant 5). Therefore, increased awareness of the need for alternatives to smoking for managing emotions appears to be a mechanism of change in reducing smoking.

Participants also described greater awareness of how smoking contributed to the maintenance of a ‘vicious cycle’ (Participant 13) of smoking and stress (Table 3, Quote 3). Several participants identified an increased awareness of the specific role of nicotine withdrawal in maintaining unpleasant feelings (Table 3, Quote 4).

This increased awareness of how smoking was contributing to difficulties in their mental health appeared to increase motivation to change. One participant described smoking to control her appetite; increased awareness of the unhelpful nature of her beliefs about her weight helped to facilitate stopping smoking (Table 3, Quote 5). This suggests that there is a range of beliefs that drive smoking, and increased awareness of these allows them to be challenged. Moreover, increased awareness that smoking appears to help with strong negative emotions in the short term, but maintains them in the long term, seemed to increase motivation to quit smoking, and understanding of the need to develop other ways to cope with emotions.

Developing individualized strategies: ‘What's in Your Toolkit?’

All participants described developing a personalized set of strategies that they found helpful in reducing smoking, described as acquiring ‘a couple of tricks up your sleeve’ (Participant 10). These strategies included goal setting, setting a quit date, problem-solving, removing access to cigarettes, communicating with others about quitting, positive self-talk, distraction, breathing and mindfulness techniques and using smoking cessation resources such as apps. It seemed particularly important to develop ‘substitutes’ (Participant 12) that fulfilled the function of smoking (Table 3, Quote 6).

Practical strategies provided by the intervention appeared to make the quit attempt feel more manageable and enhance participants' sense of psychological capability (Table 3, Quote 7). Similarly, Participant 13 specifically referred to ‘exercises for breathing and mindfulness and distraction techniques’ that helped her to feel capable: ‘No, I don't have to have a cigarette (…) I'm stronger, I can deal with that’. Several strategies also enhanced motivation, namely, goal setting, setting a quit date and using a pros and cons list, which helped to ‘cement’ motivation for change (Participant 4). Once participants noticed concrete signs of improvement resulting from these strategies, this reinforced their motivation to continue to implement them, as well as enhanced their sense of psychological capability (Table 3, Quotes 8 and 9).

Although smoking cessation medication was not viewed as helpful by everyone, some participants found it to be an important tool (Table 3, Quote 10). It seemed particularly helpful for managing the physiological nicotine cravings, which enhanced some participants' sense of their physical capability to quit (Table 3, Quote 11).

The strategy of communicating with others about quitting was viewed as important by several participants. They described being with others who were smoking as a trigger (Table 3, Quote 12). However, telling others about the quit attempt also helped (Table 3, Quote 13). This suggests that changing the social environment can facilitate behaviour change in smoking, consistent with social learning theory principles.[30]

Practitioner style as ‘Supportive but not Like Lecture-y’

Participants expressed how important the therapeutic alliance was in making changes to their smoking behaviour, particularly their sense of having the space to talk through their feelings and difficulties, with these being heard and not judged but met with compassion, curiosity and support. This contrasted with descriptions of other people in their lives who tended to adopt a more judgemental and directive approach towards their smoking and was often experienced by participants as unhelpful: ‘lecture doesn't have any impact’ (Participant 6). The IAPT practitioners' understanding and empathy seemed to enable participants' disclosure of when they had struggled with their quit attempt 1 week, as well as participants' openness and receptiveness to suggestions (Table 3, Quote 14).

Additionally, participants expressed a sense that IAPT practitioners' understanding of their mental health struggles helped them to feel more able to talk about smoking (Table 3, Quote 15). One participant expressed that the practitioner's nonjudgemental approach to setbacks was particularly important, given the participants' tendency to be self-critical (Table 3, Quote 16).

This nonjudgemental IAPT practitioner stance enabled participants to learn from setbacks that may otherwise have served as a trigger to give up on smoking cessation. Given that levels of self-criticism are often high in depression,[31] and that setbacks could be experienced as ‘an added failure’ (Participant 15), this therapeutic stance is likely to be particularly important in this population. Alongside viewing setbacks as an opportunity for learning, IAPT practitioners' encouragement enhanced participants' motivation to continue with their attempt to quit (Table 3, Quote 17).

Several participants found it helpful that the IAPT practitioner helped them to confront their avoidance of talking about smoking or attempting to quit (Table 3, Quote 18).

Participants also noted the IAPT practitioner's use of guided discovery to empower them to think through decisions themselves (Table 3, Quote 19). Sometimes, practitioner empowerment of participant decision-making took more explicit forms, such as giving options around the use of smoking cessation medication.

One participant expressed that an unhelpful experience was that he felt rushed at times, rather than the IAPT practitioner responding to him in a flexible and person-centred way (Table 3, Quote 20).

‘Having Someone that's Checking on You’: Regularity of sessions

Most participants expressed the importance of the regularity of sessions, which supported the development and maintenance of the therapeutic alliance. One helpful aspect of regular sessions was having the space to solve problems as they arose (Table 3, Quote 21). Regular check-ins seemed to enhance participants' sense of psychological capability compared to when they had tried to quit on their own due to the extra support and sense of accountability (Table 3, Quotes 22 and 23).

Feeling ‘answerable’ (Participant 4) or ‘responsible’ (Participant 11) to the IAPT practitioner each week appeared to enhance participants' motivation or even made them feel ‘obliged’ (Participant 13) to quit (Table 3, Quote 24). This sense of anticipatory guilt seemed to drive reduced smoking for some participants. This sense of accountability was not limited to the IAPT practitioner; one participant recruited friends and family to have ‘more people to check in on how I was doing’ (Participant 13).

Having the opportunity at the ‘Right Time’

Several participants talked about the combination of feeling that it was ‘the right time’ to change, and the integrated mental health intervention providing an opportunity, or the ‘push’ (Participant 5) they ‘needed’ (Participant 4). For these participants, mental health problems did not prevent it from being the right time: ‘although I was in a very bad place at that time, it was the right time’ (Participant 4). In fact, beginning therapy for their mental health in some cases appeared to signal their readiness to change (Table 3, Quote 25). However, a couple of participants expressed that although they valued the opportunity, stressful life events made quitting seem too much (Table 3, Quote 26). For another participant, it appeared that the intervention helped her to approach ‘the right time’, even though she was not there yet. This suggests that the intervention helped her shift from attempting to quit to planning to quit, a shift consistent with the SNAP model of smoking cessation[17] (Table 3, Quote 27). Thus, having the opportunity for support with smoking cessation enhanced her motivation ‘because it was offered (…) I probably was more motivated to do it, have a go’ (Participant 6).

DISCUSSIONSummary of findings

In this qualitative study, we investigated participants' subjective experiences of mechanisms underlying change in smoking behaviour. Participants reported that the integrated smoking cessation and mood intervention helped facilitate reductions in smoking through increased self-awareness of smoking patterns, and supported them in developing individualized behavioural and cognitive strategies to aid cessation. Participants stated that the regularity of support, the supportive, ‘non-lecture-y’ therapeutic style of IAPT practitioners and being offered the smoking intervention at the ‘right time’ (i.e., integrated with mental health support) all contributed to participants' sense of being able to make changes to the smoking. These findings further our understanding of the active ingredients and processes for behaviour change in this integrated intervention.

Study strengths and limitations

Participants were all white British; this limits the study's transferability to people of different backgrounds. Furthermore, the lack of a longer-term follow-up means that we could not show whether intervention mechanisms were maintained long-term. The sampling method may have contributed towards bias. Participants consisted of those from the ESCAPE trial who had attended three or more sessions, completed at least one follow-up and had self-selected to attend interviews. This population may have had more positive experiences of the intervention compared to those not sampled, which may not represent the experiences of all intervention arm participants. In addition, as participants were also part of a larger study that was not designed to investigate mechanisms of smoking cessation, it is unknown if these findings are transferable to the general IAPT population. However, studies of participants who were not selected to take part in an RCT are predictive of some of the findings in this study. For example, in another study of IAPT users, participants prospectively predicted that knowing about the mental health benefits of smoking cessation could help them to quit smoking.[12]

When interpreting qualitative results, it is important to acknowledge the researcher's background[32]; K. F. S. is a nonsmoker, whose primary orientation clinically is CBT. These factors are likely to have influenced the interviews and analysis, although research supervision was used reflexively throughout the process to consider the researcher's position and reflect on alternative perspectives. Finally, although persons with lived experience of

depression, anxiety and tobacco addiction contributed to the study design, the design of study materials and the debriefing process, our study would have further benefited from consulting with lived experienced persons in transcription, analysis and interpretation of the study findings. This is a limitation of our research.

This study has considerable strengths. According to Malterud's et al.[23] criteria for information power, the study had strong information power as the sample was very specific to the research question and the study had clear theoretical underpinnings. Moreover, including people with lived experience in the research design, (including the interview schedule), helped to ensure that questions were acceptable, relevant and nonjudgemental for those who did not manage to quit smoking. Past studies have shown that integrated interventions can be effective but have not explored how and why they work.[10] Our qualitative approach shed light on how participants made sense of what helped them reduce their smoking, situating these perceived mechanisms within the context of the participants' lives.

Comparison with the existing literatureIncreased awareness of smoking patterns

This theme reflects one mechanism underlying several behaviour change techniques identified in Michie et al.'s[33] taxonomy of behaviour change techniques for smoking cessation, such as prompting self-recording, facilitating an understanding of how lapses occur and identifying barriers to change. The current study improves our understanding of why these techniques can facilitate reductions in smoking. First, increasing awareness of the function of smoking for an individual can facilitate awareness of alternative ways of fulfilling this function. Second, increasing awareness of how smoking can contribute to a vicious cycle that worsens mental health by maintaining difficulties such as stress (through nicotine withdrawal), or worries about health in the long term, appeared to increase motivation to change. Participants expressed how the experience of smoking relieving the unpleasant symptoms of nicotine withdrawal can become overgeneralized, leading to the misattribution that smoking can relieve stress, consistent with the misattribution hypothesis.[9,18,19] Increased awareness of this resulted in a shift towards viewing smoking as exacerbating stress rather than relieving it, leading to an awareness of the need for alternative ways to cope with unpleasant emotions. Third, there are a range of beliefs that may drive smoking (including weight concerns), and that increasing awareness of these beliefs is an important first step towards addressing them. This theme is consistent with the traditional CBT theory, which emphasizes the role of increasing awareness of triggers and maintenance cycles,[34] and third-wave CBT approaches, such as Mindfulness-based Cognitive Therapy, which highlights the centrality of awareness in behaviour change.[35]

Developing individualized strategies: ‘What's in your toolkit?’

Most strategies mentioned by participants are included in Michie et al.'s[33] taxonomy of behaviour change techniques in smoking cessation interventions. However, some participants also found additional techniques, such as breathing and mindfulness, to be helpful. This suggests that incorporating some ‘third wave techniques’ into standard CBT for smoking cessation may also be helpful. This theme highlights why the inclusion of practical, individualized strategies is important. First, they help participants to find alternative ways to fulfil the function of smoking (e.g., relaxation), consistent with the behavioural theory.[36] Second, strategies can enhance participants' sense of psychological capability to achieve the desired change in behaviour. Third, once participants found individualized strategies that were effective, this provided an experience of success. This reinforced their use of the technique and increased their motivation to continue with the quit attempt.

Practitioner style as ‘Supportive but Not Like Lecture-y’

The therapeutic style was an important factor in facilitating perceived smoking behaviour change. Rapport may have been enhanced as practitioners delivering the integrated intervention were experienced in promoting a therapeutic environment. Building ‘general rapport’ is included as an intervention component in Michie et al.'s[33] taxonomy of behaviour change techniques. Results highlight how therapeutic style influences perceived treatment outcomes by enhancing or diminishing participants' motivation and sense of capability to change. Consistent with a motivational interviewing stance, participant descriptions of helpful aspects of the IAPT practitioner style included eliciting thoughts from the client rather than imposing their own views and empathic listening,[37] whereas one unhelpful aspect of the practitioner style was the perception of being rushed, and of the practitioner paying more attention to manual pro-forma than to the participant. This indicates the importance of allowing sufficient time and flexibility in the

implementation of the intervention and maintaining focus on the client's needs during sessions.

The importance of the therapeutic alliance to promote behaviour change is consistent with past research showing that the strength of the therapeutic alliance predicted reduced cannabis use in young people at 3- and 6-month follow-up assessments.[14] Furthermore, in the UK Alcohol Treatment Trial, the therapist characteristics and therapeutic alliance were the most common reasons given by participants for their successful reduction of alcohol use.[15] This study, therefore, extends the finding that the therapeutic allliance can play a role in behaviour change to smoking cessation and is relevant, given Orford's criticism of addiction research as 'neglecting relationships in favour of techniques'.[38]

'Having Someone that's Checking on You': Importance of regularity of sessions

This theme highlights the importance of providing the opportunity for regular IAPT practitioner check-ins and encouraging individuals to ask others in their network to check in on them, to enhance the sense of feeling 'answerable'. This suggests that interventions should aim to keep sessions regular and could explore recruiting social networks for check-ins with clients. The importance of feeling 'answerable' is consistent with the literature on the role of social processes in addiction mutual help organizations such as Alcoholics Anonymous.[39]

Having the opportunity at 'the Right Time'

Participant ability to quit smoking coinciding with the opportunity to access support for smoking cessation whilst receiving mental health therapy is consistent with the COM-B model.[16] The presence of mental health problems did not prevent participants from feeling ready to make changes to their smoking. This is important because individuals with mental health problems are not given the same opportunities for smoking cessation support as the general population,[40] and there is a widely held belief among healthcare professionals that smoking cessation should only be attempted after mental health has improved.[7,8] These findings reinforce previous research suggesting that individuals with depression or anxiety can be motivated and supported to successfully reduce or quit smoking when given the opportunity.[10,41] However, this did not apply to all participants, with some expressing that life felt too difficult to quit. Nevertheless, the intervention appeared to help some participants move closer to 'the right time', into the 'preparing to quit' stage of smoking cessation,[17] suggesting that the intervention may assist individuals to progress in their change process.

Research and practice implications

These findings further our understanding of the active ingredients and processes for behaviour change in integrated smoking and mental health interventions. In terms of clinical practice, we have outlined the strategies and the therapeutic stance that could be embedded and emphasized in manualized interventions to optimize therapeutic benefits. Findings indicate that practitioners should not assume that having anxiety or depression means that individuals are not ready or motivated to quit smoking. These processes are currently being implemented in the ESCAPE trial; however, a full-scale trial is required to investigate the effectiveness of these components. Future research in this area should strive to recruit people from diverse cultural backgrounds and include a longer-term follow-up.

CONCLUSIONS

Several key factors were identified by participants to be important in quitting smoking: gaining an increased awareness of smoking patterns; developing individualized strategies; having a practitioner with an empathic, nonjudgemental and motivational stance; regular sessions to promote a sense of accountability and solve problems as they arise; and being given the opportunity at a time when individuals feel ready to change. If similar results are found in more diverse samples, these aspects should be embedded and emphasized within practitioner training and integrated interventions for smoking cessation and depression/anxiety. Future research could use the current findings to inform the constructs that should be targeted for study in research exploring mechanisms of change within a randomized-controlled trial design.

AUTHOR CONTRIBUTIONS

Kim Fredman Stein: Conceptualization; data curation; formal analysis; investigation; methodology; project administration; validation; visualization; writing –original draft; writing –review and editing. **Gemma Taylor**:

Conceptualization; data curation; formal analysis; funding acquisition; investigation; methodology; project administration; resources; supervision; validation; writing –review and editing. **Jennifer Allen**: Supervision; writing –review and editing. **Katherine Sawyer**: Data curation; formal analysis; investigation; methodology; project administration; validation; writing –review and editing. **Shadi Daryan**: Project administration; writing –review and editing.

ACKNOWLEDGEMENTS

We would like to thank the participants in this research and gratefully acknowledge CRUK for funding this research. Kim Fredman Stein was funded by Health Education England's DClinPsy Training Programme during the conduct of this study. Gemma Taylor is funded by a Cancer Research UK Population Researcher Postdoctoral Fellowship award (reference: C56067/A21330) and a Cancer Research UK project award (reference: PPRCPJT\100023). Katherine Sawyer is funded by the Cancer Research UK Population Researcher Postdoctoral Fellowship award (reference: C56067/A21330). Shadi Daryan is funded by the Cancer Research UK project award (reference: PPRCPJT\100023).

CONFLICT OF INTEREST

G. T. has previously received funding from Pfizer, which manufactures smoking cessation products, for research unrelated to this study. The remaining authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The anonymized transcript data are available for free via application to the University of Bath's Research Data Service https://www.bath.ac.uk/guides/research-data-service/.

ETHICS STATEMENT

Ethics approval for this study was received from the NHS Research Ethics Committee on 12 July 2019 (Reference 19–245). Ethical approval for the IntEgrating Smoking Cessation treatment As part of routine Psychological care for dEpression and anxiety (ESCAPE) trial was obtained from a National Health Service Research Ethics Committee (19/03/2018, IRAS ID: 239339).

DETAILS

Subject:	Mental health; Anxiety; Cognitive-Behavioural factors; Mortality; Intervention; Therapeutic alliances; Mental depression; Cognitive behavioral therapy; Feasibility; Tobacco; Debriefing; Psychological intervention; Smoking cessation; Smoking; Morbidity; Investigations; Drug addiction; User experience; Interviews; Access; Consent; Questions; Change agents; Individualized; Addictions; Tobacco smoking; Cigarette smoking; Cognitive ability; Acceptability; Behavior change
Location:	United Kingdom--UK
Identifier / keyword:	anxiety; behaviour change; depression; IAPT; integrated interventions; mechanisms of change; smoking cessation
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	498-509
Publication year:	2023

Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-12-08
Milestone dates:	2022-11-23 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-07-27 (manuscriptReceived); 2022-12-08 (publishedOnlineEarlyUnpaginated); 2022-11-25 (manuscriptAccepted)
Publication history :	
First posting date:	08 Dec 2022
DOI:	https://doi.org/10.1111/hex.13684
ProQuest document ID:	2767102230
Document URL:	https://www.proquest.com/scholarly-journals/service-user-experiences-integrated-psychological/docview/2767102230/se-2?accountid=211160
Copyright:	
Last updated:	2023-11-08
Database:	Public Health Database,Publicly Available Content Database

Children, young people and parent engagement in health intervention design and implementation: A scoping review

Crowther, Daniel [1]
; McCulloch, Holly [1] ; Wong, Helen [2] ; Mackay, Rebecca [1] ; Johnson, Catie [1] ; Chorney, Jill [3] ; Ritchie, Krista [4] ; Logan, Lawrence [5] ; Bishop, Andrea [6] ; Helwig, Melissa [7] ; Curran, Janet [8] [1] Strengthening Transitions in Care Lab, IWK Health, Halifax, Nova Scotia, Canada [2] Faculty of Health, Dalhousie University, Halifax, Nova Scotia, Canada [3] Department of Psychiatry I Department of Psychology and Neuroscience, Dalhousie University, Halifax, Nova Scotia, Canada [4] Faculty of Education, Mount Saint Vincent University, Halifax, Nova Scotia, Canada [5] Research and Innovation, Nova Scotia Health, Halifax, Nova Scotia, Canada [6] Policy Development and Research, Nova Scotia College of Pharmacists, Halifax, Nova Scotia, Canada [7] Research &Scholarly Communications, Dalhousie University, Halifax, Nova Scotia, Canada [8] Strengthening Transitions in Care Lab, IWK Health, Halifax, Nova Scotia, Canada; Faculty of Health, Dalhousie University, Halifax, Nova Scotia, Canada

ABSTRACT (ENGLISH)

Introduction
Engaging children and young people (CYP) with and without their parents in health research has the potential to improve the development and implementation of health interventions. However, to our knowledge, the scope of engagement activities used with this population and barriers to their engagement is unknown. The objective of this review was to identify and describe CYP engagement with and without their parents in the development and/or implementation of health interventions.
Methods
This scoping review included any primary research studies reporting on engaging CYP, with or without parents, in the design and/or implementation of health interventions. Healthcare professionals had to be involved over the course of the study and the study had to take place in either community, primary or tertiary care settings. The following databases were searched in May 2017, May 2020 and June 2021: Medline (OVID), CINAHL (EBSCO) and Embase (Elsevier). Two independent reviewers screened titles, abstracts and full-text articles and used a previously piloted extraction form to extract and summarize information from the included articles.
Results
Twenty-eight articles discussing twenty-four studies were included. CYP engagement throughout the research cycle was limited. There were no observed differences in the reported presence of engagement, types of interventions or outcomes of engagement between studies engaging CYP or CYP and parents. Studies engaging CYP and parents contained limited information on how these relationships affected outcomes of engagement. Engagement was enabled primarily by the maintenance of resources and relationships among stakeholders.
Conclusions
Although CYP engagement often influenced health intervention and implementation design, they are inconsistently engaged across the research cycle. It is unclear whether parental involvement enhances CYP engagement. Future research should consider reporting guidelines to clarify the level of CYP and/or parent engagement, and enhance CYP engagement by fostering synergistic and sustainable partnerships with key stakeholders.
Patient or Public Contribution
A parent partner with codesign experience contributed to the creation of the research questions, screened titles,

abstracts and full texts, helped with data extraction and provided feedback on the manuscript.

FULL TEXT

INTRODUCTION

Patient engagement in health research refers to a collaborative relationship between patients and researchers, where patients with lived experience are actively involved in health research decisions.[1,2] Emerging evidence has shown that patient engagement is a key practice for successful health research.[1] Patient input has the potential to improve the overall quality of outcomes and uptake of new knowledge.[3,4] As a result, several initiatives have emerged to encourage patient-oriented research in North America and Europe.[1,5,6] Although patient engagement in health research has gained momentum over the past decade, we continue to strive for a greater understanding of how engagement occurs throughout all phases of the research process, more formal evaluations of engagement activities and stronger data to support the value of partnering with these stakeholders.[7,8]

The abilities of children and young people (CYP) (0–24 years of age)[9] to participate in paediatric health research topics has been acknowledged in the literature over the past decade.[10–13] While there has been a shift in the methodological approach towards transforming CYP into active research partners, further research is required to determine how best to engage and involve CYP in actual practice.[14] Current evidence demonstrates the benefits of involving both CYP and parents as important stakeholder partners in health research. Three scoping reviews have separately described the engagement process along with the associated benefits and challenges of working with either CYP, parents or both in engagement approaches.[14–16] Together, these reviews provide a broad range of evidence related to engagement of CYP and parents in health research.

However, to our knowledge, there remains a gap in describing the scope of literature related to the health research subfield of CYP and parent engagement in the development, design and/or implementation of health interventions. Neither does there appear to be a synthesis of evidence describing differing engagement practices between CYP engagement with and without parents. Building on previous work, this review aimed to provide a comprehensive and systematic overview of published literature on CYP engagement in health research with a specific focus on intervention design and/or implementation in the presence and absence of parents. The following research questions were addressed:

- 1. How does CYP engagement in health intervention design and/or implementation differ with and without parental involvement?
- 2. What are the characteristics of the studies and health interventions that engage CYP with and without parental involvement in the design and/or implementation of the interventions?
- 3. How are engagement outcomes reported, including the enablers and barriers of engaging CYP with and without parental involvement in the design and/or implementation of health interventions?

METHODS

This review was conducted in accordance with the JBI methodology for scoping reviews.[17] A integrated knowledge translation approach was used.[18] While there is no published protocol for this review, an a priori protocol was developed by the research team.

Inclusion criteriaParticipants

This review considered studies that involved CYP who were between the ages of 0 and 24 years[9] and were engaged in the codesign and/or implementation of a health intervention. Parental involvement in the codesign and/or

implementation process was not a requirement for inclusion; however, if parents were involved, the article was included. Articles that engaged only the parents were excluded.

Concept

Any health intervention and/or implementation strategy—including programmes, tools or frameworks—that were codesigned with CYP to improve any facet of CYP health were included. Informed by patient engagement hierarchies,[19,20] we defined engagement as CYP or CYP and parents who were consulted and informed about the research project and were directly involved in decisions related to the design of intervention and/or implementation components. Interventions that did not target CYP health outcomes (i.e., targeted parent health outcomes) and that were delivered by non-health care providers (i.e., teachers) were excluded. Articles that reported outcomes, including qualitative outcomes, related to the process of CYP engagement and health outcomes of intervention were included. Articles without any reported outcome measures related to the process of engagement were excluded.

Context

Community, primary and tertiary healthcare settings were considered for this review.

Types of sources

All primary research study types were considered for inclusion. Experimental and quasi-experimental study designs, including randomized-controlled trials, nonrandomized-controlled trials, pre–post trials and interrupted time series, were considered. While examining health intervention efficacy was beyond the scope of this review, these types of study designs were included in the event that study authors reported details relating to the engagement of CYP or CYP and parents in intervention design and/or implementation. Observational studies including prospective and retrospective cohort studies, case–control studies and cross-sectional studies were considered. Qualitative and mixed-methods studies were also considered. Systematic reviews and meta-analyses were not included; however, relevant evidence syntheses identified in our search were reviewed for relevant articles. Text, commentary and opinion articles were excluded.

Search strategy

The research team established search parameters in partnership with a library scientist. A mix of controlled vocabulary such as Medical Subject Headings or Emtree terms was used in combination with keywords. The search strategy, including all identified keywords and index terms, was peer-reviewed by a JBI-trained information specialist, and was adapted for each included database and information source (Supporting Information Appendix: Tables S1–S3). No date limit was set for the included articles.

Information sources

The databases searched include Medline (OVID), CINAHL (EBSCO) and Embase (Elsevier). Database searches were conducted on 29 May 29 2017, 22 May 2020 and 23 June 2021. A manual search of the table of contents from the last 5 years was also conducted for the following relevant journals: *Implementation Science*, *Journal of Pediatrics*, *BMC Health Services Research and Paediatrics* and *Child Health*. Given the range and breadth of primary sources identified through our search of the published literature, our team was not confident that a grey literature search would yield significant value to warrant expenditure of our limited resources.

Study/source of evidence selection

Following the search, all identified citations were collated and uploaded into Covidence systematic review software (Veritas Health Innovation) and duplicates were removed. Titles and abstracts were screened by two or more independent reviewers for assessment against the inclusion criteria for the review. Potentially relevant sources were retrieved in full, and their citation details were imported into Covidence systematic review software. The full text of selected citations was assessed in detail against the inclusion criteria by two or more independent reviewers.

Reasons for exclusion of sources of evidence at full text that did not fulfil the inclusion criteria were recorded. Any disagreements between the reviewers at each phase of the selection process were resolved through discussion, or with an additional reviewer. The results of the search and the study inclusion process are presented in a Preferred Reporting Items for Systematic Reviews (PRISMA) and Meta-analyses extension for scoping review flow diagram.[21]

Data extraction

Data were extracted from articles included in the scoping review by two or more independent reviewers using a data extraction tool developed by the reviewers. The data extraction tool was designed to capture information about the source (author, year of publication, country of study), study design, type of intervention, health topic and outcome measure of interest. Pilot extraction was undertaken with three included studies.

Data analysis and presentationAssessment of engagement

The reported presence of both CYP and parent engagement in research involving the design and/or implementation of health interventions were categorized based on five key phases of research, which were developed in consultation with experts: (1) generating a research question; (2) designing study methods; (3) collecting data; (4) interpreting results; and (5) reporting findings. An engagement score was coded for each study to represent the total reported phases of the research process with CYP involvement (0 = no phases of involvement reported to 5 = reported involvement in all phases). A similar process was undertaken to evaluate parental involvement in the included studies.

Assessment of interventions

The Behaviour Change Wheel (BCW) was used as a framework to characterize the included interventions according to the nine intervention function types (i.e., the proposed mechanism of the intervention).[22] The nine intervention function types are environmental restructuring, modelling, enablement, training, coercion, incentivization, persuasion and education. The BCW has been used to characterize interventions in a number of different settings.[23,24] Intervention functions types were mapped against the population that was engaged in the research process and the intervention target population. This was done to explore whether intervention approaches differed between different target and engagement populations. Two independent reviewers coded the reported intervention descriptions using the BCW. Reviewers met and came to consensus on any discrepancies in coding. If consensus could not be reached, a third reviewer was consulted.

Assessment of barriers and enablers

Author-reported barriers and enablers to engaging CYP in the development or implementation of health interventions were categorized using the determinants of partnership synergy, a component of the partnership synergy framework.[25] The partnership synergy framework is a theory-based framework designed to study and optimize the effectiveness of partnerships.[25] The determinants of partnership synergy operationalize five determinants that contribute to high levels of synergy: (1) resources; (2) partner characteristics; (3) relationships among partners; (4) partnership characteristics; and (5) external environment. Enablers and barriers were categorized into these determinants, indicating a presence or absence, respectively. Two independent reviewers coded enablers and barriers, following similar methods utilized for the assessment of engagement and intervention functions.

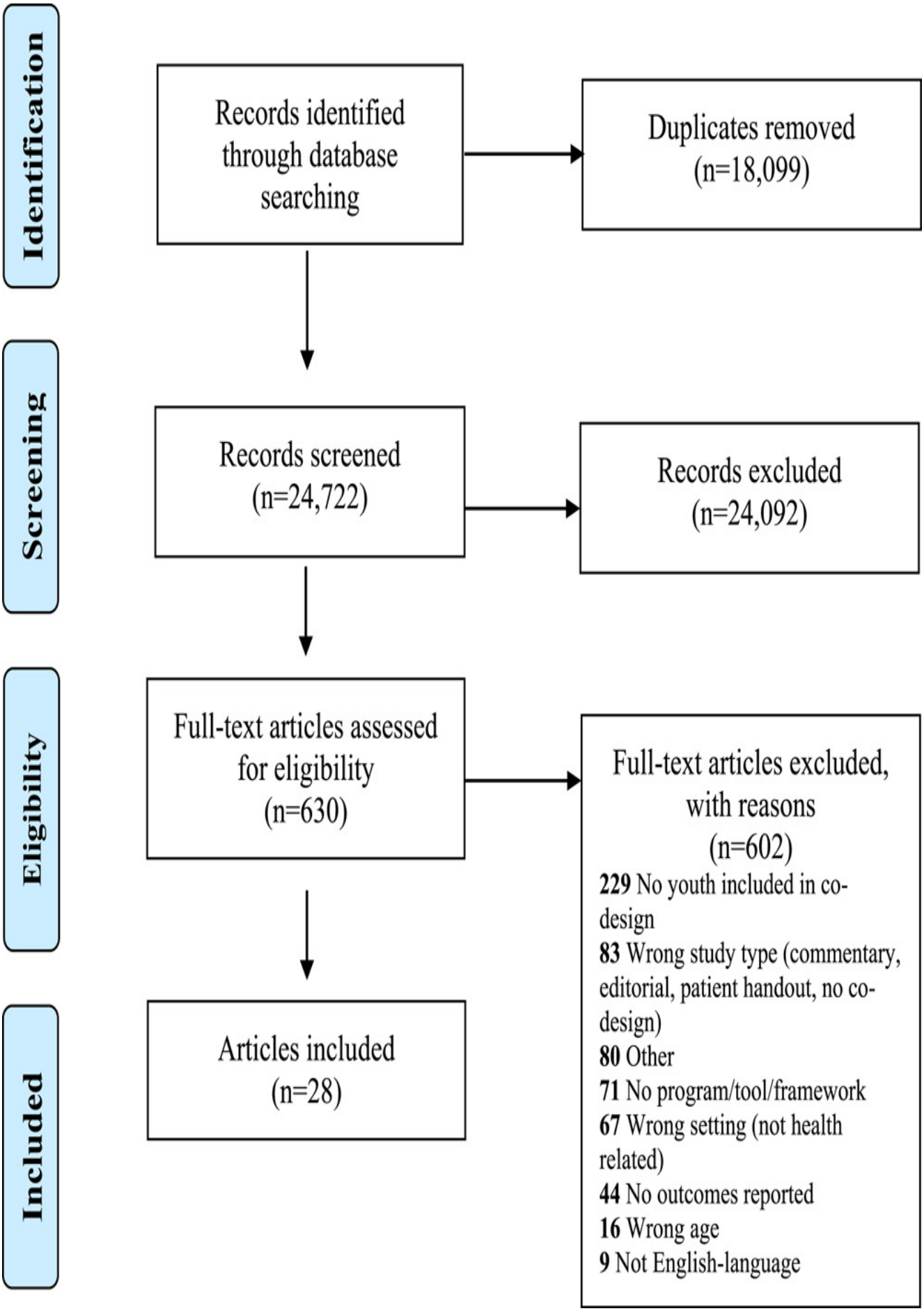
Parent involvement

A parent partner with codesign experience was involved throughout the research process. The aim of their involvement was to inform the framing of our research question and interpretation of our results from the perspective of someone with experience in intervention design. They contributed to the creation of the research questions, screened titles, abstracts and full texts, helped with data extraction and synthesis and provided feedback on the

manuscript. This review adhered to the patient involvement reporting standards outlined in the short form of the Guidance for Reporting Involvement of Patients and the Public-2 (GRIPP-2).[26]

RESULTSCharacteristics of included studies

Reviewers screened 42,722 titles/abstracts and reviewed 631 full-text articles for eligibility (Figure 1). Our parent partner screened 891 abstracts and 55 full texts and extracted data from 10 articles. Twenty-eight articles describing twenty-four studies fulfilled the inclusion criteria. The general characteristics of the included studies are described in Table 1. Sixteen studies engaged only CYP,[27–44] while the remaining eight engaged both CYP and parents.[45–54] The research took place in community, primary and tertiary care settings and addressed a variety of health topics (e.g., sexual health, asthmas, obesity, mental health, cancer, limited mobility, visible difference). Qualitative and mixed-methods study designs were observed most often (n = 22),[27,28,30–36,38–54] and all studies were guided by a diverse set of frameworks, with community-based participatory research (n = 5)[29,36,37,49,51] and participatory action research (n = 8)[27,28,32–35,39,44,51] being the most common.



Enlarge this image.

Table 1 General characteristics of included studies (*n* = 20)

Author (publication year)	Country	Setting	Health topic	Age of CYP	Study design	Frameworks used to guide study
Studies involving CYP and parents						
Eberhart et al. (2019)[45]	USA	Community	Asthma	≥12	Qualitative	Human-centred design
Harrington et al. (2021)[46]	UK	Community	Diabetes—type II	12–14	Qualitative	A theoretical framework based on self-efficacy theory and the capability, opportunity, motivation, behaviour (COM-B) model
Loyd et al. (2017)[47]	UK	Community	Obesity	9–10	Mixed methods	Intervention mapping
Morales et al. (2018)[48]	Canada	Community	Limited mobility	12–21	Qualitative	User-centred design
Morales-Campos et al. (2015)[49]	USA	Community	Obesity	11–14	Qualitative	CBPR, Social cognitive theory
Pembroke et al. (2021)[50]	Ireland	Tertiary Care	Diabetes—type I	11–17	Qualitative	Social cognitive theory
Radovic et al.(2016)[51]	USA	Community	Mental health—depression	13–21	Mixed methods	CBPR, Obesity-related behavioural intervention trials model
Ruland et al. (2006, 2007, 2008)[52–54]	Norway	Tertiary care	Cancer	9–11	Qualitative	Participatory design
Studies involving CYP						
Anselma et al. (2019, 2020)[27,28]	Netherlands	Community	Obesity	9–12	Qualitative	Youth-led Participatory Action Research, Intervention mapping
Bauermeister et al. (2015)[29]	USA	Community	HIV/STIs	17–24	Quantitative	CBPR, Integrated behavioural model
Braun et al. (2020)[30]	Austria	Community	Mental health—suicide	15–19	Qualitative	Suicide Awareness and Voices of Education in the United States

Chaniang et al. (2019)[31]	Thailand	Mixed methods	Mental health—suicide	12–18	Mixed methods	Action research
Dunn (2017)[32]	UK	Tertiary care	Mental health	16–22	Qualitative	Participatory research approaches
Hawkins et al. (2017)[33]	UK	Community	Mental health—substance misuse	13–19	Qualitative	Transdisciplinary Action Research
Jaume et al. (2015)[34]	UK	Community and tertiary care	General health	4–14	Qualitative	Participatory research
Lane et al. (2019)[35]	USA	Community	Obesity/diabetes—type II	11–14	Mixed methods	Youth participatory research
Livingood et al. (2017)[36]	USA	Community	Obesity	15–19	Qualitative	CBPR
Mance et al. (2010)[37]	USA	Community	Mental health	16–24	Quantitative	CBPR, Cognitive-behavioural and stress exposure conceptual models
Patchen et al. (2020)[38]	USA	Community	Sexual health	15–21	Mixed methods	Social cognitive theory, Problem-solving theory
Povey et al. (2020)[39]	Australia	Community	Mental health	10–18	Mixed methods	Participatory research approaches
Saini et al. (2020)[40]	Canada	Community	Acute gastrointestinal illness	11–12	Qualitative	Community engagement methods
Versnel (2011)[41]	Canada	Tertiary care	Chronic health conditions	13–15	Qualitative	Youth engagement
Watson et al. (2017)[42]; Brady et al. (2018)[43]	UK	Community	Mental health—substance misuse	16–21	Qualitative	Young People's Advisory Group

Williamson et al. (2015)[44]	UK	Community	Visible difference	12–19	Mixed methods	Participatory intervention model, participatory action research, Kent's Model of Psychosocial Distress and intervention for individuals with visible differences

Abbreviations: CBPR, community-based participatory research; CYP, children and young people; HIV, human immunodeficiency viruses; NR, not reported; STDs, sexually transmitted diseases; STIs, sexually transmitted infections; UK, United Kingdom.

Presence of engagement

The CYP engagement score in research involving the design and/or implementation of health interventions ranged from 1–5. The majority of studies only reported engaging CYP during one (n = 13)[30,32–34,37,38,44,46–48,50–54] or two (n = 6)[29,31,35,39–41] phases of the research process. Developing the research question and interpreting results had the lowest reported engagement of CYP, while designing methods had the highest reported engagement of CYP (Table 2). Studies that did not engage CYP in forming the research question recruited them after the research question was generated.

Table 2 Research phases in which CYP were engaged (n = 24)

Author (publication year)	Research question	Methods	Data collection	Interpretating results	Reporting results	Engagement score
Studies involving CYP and parents						
Eberhart et al. (2019)[45]	1	1	1	0	0	3
Harrington et al. (2021)[46]	0	0	0	1	0	1
Loyd et al. (2017)[47]	0	1	0	0	0	1
Morales et al. (2018)[48]	0	0	1	0	0	1
Morales-Campos et al. (2015)[49]	1	1	0	1	1	4
Pembroke et al. (2021)[50]	0	0	0	1	0	1
Radovic et al. (2016)[51]	0	1	0	0	0	1
Ruland et al. (2006, 2007, 2008)[52–54]	0	1	0	0	0	1
Studies involving CYP						
Anselma et al. (2019, 2020)[27,28]	1	0	1	1	0	3

Bauermeister et al. (2015)[29]	0	1	1	0	0	2
Braun et al. (2020)[30]	0	1	0	0	0	1
Chaniang et al. (2019)[31]	0	1	0	0	1	2
Dunn (2017)[32]	0	0	0	0	1	1
Hawkins et al. (2017)[33]	0	0	0	1	0	1
Jaume et al. (2015)[34]	0	1	0	0	0	1
Lane et al. (2019)[35]	0	1	0	0	1	2
Livingood et al. (2017)[36]	0	1	1	1	1	4
Mance et al. (2010)[37]	0	1	0	0	0	1
Patchen et al. (2020)[38]	0	1	0	0	0	1
Povey et al. (2020)[39]	0	0	1	1	0	2
Saini et al. (2020)[40] 2020	0	1	0	0	1	2
Versnel (2011)[41]	0	1	0	0	1	2
Watson et al. (2017)[42]; Brady et al. (2018)[43]	1	1	1	1	1	5
Williamson et al. (2015)[44]	0	1	0	0	0	1

Note: 1 = engaged, 0 = not reported.

Abbreviation: CYP, children and young people.

Only one study, which was discussed in two articles,[42,43] reported engagement across all five phases of the research process. The youth social behaviour and network therapy study established a young people's advisory group to guide engagement, which emphasized both consultation and coproduction to facilitate opportunities for young people at each phase of the research.[42,43]

Among the studies that engaged both CYP and parents, the engagement score in research involving the design and/or implementation of health interventions for parental involvement ranged from 1–3 (Table 3), with most reporting parent engagement during one phase of the research process (n = 6).[46–48,50–54] Parents were reported as being engaged in an equal or lower number of phases than CYP. For instance, Morales-Campos et al.[49] reported engaging CYP during the research question, methods, interpretation of results and reporting of findings, while there were only descriptions of parent engagement during two phases. Parents were always reported as being engaged concurrently with CYP, and parents were never engaged in phases in which CYP were not.

Table 3 Research phases in which parents were engaged (*n* = 10)

Author (publication year)	Research question	Methods	Data collection	Interpretating results	Reporting results	Engagement score
Eberhart et al. (2019)[45]	1	1	1	0	0	3
Harrington et al. (2021)[46]	0	0	0	1	0	1
Loyd et al. (2017)[47]	0	1	0	0	0	1
Morales et al. (2018)[48]	0	0	1	0	0	1
Morales-Campos et al. (2015)[49]	0	1	0	1	0	2
Pembroke et al. (2021)[50]	0	0	0	1	0	1
Radovic et al. (2016)[51]	0	1	0	0	0	1
Ruland et al. (2006, 2007, 2008)[52–54]	0	1	0	0	0	1

Note: 1 = engaged, 0 = not reported.

There appeared to be no differences in the reported presence of engagement in studies engaging only CYP versus studies engaging CYP and parents.

Target and types of interventions

Of the 10 studies that included CYP and parents, four studies reported on interventions that targeted both CYP and parents,[31,40,45,51] with two of these engaging CYP and parents for more than one research phase.[45,51] The remaining six studies designed interventions for CYP only.[46–50,53] There was no noticeable difference between the intervention function types used when both CYP and parents were engaged compared to when only CYP were engaged (Table 4). However, the training intervention function type was only used with interventions designed to target CYP.[37,42–44] Incentivization, coercion and restriction function types were not used in any of the studies.

Table 4 Health intervention characteristics (*n* = 24)

Authors (publication year)	Description of the intervention	Target of the intervention	CYP engagement score	Identified BCW domains

				Education	Persuasion	Training	Modelling	Environmental restructuring	Enablement
Studies involving CYP and parents									
Eberhart et al. (2019)[45]	Support system, asthma activity sheet, conversation starter pack.	CYP and parents	3					✗	
Harrington et al. (2021)[46]	An interactive lifestyle training programme.	CYP	1	✗		✗			
Loyd et al. (2017)[47]	Health education programme.	CYP	1	✗					
Morales et al. (2018)[48]	Mobility devices and environmental design solutions.	CYP	1					✗	
Morales-Campos et al. (2015)[49]	Community physical activity programme.	CYP	4						✗
Pembroke et al. (2021)[50]	Educational videos of common questions asked during hospital visits.	CYP	1	✗					
Radovic et al. (2016)[51]	Educational websites designed to increase treatment engagement and access to an online community.	CYP and parents	1	✗	✗				

Ruland et al. (2006, 2007, 20087)[52–54]	Phone/tablet application designed to improve communication between youth and clinicians.	CYP	1					✗	
Studies involving CYP									
Anselma et al. (2019, 2020)[27,28]	Cooking workshops, after school activities, sports events, installation of a water fountain.	CYP	3						✗
Bauermeister et al. (2015)[29]	Web application promoting HIV/STI testing.	CYP	2	✗	✗				
Braun et al. (2020)[30]	Suicide prevention videos.	CYP	1	✗					
Chaniang et al. (2019)[31]	Suicide prevention promotion and education programme.	CYP and Parents	2	✗			✗		
Dunn (2017)[32]	Transition programme for transitions from mental health services.	CYP	1	✗					
Hawkins et al. (2017)[33]	Informal peer-led drug prevention programme.	CYP	1		✗	✗			
Jaume et al. (2015)[34]	Animated tool designed to collect health information from children.	CYP	1					✗	
Lane et al. (2019)[35]	Skill-building and community changes to reduce sugar-sweetened beverage intake.	CYP	2						✗
Livingood et al. (2017)[36]	A digital health promotion programme.	CYP	4	✗				✗	
Mance et al. (2010)[37]	Modified Psychotherapy for Adolescents Responding to Chronic Stress.	CYP	1			✗			
Patchen et al. (2020)[38]	A sexual health education mobile-based video game.	CYP	1	✗					

Povey et al. (2020)[39]	An interactive care planning e-mental health mobile app.	CYP	2						✗
Saini et al. (2020)[40]	Educational video about acute gastrointestinal illness.	CYP and parents	2	✗					
Versnel (2011)[41]	Youth wellness support networks.	CYP	2					✗	
Watson et al. (2017)[42]; Brady et al. (2018)[43]	Youth social behaviour and network therapy.	CYP	5			✗		✗	
Williamson et al. (2015)[44]	Web application that integrates cognitive behavioural therapy and social skills training.	CYP	1			✗			

Abbreviations: BCW, Behaviour Change Wheel; CYP, children and young people.

Reported outcomes of engagement

CYP or CYP and parent engagement outcomes were primarily captured through informal qualitative feedback from CYP, parents or observations made by the study team. No validated quantitative outcome measures related to engagement or engagement frameworks/hierarchies were used. As such, reported outcomes of engagement were entirely narrative descriptions. A summary of reported author anecdotes is presented in Table 5. Reported outcomes of engagement fell into two categories: (1) CYP or CYP and parents benefited from being engaged (i.e., gained research experience, knowledge, confidence, opportunities and changes in attitudes and behaviours) and (2) the intervention design and/or implementation were improved by CYP engagement with or without parental involvement (Table 5).

Table 5 Outcomes of engagement (*n* = 24)

Author (publication year)	Outcome of engagement
Studies involving CYP and parents	
Eberhart et al. (2019)[45]	Stakeholder involvement resulted in the development of unique and ready to implement interventions.
Harrington et al. (2021)[46]	Stakeholder involvement provided valuable feedback of session content, format and delivery.
Loyd et al. (2017)[47]	High retention rates were attributed to the involvement of children in the development of the intervention.
Morales et al. (2018)[48]	Engaging stakeholders provided a novel perspective that yielded new design solutions.

Morales-Campos et al. (2015)[49]	Girls gained new insights into better understanding their community and the issue of increasing PA among girls their age.
Pembroke et al. (2021)[50]	Stakeholder involvement allowed for the identification of relevant intervention priorities and made involved adolescents feel empowered.
Radovic et al. (2016)[51]	Without stakeholders, investigators would have had to make major (and potentially incorrect) assumptions.
Ruland et al. (2006, 2007, 2008)[52–54]	Children contributed creative suggestions that the design team would not have thought of and improved the software.
Studies involving CYP	
Anselma et al. (2019, 2020)[27,28]	Children's perspectives improved understanding of the issues and resulted in a more relevant intervention. Children were empowered through participation.
Bauermeister et al. (2015)[29]	Youth insight was crucial to the success of the study.
Braun et al. (2020)[30]	Involving adolescents increased the relevance of the intervention and resulted in an increased sense of well-being.
Chaniang et al. (2019)[31]	Adolescent involvement was a key to successful programme development.
Dunn (2017)[32]	Young people gained the opportunity to think creatively about transition preparation.
Hawkins et al. (2017)[33]	Involving young people improved the acceptability, feasibility and quality of the intervention.
Jaume et al. (2015)[34]	Child involvement allowed the tool to be adjusted to children's needs.
Lane et al. (2019)[35]	Youth gained valuable knowledge through involvement.
Livingood et al. (2017)[36]	Youth contributed valuable insight into the development of the intervention.
Mance et al. (2010)[37]	Peer leaders added intervention material, making it more relevant for the target community.
Patchen et al. (2020)[38]	Youth engagement was essential to the success of the development of the intervention.
Povey et al. (2020)[39]	Young people assisted in tailoring the intervention to their preferences.

Saini et al. (2020)[40]	Partnering with youth was attributed to the success and cultural relevance of the intervention.
Versnel (2011)[41]	Youth leaders gained a sense of accomplishment and a desire to 'do something more meaningful with my life'.
Watson et al. (2017)[42]; Brady et al. (2018)[43]	Young advisors benefited by recognizing their ability to achieve positive change, but were worried about the stigma associated with being involved in a project about mental health.
Williamson et al. (2015)[44]	Stakeholders were empowered to develop an acceptable intervention that integrates the theoretical and current evidence base regarding intervention content, with the beliefs, motivations, language, culture and practices of potential service users and HCPs

Abbreviation: CYP, children and young people.

Although most studies (n = 20)[27,29–34,36–40,44–48,50–54] noted the positive impact of CYP with or without parental involvement on health intervention design and/or implementation, limited details were provided on how they influenced the process. Furthermore, there were no apparent differences between the outcomes reported in studies with CYP and studies with CYP and parents. In one case, the word 'stakeholders' was used to group both CYP and parents together,[51] resulting in a lack of emphasis on outcomes related to parents overall.

Reported barriers and enablers of engagement

Of the 24 included studies, most (n = 20)[27,28,30–44,46,47,49,51–54] reported barriers and/or enablers to engagement (Table 6). A prevalent determinant, addressed both as a barrier and as an enabler, was resources. Resources extended beyond the financial to include adequate time,[33,38,41] training[52] and involvement of key stakeholders.[27,44] Notably, involvement of community experts, healthcare experts, authority figures and policy makers was reported as an enabling factor of engagement.[27,44]

Table 6 Barriers and enablers classified by the determinants of partnership synergy (n = 24)

Determinants of partnership synergy	Barriers	Enablers
Resources	•Recruitment (i.e., lack of participation, selection bias).[39,42,49]	•Involving community and healthcare experts.[27]
	•Time limitations.[33,38,41]	•Involving authority figures and policy makers.[44]
	•Lack of resources.[35]	•Incorporating community and regional partnerships.[28,40,46]
		•Having adequate funding.[52]
		•Providing training.[52]

		•Maintaining resources and support to accommodate a variety of different backgrounds and ages.[34]
Partner characteristics	•Including youth with suicide ideation during development of a suicide intervention posed health concerns.[31]	•Involving CYP early on.[41,42]
	•Youth lacked methodological expertize.[36]	
	•Youth lacked of technological skill.[30]	
	•Ageing out of the study.[35]	
	•Steep learning curve of subject matter.[52–54]	
	•Youth and parents lacked the motivation to contribute.[28]	
Relationships among partners	•Trust building.[53]	•Maintaining contact throughout the research process (via email and social media in conjunction with group meetings).[42,43]
	•Clique formation.[52]	•Researchers encouraging responsibility of youth's own care.[32]
	•Conflict among stakeholders.[33]	•Mentorship that values the skills and ideas of youth.[30]
	•Competing priorities and goals between stakeholders.[33]	•Researchers maintaining awareness of group dynamics.[51]
		•Allowing engaged partners to share personal experiences.[53]
Partnership characteristics	•Maintaining consistent engagement over a lengthy research process.[37,49,53,54]	•Including other lines of methodology and clinical research (engaging partners is not always sufficient to ensure a valid intervention).[54]
	•Researchers balancing guidance and autonomy in terms of youth engagement.[27,37]	•Working in parallel with two different age groups.[52]
	•Involving youth slowed down the process.[34]	•Keeping flexible scheduling to accommodate engaged partners.[42]

		• Using principles of colearning and shared responsibility.[37]
		• Defining roles of engagement.[52]
		• Iterative design, with multiple opportunities for feedback.[38]
External environment	• Geographic distances between researchers and youth.[35]	
	• Within a school setting, school assessment pressures and school staffing issues.[47]	

Abbreviation: CYP, children and young people.

Relationships among partners also represented a common determinant among the studies. However, the enablers and barriers under this classification were not specifically related to the parent–CYP dyad; instead, they focused on engaged partners in general, the CYP–CYP dyad or the CYP–researcher dyad. For example, one study, which engaged both CYP and parents, reported only on the CYP/parent–researcher dyad (i.e., building trust between researchers and partners and ensuring that researchers allowed engaged partners to share personal experiences).[53] There were no discernible differences in the types of reported barriers and enablers or their frequency between articles engaging CYP and articles engaging CYP and parents.

DISCUSSION

This scoping review identified a heterogeneous body of literature and covers a wide range of health interventions. These findings are consistent with previous scoping reviews, which describe inconsistent engagement across research phases with varying levels of engagement[14,15] and inconsistent use of terminology.[16] This review builds on previous work by providing a detailed overview of current engagement practices with CYP and CYP and parents in research involving the design and/or implementation of health interventions. Our findings show that there is little evidence to support any differences between studies that engaged CYP versus CYP and parents in the presence of engagement, the types of interventions that were designed and/or implemented and the outcomes of engagement. This review also provides novel insight into the scarcity of evidence related to how relational dynamics impact engagement and summarizes the breadth of barriers and enablers to engagement unique to the context of CYP and parent involvement in health intervention design.

CYP were rarely reported to have been engaged at every phase of the research process and parents were never reported to have been engaged in more than three phases. Engagement for both CYP and parents was mostly limited to the development and design of research methods. Although similar scoping reviews have also reported a lack of consistent stakeholder engagement, Larsson et al. found CYP to be less involved during research design, implementation and data analysis phases.[14] Shen et al.[15] observed a greater range of parent participation across the research spectrum including the planning, design, collection and analysis of data, and dissemination of findings; however, no study maintained parent engagement throughout the entire research process. Current engagement guidelines and frameworks promote patient involvement in all aspects of the research process, as it is considered a feature of meaningful involvement and ensures stakeholder-oriented outcomes.[2,55–57] Yet, a disparity appears to exist between theory and the reported practice for sustaining CYP and parent engagement throughout the research

process. Whether this disparity is the result of underreporting or lack of adherence to engagement guidelines is unclear. It is possible that increasing the use of reporting guidelines for engaging patients in research could improve reporting of engagement practices and could encourage greater engagement and collaboration throughout the entirety of the research process.

Researchers struggle with determining how to authentically engage CYP as coinvestigators, and adding parents to the process can result in an additional layer of complexity.[58] While some advancements can be seen with regard to effective ways to engage CYP, there is growing support to also demonstrate the added value of incorporating parents with CYP in the design and implementation of programmes or interventions related to CYP health (e.g., parents can challenge assumptions underlying research priorities and provide first-hand perspectives).[59–63] It is recognized that engagement should extend beyond the patient to their family[64] and that relational dynamics inform engagement.[65] However, in practice, limited strides have been made toward the greater inclusion of parents within the field of codesigning health intervention for CYP.[15] Further, there remains a scarcity of literature concerning dyadic activation and engagement of patients with their caregivers.[66] Future work should examine how to synergistically bring CYP and parents together as important stakeholders within the research team.

All of the included studies reported on the benefits of engagement for CYP or CYP and parents and/or the interventions. Successful experiences were most often enabled by the presence of sufficient resources (e.g., funds, training, involvement of relevant community members)[27,44,52] and supporting relationships among partners (e.g., mentorship, awareness of group dynamics, maintaining contact throughout the research process).[30,42,43,51] Research environment, expectations, support and value have been identified by patients and their families as essential factors to ensuring meaningful engagements as partners on research teams.[67] Future engagement research should specifically plan how to fund and support engagement opportunities.

In line with the findings from Flynn et al.,[16] this review encountered substantial variation in reporting standards among the included studies, which made comparisons across papers difficult. The lack of standardization of key terms describing ‘engagement’ in research made it challenging to clearly distinguish studies that fulfilled the inclusion criteria. The inconsistent use of language is particularly pertinent to scoping and systematic reviews since diverse terminology used to define engagement can make it more difficult to find existing literature and potentially problematic when determining the level of stakeholder engagement.[15] Implementing standards in nomenclature would help in clarifying issues arising from inconsistent use of terminology and has been identified as an important next step in other scoping reviews of integrated knowledge user engagement.[15,68] In addition, adherence to the Consolidated Standards of Reporting Trials statement[69] for clinical trials, the PRISMA statement[70] for systematic reviews and meta-analyses or other applicable reporting guidelines is strongly encouraged to improve the transparency of publication in health intervention research.

Parent engagement

Our parent partner provided valuable insight into our findings, which helped contextualize and inform the presentation of our results. They held an integral role throughout this review, being involved in each stage of the process. While our parent partner was an essential aspect of this review, there were several challenges. In line with our findings, for those with little to no research experience, there can be a steep learning curve in understanding research methodology. Additionally, scientific jargon can, at times, make communication less efficient, as more time has to be spent clarifying concepts.

Limitations

This study had several limitations. The search was limited to the English language only. In addition, given the diverse use of terminology related to CYP engagement, it is possible that we missed some relevant literature.

However, we kept our search strategy broad with the intent of capturing hard to reach reports. It is possible that some relevant literature may have been omitted during screening due to CYP or parents not being mentioned in the abstract. However, our implementation of broad inclusion criteria ensured that a wide range of literature was captured and included in our review. We appreciate that the absence of reporting does not necessarily indicate the absence of engagement. Adherence to reporting standards such as the GRIPP-2 would strengthen our synthesis work related to this topic. The use of engagement hierarchies, such as Hart's Ladder of Youth Participation[20] or Shier's Pathways to Participation Model,[19] could potentially be valuable tools in defining levels of engagement for future researchers. Although our review team did include a parent and researchers and healthcare providers who work with children and young adults, a CYP was not directly involved in this review. Also, while this review pertains to more systems-level content, our review team did not include any decision-makers.

CONCLUSION

Our findings suggest that engagement of CYP, with or without parental involvement, in designing and/or implementing health interventions is limited. While CYP have been engaged in decisions regarding intervention components, they are seldom engaged throughout the research process, which may hinder meaningful involvement and the inclusion of patient-reported outcomes. Further, we do not yet know if parental engagement alongside CYP may alter the nature of CYP engagement as this was not addressed in the study reports. While more engagement can create barriers, researchers should consider how the perspectives of CYP and their parents can strengthen the research process beyond the design of research methods, as well as the impact of dyadic engagement. This scoping review provides foundational knowledge on the enablers and barriers of CYP engagement, both with and without parents, for health intervention research. Our findings serve as a valuable tool for future intervention development and offer avenues for further exploration of appropriate engagement practices of both CYP and parents during health intervention design and implementation.

AUTHOR CONTRIBUTIONS

Daniel Crowther contributed to article screening, data extraction and data analysis, and drafted the manuscript. Holly McCulloch, Helen Wong and Catie Johnson contributed to article screening and data extraction, and reviewed and edited the manuscript. Dr. Jill Chorney and Dr. Krista Ritchie developed the research questions, contributed to data analysis and reviewed and edited the manuscript. Rebecca Mackay, Dr. Logan Lawrence and Dr. Andrea Bishop developed the research questions, contributed to article screening, data extraction and data analysis, and reviewed and edited the manuscript. Melissa Helwig developed the search strategy, contributed to article screening and reviewed and edited the manuscript. Dr. Janet Curran developed the research questions, contributed to article screening and data analysis and reviewed and edited the manuscript.

ACKNOWLEDGEMENTS

This study was funded by a Nova Scotia Health Research Foundation Development and Innovation Grant.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

DETAILS

Subject:	Parents &parenting; Health promotion; Software; Intervention; Stakeholders; Parents; Co-design; Databases; Health care; Extraction; Medical research; Implementation; Young adults; Medical personnel; Information processing; Parent participation; Information sources; Partnerships; Systematic review; Children; Youth
Identifier / keyword:	children; engagement; health interventions; parent; young people
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	1-15
Publication year:	2023
Publication date:	Feb 2023
Section:	REVIEW ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Literature Review, Journal Article
Publication history :	
Online publication date:	2022-11-08
Milestone dates:	2022-07-13 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-03-02 (manuscriptReceived); 2022-11-08 (publishedOnlineEarlyUnpaginated); 2022-07-14 (manuscriptAccepted)
Publication history :	
First posting date:	08 Nov 2022
DOI:	https://doi.org/10.1111/hex.13572

Use of co-design methodology in the development of cardiovascular disease secondary prevention interventions: A scoping review

Talevski, Jason [1] ; Kulnik, Stefan T [2] ; Jessup, Rebecca L [3] ; Falls, Roman [4] ; Natali Cvetanovska [5] ; Beauchamp, Alison [6]

[1] Institute for Physical Activity and Nutrition Research (IPAN), School of Exercise and Nutrition Sciences, Deakin University, Geelong, Victoria, Australia; School of Rural Health, Monash University, Warragul, Victoria, Australia; Australian Institute for Musculoskeletal Science (AIMSS), The University of Melbourne and Western Health, St Albans, Victoria, Australia [2] Ludwig Boltzmann Institute for Digital Health and Prevention, Salzburg, Austria; Faculty of Health, Social Care and Education, Kingston University and St George's University of London, London, UK [3] School of Rural Health, Monash University, Warragul, Victoria, Australia; Academic and Research Collaborative in Health, La Trobe University, Bundoora, Victoria, Australia; Allied Health Research, Northern Health, Epping, Victoria, Australia [4] Western Centre for Health Research and Education, Sunshine Hospital, St Albans, Victoria, Australia [5] School of Rural Health, Monash University, Warragul, Victoria, Australia; Office of Research, Northern Health, Epping, Victoria, Australia [6] School of Rural Health, Monash University, Warragul, Victoria, Australia; Australian Institute for Musculoskeletal Science (AIMSS), The University of Melbourne and Western Health, St Albans, Victoria, Australia; Victorian Heart Institute, Monash University, Clayton, Victoria, Australia

ABSTRACT (ENGLISH)

Introduction

There is growing evidence to support the use of co-design in developing interventions across many disciplines. This scoping review aims to examine how co-design methodology has been used in the development of cardiovascular disease (CVD) secondary prevention interventions within health and community settings.

Methods
We searched four academic databases for studies that used the co-design approach to develop their intervention. Studies were included if consumers (adults with CVD) and key stakeholders (e.g. clinicians, service providers) were involved in the co-design process. The review focused on methodology rather than traditional study outcomes; therefore, co-design processes and activities were extracted and evaluated against a selected co-design framework.
Results
Twenty-two studies were included in this review. Studies were implemented across various settings with consumers and stakeholder groups most frequently consisting of patients and healthcare professionals, respectively. Most studies specifically stated that they used a 'co-design' approach ($n=10$); others used terms such as participatory action research ($n=3$), user-centred design ($n=3$) and community-based participatory research ($n=2$). Although there was variability in terminology, co-design processes, and participants, all studies adhered to the key principles of consumer engagement. Predominant co-design activities included semistructured interviews, focus groups, co-design/development workshops and advisory group meetings. Intervention effectiveness was assessed in eight studies showing mixed results.
Conclusions
This review provides an overview of how the co-design approach has previously been used in the development of CVD secondary prevention interventions. These findings provide methodological considerations that can guide researchers and healthcare services when implementing co-design to develop feasible and acceptable interventions that can improve outcomes for CVD populations.
Patient or Public Contribution
No patients, service users, caregivers, people with lived experience or members of the public were involved in this scoping review. This review article was written by academics who have undertaken a significant amount of co-design work with consumers and stakeholders.

FULL TEXT

INTRODUCTION

Cardiovascular diseases (CVDs) are a group of disorders of the heart and blood vessels and include coronary heart disease, stroke, heart failure and other conditions. CVD has remained the leading cause of death globally for the last 20 years and is estimated to cause more than 18 million deaths each year.[1] Significant progress has been made over the past few decades in the management and treatment of CVD, particularly in the prevention of recurrent cardiovascular events in high-risk individuals such as those with previous events or known CVD (i.e., secondary CVD prevention). Secondary prevention guidelines have been developed[2,3] that recommend lifestyle changes for ongoing management of cardiovascular risk factors, including a healthier diet (reduction of salt, eating more fruits and vegetables), regular physical activity, medications and cessation of tobacco use and harmful intake of alcohol. However, while these behaviour changes have been shown to significantly reduce the risk of secondary CVD events,[4–6] they are also shown to be difficult to sustain long term.[7–9]

Several effective approaches to support CVD patients in adopting these preventive behaviours have been established, including engagement in lifestyle modification programmes, use of technology-based interventions (e.g., telehealth, mobile health applications) and cardiac rehabilitation.[5] However, despite good evidence for their effectiveness, these programmes are not always well-utilized or accepted by the target population. For example, the benefits of cardiac rehabilitation are well-recognized and include lower mortality rate, reduced risk of hospital admissions and improved health-related quality of life.[10,11] However, cardiac rehabilitation continues to have poor attendance rates, with only 15%–30% of eligible patients engaging with the programme.[12,13] To address poor uptake, policymakers and healthcare providers need to ensure that interventions are well-accepted and adapted to the specific needs and perceptions of the target population.[14,15] One way to achieve this is for researchers to work with end-users and nonacademic stakeholders in the development of interventions; this is known as co-design.

The engagement of stakeholders in the development of interventions or public health initiatives is captured in the

literature under different terminologies such as ‘co-design’, ‘co-production’, ‘co-creation’, ‘participatory action research’ or ‘user-centred design’.[16] These terms are often used interchangeably by authors and are understood to describe equivalent approaches to stakeholder engagement. Co-design is a process in which targeted end-users and other relevant stakeholders form a partnership with researchers and work together on all aspects of intervention development, from understanding the needs of end-users to content development and pilot testing.[17] There are a number of co-design research frameworks in the literature, all describing a similar series of sequential phases and core principles, which include equity (shared decision-making across all stages), understanding experiences (co-learning with a mutual exchange of information between partners) and improving services or health outcomes (development of a programme based on the findings).[17] Co-design is a relatively new concept within healthcare, although over the past decade, researchers and healthcare providers have increasingly involved consumers and nonacademic stakeholders in the development of public health interventions to improve process and health outcomes.[18] Involving consumers in this way has been shown to increase acceptance, uptake, long-term adherence and satisfaction with interventions, as well as improve the health outcomes of end-users.[19,20] For this reason, evidence for the use of the co-design approach in the development of healthcare interventions is increasing across many disciplines.[21] However, despite an abundance of CVD intervention studies in the literature, few describe the use of co-design methodologies. Utilizing the co-design approach could address potential barriers to the uptake of an intervention and may deliver more effective and sustainable solutions to CVD secondary prevention.[22,23]

The aim of this review is to examine the nature and extent of co-design methods utilized in the development of CVD secondary prevention interventions within health and community settings. Specific review questions are:

- 1. What *approaches/concepts* to co-design have been used in CVD secondary prevention research (e.g., co-production, participatory action research, etc.)?
- 2. What *activities/methods* were used to develop these CVD secondary interventions (e.g., focus groups, co-design workshops, etc.)?
- 3. Have these co-designed CVD interventions been evaluated for *effectiveness*, and if so, what health outcomes were evaluated?

By summarizing the key processes used to develop these CVD interventions, we intend to improve the knowledge base for co-design in CVD research and provide a guide for researchers considering using these methods.

METHODS

A scoping review was undertaken to address our research questions following a methodological framework to guide the review process.[24] The review is reported in accordance with the Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) Checklist[25] and is registered with PROSPERO (CRD42021291841).

Search strategy

An electronic database search for published literature was performed in August 2022 using Ovid Medline (biomedical literature), Embase (biomedical and pharmaceutical literature), PsycINFO (psychology and behavioural sciences) and Web of Science (sciences, engineering, medicine and social sciences). A sensitive search strategy was first developed for Ovid Medline comprising a broad range of terms for ‘co-design’ (Supporting Information: File 1) and then modified for other databases. Reference lists from eligible studies and systematic reviews discovered by

the search were also reviewed for further relevant studies.

Eligibility criteriaStudy design and participants

All relevant English language original primary research studies utilizing qualitative, quantitative or mixed-methods study designs were included. Systematic reviews/meta-analyses, scoping and narrative reviews, opinion pieces, editorials, letters to the editor, government reports, conference abstracts and non-English language publications were excluded. Studies were considered eligible for the review if consumers (adults aged ≥18 years with CVD including coronary heart disease, stroke, heart failure, heart attack/myocardial infarction or peripheral vascular disease), their carers and/or key stakeholders (clinicians, service providers and relevant stakeholder organizations) were involved in the co-design process described by the study.

Interventions

The definition of co-design for this review was adopted from Boyd et al.[17] that includes six core elements (Box 1). The first three elements aim to gain an understanding of the consumer experience and needs, while the latter three focus on how to improve that experience through development and action. Studies were included in this review if the intervention in question met the following three conditions: (1) steps in the development or design of the intervention were clearly described; (2) the methods used to develop the intervention matched our definition of co-design (i.e., consumers and/or stakeholders were included in both the Exploratory and Development Phases of co-design) and (3) the intervention aimed to improve management or prevention of subsequent cardiovascular events.

1:**BoxElements of co-design**

Exploratory Phase

- 1. Engage: Establish meaningful relationships with consumers and/or relevant stakeholders to understand and improve a problem.
- 2. Plan: Work with consumers and/or stakeholders to identify ideas about goals and how to achieve them.
- 3. Explore: Learn about consumers' and/or stakeholders' experiences and identify what can be improved (i.e., needs assessment).

Development Phase

- 4. Develop: Turn the ideas from consumers and/or stakeholders into potential solutions (i.e., intervention development).
- 5. Decide: Choose improvements to make and how to make them based on further feedback from consumers and/or stakeholders.
- 6. Change: Turn improvement ideas into actions (e.g., prototype testing with end-users) and finalize the intervention.

Outcomes

The main outcomes of interest were the methods and phases of the co-design process used in each study. This

included the number and type of participants involved (consumers and key stakeholders), the degree of consumer and/or stakeholder involvement, and intervention feasibility/acceptability. If reported, data on the evaluation of intervention effectiveness were also collected.

Study selection

Two authors (J. T. and A. B.) screened abstracts of potentially relevant studies against the eligibility criteria using Covidence—a web-based platform for undertaking the steps in the study review process.[26] Of the potentially relevant studies from this initial screening, full-text articles were obtained and assessed for inclusion independently by two authors (J. T. and one other author). If there were any conflicts between reviewers, a third author was called upon to make the final decision.

Data charting

The following data were independently extracted from each study by two authors (J. T. and A. B.): lead author, publication year, country of study, study design, consumer characteristics (number, % male, mean age), stakeholder characteristics (number, % male, mean age), intervention description, methods of co-design and outcome data (feasibility, acceptability and effectiveness). If any discrepancies were observed between the extracted data, the authors met to reach a final consensus.

Critical appraisal of evidence

As this was a scoping review, a formal assessment of methodological quality was not undertaken. However, to meet the aims of the review, a sufficiency of the reporting approach was undertaken using an amended version of the Critical Appraisal Skills Programme (CASP) critical appraisal tools designed for multiple research study designs.[27] The items included:

- 1. Aim (was there a clear statement of the aims of the research?)
- 2. Setting (was it clear where the development of the intervention took place?)
- 3. Recruitment (was it clear how the study participants were recruited?)
- 4. Participants (was it clear which consumers/stakeholders were involved in the co-design process, and do you know all that you need to about the participants?)
- 5. Facilitators (was it clear who facilitated the co-design process?)
- 6. Procedure (was the description of the overall co-design process clear/complete?)
- 7. Schedule (were the interval and frequency of the co-design sessions clear?)
- 8. Results (were the results of the study clearly documented and discussed?)

•9.

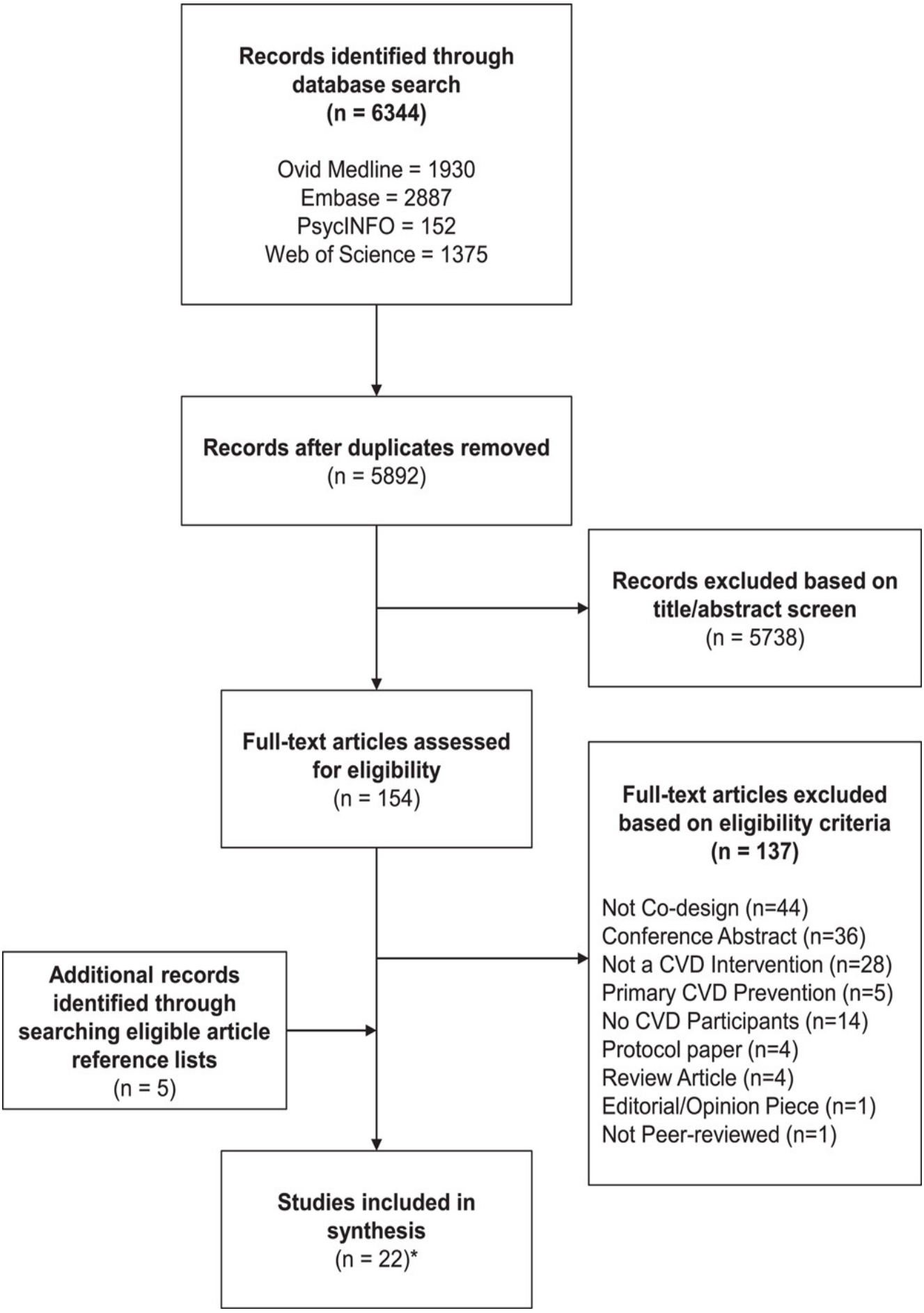
Intervention (was the final version of the intervention clearly described?)

Synthesis of results

A co-design methodology assessment tool was developed based on Boyd et al.'s[17] co-design framework. To determine how co-design has been previously implemented in CVD populations, included studies were assessed for their inclusion of Boyd et al.'s[17] six elements of co-design framework: engage, plan, explore, develop, decide and change. The ‘develop’ and ‘decide’ steps were reported together in the results as the synthesis found these steps coincided with each other within all studies. Where available, data of effectiveness were summarized thematically. Data synthesis was completed by the first author (J. T.) and checked for consistency by one other author (A. B.).

RESULTSStudy selection

Following the removal of duplicates, 5892 articles were retrieved from the electronic search. Of these, 5738 articles were excluded based on the review of the title and abstract, and the full text was obtained for the remaining 154 articles. Based on the authors' assessment, 22 unique studies[28–49] published across 45 articles met the eligibility criteria and were included in this review (Figure 1).



Enlarge this image.

Study and participant characteristics

Of the 22 included studies, 16 utilized a mixed-method approach[28-30,32,34,35,38,39,41-45,47-49] and six were qualitative.[31,33,36,37,40,46] Only one study had been published before the year 2010,[39] while the rest were published between 2014 and 2022. Multiple geographical settings were represented with the majority of studies from Australia (*n* = 7) and the United States (*n* = 4), and two studies each from the Netherlands, Sweden and the United Kingdom. Studies were implemented across various settings including hospitals, outpatient clinics, primary care centres and community centres/organizations.

All 22 studies reported the number and type of participants involved in the co-design process, although demographic information such as age, gender and socioeconomic position of consumers and stakeholders were poorly reported. Almost all of the studies included both consumers and other key stakeholders in their co-design methodology, with the exception of one study[29] that only involved consumers. Consumer perspectives were represented by intervention end-users including patients, caregivers and/or family members (range: 9–178), while key stakeholder groups mostly consisted of healthcare professionals (nurses, doctors and allied health professionals), hospital management staff or representatives from key stakeholder organizations (range: 6–282). Table 1 presents a summary of the study and participant characteristics.

Table 1 Study and participant characteristics

References	Country	Study setting	Study design	Consumer involvement		Stakeholder involvement	
				Participants	*N*	Participants	*N*
Aaby et al.[28]	Denmark	Outpatient	Mixed-methods	CR participants	178	CR staff and CR team leaders	35
Ahmed et al.[29]	USA	Outpatient	Mixed-methods	HF patients and caregivers	43	-	-
Bonner et al.[30]	Australia	Primary care	Mixed-methods	CVD patients and people with ≥1 CVD risk factor	34	GPs	282
Breeman et al.[31]	Netherlands	Outpatient/community	Qualitative	CVD patients, patient representatives and CR participants	35	Nurses, cardiologists, physical therapists, GPs, psychologists, neurologists, lifestyle coaches	58

Cornet et al.[32]	USA	Outpatient	Mixed-methods	HF patients and family/friends	73	Cardiologists, clinic supervisors and managers, technicians, nurses	7
Dorri et al.[33]	Iran	Hospital	Qualitative	ACS patients and family members	38	Nurses, faculty members, cardiologists, educational supervisors	10
Driver et al.[34]	USA	Hospital	Mixed-methods	Stroke patients and carers	32	Physiotherapists, occupational therapists, speech therapists, exercise specialists, dieticians, a stakeholder organization representative	22
Hjelmfors et al.[35]	Sweden	Outpatient	Mixed-methods	HF patients and family members	11	Physicians and nurses	25
Kjork et al.[47]	Sweden	Outpatient/community	Mixed-methods	Stroke patients	22	Nurses, occupational therapists, physicians, physiotherapists, neuropsychologists, speech therapists	11
Lalonde et al.[36]	Canada	Primary care	Qualitative	CVD patients and family members	20	Physicians, pharmacists, nurses, nutritionists, psychologists	52
Pekmezaris et al.[37]	USA	Community	Qualitative	Patients, caregivers and patient advocates	10	Cardiologists, geriatricians, nurses, pharmacists, health policy workers	8
Prick et al.[48]	Netherlands	Hospital	Mixed-methods	Stroke patients and carers	77	Neurologists, rehabilitation specialists, geriatricians, nurses, occupational therapists, physiotherapists, speech therapists	111
Ramage et al.[49]	Australia	Community	Mixed-methods	Stroke patients and carers	16	Healthcare workers	21
Raynor et al.[38]	UK	Hospital/primary care	Mixed-methods	HF patients and carers/family members	75	GPs, heart failure nurses, heart failure consultants, pharmacists, hospital managers	103

Redfern et al.[39]	Australia	Outpatient	Mixed-methods	Angina/MI patients and people with ≥1 CVD risk factor	52	Allied health professionals, medical staff, cardiologists, nurses	6
Sabater-Hernandez et al.[40]	Australia	Community	Qualitative	AF patients and patients with hypertension	9	Pharmacists, GPs, cardiologists, cardiac/research nurses, a stakeholder organization representative	11
Toledo-Chavarri et al.[41]	Spain	Community	Mixed-methods	IHD Patients	25	GPs, nurses, cardiologists	10
Tongpeth et al.[42]	Australia	Hospital/community	Mixed-methods	ACS patients	20	Cardiologists, cardiac nurses, cardiac researchers	12
Triantafyllidis et al.[43]	UK	Hospital/community	Mixed-methods	HF patients and caregivers	78	Cardiologists, nurses, GPs, hospital administrators	23
Walsh et al.[44]	Ireland and Belgium	Hospital	Mixed-methods	CVD Patients	72	Community, representatives, GPs, nurses, cardiologists, CR coordinators	31
Woods et al.[45]	Australia	Hospital/community	Mixed-methods	HF patients and carers	13	Nurses, cardiologists, allied health professionals	20
Zacharia et al.[46]	Australia	Outpatient/community	Qualitative	Stroke patients and carers	14	Stroke rehabilitation clinicians, dietitians	15

Abbreviations: ACS, acute coronary syndrome; AF, atrial fibrillation; CR, cardiac rehabilitation; CVD, cardiovascular disease; GP, general practitioner; HF, heart failure; IHD, ischaemic heart disease; UK, United Kingdom; USA, United States of America. **Intervention characteristics**

The interventions developed across studies were broad, but all aimed to improve the management of CVD or prevent further cardiovascular events. Five interventions focused on the broad area of CVD secondary prevention, whereas other interventions focused on specific cardiac disorders such as heart failure (*n* = 7), stroke (*n* = 5), acute coronary syndrome (*n* = 2), coronary heart disease (*n* = 1), atrial fibrillation (*n* = 1) and ischaemic heart disease (*n* = 1). Fourteen studies described the development of an e-health intervention such as health data dashboards or

online platforms,[29–31,41,44,47,48] mobile/tablet applications[32,42,43,45] and telehealth programmes.[37,46,49] Two interventions aimed to improve or address the health literacy of patients,[28,41] three focused on improving patient self-empowerment and behaviour change,[34,41,49] and there were two exercise-based interventions.[44,49] Almost all interventions (*n* = 20) included patient education on the importance of healthy behaviours for cardiovascular health or information on self-management of CVD risk factors (medications, exercise, diet, etc.). The interventions of the included studies are described in detail in Supporting Information: File 2.

Co-design methodology

Although study authors used various terminology and described different definitions of co-design research, all the included studies adhered to the principles of consumer engagement, where consumers and relevant stakeholders formed a partnership with researchers and took an active role in intervention development. Most of the studies stated that they used a 'co-design' approach (10 studies), while other studies identified their approach as participatory action research (3 studies), user-centred design (3 studies), community-based participatory research (2 studies), participatory research (2 studies) or co-production (*n* = 1). One study did not report using co-design methodology but instead described their approach as 'gaining patient and healthcare professional input'.[39] Many different roles for participants were also described. This varied from advisors and committee members who provided advice on co-design methodology, reviewers who examined and assessed the implications of findings from co-design tasks, and co-designer roles where the participants were integrally involved in intervention development. Research methods and co-design activities varied across studies: the predominant co-design activities conducted with consumers and stakeholders across studies were focus groups (14 studies), semistructured interviews (13 studies), co-design/development workshops (11 studies), advisory group/working group meetings (7 studies) and ideas/brainstorming meetings (5 studies). Eight studies tested the intervention in a 'real world' feasibility study that provided feedback on usability, acceptability or suitability. Table 2 details the co-design methodologies utilized in the included studies.

Table 2 Co-design methodology of included studies

References	Co-design methodology		Boyd et al.[17] elements of co-design					
	Approacha	Methods/activities	Engage	Plan	Explore	Develop	Decide	Change
Aaby et al.[28]	Co-design	User health literacy assessments; Semistructured interviews; Focus groups; Ideas workshops	X	✓	✓	✓	✓	✓
Ahmed et al.[29]	Participatory research	Focus groups; Design workshops; Feasibility study	X	X	✓	✓	✓	✓
Bonner et al.[30]	Co-design	'Think aloud' interviews; Semistructured interviews; Design workshops; Feasibility study	X	✓	✓	✓	✓	✓

Breeman et al.[31]	User-centred design	Semistructured interviews; Usability workshop; Focus groups; Stakeholder workshop; 'Think aloud' interviews	X	✓	✓	✓	✓	✓
Cornet et al.[32]	User-centred design	Critical incident interviews; Scenario-based cognitive interviews; Usability evaluation workshops; Advisory group meetings	X	X	✓	✓	✓	✓
Dorri et al.[33]	Participatory action research	Brainstorming meetings; Semistructured interviews; Focus groups	✓	✓	✓	✓	✓	✓
Driver et al.[34]	Community-based participatory research	Focus groups; Advisory board meetings	✓	✓	✓	✓	✓	✓
Hjelmfors et al.[35]	Co-design	Ideas workshops; Feasibility study	X	✓	✓	✓	✓	✓
Kjork et al.[47]	Co-design	Expert panel meetings; Focus groups; Interviews; Surveys; Co-design workshops	✓	✓	✓	✓	✓	✓
Lalonde et al.[36]	Participatory research	Needs assessment workshop; Focus groups; Appropriateness surveys; Working-group meetings; Semistructured interviews	X	✓	✓	✓	✓	X
Pekmezaris et al.[37]	Community-based participatory research	Focus groups; Co-design workshops	✓	✓	✓	✓	✓	✓
Prick et al.[48]	User-centred design	Steering group meetings; Surveys; Focus groups; Co-creation workshops; Think-aloud interviews	X	✓	✓	✓	✓	✓
Ramage et al.[49]	Co-production	Co-production team meetings; Ideas workshops; Development workshops; Interviews	✓	✓	✓	✓	✓	✓
Raynor et al.[38]	Co-design (experience-based)	Hospital ward observations; Semistructured interviews; Co-design meetings; Feasibility study	✓	✓	✓	✓	✓	✓
Redfern et al.[39]	-	Semistructured interviews; Readability and suitability questionnaires	X	X	✓	✓	✓	✓
Sabater-Hernandez et al.[40]	Co-design	Focus groups; Semistructured interviews	X	X	✓	✓	✓	✓

Toledo-Chavarri et al.[41]	Co-design	Listening labs; Focus groups; Development workshops; Online feasibility activities	X	✓	✓	✓	✓	✓
Tongpeth et al.[42]	Participatory action research	Focus groups; Co-design workshops; Feasibility study	X	✓	✓	✓	✓	✓
Triantafyllidis et al.[43]	Participatory action research	Semistructured interviews; Co-design workshops; Feasibility study	X	✓	✓	✓	✓	✓
Walsh et al.[44]	Co-design	Stakeholder expert panel; Semistructured interviews; Focus groups; Co-design workshops; Usability testing workshops and questionnaires	X	✓	✓	✓	✓	✓
Woods et al.[45]	Co-design	Semistructured interviews; Co-design workshops; Development focus groups; Feasibility study	X	✓	✓	✓	✓	✓
Zacharia et al.[46]	Co-design	Collaboration meetings; Co-design workshops	✓	✓	✓	✓	✓	✓

a

As stated by the study authors.

Step 1: Engage

Seven studies reported establishing meaningful relationships with consumers and stakeholders, which was the least reported co-design process utilized across the included studies (Table 3). Five studies[34,37,46,47,49] developed a type of community advisory board or expert panel comprised of patients, healthcare professionals and key stakeholders that either defined the research questions, devised notions to engage participants in the co-design process or participated in the co-design process to identify the essential elements of the intervention. One study undertaken in a hospital setting[38] involved meeting with hospital management staff to identify key services and/or staff who could act as champions for the co-design process. One study[33] engaged nurses and nurse managers of the cardiac care unit via several informal meetings throughout the co-design process to identify ways to address readmissions.

Table 3 Summary of co-design elements

Phase	Element	Number of studies including phase (%)
Exploratory Phase	Engage	7 (32)
	Plan	18 (82)
	Explore	22 (100)
Development Phase	Develop	22 (100)

	Decide	22 (100)
	Change	21 (95)
Implementation Phasea	Evaluation	10 (45)

a

This phase is not part of Boyd et al.'s[17] framework for co-design.

Step 2: Plan

Eighteen studies reported working with consumers and/or stakeholders to generate ideas about study goals and how to achieve them. The most common approach used in the planning phase was organizing stakeholder/expert or community advisory board meetings utilized in eight studies.[31,37,38,42,46–49] Group discussions/focus groups with participants were used in six studies[30,34,36,41,43,44]; and four studies[28,33,35,45] ran ideas/brainstorming workshops to generate ideas/goals and devise plans on how to achieve them.

Step 3: Explore

All 22 studies reported learning about consumers' or stakeholders' experiences with CVD to identify the challenges and priorities to improve CVD secondary prevention. Most studies undertook focus groups or interviews with consumers to better understand patient needs for healthcare or knowledge/awareness of cardiovascular health, or interviews with healthcare professionals to explore their experiences of working in cardiac healthcare services or to discover professionals' needs regarding helping patients with CVD. Only two studies used quantitative approaches in their need assessment by means of a cross-sectional survey analysis.[28,48]

Steps 4–5: Develop and decide

All 22 studies included an intervention development phase where ideas from consumers and stakeholders were discussed and turned into potential solutions. These steps were described differently across all studies (focus groups; co-design workshops; development workshops; advisory group meetings and working group meetings); however, generally included consumers and stakeholders coming together multiple times to discuss intervention ideas based on the needs of consumers, prioritize ideas for interventions and design an intervention prototype(s).

Step 6: Change

Twenty-one studies conducted pretesting of intervention prototypes with end-users to gather further feedback on core attributes of the intervention design and to assess the acceptability and usability of the intervention. The majority of studies (n = 17) utilized a mixed-method approach in gathering feasibility data, while the other four studies used qualitative approaches such as design workshops to elicit feedback before the intervention was finalized.

Intervention effectiveness

Intervention effectiveness was assessed in 10 studies[30,33–35,42–44,46,48,49] showing mixed results (Supporting Information: File 2). In a pre-post feasibility study with 98 GPs, Bonner et al.[30] found that the use of a newly designed CVD online platform increased the capacity for GPs to correctly identify the CVD risk categories of their patients (16% for low-risk cases, 32% for moderate-risk cases and 50% for high-risk cases). The educational intervention developed by Hjelmfors et al.[35] improved the self-reported knowledge of heart failure, confidence and skills of 13 cardiac nurses (patients were not included in the evaluation). Dorri et al.[33] also employed a pre-post study to evaluate a nurse-led, hospital-based intervention in 31 acute coronary syndrome patients, which resulted in hospital

readmission rates reducing from 32.2% to 12% at 6 months. Three studies[42–44] conducted a randomized controlled trial to test the intervention created by the co-design process. The patient education Avatar app developed by Tongpeth et al.[42] was found to significantly improve knowledge, attitudes and beliefs of heart attack symptoms in intervention participants compared to controls at a 6-month follow-up (n = 70 patients), but no significant differences between groups were observed in healthcare utilization (GP visits, ED visits or 30-day readmissions). In a randomized trial with 202 heart failure patients that tested the efficacy of a digital home monitoring system with an integrated risk prediction and disease management service, Triantafyllidis et al.,[43] reported no statistically significant between-group differences in treatment opportunity or health-related quality of life at a 6-month follow-up. The randomized controlled trial by Walsh et al.[44] evaluated an e-health intervention for the self-management of CVD risk factors in 120 CVD patients and found an increase in moderate to vigorous physical activities and a stable CVD risk score in intervention participants compared to controls at a 6-month follow-up. However, there were no significant differences between groups regarding most physical fitness outcomes, health-related quality of life, mental health, exercise self-efficacy, medication adherence or diet. Four studies[34,46,48,49] reported commencing a randomized controlled trial, and therefore, results were not available at the time of undertaking this review.

Sufficiency of reporting

The median score of the sufficiency of reporting using our amended CASP checklist was seven out of a maximum score of nine (range: 4–9), indicating that most studies described their co-design methodology adequately. Authors of all studies reported the aims of the project clearly and the majority of studies reported the study results, including the design of the final intervention, and co-design study schedule/procedures. Common omissions of information identified across studies included not reporting the study setting clearly and inadequately describing the characteristics of participants or how they were recruited (Supporting Information: File 3).

DISCUSSION

This is the first review on the use of co-design methodology in the development of CVD secondary prevention interventions. Our findings confirmed that the co-design approach is still an emerging field in CVD research, as evidenced by the small number of studies found in the literature with the majority of these published in the past 5 years. Studies were implemented across various settings with consumers and stakeholder groups most frequently consisting of patients and healthcare professionals, respectively. Consistent with co-design literature,[50] varying models of co-design methodology utilizing different research activities were reported by studies. Findings from this review suggest it is feasible to apply the principles of co-design in various settings and CVD population groups.

The lack of a singular consistent definition of ‘co-design’ made it difficult to retrieve relevant literature for this review. Many research co-design approaches were identified within the literature, with extensive variability in the methods, research phases, participants and levels of involvement. Based on the sufficiency of reporting checklist, most studies in this review described their co-design methodology clearly and adequately, making it easier for others to replicate co-design in their own research. Conversely, previous reviews of co-design have reported that studies provided insufficient details of their co-design activities in their methods to establish what was actually involved.[18,50,51] This difference in reporting may be explained by our comprehensive inclusion criteria to draw together the varying definitions and terminology used, which was guided by a co-design framework.[17] During the full-text review stage of the literature search, study methodology was evaluated against Boyd et al.'s[17] co-design framework to determine if the intervention was developed using established co-design methodology; that is, consumers and/or relevant stakeholders were involved in most aspects of intervention development, from the needs assessment through to content development and pilot testing. Over 40 studies were excluded based on this definition, including 6 studies that specifically stated they used a co-design or participatory action research approach. After closer inspection of

these six studies, consumers and stakeholders either participated only in a need assessment to guide researchers in the development of the intervention (Exploratory Phase) or were asked to provide feedback on an intervention that was already developed by the researchers (Development Phase). For the purposes of this review, this was not considered a 'true' co-design approach. The heterogeneity in approaches, variable use of terminology and gaps in reporting indicate that this area of research may benefit from a framework that sets out the core principles for cardiac services seeking to use co-design.

Engagement with consumers and stakeholders is critical to true and successful co-design.[17] Although all 22 studies reported learning about consumers' or stakeholders' experiences with CVD (i.e., performed a needs assessment) and included them in the development phase of the intervention, few studies reported methods of establishing meaningful relationships with consumers and stakeholders. This involved strategies such as developing a community advisory board comprised of key stakeholders, identifying local champions to oversee the study, and engaging with key staff before beginning the research to help guide the co-design process. Previous studies have identified that selecting stakeholders strategically to fit project needs, adapting the project to the practical needs of stakeholders and clearly defining roles and expectations for stakeholders, as well as responsibilities and powers, are key strategies to establishing meaningful partnerships in research.[52] Broad engagement principles such as these need to be present when using co-design methods to develop effective and participatory research relationships, rather than engaging stakeholders merely to fulfill a requirement. Additionally, having stakeholders involved early means that their experiences and requirements can be taken into account at the start of the process rather than researchers presuming to know what is required.[53] Researchers and policymakers who are new to the co-design approach should consider how involving end-users and stakeholders in the study planning phase can assist in prioritizing research topics, setting research agendas and helping to refine research design and processes.

Assessing the effectiveness of co-designed interventions in formal evaluations and clinical trials is important to determine the efficacy of this method.[18] There is minimal literature that has evaluated the direct links between co-designed CVD interventions and improved patient experiences or outcomes. In this review, intervention effectiveness was only assessed in 10 studies and mixed results were reported across studies. While few studies to date have investigated the effectiveness of co-designed interventions in CVD secondary prevention, a recent systematic review and meta-analysis found that co-designed public health interventions for various health conditions can improve short-term health outcomes such as self-efficacy, healthy behaviours and lifestyle changes, health service access and physical health.[54] Further to this, the majority of studies included in this review can be considered complex interventions (i.e., having multiple interacting components or target groups and settings).[55] An important element of complex interventions is understanding 'how' and under what circumstances an intervention can achieve its desired effect, often termed the 'mechanism of change'. These mechanisms, or causal links, are more transparent when 'theory' is applied to the development of an intervention, also facilitating meaningful evaluation of outcomes.[55] Of the included studies, only seven applied a theoretical framework during the intervention development. These included the Behaviour Change Wheel,[30,46] the Social Cognitive Theory,[44] the Medical Research Council (MRC) framework,[38] the Ophelia approach[28] and the Integrated Knowledge Translation theoretical approach.[46,49] However, the way in which these theories informed evaluation was not clearly articulated. In other studies, authors proposed suggestions for mechanisms of change, such as tailoring interventions to the needs of end-users,[35,40–42] but these were not underpinned by a theoretical framework. To advance robust reporting of co-designed interventions, authors are encouraged to consider the mechanisms of action in both the design and evaluation of their interventions and to explicitly describe these to readers using public health frameworks such as the Template for Intervention Description and Replication (TIDieR) checklist.[56]

The issue of implementation is increasingly understood to be an integral point of attention throughout the course of intervention development, from conceptualization to evaluation. During the pilot testing/evaluation phase, aspects of implementation are typically addressed through process evaluations,[57] and more recently, through innovative hybrid effectiveness-implementation study designs that provide outcomes on both intervention effectiveness and implementation (e.g., implementation fidelity, the proportion of reach and barriers to implementation).[58] In contrast, issues of implementation are generally addressed within the stages of the co-design approach through dedicated stakeholder engagement. The MRC framework for complex interventions highlights the value of stakeholder engagement throughout all stages of intervention development and evaluation: prioritizing research questions, development of the programme theory, intervention refinement and practical support for evaluation.[55] Engagement of stakeholders can also help to clarify which contextual factors might influence both the implementation and effectiveness of an intervention.[59] Understanding the role of these contextual factors is particularly important when considering scalability (i.e., implementing a locally designed intervention in multiple settings where operating systems and delivery of care are likely to vary).[55] Involving stakeholders who hold relevant, lived experience and including a dedicated focus on implementation in the co-design process may support successful upscaling and dissemination of interventions to a wide range of settings.

Although co-design has been used frequently to develop health interventions for 'underserved' population groups such as people with disabilities or from culturally and linguistically diverse communities,[60–62] it was interesting to find that only one study involving these populations was identified in this review. Pekmezaris et al.[37] included Black and Hispanic patients and disparity experts in their study to create a telemonitoring intervention for heart failure that is acceptable and feasible for use with a lower-income, Black and Hispanic patient population. More studies in this review may have included participants representing diverse groups; however, our sufficiency of reporting analysis found that studies often lacked an adequate description of participant characteristics. The burden of CVD is higher among minority groups and people living with socioeconomic disadvantage, which and is associated with increased mortality rates and a greater risk of subsequent cardiac events.[63] Evidence also shows that these populations receive suboptimal healthcare access compared to other population groups.[64] Healthcare services have an important role to play in addressing this social gradient,[65] by ensuring that the care they provide is accessible for all, including culturally and linguistically diverse communities, minority groups and those with lower socioeconomic status.[66] Use of the co-design approach can take context-specific challenges of 'underserved' population groups into consideration to provide a culturally appropriate and logistically sound CVD intervention that is feasible and acceptable.

Strengths and limitations

The strengths of this review include were our rigorous methodology and comprehensive search strategy. We have confidence that we identified all published studies that met our inclusion criteria as we used various synonyms of 'co-design' in our search strategy. Furthermore, we excluded studies that were assessed as not genuinely utilizing the co-design approach so we could accurately examine the use of co-design in CVD secondary prevention research. By summarizing the co-design processes across multiple studies, we have improved the knowledge base for co-design in CVD research and provided a guide for future CVD researchers considering using these methods.

Limitations of our review should also be considered. Searches were limited to published studies, subjecting this review to the possibility of publication bias. Further to this, because we restricted our search to studies published in English, we may have omitted relevant research written in other languages. It would have been useful to understand the link between specific co-design methods and research outcomes; however, this was not possible due to the heterogeneity of co-design activities in the included studies, although determining intervention effectiveness was not

the primary aim of this review. Lastly, no consumers (i.e., people with lived experience) or stakeholders were involved in undertaking this scoping review, which may have supported a more critical interpretation of the implementation of co-design approaches in the current CVD literature. However, as this was an unfunded review project, we had no resources to conduct consumer involvement activities such as offering adequate education and training (scoping reviews require research knowledge or training that consumers commonly do not hold) and remuneration to consumers for their time.

CONCLUSION

We conducted a scoping review to examine how studies have undertaken co-design to develop CVD secondary prevention interventions. In doing so, we make several practical contributions to the literature. First, the findings from this review provide methodological considerations that can guide researchers and healthcare services in understanding how the co-design approach may be implemented to develop locally feasible, acceptable and sustainable strategies to improve outcomes for CVD populations. Second, by adapting Boyd et al.'s[17] co-design framework, we mapped the co-design activities used in studies to the six steps of co-design to ensure sufficient reporting of methods for future research. Finally, we identified the knowledge gaps in CVD co-design research (e.g., limited research in post-design evaluation, inadequate reporting of levels of consumer involvement), highlighting the importance of assessing the effectiveness of co-designed interventions in future research.

AUTHOR CONTRIBUTIONS

The study was conceived by Jason Talevski and Alison Beauchamp. Jason Talevski conducted the literature search and all authors contributed to the study selection. Jason Talevski and Alison Beauchamp extracted, analysed and interpreted the data. Jason Talevski, Roman Falls, Natali Cvetanovska and Alison Beauchamp conducted the critical appraisal of studies. Jason Talevski drafted the manuscript, and all authors critically revised the manuscript and approved the final version.

ACKNOWLEDGEMENTS

J. T. was supported by an Alfred-Deakin Postdoctoral Research Fellowship. A. B. was supported by a National Health &Medical Research Council Grant (Number: 1198006).

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

No new data were generated or analysed in support of this research.

DETAILS

Subject:	Community based action research; Intervention; Databases; Prevention programs; Participatory action research; Participatory research; Secondary prevention; Telemedicine; Health care; Heart failure; Cardiovascular diseases; Academic staff; Content analysis; Rehabilitation; Caregivers; Disease prevention; Health services; Prevention; Co-design; Workshops; Heart diseases; Consumers; Advisory groups; Terminology; Stakeholders; Community involvement; Design engineering; Research methodology; Cardiovascular disease; Public health; Community participation; Medical personnel
Business indexing term:	Subject: Consumers Stakeholders

Identifier / keyword:	cardiovascular disease; co-design; community-based participatory research; consumers; secondary prevention; stakeholders
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	16-29
Publication year:	2023
Publication date:	Feb 2023
Section:	REVIEW ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Countryof publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Literature Review, Journal Article
Publication history :	
Online publication date:	2022-11-10
Milestone dates:	2022-08-29 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-06-15 (manuscriptReceived); 2022-11-10 (publishedOnlineEarlyUnpaginated); 2022-10-02 (manuscriptAccepted)
Publication history :	
First posting date:	10 Nov 2022
DOI:	https://doi.org/10.1111/hex.13633
ProQuest document ID:	2767102211

Evaluating the role and effectiveness of co-produced community-based mental health interventions that aim to reduce suicide among adults: A systematic review

Hanlon, Claire A [1] ; McIlroy, David [1] ; Poole, Helen [1] ; Chopra, Jennifer [1] ; Saini, Pooja [1] [1] School of Psychology, Faculty of Health, Liverpool John Moores University, Liverpool, UK

ABSTRACT (ENGLISH)

Background

Suicide is a major public health risk requiring targeted suicide prevention interventions. The principles of co-production are compatible with tailoring suicide prevention interventions to meet an individual's needs.

Aims

This review aimed to evaluate the role and effectiveness of co-produced community-based suicide prevention interventions among adults.

Methods

Four electronic databases (PsycInfo, CINAHL, MEDLINE and web of science) were systematically searched. A narrative synthesis was conducted.

Results

From 590 papers identified through searches, 14 fulfilled the inclusion criteria. Most included studies elicited the views and perspectives of stakeholders in a process of co-design/co-creation of community-based suicide prevention interventions.

Conclusion

Stakeholder involvement in the creation of community-based suicide prevention interventions may improve engagement and give voice to those experiencing suicidal crisis. However, there is limited evaluation extending beyond the design of these interventions. Further research is needed to evaluate the long-term outcomes of co-produced community-based suicide prevention interventions.

Patient and Public Involvement

This paper is a systematic review and did not directly involve patients and/or the public. However, the findings

incorporate the views and perspectives of stakeholders as reported within the studies included in this review, and the findings may inform the future involvement of stakeholders in the design, development and delivery of community-based suicide prevention interventions for adults.

FULL TEXT

INTRODUCTION

Co-production is advocated within mental health policy and has garnered increasing attention.[1–3] This is highlighted within health care initiatives including person-centred care,[4] the 'Five Year Forward View for Mental Health' policy strategy[5] and more recently 'The Community Mental Health Framework for Adults and Older Adults—Support, Care and Treatment. Part 1 &2'.[6,7] Within a co-production framework, multiple stakeholders work in collaboration, including commissioners, service providers and service users.[8,9] Emphasis is placed upon shared decision-making and information exchange within a mutually equitable relationship.[2] Subsequently, equal value is placed upon contributions by service users, and service providers and professionals.[2,3]

It is argued that co-production produces meaningful knowledge within the context to which it is to be applied.[9,10] This creates services that are more contextually specific, promoting engagement and bridging the translational gap between research evidence production and real-world implementation.[9,11] Relatedly, co-production improves quality of care,[3,12] having considered service user needs and priorities during the co-production process[1,13] leading to cost-efficient and cost-effective services.[14]

Despite the highlighted benefits of co-production, several limitations have been identified. There remains a lack of consensus in how co-production is defined, leading to interchangeable language used to describe co-production processes.[2,13,15,16] For example, undefined collaborative roles have led to a plethora of collaborative working activities marketed under a co-production umbrella including co-creation and co-design.[13,17,18] This 'one size fits all' approach is attributed to different interpretations in how co-production is operationalized within policy, knowledge creation and subsequently implemented in practice within service delivery.[2,19,20] There is a paucity of evaluation considering the extent to which co-productive approaches cultivate meaningful outcomes[20–22] and whether positive outcomes associated with co-production are sustained over time.[23] Further, reluctance to relinquish professional roles and responsibilities, such as those held by researchers or practitioners, may lead to a power imbalance that could threaten the integrity of the mutually equitable relationship.[9,12]

Mental health services have striven to harness the innovative and transformative potential of co-production in a quest to improve service user inclusivity in decision-making, and service delivery and experience.[1] Suicide is a major public health problem, accounting for over 700,000 deaths worldwide.[24] Help-seeking remains a significant barrier for those at risk of suicide, with fewer than one-third of individuals seeking help for their mental health.[25] The reasons why individuals experiencing suicidal thoughts and behaviours do not seek help from mental health services vary but include high self-reliance, a low perceived need for treatment and stigmatizing attitudes towards suicide and/or mental health problems and seeking professional help.[26] In recognition of such barriers, there has been a call for suicide prevention interventions to be tailored to improve reach and increase effectiveness.[27]

The principles of co-production are congruent with tailoring suicide prevention interventions to suit the needs of individual service users and are aligned to recovery-orientated services that emphasize individualized care and recognize the value of experiential knowledge.[6,7,28] Research is emerging that supports implementation of co-produced mental health service provision. For example, studies evaluating the impact of recovery colleges featuring co-production have reported positive outcomes upon service-user well-being such as improved self-esteem or confidence,[29] improved employment opportunities[30] and reduced use of mental health services.[31] Additionally, applying co-production to tailor delivery of mental health services such as the Improving Access to Psychological Therapies to improve reach among black and minority ethnic communities has shown increased accessibility and retention.[32] Further, Pocobello et al.[33] reported a 63.2% reduction in hospitalizations and a 39% decrease in psychiatric medication use or withdrawal among service users of an experimental co-produced mental health service versus traditional mental health services. Findings such as these are encouraging; however, qualitative findings

pervade this field and there remains a paucity of quantitative research assessing the impact of co-production within mental health service provision,[34] even less so in relation to suicide prevention. While studies focusing upon the preventative aspect of co-produced mental health services assert that they prevent service user mental health from reaching crisis point,[34] validated assessment of this impact is lacking.

As highlighted, co-production does have its limitations, which need to be mitigated for the potential of co-production in suicide prevention to be fully embraced. Key to furthering understanding of the role of co-production within suicide prevention relies upon understanding the language used to define co-production; evaluating how and to what extent service providers and service users contribute to the co-produced service and how information is synthesized, and outcomes are assessed. Therefore, this review aims to evaluate the role and effectiveness of co-produced, community-based suicide prevention interventions for adults that aim to reduce suicide to:

•1.

Understand how co-production is defined and operationalized.

•2.

Examine evidence for the role of co-production in these interventions.

•3.

Identify and evaluate co-production-related outcomes associated with these interventions.

•4.

Identify and evaluate intervention components associated with a reduction in suicide-related outcomes.

METHODS

The protocol for this review was registered on the University of York, Systematic Review database PROSPERO (CRD42020221564).[35] The research questions and inclusion and exclusion criteria were generated using the patient/problem or population, intervention, comparator and outcome (PICO) framework.

Eligibility criteria

Studies were eligible for inclusion if they fulfilled the following criteria:

•1.

Population: Adults aged 18 years or older.

•2.

Intervention: Co-produced community-based mental health interventions that aim to reduce suicidal risk, thoughts and/or behaviour and/or those that include subanalyses for participants described as experiencing suicidal crisis or at risk of suicide were included. Treatment studies focusing upon clinical populations were excluded; however, co-produced community-based studies examining the effects of prevention interventions to reduce suicide risk (e.g., self-harm, depression) were included if these data were reported as separate subanalyses. In addition, studies that broadly focussed upon mental health but clearly reported co-produced outcomes and suicide prevention outcomes were included.

•3.

Comparator: It was unnecessary for included studies to have control group comparators. However, it was expected that some studies such as randomized-controlled trials that fulfilled the inclusion criteria would compare intervention outcomes with a control group (e.g., usual care). Therefore, comparators could be no intervention or control group, or comparison with a different intervention group.

•4.

Outcomes: As the goal of suicide prevention interventions is to prevent suicide, changes in suicide risk and/or suicide-related behaviours (e.g., suicide ideation) comprised the primary outcome. Both qualitative and quantitative studies (including cross-sectional and longitudinal studies) that assessed changes in suicidal risk and behaviour were assessed against the eligibility criteria. Quantitative studies using both standardized and nonstandardized measures were eligible for inclusion. Intervention-based studies measuring outcomes over a period of follow-up were included only if suicide risk was reported (e.g., self-reported) at baseline and at each follow-up point and were re-revaluated at follow-up at least 1 week beyond baseline. Number of follow-ups and type of suicide risk behaviour assessed were not determinants for inclusion. A narrative evaluation of service features of interest (e.g., co-production definition and operationalization) was reported. Secondary outcomes were changes in psychological well-being and quality of life.

Only studies published in English were included and no geographical or publication date restrictions were imposed. This was to capture the breath of co-production-based studies within the literature.

Search strategy

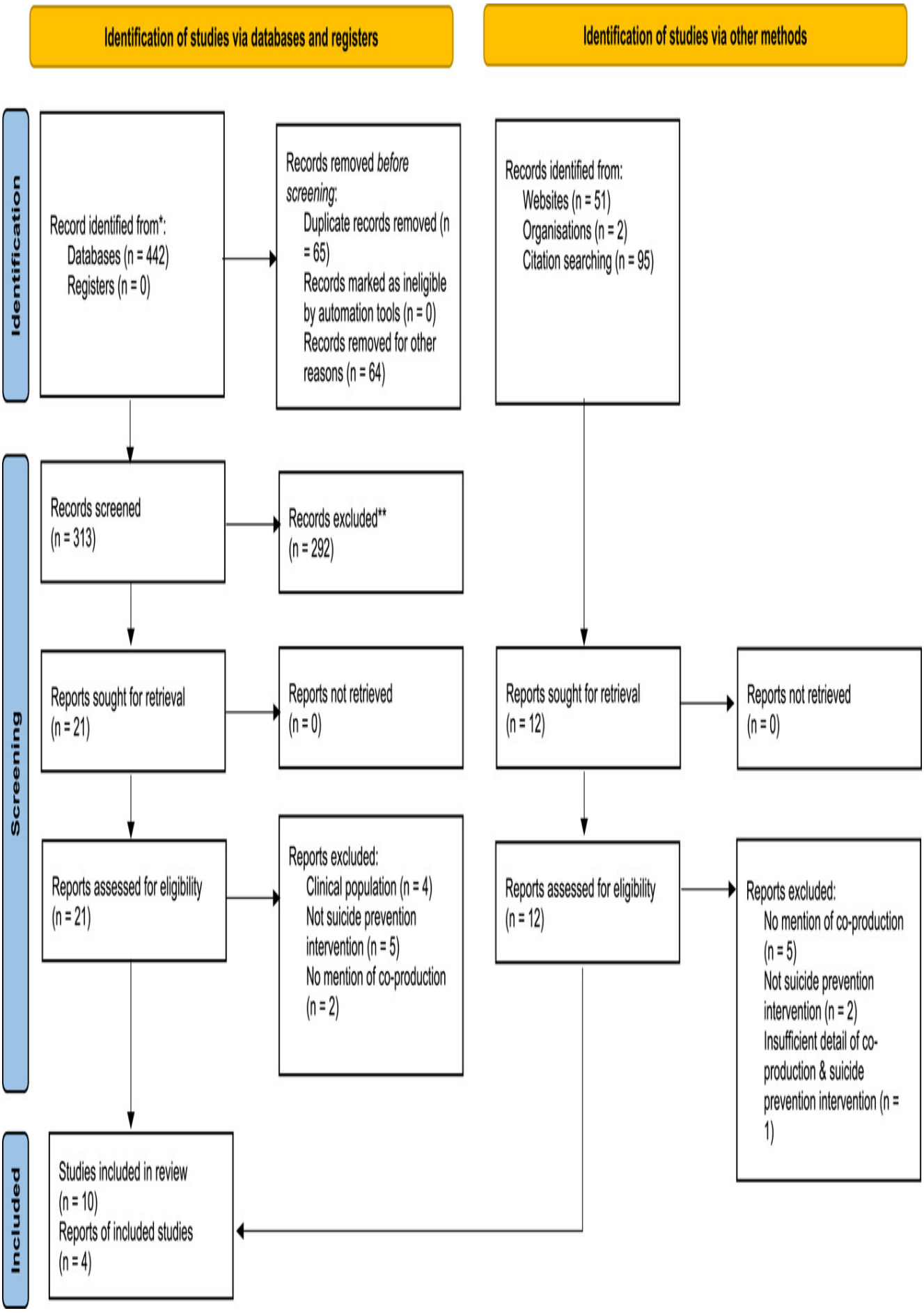
Four electronic databases (PsycINFO, CINAHL, MEDLINE, Web of Science) were searched. Studies published in English to the 21 March 2022 were eligible for inclusion. Filters were not applied during the search for type of study. Systematic reviews were excluded, but back searches of reference lists were checked for additional relevant studies that fulfilled the inclusion criteria.

Search terms

Scoping of the literature was undertaken in the development of the search terms exploring the extent of co-production in the context of community mental health. Consequently, a broad search strategy was developed to ensure that all relevant papers were captured. The search strategy utilized relevant terms for co-production (e.g., ‘co-product*’, ‘co-design*’, ‘co-create’), suicide (e.g., ‘sucid*’) and community mental health (e.g., ‘community mental health’) (see Appendix A, e.g., search terms).

Study selection

The primary author removed duplicate studies from the final search and independently screened the titles and abstracts of the remaining studies against the eligibility criteria. The co-authors also independently screened titles and abstracts according to the inclusion and exclusion criteria. Full-text studies meeting the eligibility criteria were retrieved and reviewed for inclusion by the primary author. Two co-authors reviewed all full-text papers for comparison. Disagreements were resolved through discussion within the team at the title and abstract stage and by one co-author at the full-text screening stage. The PRISMA flowchart documents the screening process (see Figure 1). Fourteen papers were identified as eligible for inclusion.



Enlarge this image.

Data extraction and quality assessment

Data were extracted by the primary author and transferred onto a data extraction sheet that was created and piloted before use. The following details were extracted: (1) study characteristics including study design and co-production definition if included (Table 1) and (2) intervention characteristics including intervention type and study outcomes (Table 2).

Table 1 Study characteristics

References	Study aims/purpose	Design and methods (inc. measures used to assess suicide risk/behaviour)	Focus population of intervention	Age range	Community setting	Quality assessment rating

Bruce and Pearson,[44] Country: US	To describe the aims and methodology to be used to test and evaluate the PROSPECT (Prevention of Suicide in Primary Care Elderly: Collaborative Trial) intervention, a model of depression recognition and treatment aimed at preventing and reducing suicide among older adults.	Descriptive paper, including a fictional case study, which describes a longitudinal study design planned to be used to test and evaluate the PROSPECT intervention. Proposed use of the Centers for the Epidemiologic Studies Depression (CESD) scale to screen potential participants for depression during recruitment. Eligible participants would undergo further in-person assessment for depression and other clinical, neuropsychological and social variables. Telephone follow-ups at 4 and 8 months and bi-annual administration of the full research assessment battery are proposed. It is unclear what measures would determine depression- and suicide-related risk/behaviours	Community-dwelling elderly depressed primary care patients from 18 sites within 3 geographical areas in the US were the focus population, with collaborative working between physicians and health care specialists.	Focus population age range: 780 aged 60–74 years and 600 aged 75 years and older.	18 primary care sites located in 3 geographical areas	MMAT = 20% QuADS Q = 1

		beyond screening participants for inclusion.				
Buus et al.,[49] Country: Australia and Denmark	To examine stakeholders' suggestions and contributions to the design, function and content in the development of an existing app called MYPLAN aimed towards individuals in or at risk of suicidal crisis.	An instrumental case study involving a qualitative study using focus groups and participatory workshops.	People in or at risk of suicide crisis. Study participants, including MYPLAN app users, relatives and clinicians, worked collaboratively with the researchers and software developers revised the app.	Reported mean age range of participants: 16–46 years.	Online—A Safety planning mobile phone app	MMAT = 80% QuADS Q = 2

Cheng et al.,[50] Country: Hong Kong	Aimed to investigate the impacts of promoting suicide prevention using social media and to evaluate the co-creation process involving a popular YouTuber.	Mixed methods. Qualitative analysis of the co-creation process in the development of a YouTube suicide prevention short film. Video statistics (e.g., views) generated online, an online survey and online public comments evaluated video impact and effectiveness. Suicide risk/behaviours assessed within the online survey using two questions about suicide thoughts in the past 12 months and help-seeking.	Social media users (e.g., YouTube).	Viewers of the YouTube short film ages ranged from 13 to 44 years. Respondents filled in an online survey—ages are reported to have ranged from 12 to below 65 years.	Online—Youtube video	MMAT = 80% QuADS Q = 1

Chopra et al.,[38] Country: UK	Aimed to evaluate the effectiveness of James' Place Model and to conduct a social value assessment of the service to provide an understanding of the potential social, economic and environmental impact of James' Place.	Case series study involving quantitative assessment of James' Place Model effectiveness. Suicide risk assessment conducted collaboratively between a therapist and service user with a safety plan, a CORE-OM self-report questionnaire, referrer evaluation of precipitating factors (e.g., relationship breakdown) and therapist assessment of various psychological, motivational and volitional factors (e.g., entrapment, perceived burdensomeness).	Adult men experiencing suicidal crisis.	Adults aged 18 years and older.	Community-based, face to face	MMAT = 80% QuADS = 1

Ferguson et al.,[39] Country: Australia	This study aimed to explore the perspectives and experiences of workers providing case management, support or counselling to refugee and asylum seeker clients on co-developed personalized safety plans.	Qualitative study involving semistructured interviews with workers from nongovernment organizations providing case management, support or counselling to refugees and asylum seekers.	Refugees and asylum seeker clients.	Age not given	Unclear	MMAT = 100 % QuADS = 1
Hetrick et al.,[48] Country: Australia	This study aimed to Co-design with young people a mobile phone app-based self-monitoring mood tool that facilitates communication of this with a clinician.	Participatory design and studio design method were used in the development of the app, which followed human-centred principles. This involved workshops and focus groups with young people and clinicians.	Young people experiencing depression	Young people aged 18–24 years.	Online community	MMAT = 100 % QuADS = 3

Richardson et al.,[40] Country: Ireland (both Northern &Southern Ireland)	The Young Men and Suicide Project (YMSP) aimed to develop a range of mental health initiatives to promote positive mental health among young men in Ireland and to assess the efficacy of these.	Mixed methods involving a literature review to identify best practice, online surveys with stakeholders including community-based services, education services and prisons and focus groups service providers and men to understand what works with young men in mental health service provision. Findings informed the development and piloting of two initiatives called 'Mind Yourself' and 'Work out'. Pre- and postmeasures of self-esteem, depression and resilience were assessed in the Mind Yourself programme. Validated psychometric tests (e.g., six items from the General Health Questionnaire-12 [GHQ-12]) taken pre-, during and postintervention	Young men	Northern Ireland initiative targeted adolescents (age not specified). Southern Ireland initiative targeted young men (age not specified).	School Online	MMAT = 60% QuADS = 2

		in the ‘work out’ programme assess changes in mental fitness.				

Saini et al.,[41] Country: UK	This study aimed to evaluate the effectiveness of the James' Place Mode in reducing suicidality in men using the service and to conduct a social value assessment of the service to provide an understanding of the potential social, economic and environmental impact of James' Place.	Mixed methods. Qualitative methods included semistructured interviews with men who had used the service and written responses to interview questions from a GP. Quantitative analyses of pre- and postoutcome data. Quantitative and qualitative findings were triangulated to understand the wider social value of James' Place. Suicide risk assessment conducted collaboratively between a therapist and service user with a safety plan, CORE-OM self-report questionnaire, referrer evaluation of precipitating factors (e.g., relationship breakdown) and therapist assessment of various psychological, motivational and volitional factors (e.g.,	Adult men experiencing suicidal crisis.	18 years and older	Community-based, face-to-face delivery of a suicide prevention model	MMAT = 100 % QuADS = 1

		entrapment, perceived burdensomeness).				
Saini et al.,[42] Country: UK	This study aimed to evaluate the effectiveness of the James' Place Model in reducing suicidality in men over a 2-year period and to compare the findings pre- and post-COVID-19 pandemic.	Mixed methods. Semistructured qualitive interviews with therapists. Quantitative analyses of pre- and post-CORE-OM outcome data to assess the effectiveness of the James' Place Model. Suicide risk assessment conducted collaboratively between a therapist and service user with a safety plan, CORE-OM self-report questionnaire, referrer evaluation of precipitating factors (e.g., relationship breakdown) and therapist assessment of various psychological, motivational and volitional factors (e.g., entrapment, perceived burdensomeness).	Adult men experiencing suicidal crisis.	18 years and older	Community-based, face-to-face service temporarily moved to online delivery during the COVID-19 pandemic	MMAT = 100 % QuADS = 1

Saini et al.,[43] Country: UK	Aimed to evaluate an innovative suicidal crisis intervention for younger men (18–30 years) versus older men (31 years and older).	Case series study involving quantitative assessment CORE-OM scores and clinical records of psychological, motivational and volitional factors associated with participants' suicidal crisis and CORE-OM scores. Suicide risk assessment conducted collaboratively between a therapist and service user with a safety plan, CORE-OM self-report questionnaire, referrer evaluation of precipitating factors (e.g., relationship breakdown) and therapist assessment of various psychological, motivational and volitional factors (e.g., entrapment, perceived burdensomeness).	Adult men experiencing suicidal crisis.	18 years and older (age range 18–66 years)	Community-based, face-to-face delivery of a suicide prevention model.	MMAT = 40% QuADS = 3

Thorn et al.,[51] Country: Australia	This study aimed to improve dissemination of and engagement with the #Chatsafe guidelines by including young people in the design and development of a social media campaign to promote safe web-based communications about suicide. Objectives of the study were to document key elements of the co-design process, evaluate young people's experiences of the co-design process and capture young people's recommendations for the #Chatsafe suicide prevention campaign.	Mixed methods. Participatory co-design approach involving 11 workshops with young people. Workshop activities included a warm-up, co-design activities evaluation and cooldown. At the end of each workshop, participants were invited to complete a quantitative evaluation survey including questions on demographics, perceived benefits from participation and workshop acceptability and safety. Safety protocols (e.g., wellness plan) and monitoring (e.g., workshop evaluation survey/debrief) were included.	Young people accessing the web	17–25 years	Online community	MMAT = 80% QuADS = 3

Wilcock et al.,[45] Country: UK	Evaluation of the Offload programme, a men's rugby-league community-based mental health programme.	Mixed methods involving pre- and post-intervention questionnaires (n = 699) exploring aspects related to health and well-being (e.g., resilience, social support). Also, focus groups and case studies with men who engaged with the Offload programme. Provision was available to assess men using the Patient Health Questionnaire-9 (PHQ9) and/or the General Anxiety Disorder scale (GAD7) if facilitators delivering the intervention were concerned about a participant's well-being. Facilitators were also able to seek advice from a mental health clinician. These measures were not routinely given for the assessment of suicidal risk/behaviours. Men did, however, self-report mental	Community, sport-based intervention for men experiencing mental health illness (anxiety and depression) to prevent development of complex mental illness and suicide.	Men aged 16 years or older	Community-based	MMAT = 60% QuADS = 3

		health conditions/diagnoses.				
Wilcock et al.,[46] Country: UK	This study aimed to explore stakeholder perspectives of the key design characteristics and the roles played by delivery staff in the conception and development of a community-based men's rugby mental health programme called Offload.	Qualitative study involving one-to-one semistructured interviews with 18 programme designers and delivery staff.	Community, sports-based intervention for men experiencing mental health illness (anxiety and depression) to prevent development of complex mental illness and suicide.	Intervention targets men aged 16 years or older.	Community-based	MMAT = 100 % QuADS = 2

Zealberg et al.,[47] Country: US	To describe the development of the collaboration between emergency psychiatric services and the police.	Descriptive paper outlining development of a mobile crisis programme involving collaboration between emergency psychiatric services and the police, which includes case studies to illustrate collaboration. It is unclear how suicidal risk/behaviours were determined. However, it appears that this involved a subjective or clinical assessment (e.g., a clinical history) of the situation made by police and/or psychiatric team members responding to incidents.	Community population experiencing psychiatric crisis.	Age of the focus population not specified.	Community-based	MMAT = 40% QuADS = 1

Note: MMAT refers to the Mixed Methods Appraisal Tool.[36] QuADS Q refers to the question derived from the Quality Assessment with Diverse Studies quality assessment tools.[37]

Table 2 Intervention characteristics

References	Intervention details	Co-production methodological approach	Co-production and/or suicide-related outcomes

Bruce and Pearson[44]	Delivery of a comprehensive treatment algorithm for depression adapted from the Agency for Health Care Police and Research (AHCPR) guidelines. Antidepressant therapy or Interpersonal Therapy (IPT), if antidepressants were unwanted by the patient, was to be recommended. A health specialist (e.g., nurse, social worker or clinical psychologist) was to 'prompt' physicians to facilitate timely and recommended treatment decisions by advocating for patients (e.g., obtaining and providing feedback of information on patient symptoms and treatment experiences to the physician). Education was also to be provided to patients, families and physicians on depression and suicide ideation. However, it is unclear who delivered this aspect of the intervention.	Collaboration between a health specialist (e.g., nurse, social worker or clinical psychologist) and physician to facilitate timely and targeted identification and treatment of depression among older adults. It was proposed that the health specialist would liaise with the patient, help the physician to recognize depression and make treatment recommendations within the remit of the PROSPECT intervention guidelines based upon patient information/monitoring and encourage treatment adherence among patients.	No co-production outcomes(s) provided. Outcomes proposed to assess the effectiveness and impact of the intervention relate to depressive symptomatology (e.g., suicide ideation, hopelessness, depression and suicidal risk behaviours including substance abuse and disturbed sleep). Authors estimated that 18% of participants would experience depression at baseline. No evaluation of suicide-related outcomes provided.

Buus et al.[49]	App-based intervention called MYPLAN combining three preventative strategies around safety planning, help-seeking from peers and professionals and restriction of access to lethal means. An additional feature promotes help-seeking behaviour by including a map and directions to an emergency room nearest to the users' location.	Focus groups and participatory workshops were used to further develop the MYPLAN intervention. This involved engagement between participants, software developers and researchers in the design, evaluation and revision of MYPLAN app prototypes in response to participant feedback. Emphasis was placed upon personal experiences of using MYPLAN and evaluation of its wireframe, functionality and whether the app was culturally suited to an Australian user audience. Software developers revised and developed prototypes in response to user feedback.	Thematic analysis led to the development of 3 phases of user involvement in the development of the MYPLAN app relating to ‘suggestions of core functions’, ‘refining functions’ and ‘negotiating finish’. Increased participant engagement with researchers and software developers during the later stages of user-involving processes as the app became increasingly revised. The revised MYPLAN app included the suicidal ideation attributes scale (SIDAS) to measure suicide ideation, a mood ratings tracker and a customizable list of personal warning signs of crisis. No evaluation of the impact of the intervention upon suicidal risk/behaviours reported.

Cheng et al.[50]	Short film designed to reduce suicidality and promote help-seeking behaviours. The storyline of the film focused upon a suicidal university student and a taxi driver who encourages the former to seek help. Also featured is an obscured scene of a suicide method (hanging).	Co-creation of a YouTube short film involving a popular YouTuber and researchers. To inform this process, the YouTuber engaged with literature, online material and staff and clients from a local suicide survivor service.	Thematic analyses of the co-creation process identified three facilitating factors of 'shared concern about youth suicide prevention', 'enriched knowledge of lived experience with suicide' and 'preserve the uniqueness of the YouTuber', and one barrier: 'the balance between realism and appropriateness of content'. Overall, positive perceived changes in audience suicide prevention knowledge, attitudes and behaviours reported. Mixed views received from qualitative feedback and public comments. Some respondents who had suicidal thoughts and provided qualitative feedback (*n* = 22) reported that the storyline resonated with their situation (e.g., academic and life stress; *n* = 6), one felt that the film helped to alleviate stress and another felt that it motivated them to live. Three respondents criticized the film. Public comments (*n* = 164) generally supported the film (e.g., 10.8% showed support to people in distress). Eight commentators reported past suicidal thoughts; four had attempted suicide. Two commentators with suicide intent reported abandoning their suicide plans after watching the film. One commentator displayed current suicidal thoughts and another endorsed suicide as an option.

Chopra et al.[38]	A community-based suicide prevention intervention underpinned by three prominent suicidal theories (interpersonal theory of the suicide, collaborative assessment and management of suicidality and the integrated motivational–volitional theory of suicide). Emphasis is on the therapist and service user co-producing the therapeutic intervention together. Brief therapeutic approaches and interventions (e.g., behavioural activation, sleep hygiene) focussed upon reducing suicidal distress and developing resilience and coping are delivered.	Co-production of the suicide prevention intervention and safety planning with men engaged in the service and therapists delivering the James' Place Model. Co-production with stakeholders (including academics, clinicians, commissioners, therapists and experts-by-experience) also informed service inception, design and delivery.	Feedback evaluations completed by 18% of men (39/212) indicated that the James' Place service was perceived as a safe and welcoming therapeutic setting and improved overall mental well-being and coping. No formal evaluation of co-production reported. Significant mean reduction in CORE-OM scores for men who completed assessment and discharge questionnaires. No relationship found between the precipitating factors and levels of general distress, or between those with or without each precipitating factors.

Ferguson et al.[39]	To explore the perspectives and experiences from workers who provide case management, support or counselling to refugee and asylum seeker clients on co-created personalized safety plans.	Co-production discussed in the context of co-creating safety plans. The theme from worker interviews, ‘safety planning as a co-created, personalised activity’, highlights the workers' perspectives that safety planning should be a collaborative process and personalized to the individual.	Four themes developed: ‘Safety planning as a co-created, personalised activity for the client’; ‘therapeutic benefits of developing a safety plan’; ‘barriers to engaging in safety planning’ and ‘strategies to enhance safety planning engagement’. Overall, these highlight the perceived facilitators, barriers and strategies to enhance safety planning as a suicide prevention intervention for refugees and asylum seekers. Benefits of co-production reported included equitable working relationship between the client and the worker, recognition of the client's expertise and flexibility and creativity to tailor and co-creation safety planning using alternative modes (e.g., photographs, drawings). Perceived therapeutic benefits of co-created safety planning included increased awareness of distress triggers among clients and coping strategies, use of personalized strategies to interrupt suicidal thoughts and normalization of their suicidal experience. No formal evaluation of suicide-related outcomes provided.

Hetrick et al.[48]	Development of a mobile phone app designed to enable monitoring of mood with feedback for users and clinicians. Users able to customize the app to suit their preferences. Features included mood monitoring (named 'well-being checker') with space to record factors influencing users' mood; brief personalized interventions to support young people in the time between face-to-face appointments linked to the well-being tracker such as distraction techniques to reduce stress (e.g., meditation, games and breathing techniques) and a photo album to promote positive emotion (e.g., photos, supportive messages from friends and loved ones, music playlists); lastly, a one-touch safety feature enabling users to contact emergency services and their supporters.	Co-design workshops with young people and two focus groups with clinicians designed to elicit information sharing and generation of concepts for the app. Young people sketched design features of the app and gained feedback from the group on their individual design. The group created a design using the best ideas from individual designs in a process called feature prioritization. This informed subsequent co-design rounds until consolidation of the best ideas resulted in the final design. Clinicians proposed their needs and concerns of monitoring young people using an app before the co-design workshops took place. In a second focus group with clinicians, a young person involved in the co-design workshops presented the app wireframes and clinician feedback gained on the app design and its use in practice.	Various app features supported co-production between the app user and clinician (e.g., the onboarding process, tailoring of trigger points within the well-being checker). The well-being tracker mood rating function incorporated trigger points for high distress to assess suicide risk/behaviours. No formal evaluation of the effectiveness of the app in reducing suicidal risk/behaviours was reported, but it was proposed that it could enhance help-seeking.

Richardson et al.[40]	Northern Ireland: 'First Instinct' a whole community approach, aimed to encourage help-seeking among the young men. This involved development of the 'Mind Yourself' brief mental health intervention; young men's advisory/reference group; training programmes for practitioners focused upon developing work with men and creation of a 'working with men' resource library offering off-the-shelf resources for practitioners. Southern Ireland: 'Work Out', a mental fitness app, was developed that aimed to improve help-seeking, social connectedness and mental health literacy. Comprised of a series of brief online interventions (called 'missions') underpinned by cognitive behavioural therapy principles that aimed to address four areas: being practical, building confidence, taking control and being a team player.	Various components of intervention design, development and delivery involved co-production. An advisory group of key men's health and suicide prevention representatives supported and oversaw intervention development. Local stakeholder (e.g., from community-based services, education services, prisons and young men) views on the extent and nature of mental health/suicide prevention initiatives for young men in Ireland and the perceived facilitators and barriers of working with young men elicited through surveys and focus groups informed intervention development. Northern Ireland: Local community members delivered the Mind Yourself programme. A young men's advisory forum/reference group was set up by staff from a local organization and involved local youth leaders as 'co-workers' and facilitators in its delivery. Southern Ireland intervention development involved collaborative working between developers of the Irish version of 'work out' and developers of the Australian version through data sharing. Focus groups involving young men provided feedback on 'Work out' during intervention development and testing.	Facilitators of Mind Yourself perceived the programme as effective, but some barriers were identified (e.g., literacy issues hindering questionnaire completion). Positive feedback from the young men advisory/reference group reported suggested that participants reflected positively upon their involvement (e.g., welcomed the opportunity to focus on issues affecting men in an equitable way with other stakeholders). Mind Yourself evaluation showed no significant change in pre- and postmeasures of self-esteem, depression and resilience. Feedback-suggested Work Out was perceived as acceptable and accessible. No suicide-related outcomes reported.

Saini et al.[41]	A community-based suicide prevention intervention underpinned by three prominent suicidal theories (interpersonal theory of the suicide, collaborative assessment and management of suicidality and integrated motivational-volitional theory of suicide). Emphasis is on the therapist and service user co-producing the therapeutic intervention together. Brief therapeutic approaches and interventions (e.g., behavioural activation, sleep hygiene) focussed upon reducing suicidal distress and developing resilience and coping are delivered.	Co-production of the suicide prevention intervention and safety planning with men engaged in the service and therapists delivering the James' Place Model. Co-production with stakeholders (including academics, clinicians, commissioners, therapists and experts-by-experience) also informed service inception, design and delivery.	Elements of co-production were evident in the design and delivery of the James' Place Model. For example, men spoke of the utility of the 'lay your cards on the table' component for exploring factors underpinning their suicidal crisis and for exploring coping strategies, and described improved mood, motivation and family relationships. No formal evaluation of co-production provided. Impact of the intervention on suicidal crisis evaluated using CORE-OM scores. The initial overall mean CORE-OM score on entry to the service was reported as 85.5 (*n* = 137) and the mean overall discharge score was reported as 38.9 (*n* = 60). The mean reduction in CORE-OM scores was reported as 46.6. Psychological factors related to men's suicidality (e.g., impulsivity, thwarted belonginess, hopelessness) reported. No relationship between precipitating factors and general distress levels found at initial assessment, or between those with and without each precipitating factors found.

Saini et al.[42]	A community-based suicide prevention intervention underpinned by three prominent suicidal theories (interpersonal theory of the suicide, collaborative assessment and management of suicidality and integrated motivational–volitional theory of suicide). Emphasis is on the therapist and service user co-producing the therapeutic intervention together. Brief therapeutic approaches and interventions (e.g., behavioural activation, sleep hygiene) focussed upon reducing suicidal distress and developing resilience and coping are delivered.	Co-production of the suicide prevention intervention and safety planning with men engaged in the service and therapists delivering the James' Place Model. Co-production with stakeholders (including academics, clinicians, commissioners, therapists and experts-by-experience) also informed service inception, design and delivery.	Co-production evidenced within therapist interviews in the management of men engaged in the service during remote delivery of the James' Place Model. Formal evaluation of co-production was not performed. Impact of the intervention on suicidal crisis evaluated using CORE-OM scores. Evaluation of 2-year intervention effectiveness showed an initial overall mean CORE-OM score on entry to the service of 86.56 (n = 322) and a mean overall discharge score of 35.45 (n = 145). The mean reduction in CORE-OM scores was reported as 50.9. Evaluation of CORE-OM scores suggested that the James' Place model was as effective, if not more, during COVID-19.

Saini et al.[43]	A community-based intervention underpinned by three prominent suicidal theories (interpersonal theory of the suicide, collaborative assessment and management of suicidality and integrated motivational–volitional theory of suicide). Emphasis is on the therapist and service user co-producing the therapeutic intervention together. Brief therapeutic approaches and interventions (e.g., behavioural activation, sleep hygiene) focussed upon reducing suicidal distress and developing resilience and coping are delivered.	Co-production of the suicide prevention intervention and safety planning with men engaged in the service and therapists delivering the James' Place Model. Co-production with stakeholders (including academics, clinicians, commissioners, therapists and experts-by-experience) also informed service inception, design and delivery.	A clinically significant reduction in the mean CORE-OM scores between assessment and discharge for both younger and older men engaged with the James' Place Model intervention reported. No significant difference in distress scores between younger versus older men at assessment and discharge. However, younger men showed lower levels of distress compared to older men at initial assessment and lower levels of wellness than older men at discharge. No formal evaluation of co-production. Assessment of psychological, motivational and volitional factors reported. Younger men were less affected by entrapment, defeat not engaging in new goals and had positive attitudes towards suicide than older men at assessment. Older men at discharge were significantly more likely to have an absence of positive future thinking, less social support and entrapment than younger men.

Thorn et al.[51]	A social media campaign aiming to promote safe web-based communication about suicide.	An iterative process of co-design whereby learning from workshops informed the next workshop. Workshop facilitators (e.g., researchers and designers) guided design activities. Co-design activities facilitated peer-to-peer mapping of young people's social media usage and communication of suicide on the web, idea generation (e.g., campaign themes and content) and testing of and feedback on the design protocol for the campaign. Three key elements comprised the co-design process: 1. 'Define' involved mapping young people's social media usage, their communication about suicide and determined how young people wanted #Chatsafe guidelines to be integrated into the campaign; 2. 'Design' involved integrating young people's perspectives and addressing their wants and needs in the campaign development including campaign themes and delivery methods; 3. 'User-testing' involved prototype testing and gaining feedback. A collaborative approach ensured participant safety (e.g., a researcher accompanied distressed participants to a private space to enact the young person's wellness plan).	Overall, co-design workshops were perceived by participants as acceptable, beneficial and safe, although some participants reported feeling suicidal (n = 8) or unsure whether they felt suicidal (n = 6) after workshops. Findings support the feasibility of safe involvement of young people in the development of co-designed recommendations (e.g., content and format) for a web-based suicide prevention campaign to enhance its acceptability among young people. Positive outcomes of feelings of improved ability to communicate online about suicide and to identify others who may be at risk of suicide were reported.

Wilcock et al.[45]	Ten-week, education-based intervention that uses the rugby league brand to address low-level mental health problems (e.g., low self-esteem, depression and anxiety). Rugby-related language is used to normalize mental health, promote intervention accessibility, acceptability, engagement and adherence. Comprised of 10 sessions (called 'fixtures') aimed at raising awareness of mental health problems (e.g., low self-esteem, anxiety, depression), tackling stigma and encouraging the development of coping strategies. Sessions were comprised of two, 40-min halves.	Coproduction is evident in the design and delivery of Offload. The design phase involved collaborative working partnerships between Rugby League Cares, State of Mind, three Rugby League Club's charitable foundations (Salford Red Devils Foundation, Warrington Wolves Foundation and Vikings Sports Foundation) and over 200 men from the targeted population who participated in interviews, focus groups and questionnaires exploring their views of mental health intervention provision. Findings from men's participation informed the intervention name, where (i.e., from rugby stadiums) and when the intervention is delivered, the language used (i.e., rugby-centric) and the content of the intervention (e.g., type of self-care tools to use). Foundation managers/lead, former players and coaches, officials, mental health and mindfulness specialists were involved in the delivery of Offload.	The co-produced programme content was perceived as more relatable. Accessibility, use of nonclinical language and informal setting (i.e., rugby league stadiums) were perceived to encourage help-seeking and to remove stigma. Additional reported benefits include increased confidence and self-esteem, improved coping, social connectedness, increased social support, willingness to talk about mental health and reduced suicide ideation and/or attempts. Pre- and postintervention questionnaire findings showed positive improvement in nine outcomes reported relating to areas including coping, resilience, engagement in sport and identification of support around the men. For example, approximately three-quarters of participants reported improved awareness of how to look after their health and well-being, coping and better able to manage setbacks and challenges.

Wilcock et al.[46]	Ten-week, education-based intervention that uses the rugby league brand to address low-level mental health problems (e.g., low self-esteem, depression and anxiety). Rugby-related language is used to normalize mental health, promote intervention accessibility, acceptability, engagement and adherence. Comprised of 10 sessions (called 'fixtures') aimed at raising awareness of mental health problems (e.g., low self-esteem, anxiety, depression), tackling stigma and encouraging the development of coping strategies. Sessions were comprised of two, 40-min halves.	Coproduction is evident in the design and delivery of Offload. The design phase involved collaborative working partnerships between Rugby League Cares, State of Mind, three Rugby League Club's charitable foundations (Salford Red Devils Foundation, Warrington Wolves Foundation and Vikings Sports Foundation) and over 200 men from the targeted population who participated in interviews, focus groups and questionnaires exploring their views of mental health intervention provision. Findings from men's participation informed the intervention name, where (i.e., from rugby stadiums) and when the intervention is delivered, the language used (i.e., rugby-centric) and the content of the intervention (e.g., type of self-care tools to use). Foundation managers/lead, former players and coaches, officials, mental health and mindfulness specialists were involved in the delivery of Offload.	Thematic analysis generated three themes reflecting the importance of co-production in the co-design of the intervention: 'tacit forms of knowledge are essential to initial programme designed'; 'stigma-free and non-clinical environments appeal to and engage men' and 'lived experience and the relatability of personal adversity'. Co-production was perceived to improve intervention reach and engagement by using nonstigmatizing language and delivering the intervention in a nonjudgmental, nonclinical environment. Delivery of solution-focused activities provided by men with lived experience was perceived to promote relatability and trustworthiness. Suicide-related outcomes were not formally evaluated. Delivery of the intervention by former professional sportspeople who recalled their lived experience of mental illness/adversity was perceived to possibly promote modelling of alternative masculine behaviours that could potentially enhance mental health and help-seeking.

Zealberg et al.[47]	An emergency psychiatry-mobile crisis programme linking key professionals, specifically mental health professionals (e.g., Master's-level clinicians in nursing, counselling, psychology, social work) with the police to provide mobile, crisis intervention. Clinicians supported police officers in a consultative role during police incidences involving people experiencing serious mental health illness. Clinicians would obtain a history from the individual, neighbours, family and friends, drug and alcohol use and establish trust and a therapeutic alliance with the individual. Details on three case studies are provided and intervention techniques, for example developing a rapid therapeutic alliance with a woman threatening to jump from a ledge and holding her there while police assembled a safety net below.	Collaboration between the police and clinicians allowed clinicians to liaise with the individual experiencing crisis to encourage a peaceful resolution to specific situations. This was facilitated through regular meetings with law enforcement officials, reclarification of mutual responsibilities and expectations and reviewing of critical situations. This partnership was further affirmed through debriefing of police officers following incidents, providing mental health referrals for police officers and being informal consultants.	Outcomes reported relate to three case studies and involve de-escalation of police incidents with individuals experiencing crisis.

RESULTS

The PRISMA diagram (Figure 1) illustrates the screening process. Five hundred and ninety papers were identified by searching databases (*n* = 442) and other methods (148). After the removal of duplications and nonrelevant papers (e.g., book titles, conference submissions), 449 titles and abstracts were screened. Of these, 33 papers were retrieved for full-text screening. Fourteen studies fulfilled the inclusion criteria.

Description of studies

Table 1 presents a description of the characteristics of the included studies. Studies either had a qualitative (*n* = 6), mixed methods (*n* = 6) or quantitative design (*n* = 2). Notably, some studies (*n* = 5) focused upon the delivery of suicide prevention interventions online, including via apps (e.g., mobile phone apps) (*n* = 3), YouTube (*n* = 1) or to inform safe online web-based communications (*n* = 1). Most of the remaining studies were community-based and delivered the intervention face-to-face (*n* = 9). Most studies focussed upon suicide prevention among younger to older adults aged 16 years or older (*n* = 10). One study targeted older adults aged 60 years or older (*n* = 1), another focussed upon intervention delivery for adolescents and young men (*n* = 1) and two studies did not stipulate the age of the target population (*n* = 2).

Methodological quality

The Mixed Methods Appraisal Tool (MMAT)[36] and an additional question taken from the Quality Assessment with Diverse Studies (QuADS) quality assessment tool[37] to evaluate stakeholder inclusion through co-production, were used to assess methodological quality. All studies were independently assessed by the first author (C. A. H.) and the last author (P. S.) independently assessed the quality of 10% of the included studies. MMAT revealed a range in

methodological quality assessment (see Table 1). However, most studies assessed were of high quality, with nine studies scoring 80%–100%. Studies scored low to moderate in quality in terms of co-production inclusion, appraised using the QUADS as described. No studies were excluded from this review based on quality assessment.

Synthesis of findings

Findings were synthesized to produce a narrative summary describing the role of co-production in community-based suicide prevention interventions.

Definition and operationalization of co-production

Half of the studies directly refer to co-production as a methodological approach in the design of the suicide prevention intervention.[38,39,41–43,45,46] None of the studies provide an explicit definition of co-production. Rather, most individual studies were found to integrate key elements of co-production within the design and/or delivery of an intervention by involving stakeholders, representing the diverse modes in which co-production can be applied. All studies featured stakeholders working collaboratively towards some shared goal as a function of co-production. Most studies mention stakeholder involvement in the development and design of suicide prevention interventions (*n* = 13). In five studies[40,44–47] stakeholders, including health professionals and those with lived experience, delivered the suicide prevention interventions. Also, in five studies, those trained to deliver the suicide prevention intervention worked collaboratively with the recipient, adapting the intervention (e.g., safety plans and talk therapy) to suit their individual needs.[38,39,41–43] A diverse range of stakeholders participated in the studies. Stakeholders included health professionals, clinicians, mental health specialists, police officers,[38–49] community representatives including sporting representatives (e.g., ex-rugby players) and community leaders,[38,40–43,45,46] YouTubers,[50] those who are representative of theor with lived experience/or with lived experience.[38,40–43,45,46,48–51]

Facilitators of co-production

Stakeholders mainly engaged through an iterative process to elicit their perspectives on functional aspects and/or the content of the design and development of the suicide prevention intervention (*n* = 13). This was facilitated either through focus groups/workshops[40,45,46,48,49,51] and/or one-to-one discussions with stakeholders including researchers, those with lived experiences and a YouTuber.[38,39,41–43,45,46,50] Seven studies[38,39,41–44,47] integrated co-production that was discursive in nature between key partners during the delivery of the suicide prevention intervention. In Bruce and Pearson's[44] study, a health professional was nominated to advocate for the patient and to assist physicians in the recognition of depression to allow timely intervention. In contrast, discussions around the intervention and to troubleshoot potential problems that may occur during implementation were held between local police agencies before and during intervention delivery in Zealberg et al.[47] Conversely, co-production informed service design and delivery of four studies focusing upon a suicide prevention intervention for men experiencing suicidal crisis.[38,41–43] Co-production was integrated in the creation of personalized safety plans for asylum seekers and refugees.[39] Discussions acted as a forum for rapport building, enabling improved collaboration between diverse professional disciplines and people with lived experience. For example, Zealberg et al.[47] attribute ‘prior working discussions’ with local police agencies to redressing problems and building trust within the collaborative working relationship, a key factor in the successful implementation of their suicide prevention intervention. Studies identified that discussions among stakeholders provided an opportunity for negotiation and consensus-seeking when addressing disagreements that may arise during intervention development or delivery.[40,47–50] Cheng et al.[50] report that researchers expressed concern over the inclusion of a suicide scene of hanging in the co-creation of a suicide prevention video with a YouTuber for example. The YouTuber felt that the inclusion of this scene was imperative to maintaining the authenticity of the video's storyline. However, the YouTuber adapted the scene once the researchers explained the potential for contagion effects.

Challenges of co-production

The evidence highlights some challenges that may hinder the inclusion of co-production in the design and/or implementation of suicide prevention interventions. During co-production, both parties must be willing to engage when working collaboratively. This issue is highlighted in Ferguson et al.'s[39] study exploring the views and perspectives of workers supporting asylum seekers and refuges in the co-creation of safety planning. Workers perceived a lack of 'client readiness' to engage in safety planning (e.g., unwillingness to write a safety plan down) as a potential barrier hindering the co-production of personalized safety planning.

A reluctance of professionals to relinquish power was evident. Hetrick et al.[48] reported clinician resistance towards the inclusion of service users in shared decision-making and accessing a mobile App (mApp). Similarly, Buus et al.[49] reported that software designers included a suicidality rating scale against the wishes of stakeholders involved in the design and development of an mApp. Conversely, three studies emphasize the importance of each stakeholder maintaining the boundary of their individual area of expertise when working in partnership.[47–49] Failure to do so could affect the safety of professionals and service users during intervention delivery[47] and unduly burden parents/clinicians with notifications alerting them to the suicidality risk of their child/patient,[49] particularly out of working hours.[48] Some safeguarding concerns were highlighted. These centred around whether participation may have induced suicidal feelings and[50,51] also the implications of clinicians being alerted to client suicidality out of hours and not being able to respond to this.[48] Similarly, Thorn et al.[51] highlight some challenges of gaining ethical approval to undertake co-productive methodologies in suicide prevention research, and the additional burden on resources that safety protocol development and the monitoring of stakeholder well-being may have.

Benefits of co-production

Integrating co-production within the methodological approaches provided opportunity for knowledge sharing between partners to create new knowledge that could be applied to shape aspects of the suicide prevention intervention design and/or delivery. Areas of new knowledge included the identification of gaps in existing suicide prevention approaches, the adaptation of suicide prevention interventions to better suit intervention user needs and to improve reach among the targeted population. For example, Thorn et al.[51] used new learning generated in stakeholder workshops to inform the schedule of subsequent workshops during the design and development of a suicide prevention campaign associated with the #Chatsafe project to improve reach among the targeted population. The consultation of stakeholders, whether they have professional or lived experience expertise, encourages consideration of suicidality and suicide-related risk factors through a different lens. Including stakeholders with lived experience promotes reaching back to gain a deeper understanding of the issues that matter, informing the adaptation of suicide prevention interventions to suit the needs and preferences of their targeted population. This effect is reported in 12 studies.[38–43,45,46,48–51] Richardson et al.[40] undertook an extensive consultative process involving an advisory group, with the views of service providers and young men considered. This revealed to the researchers the issues that men experience that may place them at risk of suicide such as 'resistance to connection' and 'stigma attached to mental illness and mental health' and ways to better engage and reach young men within community settings. This acquired new learning-informed intervention development that engaged community partnerships and young men from the targeted population. For example, 'train the trainer' within the Mind Yourself intervention enabled facilitators to consider different ways of engaging the targeted population before formal delivery. Similarly, in setting up a suicide prevention service for men, diverse stakeholder views informed service inception, design and delivery of James' Place reported in Chopra et al.[38] and Saini et al.[41–43]

New knowledge acquired through stakeholder involvement led to intervention development with content adapted to suit the targeted population. Buus et al.[49] described how participants involved in the co-design adapted features of

their mApp-based suicide prevention intervention. This included mood descriptors that could be customized by the user and change nonclinical language used to describe core functions of the app (e.g., 'warning signs' was changed to 'well-being checker'). This is also evident in the delivery of the James' Place Model, where co-production is used to tailor the suicide prevention intervention to suit the individual needs of men.[38,41–43] Similarly, Ferguson et al.[39] reported that participants in their study recognized individuals as being the expert of their own life when co-creating and co-developing safety plans with refugees and asylum seeker clients. Also, the rugby-themed Offload programme[45,46] was perceived as more relatable as it was delivered by those with lived experience of mental health conditions, used nonclinical language and was implemented within an informal, nonclinical environment (i.e., Rugby stadiums). In this sense, co-production provides voice and autonomy in decision-making for individuals accessing a suicide prevention intervention.

Outcomes associated with co-produced community-based suicide prevention interventions

Eleven studies reported participants gaining positive and enriching experiences from their involvement in co-production-based methodologies irrespective of the nature of this involvement (e.g., co-design, co-production of the suicide prevention intervention, etc.). These included beneficial/suicide literacy,[51] enthusiasm,[48] therapeutic benefits including normalizing suicidal experiences and being able to identify unique triggers and coping strategies,[39] rapport and trust building,[47] an enriching process,[50] sharing of experiences in focus groups/debrief,[49] receiving psychological support within a safe and supportive therapeutic environment,[41] improved relationships, coping and understanding of health and well-being needs[45] and being involved in the decision-making process alongside the therapist during the co-production of therapy.[38,41,42]

A lack of formal evaluation of outcomes associated with the suicide prevention intervention is evident. This is likely in part due to the type of studies included, the majority of which focused upon the co-design of the intervention. Nine studies[38,40–45,47,50] propose or report some evaluation of the intervention impact. However, only half embedded formal evaluation of outcomes pre- and postdelivery of the intervention.[38,40–45] Bruce and Pearson[44] proposed baseline measurement of various measures in their study, including depression and social variables to allow monitoring by health professionals, and anticipated that approximately 18% of their cohort would present at baseline with suicide ideation. They go on to report that these measures would be repeated at two annual follow-up interviews and anticipated a reduction in depressive symptomatology and suicide ideation and behaviour. Cheng et al.[50] report that participants gained improved web-based suicide literacy skills. Zealberg et al.[47] provide case studies to illustrate how three lives were saved by their emergency crisis support team intervention. Richardson et al.[40] found no significant change in self-esteem, depression and resilience in their 'Mind Yourself' suicide prevention intervention. However, they report gaining a valuable understanding of barriers related to procedural aspects of intervention delivery including extending the programme duration and the need to consider literacy levels among the target population. Lastly, four studies evaluating a suicide prevention intervention specifically for men assessed pre- and postintervention changes using the CORE-OM clinical assessment tool.[38,41–43]

Mechanisms of behaviour change associated with co-production

None of the included studies explicitly identify the mechanisms of behaviour change associated with the inclusion of co-production. Subsequently, it is impossible to determine whether any potential behaviour change related to suicide and/or mental health can be definitively attributed to the inclusion of co-production. Nevertheless, all studies link reported outcomes to positive changes engendered by engagement in the suicide prevention intervention such as self-monitoring of mood/well-being,[48] improved help-seeking,[39–42,45,46,48–50] rapid access [41–42,44–48] and improved coping strategies.[38–42,45,46,48–49]

Most studies do not specifically report on the theory underpinning suicide prevention interventions, despite a wide

range of techniques being used to reduce suicidality. Four studies describe three models of suicide underpinning the suicide prevention intervention,[38,41–43] namely, the interpersonal theory of suicide,[52] the collaborative assessment and management of suicidality[53] and the integrated motivational–volitional theory of suicide.[54,55] However, these studies each focus upon evaluating the same suicide prevention intervention, the James' Place Model. Similarly, Hetrick et al.[48] link the functionality of the content of their mApp to Dialectical Behavioural Therapy and Thorn et al.[51] relate features of their #chatsafe to the resilient-focussed Papageno effect. In addition, while not explicitly theory-based, Buus et al.'s[49] mApp and the safety planning intervention used by Ferguson et al.[39] are based upon Stanley and Brown's[56] safety planning tool.

DISCUSSION

This review has synthesized research evidence to understand how co-production is defined and operationalized, and to examine how co-production is implemented. In addition, the aim was to evaluate the outcomes assessed and to identify core components within community-based suicide prevention interventions that aim to reduce suicide among adults. The study findings show that most included studies were qualitative (or were mixed methods including a qualitative element), aiming to elicit the perspectives and opinions of service users to inform the design and development of community-based suicide prevention interventions. Few studies reported quantitative findings. The rationale for why and how a co-productive approach was to be implemented was mostly explained (e.g., to elicit stakeholder perspectives to inform intervention development). However, some studies omitted a clear definition of the nature of co-production applied. This finding is consistent with the literature, where an agreed definition of co-production is yet to be determined.[2,17,18] As a result, the concept of co-production is interpreted to mean different forms of activities, commanding different levels of involvement, responsibility and resources within shared decision-making that are couched under the umbrella of co-production.[16,18,19] This points to a wider issue within the field of co-production research as a lack of consensus in how to define co-production means there is no clear metric against which to evaluate the multilevel components of co-production. Smith et al.[13] argue that researchers should abandon efforts to define co-production in favour of embracing heterogeneity co-production offers within research and instead provide a contextually specific definition suited to their research objectives. Others echo this and go further by advocating the abandonment of the pursuit for a gold standard definition of co-production arguing that different approaches are needed to allow tailoring of the co-productive approach to suit the context in which it is implemented.[57] Instead, they urge researchers to be more reflective upon their application of co-productive approaches and be more explicit in their reporting to overcome issues associated with poor operationalization of co-production.[57] Indeed, co-production has been applied across different health-related contexts including mental health.[58] However, it is important for researchers to identify distinct measurable components of the co-production approach used to facilitate the evaluation of any potential outcomes associated (i.e., you need to know you are evaluating to evaluate it).[2]

Involvement of stakeholders from diverse disciplines and backgrounds, and the collaborative working relationships formed were viewed as positive. Iterative discussions between stakeholders were the lynchpin to the success of this collaborative working partnership, giving voice to stakeholders in shaping the suicide prevention interventions. Equity within collaborative working partnerships in co-production is the cornerstone of this approach.[11,34,59] Yet, resistance from some researchers, developers and clinicians towards relinquishing power was evident. For example, a software developer in Thorn et al.'s[51] study included a safety feature despite the users explicitly expressing that they wished for this feature to be omitted. This power differential is common within the co-production literature[59–61] and can lead to tokenistic approaches in co-production-based research.[59,62,63] Redressing power imbalances is important for promoting a culture that empowers stakeholders, particularly service users, to share their knowledge.

Failure to do so risks undermining equity within the collaborative relationship, leading to professional knowledge being prioritized over lay knowledge.[63] However, methods to integrate key values of co-production to avoid potential pitfalls, including power in-balance, have been proposed (e.g., INVOLVE).[10]

Within this review, participants' preferences of intervention content challenged researchers' and clinicians' preconceived ideas of what intervention elements should be included (e.g., Hetrick et al., study).[48] A shift away from 'one size fits all' approaches in suicide prevention interventions towards a tailored approach has been called for.[27,64] Co-production offers an opportunity to work with the individual to identify and address their unmet needs in developing a tailored intervention approach to suicide prevention. Research evidence supporting the implementation of a co-productive approach within service design and delivery of a suicide prevention intervention is emerging. This is highlighted by studies involving the James' Place Model, which aims to support men experiencing suicidal crisis and has been found to significantly reduce suicidal distress.[38,41–43] Relatedly, participants in Ferguson et al.'s[39] study noted the value of co-creation in formalizing personalized safety planning with their clients for the recognition of unique triggers of distress and coping strategies to mitigate this.

The focus of this review was upon co-production within community-based suicide prevention interventions for adults. Several papers identified within the search referred to mobile app or online suicide prevention interventions. The authors determined it to be appropriate to include these studies as technological advancement towards web-/app-based suicide prevention highlights a new, burgeoning community that warrants further research to understand the potential effectiveness of these types of interventions. Web-/app-based suicide prevention could facilitate rapid access to support for individuals experiencing suicidal crisis. However, increased accessibility may add an additional burden to those who monitor such interventions as highlighted by some included studies (e.g., Hetrick et al., study).[48] Additionally, the very nature of web-/app-based suicide prevention interventions requires users to have the relevant access to technology to support their ability to access such interventions. Therefore, whilst web-/app-based technology provides a conduit for remote delivery of rapid suicide prevention intervention, it also may further widen health inequalities for the most vulnerable including those of low socioeconomic status and the elderly.[65,66]

A key strength of this review was the broad inclusion criteria used to capture multiple modes of co-production implementation (e.g., co-design, co-create, co-production). Second, the PRISMA reporting guidelines have also been followed. Thirdly, a second reviewer has been involved during each phase of this review, thus reducing risk of bias within the results. The findings of this review should be interpreted with caution due to the small number of included papers, inclusion of only papers published in English and the homogeneity of the study populations (i.e., westernized populations). Last, while multiple modes of co-production were included in the search criteria, the searches of databases were limited to title searches that may have led to some studies being inadvertently omitted.

Implications for policy and practice

The present review findings provide some evidence that co-production can work in practice to engender positive outcomes. However, a lack of universal definition and established model for co-production implementation may pose some problems when creating policy and practice guidance for the implementation of co-production within suicide prevention interventions. For example, different modes and levels of stakeholder involvement in co-production activities were evident within the included studies, but their involvement was predominantly limited to the co-design aspect of the intervention. Stakeholder involvement generally did not extend to other stages of the research process. This finding has been reiterated in other reviews within a health-related context,[58] including suicide prevention.[67] Inclusion of stakeholders within the research process before implementation of suicide prevention intervention may allow tailoring of the intervention to suit a specific service user's needs and preferences.

[67] Yet, exclusion beyond these formative stages removes the stakeholder from decision-making processes that may be pertinent to implementation aspects of the suicide prevention intervention (e.g., delivery and intervention evaluation and impact).[67] Co-produced related outcomes are often context-specific.[57] Therefore, involvement of stakeholders within the latter stages of the research process, including the evaluation of research findings, is warranted.[67] This could prevent tokenistic involvement of stakeholders by legitimizing the translation of their knowledge and expertise into research evidence that meets the intervention objectives, and the creation of evaluation approaches that measure meaningful impacts associated with co-produced suicide prevention interventions.[67]

Implications for future research

Future research should clearly define how co-production is implemented and formally evaluate corresponding outputs from co-production in the delivery of suicide prevention interventions. This is important for understanding the impact on potential outcomes, if any, associated with a co-production approach. While it is likely that there are wider impacts associated with co-produced community-based suicide prevention interventions, further research is needed to understand the theoretical components of co-produced community-based suicide prevention interventions. This would allow for the development of validated evaluation measures that can determine the intervention effects on suicide.

While some positives were reported for the inclusion of co-production in community-based suicide prevention interventions, particularly from the perspective of participants, there is some evidence that some professionals (e.g., clinicians) are reticent to relinquish their paternalistic roles. Future research should seek to understand the views/perspectives of those implementing co-produced services to understand any potential barriers and facilitators to intervention delivery.

CONCLUSION

The present review found that most studies fostering a co-productive approach within community-based suicide prevention interventions elicit the views and perspectives of stakeholders in a process of co-design/co-creation. Positive evaluation attributed towards this co-productive approach indicates some benefits in the creation of suicide prevention intervention that recognizes and values each stakeholder and redress potential power imbalances within the therapeutic relationship. This may improve engagement and give voice and control to those experiencing suicidal crisis. However, there is limited evaluation extending beyond the design aspects of the co-productive approach to understand its effects within community-based suicide prevention interventions.

AUTHOR CONTRIBUTIONS

Substantial contributions to the conceptualisation and design of the study: Claire Hanlon, David McIlroy, Jennifer Chopra, Helen Poole and Pooja Saini. All searches: Claire Hanlon. *Analysis and interpretation of data*: Claire Hanlon, David McIlroy, Jennifer Chopra, Helen Poole and Pooja Saini. *Revising of the work critically for intellectual content and approval of the version for publication*: Claire Hanlon, David McIlroy, Jennifer Chopra, Helen Poole and Pooja Saini. All authors have read and agreed to the published version of the manuscript.

ACKNOWLEDGEMENT

No grant or funding was provided for this study.

CONFLICT OF INTEREST

The authors declare there is no conflict of interest.

DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no data sets were generated or analysed during the current study.

AAPPENDIX

See Table A1.

Table A1 Example of search terms

	Search term	Search field
1.	co-product*	Title search
2.	collaborat*	Title search
3.	‘collaborative approach’	Title search
4.	co-design*	Title search
5.	co-creat*	Title search
6.	co-develop*	Title search
7.	co-evaluat*	Title search
8.	‘action research’	Title search
9.	‘lived experience’	Title search
10.	‘user experience’	Title search
11.	‘user involvement’	Title search
12.	‘patient involvement’	Title search
13.	‘patient participation’	Title search
14.	‘patient engagement’	Title search
15.	‘patient cent* care’	Title search
16.	‘person cent* care’	Title search
17.	‘shared decision making’	Title search
18.	MH suicide [MESH]	Title search
19.	suicid*	Title search
20.	Suicide [keyword]	Title search

21.	MH ‘community mental health services’ [MESH]	
22.	‘community mental health services’ [keyword]	Title search
22.	1 OR 2 OR 3 OR 4 OR 5 OR 6 OR 7 OR 8 OR 9 OR 10 OR 11 OR 12 OR 13 OR 14 OR 15 OR 16 OR 17	
23.	18 OR 19 OR 20 OR 21 OR 22	
24.	23 AND 24	

DETAILS

Subject:	Behavior; Intervention; Mental health; Databases; Public health; Collaboration; Prevention programs; Citizen participation; Health services; Mental health care; Systematic review; Suicide; Community mental health services; Prevention; Co-design; Interest groups; Public involvement; Stakeholders; User needs; Health risks; Decision making; Effectiveness; Adults; Health education; Suicides &suicide attempts
Identifier / keyword:	adults mental health; community-based; co-production; suicide; suicide prevention; systematic review
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	64-86
Publication year:	2023
Publication date:	Feb 2023
Section:	REVIEW ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625

Source type:	Scholarly Journal
Language of publication:	English
Document type:	Evidence Based Healthcare, Journal Article
Publication history :	
Online publication date:	2022-11-14
Milestone dates:	2022-10-06 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-08-03 (manuscriptReceived); 2022-11-14 (publishedOnlineEarlyUnpaginated); 2022-10-25 (manuscriptAccepted)
Publication history :	
First posting date:	14 Nov 2022
DOI:	https://doi.org/10.1111/hex.13661
ProQuest document ID:	2767101992
Document URL:	https://www.proquest.com/scholarly-journals/evaluating-role-effectiveness-co-produced/docview/2767101992/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the “License”). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-05-04
Database:	Public Health Database,Publicly Available Content Database

Co-designing an intervention to improve the process of deprescribing for older people living with frailty in the United Kingdom

Silcock, Jonathan [1]
; Marques, Iuri [2] ; Olaniyan, Janice [1] ; Raynor, David K [3] ; Baxter, Helen [4] ; Gray, Nicky [5] ; Zaidi, Syed T R [6] ; Peat, George [7] ; Fylan, Beth [1]
; Breen, Liz [1]
; Benn, Jonathan [8] ; Alldred, David P [9] 1 School of Pharmacy and Medical Sciences, Faculty of Life Sciences, University of Bradford, Bradford, UK; NIHR Yorkshire and the Humber Patient Safety Translational Research Centre, Bradford Institute for Health Research, Bradford, UK 2 School of Pharmacy and Medical Sciences, Faculty of Life Sciences, University of Bradford, Bradford, UK 3 School of Healthcare, University of Leeds, Leeds, UK 4 Alliance Manchester Business School, Faculty of

Humanities, University of Manchester, Manchester, UK [5] Department of Pharmacy, School of Applied Sciences, University of Huddersfield, Huddersfield, UK [6] HPS Pharmacies, EBOS Group, Docklands, Victoria, Australia [7] Department of Health Sciences, University of York, York, UK [8] NIHR Yorkshire and the Humber Patient Safety Translational Research Centre, Bradford Institute for Health Research, Bradford, UK; School of Psychology, University of Leeds, Leeds, UK [9] NIHR Yorkshire and the Humber Patient Safety Translational Research Centre, Bradford Institute for Health Research, Bradford, UK; School of Healthcare, University of Leeds, Leeds, UK

ABSTRACT (ENGLISH)

Background

In older people living with frailty, polypharmacy can lead to preventable harm like adverse drug reactions and hospitalization. Deprescribing is a strategy to reduce problematic polypharmacy. All stakeholders should be actively involved in developing a person-centred deprescribing process that involves shared decision-making.

Objective

To co-design an intervention, supported by a logic model, to increase the engagement of older people living with frailty in the process of deprescribing.

Design

Experience-based co-design is an approach to service improvement, which uses service users and providers to identify problems and design solutions. This was used to create a person-centred intervention with the potential to improve the quality and outcomes of the deprescribing process. A ‘trigger film’ showing older people talking about their healthcare experiences was created and facilitated discussions about current problems in the deprescribing process. Problems were then prioritized and appropriate solutions were developed. The review located the solutions in the context of current processes and procedures. An ideal care pathway and a complex intervention to deliver better care were developed.

Setting and Participants

Older people living with frailty, their informal carers and professionals living and/or working in West Yorkshire, England, UK. Deprescribing was considered in the context of primary care.

Results

The current deprescribing process differed from an ideal pathway. A complex intervention containing seven elements was required to move towards the ideal pathway. Three of these elements were prototyped and four still need development. The complex intervention responded to priorities about (a) clarity for older people about what was happening at all stages in the deprescribing process and (b) the quality of one-to-one consultations.

Conclusions

Priorities for improving the current deprescribing process were successfully identified. Solutions were developed and structured as a complex intervention. Further work is underway to (a) complete the prototyping of the intervention and (b) conduct feasibility testing.

Patient or Public Contribution

Older people living with frailty (and their informal carers) have made a central contribution, as collaborators, to ensure that a complex intervention has the greatest possible potential to enhance the experience of deprescribing medicines.

FULL TEXT

INTRODUCTION

Older people living with frailty are vulnerable to harm because of age-related breakdown in physiological systems and the failure of homoeostasis.[1] Specifically, nonfrail older people have greater tolerance to adverse drug effects

(ADEs),[2] and frailty is a better predictor of medicines-related harm than chronological age.[3] Therefore, targeting frail older people has become an important focus internationally for medicines optimization and deprescribing interventions to reduce polypharmacy.[2]

In this context, polypharmacy is the concurrent use of multiple medicines, usually defined as the use of five or more medicines daily.[4,5] The King's Fund refers to 'appropriate' and 'problematic' polypharmacy to differentiate between safe and potentially harmful combinations of medicines.[6] Polypharmacy is problematic when the potential risks of use outweigh the expected benefits. In older people living with frailty, this can lead to higher healthcare costs and preventable harm such as adverse drug reactions,[7–11] hospitalizations,[12–14] falls,[15–17] lower levels of adherence[6,18] and mortality.[19]

The World Health Organization (WHO) has a Medication Without Harm initiative, with targets to reduce harm from problematic polypharmacy.[20] In the United Kingdom, the new general practice (family doctor) contract is tackling polypharmacy by ensuring that periodic Structured Medication Reviews (SMRs) are conducted in line with the National Health Service (NHS) Long-term Plan and there is a clear process for deprescribing.[21]

Deprescribing is defined as: 'the systematic process of identifying and discontinuing medicines in instances in which existing potential harms outweigh existing or potential benefits within the context of an individual patient's care goals, the current level of functioning, life expectancy, values, and preferences'.[22] It is increasingly recognized as a strategy to reduce problematic polypharmacy.[23,24] There are, however, professional and service user reported barriers to deprescribing.[10,25] Stopping prescribed medicines is often complex so it requires shared decision-making and mutual understanding.

Guidelines for shared decision-making recommend actions before, during and after the clinical consultation to ensure full service-user engagement in their care. These actions may be enhanced by: the involvement of a supporting person (e.g., family member) and links to reliable health information.[26] A person-centred approach to deprescribing ensures successful therapeutic change.[5] There are reasons to believe that greater sensitivity to lived experience can enhance service delivery for people living with frailty.[27] There are also moral reasons to engage vulnerable people in the design of public services,[28] which include an assumption of capacity to make informed decisions and a desire for the experience of service delivery to be positive. In a health context, people need information about the potential risks and benefits of treatment options at a level of detail that helps them to make an informed choice with professional guidance.[29]

One person-centred approach to improving healthcare services is experience-based co-design (EBCD).[30–38] EBCD is a narrative-based participatory method, which brings together professionals and service users to collaboratively co-design local services. This can be used instead of (or alongside) more traditional approaches to service improvement (e.g., Plan Do Study Act cycles) with a particular focus on the user experience of service delivery.[39] Traditionally, EBCD is conducted in one organization that initiates and implements the process, and then solutions are implemented locally. There is variation in the use of EBCD,[40] and co-design alone does not necessarily solve all healthcare delivery problems.[41] We have previously designed an intervention using researcher-driven EBCD and implemented this in a clinical trial to improve medicines management during discharge for heart failure patients.[30,42] The work described in this paper had similar intentions in a different context.

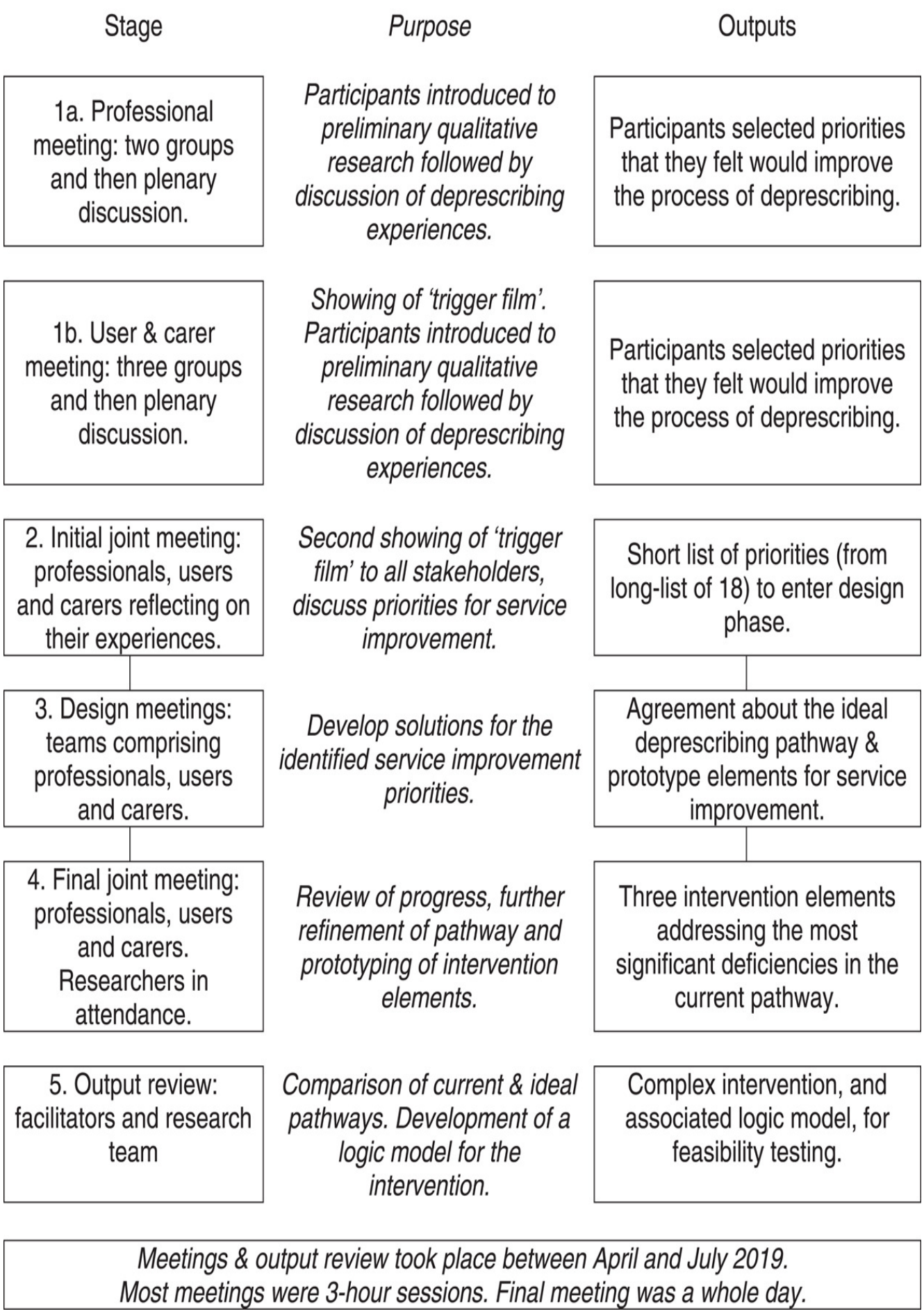
In preparation, we had already explored processes for, and the lived experience of, deprescribing in interviews with older people, their informal carers and professionals. Drawing on this work, we identified six themes related to barriers and facilitators of deprescribing.[43] This is analogous to the first phase of EBCD. In response to these themes, in this current phase of work, we completed a modified EBCD process to develop an intervention to address problematic polypharmacy in primary care. This approach builds on the strengths of traditional EBCD to develop a new intervention based on an agreed model of service delivery.

The value of careful engagement with vulnerable people to improve services has been recognized,[44] but there have been no published studies drawing on the experiences of users and professionals to collaboratively design and evaluate a process for deprescribing in primary care settings. This study aimed to co-design an intervention to improve deprescribing processes for older people living with frailty who were receiving primary care in the English

NHS.

METHODS

Usually, EBCD includes thematic analysis of interviews with stakeholders; preparation of a trigger film to use in stakeholders' meetings; collaborative problem identification and priority setting and the co-design of solutions (Figure 1). Elsewhere, we have described our qualitative research that forms the first phase of EBCD in this case, that is, gathering information from service users and professionals about deprescribing experiences.[43]



Enlarge this image.

In this phase, we completed the EBCD process and also integrated it with Medical Research Council (MRC) guidelines for the development of complex research interventions.[45] This modification of EBCD is in line with our

previously described process for the design of complex interventions.[30,42] The link between the phases of work is a trigger film summarizing the emotional touchpoints in the process of deprescribing from an older person's perspective. This film is used to focus discussions in the stakeholder meetings described below.
The objectives of this phase of work were to:

- (1) hold a series of stakeholder meetings for service users and professionals to further discuss problems with deprescribing and identify priorities for service improvement;
- (2) run design meetings to work from the agreed priorities and towards solutions (changes in process) that will enhance the experience of deprescribing with the intention to improve safety and effectiveness;
- (3) build these solutions into a complex intervention that can be tested in further phases of work;
- (4) produce a logic model that will support the future implementation and evaluation of the complex intervention.

Planning for stakeholder meetings

The inclusion criteria for stakeholders were:

- (1) people 65 years and older identified as living with frailty or at risk of frailty (a recorded diagnosis and/or electronic Frailty Index [eFI] >0.12) who were experienced in the daily management of multiple medicines for co-morbidities [43,46];
- (2) family members, or others, supporting these people on a daily basis in an unpaid or voluntary capacity;
- (3) primary care healthcare professionals—pharmacists, general practitioners and nurses, with experience in deprescribing for older people.

In the United Kingdom, primary care pharmacists now work as part of a multidisciplinary team in medical general practice. We did not engage (in this study) with community (retail) pharmacists. To supplement the core research team, an experienced EBCD trainer (H. B.) and a group-work facilitator (N. G.) were commissioned. An EBCD training event was held for the whole team with the trainer (H. B.) who was accredited by the Point of Care Foundation. Users, carers and professionals were approached using a number of channels: Yorkshire and Humber Academic Health Science Network (AHSN); West Yorkshire NHS Research &Development office; Yorkshire and Humber Patient Safety Translational Research Centre; citizens' groups in Bradford, for example, Age UK; Community Pharmacy West Yorkshire (CPWY); the service user group in the University of Bradford's Faculty of Health Studies, and participants from the qualitative interview stage, for example, professionals from two local medical general practices.
Letters inviting people to separate initial meetings were circulated to users and informal carers; and professionals. Before the meetings, any questions were answered by telephone. At the initial meetings, the whole design process was described in detail and agreements to continue with participation were confirmed. Participants were free to

attend as many sessions as they were able. The initial professional meeting was held on 29th April 2019 and the final prototyping with stakeholders was held on 3rd June 2019. All other meetings were held in this window.

Priority setting meetings

At the initial professional meeting, two working groups shared information about their workload and clinical case studies. At the initial user and carer meeting, discussions were facilitated in three working groups. The trigger film showing older people talking about their experiences was viewed then experiences of deprescribing were shared and discussed. Each of these initial (segregated) stakeholder meetings produced a long list of priorities for service improvement.

At a joint meeting for all stakeholders, the trigger film was reshown and the long lists of priorities previously agreed upon were shared. People were set the task of jointly agreeing, by facilitated discussion and debate, a short list of priorities.

Intervention design meetings

From this point onwards the users, informal carers and professionals met as a design team. Volunteers to join the design team were sought and confirmed by the group at the end of the joint meeting. At the first design team meeting, facilitators acted as scribes for two working groups. Thoughts and ideas were captured on sticky notes so that they could be sorted and arranged. At the second design team meeting, notes from the first meeting were presented for validation and any disagreements were resolved by discussion. Existing elements of good practice were noted and the participants worked towards interventions that would improve care.

In the original plan, a final meeting of users, carers and professionals was intended as a celebration and a space to present intervention prototypes. However, progress made to that point was presented to the whole group (those who had also attended the second design meeting) and then prototyping of interventions continued in three subgroups with facilitators. Various creative resources were made available including coloured pens, paper/card, plastic building blocks, blank speech bubbles, sticky notes, flip charts and wall space. Printed summaries of prior outputs were available for reference.

Since this work was a development activity (leading to future research) the people involved were considered to be collaborators or co-investigators and therefore, ethical approval was not sought. Personal information about people was not captured. Discussions were not recorded or subjected to thematic or content analysis. Prior ethical approval had covered activities up to and including the creation and viewing of the trigger film.

RESULTS

The number and type of people contributing are summarized in Table 1. Table 1 aligns with Figure 1, which shows how the outputs of each EBCD stage fed into the next stage. Note that EBCD is iterative and incorporates ongoing participant validation. The ending of one stage is the beginning of the next. In this way, a consensus is built, and peoples' ideas are taken forward with fidelity. However, the initial professional and user meetings are not linked.

Table 1 Number and type of participants at each stage

Stage of process	Participants
Professional meeting	1 medical general practitioner, 1 hospital pharmacist, 7 primary care pharmacists. 2 facilitators.
User and carer meeting	5 informal carers and 9 older people. 3 facilitators.

Initial joint meeting	3 primary care pharmacists, 3 informal carers and 5 older people. 3 facilitators.
Design meetings	3 primary care pharmacists and 9 older people or informal carers. 2 facilitators.
Final joint meeting	3 facilitators. Participants from the Design Meetings.
Output review	Meeting facilitators and members of the academic team.

Priority setting meetings

In the initial professional meeting, the importance of good relationships with older people was agreed and some high-risk medicines were identified. Professionals also noted risks associated with different parts of the care pathway and how significant safety incidents were managed. The risks associated with making and monitoring deprescribing decisions were noted, as was the need for appropriate record keeping.

It was clear that the professionals had a shared experience of problems when deprescribing and were keen to help colleagues in similar situations. Professionals noted that older people could be resistant to deprescribing if they trusted the initial prescriber and that therapeutic alternatives were sometimes lacking. Exacerbations or new diagnoses could provide opportunities for a medication review. Professionals agreed on a long list of 12 priorities which they felt would improve the process of deprescribing:

•1.

Team-based approach and clarity of roles and responsibilities.

•2.

Information available to healthcare professionals.

•3.

Information available to older people and informal carers.

•4.

Service user engagement and empowerment.

•5.

Implementation of triggers in the system to identify opportunities to optimize medicines, of which deprescribing is one component, for example, admission to hospital or discharge.

•6.

Communication at transitions of care (with other professionals and with older people): handovers.

•7.

Follow-up after medicines are stopped.

•8.

Standardization of guidelines for deprescribing.

•9.

Clear plan for each medicine prescribed when they are prescribed: agreeing on goals.

•10.
Skills of healthcare professionals: opportunities to reflect and learn about the process, what went well and what could be improved.

•11.
Skills of older people: opportunities to reflect on the process, what went well and what could be improved.

•12.
Time required to stop medicines.

In the initial user and carer meeting, difficulties in working relationships between hospital consultants, GPs and community pharmacists were noted. Older people were sometimes unsure about who to ask questions to or where to seek clarification about plans for care. Older people were aware that medicines had both risks and benefits. Older people and carers agreed to a long list of six priorities:

•1.
Two-way discussions incorporating personal views and priorities.

•2.
Following-up and monitoring of changes should be organized.

•3.
User-professional relationships and familiarity with professionals should be improved: ‘no decision about us without us’.

•4.
Advance information should be provided about medicines and proposed changes.

•5.
Alternatives to medicines should be considered.

•6.
User access to peer support should be noted and carer views on change considered.

At the joint meeting, the trigger film had a powerful effect on professionals who were viewing it for the first time and in the presence of older people. The highlighted themes, spoken about by older people in the film, included clarity of technical information, transparency of processes and the need for trust in consultations. Older people and carers at the meeting expressed the need for some flexibility around decision-making to account for uncertainties, domestic circumstances and the variability of health status. Through discussion at the joint meeting, the overall long list of 18 priorities was reduced to a short list of two priorities, which the design groups then addressed. These were:

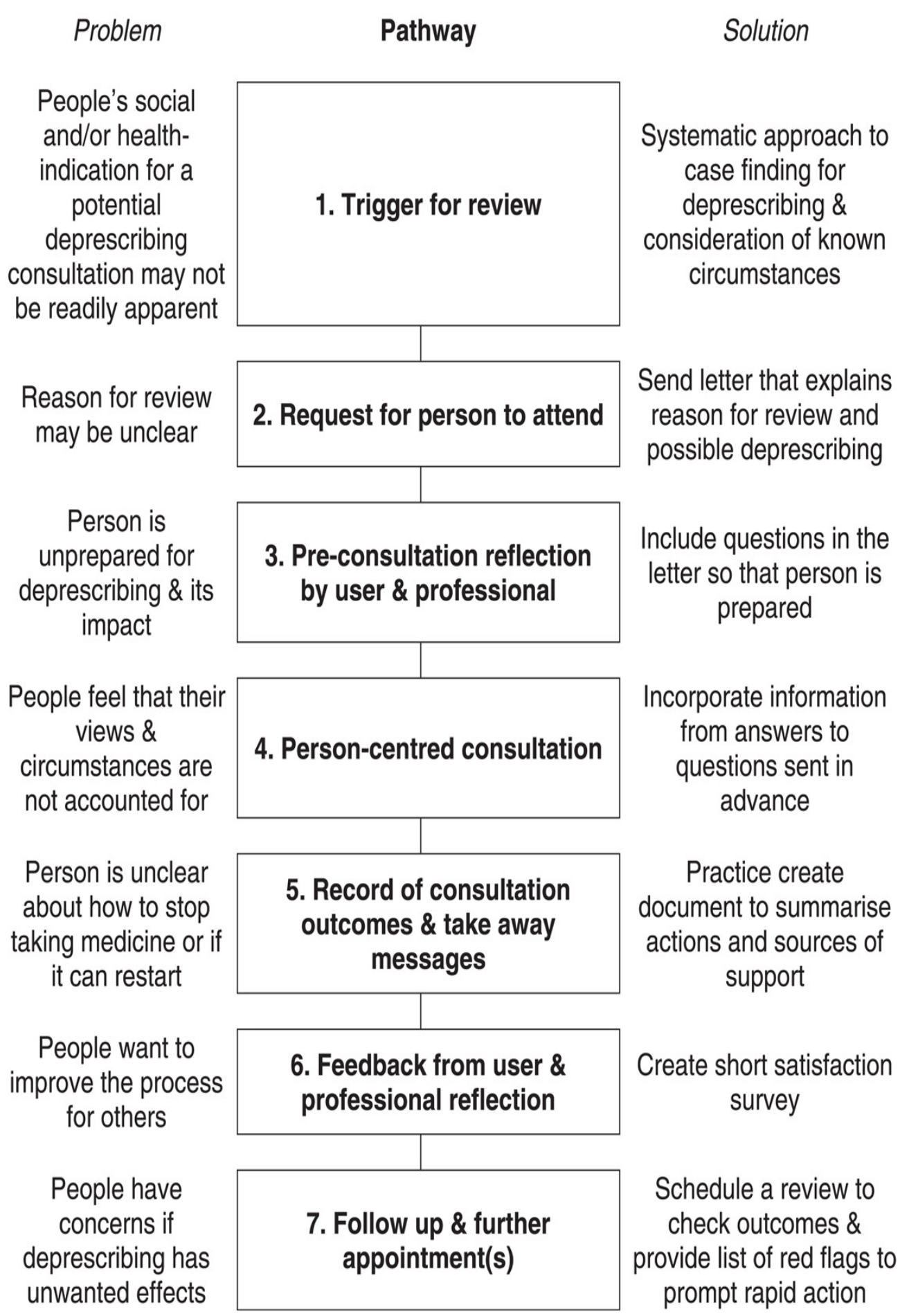
•Two-way conversations/discussions: attitudes, prior knowledge, preparation, skills, expectations—described as ‘general culture changes needed to form the context to a deprescribing decision’.

•The process of stopping medicines: the steps to take before, during and after consultation including follow-up.

Intervention design meetings

In the design meetings, the initial discussion was free ranging around preparation for deprescribing, the actual consultation and follow-up. Two planned working groups were quickly merged and their outputs linked because they

were converging. An ideal deprescribing process was outlined (simplified in Figure 2). The resources available for prototyping interventions were limited, so the co-design groups tried to made progress where they thought the capacity for benefit was greatest.



Enlarge this image.

More time for prototyping was arranged by repurposing the proposed celebration meeting to some extent. People

developed prototypes for three interventions that were suggested enhancements to the deprescribing process:

•(1)

An invitation letter to a deprescribing consultation providing information about the purpose of the consultation and encouraging older people to prepare questions for professionals.

•(2)

A ‘take-away’ for the end of the consultation listing: the agreed next steps, monitoring to ensure the safety of deprescribing, and how the effectiveness of any decisions made would be reviewed.

•(3)

A satisfaction survey for older people to complete and provide feedback for professionals about the consultation process.

Review of intervention design outputs

In subsequent researcher meetings, differences between the current deprescribing pathway and the ideal pathway (Figure 2) were further explored and discussed. Seven major differences were noted between the current deprescribing pathway and the ideal pathway that had been generated. Since prototyped interventions addressing three of these differences had already been developed, this left a further four interventions that still required initial prototyping:

•(1)

Professional training: focussed on consultation skills and shared decision-making.

•(2)

Ensuring the consultation has a clear agenda—the older person's initial feelings and any concerns about action points should both be addressed.

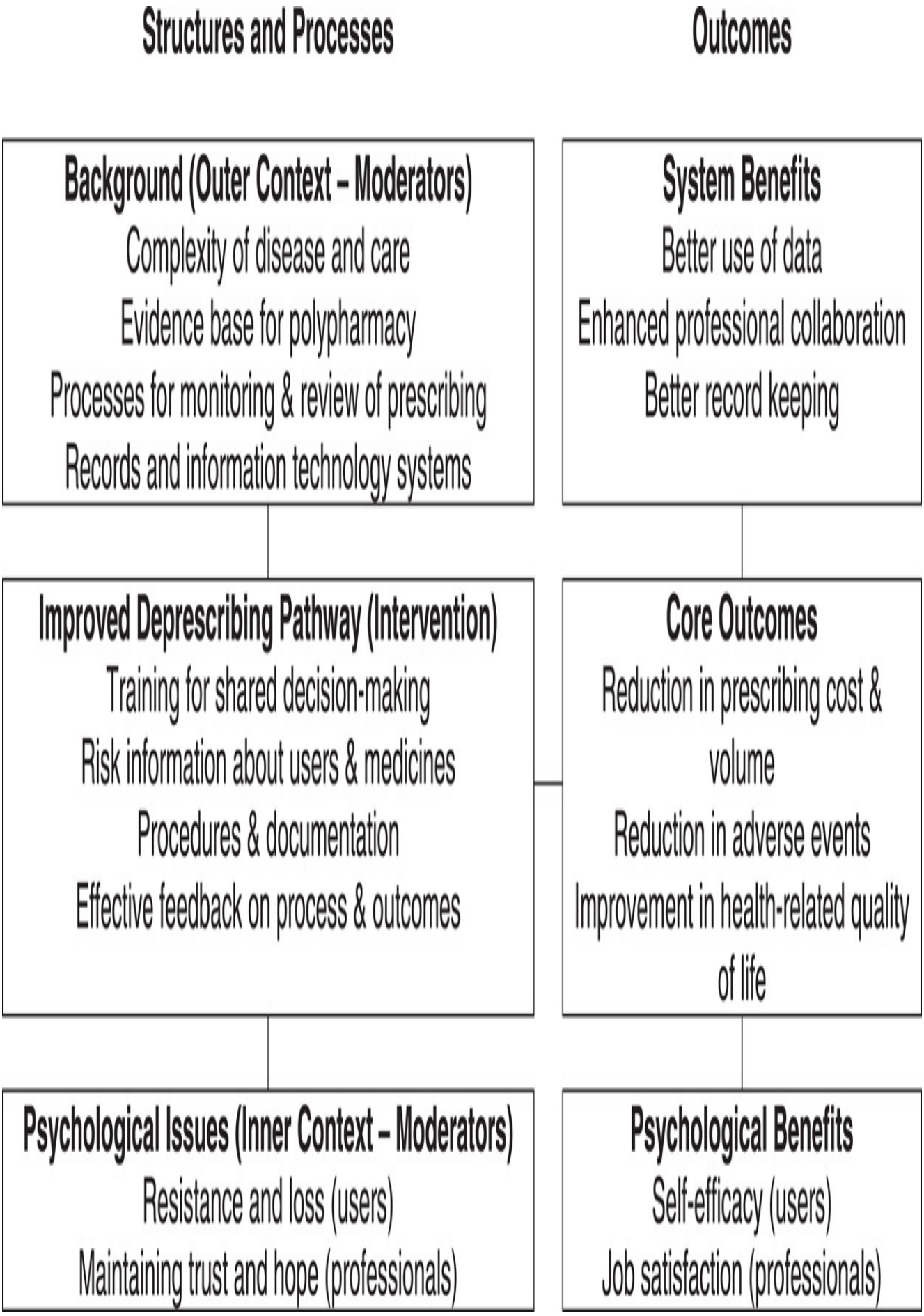
•(3)

Signposting information for further postconsultation support and updating any user-held records.

•(4)

Giving the older person a list of triggers (red flags) that would require rapid follow-up and monitoring repeat prescription requests or missed appointments.

We refined the existing prototypes and generated an overall logic model[47] for a complex intervention (combining the seven simple interventions) structured around the ideal pathway (Figure 3). Usually, EBCD outputs would be implemented locally and refined by iterative cycles of service improvement. We recognized that elements of the ideal pathway were present in current NHS practice even if not fully expressed. Pathway improvements could also be implemented in different ways, for example, written materials may be physical or electronic. Therefore, we proposed that prototyping continued (after this co-design process) with partners in primary care to develop tools and resources that could be implemented flexibly to meet local needs and create the ideal pathway.



Enlarge this image.

DISCUSSION

The final priorities (most important problems) identified by our collaborators were the clarity of all stages in the deprescribing processes and the quality of one-to-one consultations. Three simple interventions were prototyped to address these priorities and four more simple interventions were identified. Working from these simple interventions, a complex healthcare intervention (Figure 2) and a supporting logic model (Figure 3) were created. Further work is ongoing to conduct feasibility testing with primary care partners before full evaluation.

Our work has been conducted in close partnership with older people living with (or at risk of) frailty and their informal carers. In the NHS, the eFI is used as a risk stratification tool to identify if people are likely to be fit or living with mild, moderate or severe frailty.[47] This risk stratification allows body system reviews and medication reviews to be targeted for people who are at the highest risk of adverse events and, therefore, most likely to benefit from deprescribing.

If primary care professionals are working efficiently to improve the safety of medicines used for those identified as living with (or at risk of) frailty, then the incidence of deprescribing events should appropriately increase. The intention of deprescribing is to reduce problematic polypharmacy, however, this co-design work has shown that the process of deprescribing is itself problematic: requiring further optimization and evaluation. Any changes to medicines that have been prescribed for a long period can carry risks as well as benefits, such as adverse drug withdrawal events.[48]

Older people and carers understand the need for deprescribing in general, but the six long-listed user priorities (from the initial user and carer meeting) in this study focused on the themes of information and relationships. Older people and carers want to be told the rationale for medicine changes and have the opportunity to express how these changes will influence their daily lives. People living with frailty also want reliable access to peer and professional support. These user priorities (from co-design) reflect the facilitators of deprescribing identified in preparatory work.[43]

The professional priorities identified often supported, and do not fundamentally conflict with, the user priorities. However, professionals also identified the importance of: skill mix; clear roles and responsibilities; triggers to action; planning and guidelines. Professionals seek constructive engagement with users and carers. However, the working environment is already complex and there are competing demands on professional time. The proposed ideal pathway (Figure 2) seeks to structure a process around some elements that already exist in practice but may not be consistently delivered. These professional priorities (identified in stakeholder meetings) reflect more of the barriers to deprescribing identified in preparatory work.[43]

Structured and routine engagement with users, which privileges the lived experience, brings them into the healthcare system as self-managers and monitors. Those living with frailty are (by definition) vulnerable, however, they are not helpless or hopeless. Here they have made an important contribution, as collaborators and co-designers, to ensure that a complex intervention has the greatest possible potential to enhance the experience of healthcare delivery.

In the ideal pathway, a flow of information is created, integrated and managed. There are stages of information gathering, clinical decision-making and information giving; akin to consultation skills guides such as the Calgary-Cambridge model[49] and the derived Medicines Related Consultation Framework (MRCF).[50] However, the flow of information includes checks, balances and feedback loops, meaning that the deprescribing process could be paused or reviewed. One key characteristic of this pathway is that at each stage, older people are actively engaged, shared decisions are made and intended outcomes are clarified. The pathway also recognizes that, for users, medicines taking and deprescribing take place in a psychological and social context. A process that potentially addresses concerns and integrates social support is more likely to be effective and enhance user satisfaction. Co-design has allowed us to build on the current (implicit) deprescribing process that preparatory work had previously mapped out.

The ideal pathway now shown (Figure 2) is transparent, fully defined and designed to enhance collaboration at each step. In England, SMRs will be undertaken by pharmacists working in Primary Care Networks (PCNs) and our intervention will be feasibility tested within this context.[51] A study has shown that deprescribing for older people with type 2 diabetes is feasible, safe and may improve quality of life.[52] The risks and benefits of deprescribing will vary condition by condition and we have only developed a generic pathway. When people lack the capacity to engage, then their carers' views may also be considered.[53]

A more diverse mix of healthcare professionals could have strengthened our study; however, as primary care pharmacists will take the lead role in SMRs the relatively high number of pharmacists who were part of the EBCD process has strengthened our process model for adoption. It was recognized that a relatively small group of participants may miss opportunities for innovation and creativity, especially in a short timescale. However, the overarching aim of EBCD is not necessarily one of generalizability. Rather, our findings will support the feasibility testing of flexible tools and processes to enhance existing consultation processes in primary care.

CONCLUSION

Previous work demonstrates that deprescribing of potentially inappropriate medicines in older people living with frailty has the potential to prevent ADEs and improve peoples' quality of life worldwide. However, we show that deprescribing itself must be carefully managed to optimize effectiveness and minimize risks. Our pathway outlines a person-centred, clinician-facilitated approach to deprescribing consultations in primary care, which is also supported by a recent systematic review.[54] Our study further demonstrates that EBCD can work across multiple general practices as part of a programme of research to develop a person-centred deprescribing process. This has the potential to improve service efficiency and user outcomes. In keeping with the unique characteristics of EBCD, users, informal carers and professionals were best placed to identify areas for improvement in the current pathway for medication reviews and deprescribing. Collaborative intervention design ensures that changes address the needs and concerns of all stakeholders.

AUTHOR CONTRIBUTIONS

Jonathan Silcock and David K. Raynor led and directed the project. Iuri Marques and Janice Olaniyan managed and delivered the project. Helen Baxter and Nicky Gray were EBCD facilitators and supported project delivery. Syed T. R. Zaidi supported project delivery, was an EBCD facilitator and also participated in intervention review. George Peat, Beth Fylan, Liz Breen and Jonathan Benn led and managed supporting projects, contributed to the review and refinement of the intervention and edited the manuscript. David P. Alldred leads the programme of work that this project is part of, contributed to the review and refinement of the intervention, and edited the manuscript.

ACKNOWLEDGEMENTS

We thank all the older people, carers and professionals that supported this co-design work. Daisy Payne provided research and administrative support. Kate Karban was a group facilitator. Naveed Riaz helped to produce the trigger film. Andrew Clegg and David Mehdizadeh provided valuable comments on the manuscript. This project was funded by the National Institute for Health and Care Research (NIHR)Yorkshire and HumberPatient Safety Translational Research Centre (NIHR Yorkshire and Humber PSTRC). The views expressed in this article are those of the author(s) and not necessarily those the NIHR, or the Department of Health and Social Care. Iuri Marques and Daisy Payne are now researchers working in the UK Government's Civil Service, the study and views expressed are unrelated to their current departments and roles. No statement of official policy is implied or intended.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

DETAILS

Subject:	Side effects; Intervention; Priorities; Context; Avoidable; Drug stores; Caregivers; Older people; Co-design; Primary care; Research &development--R &D; Working groups; Decision making; Pharmacists; Prescription drugs; Talking; Patient safety; Health care; Feasibility; Frailty; Polypharmacy; Prototyping; Meetings; Critical incidents; Complex; Group decision making; Professionals; Stakeholders; Qualitative research; Hospitalization
Business indexing term:	Subject: Drug stores Professionals Stakeholders
Location:	United Kingdom--UK
Identifier / keyword:	aged; deprescribing; frailty; polypharmacy; primary health care; referral and consultation
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	399-408
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English

Pathways to recovery model of youth substance misuse in Assam, India

Madill, Anna [1] ; Duara, Raginie [1] ; Goswami, Sangeeta [2] ; Graber, Rebecca [3] ; Hugh-Jones, Siobhan [1] 1 School of Psychology, University of Leeds, Leeds, UK 2 Institute of Positive Mental Health &Research, Guwahati, Assam, India 3 School of Humanities and Social Science, University of Brighton, Brighton, East Sussex, UK

ABSTRACT (ENGLISH)

Introduction

There are global calls for better understanding of substance use disorder (SUD) to inform prevention, risk reduction and treatment of this relapse-prone disorder. Our aim in this article is to understand the pathways to recovery of youth in Assam, India who have suffered SUD.

Methods

We recruited 15 participants (11 men and 4 women) via two rehabilitation facilities. All are addicts-in-recovery aged 19–24 years. Material was generated through photo-led interviews, analysed using an inductive variant of thematic analysis and the resulting model refined through expert and participant checks.

Results

We present a multiroute, multidirectional pathway to recovery model. It has three phases, *Recreational Use*, *Addiction* (*Relaxed*, *Chaotic*, *Strategic*) and *Supported Recovery*, each phase consisting of cycling between, or transitioning through, a series of stages.

Conclusions

The model enhances psycho-socio-cultural insights into the experience of risk and recovery, and informs prevention and treatment for youth substance misuse in Assam. This is the first model of its kind and an important public health resource. We discuss the possible transferability of the model to a wider range of contexts.

Patient or Public Contribution

The model presented was generated through analysis of interviews with addicts-in-recovery. Four of these addicts-in-recovery, and two mental health and rehabilitation service providers, conducted participant and expert checks of the model leading to its improvement.

FULL TEXT

INTRODUCTION

There are global calls for better understanding of substance use disorder (SUD) to inform prevention, risk reduction and treatment of this relapse-prone disorder.[1] Global mental health strategies, in particular, prioritize adolescents given the high prevalence of SUDs in this population, trajectory towards lifelong disadvantage and suicide risk.[2] Similarly, the India Mental Health Survey 2015–2016[3] calls for the strengthening of youth mental health research and addiction management. Subsequently, the first government-commissioned national survey was conducted in 2019[4] on the extent and pattern of substance use in India. Recommendations include legal and policy innovations, scaled-up treatment programmes and prevention interventions targeting young people.

At 14.6% of the Indian population, alcohol is the most commonly used psychoactive substance, predominating in men at 17:1, with about 19% of users deemed dependent. Cannabis (2.8%) and opioids (2.1%) are the next commonly used substances. About 0.25% of cannabis users showed dependency and 0.70% of all Indians need help with opioid use. About 0.20% of all Indians abuse sedatives, while inhalant abuse is higher in children and adolescents (1.17%) than adults (0.58%). A relatively minor problem is reported with regard to cocaine, amphetamine and hallucinogens. Earlier studies provide additional information on gender and youth. Gururaj et al.[3] report that SUD is prevalent in 22.4% of the Indian adult population and is 2.5 times more common in men than women. Thirteen percent of those abusing substances are children and adolescents,[5] with only 5% of those under 20 years old seeking treatment: the lowest by far of all age ranges.[6] Finally, excluding tobacco, the most common substances abused by young people are alcohol, cannabis and opioids, with initiation typically between 13 and 15 years.[7]

In the late 1980s, the National Institute of Social Defense (NISD) identified nongovernmental organizations (NGOs) with expertise in substance abuse prevention and treatment and conferred them status as Regional Resource Training Centres (RRTCs) with a remit to provide training and monitor treatment centres.[8] Twelve RRTCs exist and over 450 affiliated addiction treatment centres. De-Addiction Centres or Drug Treatment Centres are also available in many psychiatry departments of government medical colleges and district and general hospitals where free medications may be dispensed.[9] There are also numerous private treatment facilities. Hence, India has a comprehensive SUD treatment programme which includes ‘detoxification, pharmacotherapy, individual therapy, family therapy, group therapy, multifamily therapy, 12-Step programs’.[10] Programmes also integrate Indian cultural practices such as yoga, spirituality and an emphasis on social interdependence including support groups for families. However, more research is required into adapting the 12 steps for the Indian context,[11] and rehabilitation facilities are usually single-sex, with women underserved and disproportionately stigmatized.[12]

Defining recovery from SUD is contentious and increasingly described as a process rather than an event.[13] Although

many heterogeneous models of addiction exist,[14] there is little research on the journey from drug initiation through to recovery that is not biologically based or of known relevance to young people, women and to Low-and-Middle-Income-Countries (LMIC). In the present article, we describe the typical routes through SUD to recovery as narrated by young Indian people who have walked this path. Listening to young people and developing services *for* them *with* them is deemed best practice in global health.[15] However, this is a relatively novel approach to informing mental health services and policy in India where the value of evidence from service users and young people can be underestimated.[16]

Assam is a state in northeast India. It is geo-politically isolated, has been propelled into the 21st century from an agrarian social base and the needs of children, youth and women requires urgent attention.[17] A high stake is placed on scholastic achievement and material affluence, while structural inequalities and lack of opportunity to develop life skills contribute to college dropout, suicide and SUD. In fact, the Assam State Report of the National Mental Health Survey of India identifies adolescent SUD as an urgent public health problem.[18]

It is difficult to find statistics on adolescent SUD in Assam, particularly by gender. Hazarika et al.[19] report that of 10–19 years old (N = 63) living in a border area, 4.8% used alcohol and 3.2% (only males), used drugs such as heroin and solvents. Islam et al.[20] report that 80% of street children in Guwahati aged 5–18 years (N = 215) abused substances, 87.4% of whom abused solvents. Katoki et al.[5] report substance abuse amongst adolescents from the urban slums of Guwahati (N = 60) to predominate in males at a ratio of 4:1, with the highest rates of abuse between 16 and 19 years. A worrying level of solvent abuse and young initiation age typically of 8–13 years in Assam is also noted by Priyanka and Ankita.[21] Recommendations of the Assam State Report of the National Mental Health Survey include working closely with rehabilitation services, decreasing stigma and encouraging help-seeking through better public awareness.

Our study focuses on 19–24 years old who have suffered SUD but have not used addictive substances for at least 1 year. A sustained sobriety definition of recovery was chosen because it is the target of the rehabilitation services in Assam with whom we were working. Moreover, as a signatory of the United Nation's International Conventions (Article 47), India is obligated to 'act to *eliminate* the use of illicit drugs, to develop measures to prevent drug use and to ensure availability of treatment for people with drug use disorders'.([3],p.689, italics added) Hence, the aim of this study is to understand the pathways to recovery of youth in Assam who have suffered SUD. In so doing, we seek to enhance psycho-socio-cultural insights into the experience of risk and recovery, and inform prevention and treatment for youth SUD in the region.

METHOD

Approval was obtained from the Ethics Committee of the LGB Regional Institute of Mental Health, Assam and from the Ethics Committee of the School of Psychology, University of Leeds, UK.

Recruitment and participants

Recruitment was undertaken by two of our Partner Organizations (POs) in Assam: Nirmaan Rehabilitation Facility and Hope Foundation Rehabilitation Centre. These organizations have direct contact with the demographic of interest and are widely networked throughout the rehabilitation, voluntary and educational sectors. Both are private rehabilitation facilities, charge a fee for care, and, as is typical, serve only male clients. Nirmaan Rehabilitation Facility has 16 staff, all trained in counselling and therapy, and a visiting psychiatrist. It follows the 12-step programme along with spiritual principles and offers 90-day residential treatment. Hope Foundation Rehabilitation Centre is a satellite of a global charity and also follows the 12-step programme. It has 14 trained staff and a part-time psychiatrist. Their work includes detoxification, rehabilitation and an extended care unit for those who have completed their 90–120-day course.

Candidates for the study had to be Indian nationals, aged 19–24 years, in successful recovery from drug and/or alcohol abuse (i.e., 1 year substance-free, irrespective of tobacco use). Most participants had daily contact with our POs, some living within the premises while giving service, with records kept of client progress. Hence, clean time was assessed through face-to-face contact with counsellors and through the extended service user network dedicated to providing close support. Other participants, including all the women, were identified by our POs through

their service user network, including Alcoholic Anonymous and Narcotics Anonymous meetings. One of these women recovered with peer as opposed to professional support. These networks are based on 'good faith.' However, recovery networks overlap substantially with personal life and people 'know each other's business' to a much greater extent that in many western contexts and our POs did not recommend for the study three women and five men they were monitoring because they deemed them to have relapsed before the requisite 12 months.

The Betty Ford Institute Consensus Panel[22] identifies three timeframes of recovery associated with increasing resilience to relapse: early recovery (1–12 months), sustained recovery (1–5 years) and stable recovery (5+ years). Hence, in this categorization, our participants have entered sustained recovery. We did not include tobacco-only users given its relative social acceptance and because the Assam State Report is of the opinion that '(t)obacco use per se, is not an issue for mental disorders'.([17], p.18).

In line with Dworkin's[23] recommended sample size for this method of research, we set a minimum target of 12 participants and, given the preponderance of men with SUD, aimed for a sample of approximately three-quarters male. Hence, we commenced with a purposive sampling strategy with regard to gender. As recruitment continued, we monitored for diversity across our age range of interest and substance of addiction. Heroin was the main substance of addiction for eight participants, alcohol for five and weed and cocaine for one each. Other drugs used, as described by participants, are brown sugar, cannabis, cough syrup, inhalants, marijuana, tablets and uppers. Nine participants were working as rehabilitation service providers, three in another form of employment, two were students and one was unemployed. Saturation of key themes and concepts for the men occurred after approximately six interviews and key differences in the women's accounts as compared to the men's were being reiterated in the interview with the fourth and final female participant.[24]

Our POs identified candidates from their service user communities, by word of mouth, and by distributing information about the study within their networks. Interested candidates recommended to us by our POs were provided the information sheet and invited to discuss the study in a face-to-face meeting where the procedure and conditions of consent were explained in the candidates' preferred language. If suitable, and wishing to take part, candidates were provided guidance material on collecting images to bring to interview (see Supporting Information). Detailed guidance was provided, such as the suggestion that 'You could start by thinking about the most important issues (or times, events, or people, or experiences) that you would like to talk about in the interview and then find an image, or take a photograph, that represents this in some way. The image can be of the thing itself or it can symbolize it'.

Data collection

Audio-recorded photo-led interviews were conducted between April 2019 and October 2020 at the premises of our POs, including Mind India: a private registered society operating throughout northeast India which provides counselling, psychosocial interventions and training. To enhance anonymity, participants were asked to provide verbal consent only which was audio-recorded before interview. Interviews were conducted in a mixture of Assamese and English. After the interview, consent was rechecked based on the participant now knowing what they had disclosed.

The interview topic guide (see Supporting Information) was developed in consultation with team members with expertise in rehabilitation and counselling in Assam. Interviews commenced with background information such as current employment situation. The usual format was then to ask the participant, 'Is there a picture you would like to share first?' using prompts where appropriate such as, 'What were your relationships with other people like at this point in your life?' and the interview ended with a request for feedback on the process of collecting images. Interviews lasted between 55 and 235 min (mean = 114 min) and the number of images brought ranged from 7 to 33 (mean = 12).

Analysis was conducted iteratively with data collection and queries raised through the analysis fed-back into the interview process. In practice, this did not change the interview topic guide but helped us identify where it was useful to add prompts to elicit more information if needed, for example about current daily functioning. Interviews were transcribed in English verbatim with translations from Assamese and checked for accuracy by two members of the team.

Data analysis

We used an inductive variant of thematic analysis.[25] Each transcript was assigned to two researchers who read it carefully and made general notes on the participant's recovery narrative. The assigned pair then discussed observations in an online meeting, one taking notes on agreed phenomena of interest, tentative patterns, concepts and themes, questions raised by the analysis and provided a short summary of the participant's story. These notes were passed to the second researcher for revision until agreed upon. The team met online together several times throughout the process of analysis to discuss the observations being made. As analysis progressed, the team decided to focus on patterns in the trajectory of the participants' stories of recovery having observed similar strategies and cycles in the material. We then rotated schematics between us until a pathways model was agreed upon and then credibility was checked. Hence, for the analysis reported here, our specific thematic analytic approach is: (i) a detailed account of one particular aspect of the data set; (ii) inductive as opposed to theory-driven; (iii) content-driven as opposed to interpretative and (iv) takes a realist as opposed to constructionist epistemological stance.

We present a pathways to recovery model that made sense of the complexities of the participants' narratives. This model was refined through expert and participant checks. An expert check was conducted with a clinical psychologist at LGB Regional Institute of Mental Health, Assam and with a senior addictions counsellor at Nirmaan Rehabilitation Facility. The expert check led to: (i) clearer justification of terminologies for, and articulation of, the stages *meaningful treatment* and *strategic self-management* and, (ii) an additional pathway from *strategic self-management* to *abstinence*. The senior addictions counsellor at nirmaan rehabilitation facility discussed the model further with members of staff at the rehabilitation facility. This resulted in adding a one-way path from *meaningful treatment* to *abstinence* to recognize that necessary lifestyle changes may not be evident after treatment even if substance use has ceased. Four participants (two men; two women, including the woman who recovered through peer support) who were willing to contribute further then individually took part in checking the model. The model was presented in pictorial form to aid understanding and each asked if they could use it to track their own journey to recovery (Figure 1). Each was able to do so with relative ease, with one (see Supporting Information) suggesting that the path between *in recovery* and *abstinence* should be two-way because there can be a period during which substances are not being abused but the recovery lifestyle is collapsing.

Enlarge this image.

RESULTS

We identify three phases defined as characteristic ways of relating to addictive substances: *Recreational Use*,

Addiction (*Relaxed*, *Chaotic* and *Strategic*) and *Supported Recovery* (Table 1). Each phase consists of a cycle between, or transition through, a series of stages. *Recreational Use* cycles between the stages of *not using* and *casual use*. *Relaxed Addiction* is a unique subphase consisting of a particular nonchalant attitude to the stage *in addiction*. The subphase *Chaotic Addiction* cycles between the stages of *in addiction* and *abstinence*, while *Strategic Addiction* cycles between the stages of *in addiction* and *strategic self-management*. The phase *Supported Recovery* consists of a transition through the stage *meaningful treatment* and settles on the stage *in recovery*, but may involve many cycles and relapses into different subphases of *Addiction*. The final element of our model, *transitions*, refers to the movement between stages each of which has a more *positive* or *negative* valance with regard to progress towards recovery.

Table 1 Phases, stages and transitions on the pathways to recovery model

We now describe the phases, stages and transitions in more detail and provide evidence through quotes from across our participants. The symbol […] is used to indicate a small portion of text omitted within quote and we indicated if a quote is from a female participant.

Phase 1: Recreational Use

All participants engaged with addictive substances at the beginning for one, or a combination, of the following psychosocial reasons. First, some wanted to gain credibility with older peers: ‘They didn't call anybody else, only me. I went and they were drinking there. They offered. I did drink too’. Second, others just wanted to join-in with friends: ‘the boys who live in my neighbourhood they too use it. And they told me “We will have alcohol. Will you have?”’ Third, curiosity was a key motivation: ‘my curiosity to know about it was increasing so then they made it by mixing with cigarette and gave me’ (female). Finally, some got into the *Recreational Use* of substances because they wanted to escape boring and/or difficult life circumstances: ‘Mom always showed her sorrows in front of me. Today we don't have this much money. […] I am staying good and I am getting the crisis. I will intoxicate [refers to Image 1]’ (female).

Enlarge this image.

In the phase of *Recreational Use*, participants had the opportunity to cycle between the stage of *not using* and the stage of *casual use*. However, it often was not apparent that a transition had occurred into the stage *in addiction*

until there was difficulty securing the needed substance(s). As one female participant explained, continual opportunities to use substances may obscure the fact that addiction has occurred: ‘often there is party so our withdrawal doesn't happen’ (female). However, at this point the ability to be a ‘take it or leave it’ *casual user* is no longer possible: ‘I tried leaving but means the symptoms started showing’.

Phase 2: Addiction (Relaxed, Chaotic and Strategic)

The phase of *Addiction* is reached via a transition constituting a *negative* ‘event horizon’ from which there is no return to *Recreational Use*. *Addiction* has three distinct, possible subphases all of which share the stage *in addiction* but are inflected with different subjective experiences and behaviours. There may be cycling between the three subphases, although they have a logical progression through a subjective orientation of nonchalance, to distress, to destructive adaptation.

In the subphase *Relaxed Addiction*, needed substances are largely available and, because the positives are perceived generally to outweigh the negatives, there is little or no subjective experience of distress or motivation to quit. For example, as one participant explains: ‘my main problem was fear, social anxiety, social awkwardness. That thing was completely removed by alcohol and cannabis for a temporary time but I thought that it is a permanent solution’. Depending on circumstances, *Relaxed Addiction* can become chronic or is a transitory lull: ‘I lie around drinking alcohol. If I think about it, useless. Means what will I do by thinking?’

The likely next subphase is *Chaotic Addiction* which cycles between the stages of *in addiction* and *abstinence*. During this subphase, there are periods of not using because the needed substance(s) cannot be accessed or because there is an attempt to quit on one's own: ‘Even ma'am [teacher] got to know that I was drunk. She got the smell. I sit near her. I felt very bad and I then I quit for some days. Means I didn't have it. Then my head started aching in the morning and little means I started wanting it. Keeps giving me cravings. Then it all started again’.

Our model does provide for the possibility that *abstinence* may lead to the stage *in recovery*, and one of our female participants did indeed take this route. However, without external support the transition to *abstinence* is fragile and may lead to relapse back to *in addiction*, especially for young adults. A key reason for relapse is that no personal or lifestyle changes have been made: ‘I've stayed clean for six months. So what happened to me? A misunderstanding developed between me and my family. I too had misbehaved’. Commensurate with this position, the female participant who recovered without professional help, did attend Alcoholics Anonymous and received mentoring from an addict-in-recovery. Moreover, the main gender difference we found was the way in which women needed to consider the implications of using professional support due to the possibility of being too identifiable as an (ex-)addict: ‘my mother, grandmother and to everyone said that. “Why is it required to let her go to rehab? She will not be able to get married. Who is going to marry her?”’ (female).

The likely next subphase is *Strategic Addiction* in which there is a cycling between the stages of *in addiction* and *strategic self-management*. The essential feature of *strategic self-management* is that there is no real or sustained intention to quit. Instead, interventions are engaged with to mitigate negative impacts and to sustain the addiction. Interventions may be used to mollify other people: ‘it was like it's better for me to stay in a rehab for like two three months. Be there and not use drugs. Gain my trust back from my family […] and then after that when I am out I will find new ways to get money, drugs’. Interventions may also be engaged to manage physical symptoms in the short term and deal with temporary interruptions to supply. One participant illustrates this in relation to opioid substitution therapy (OST): ‘the day I don't get money from home I take OST then this was my mentality. Family doesn't get to know. My addiction is also sustained’. A particularly common form of *strategic self-management* is short-term, medically supervised, family financed detoxification undertaken often on multiple occasions: ‘I started making excuses about all that too that I'll do detox. I need money for medicine. Give me money […] with that money I again keep taking heroin’.

Phase 3: Supported Recovery

In the phase *Supported Recovery*, the stage of *in addiction* moves into that of *meaningful treatment* and subsequently, if fortunate, to *in recovery*. Central to the transition from *in addiction* to *meaningful treatment* is acceptance of the support needed to quit. For example, one participant reflected on his experience of reaching out

almost despite himself: ‘Don't know what happened. I phoned that day to that counsellor. Phoned him that day and just asked how he was. He asked “What are you doing?” I said “What would I do? I'm doing substances.” He talked nicely and he talked so nicely that I thought means I should try once again’. There must also be a real and sustained intention and commitment to the personal and lifestyle changes entailed: ‘If I see someone that okay he is using she is using it's her personal life. I avoid that because I understood what are my priorities, what are the things I left back, what are the mistakes I had made in my life’ (female).

Despite engaging with *meaningful treatment*, there is the possibility of relapsing back to *in addiction* before being *in recovery* or after a period of being *in recovery*, directly or via *abstinence* defined as merely ‘not using’ without commitment to the long term: ‘After coming back from the centre, at home go out with friends. Same activity in my life. No change in the activity […] Life is not on track. Things which usually I won't do I do. So after that ah I had again’. The stage *in recovery* is always a work-in-progress and not an end-point or ‘cure’ that has been reached once and for all: ‘I've even seen people who have had 15 years of clean time but they relapse […] we are real-life soldiers who are always fighting for our lives’. Hence the importance of continued contact with a recovery community: ‘they are my seniors also and they are my using friends and we all are in recovery right now together […] basically I found my family here’. Some also sustained their recovery through the meaning they found in supporting others: ‘when they called I agreed. I said “OK I will do awareness programme. I want to [refers to Image 2]”’.

Enlarge this image.

DISCUSSION

We offer a multiroute, multidirectional pathway to recovery model of youth substance misuse in Assam. Three

studies have been conducted on the pathways to care of adult men in the context of SUD in New Delhi. These indicate a delay in treatment-seeking and report family, friends and neighbours as the main sources of encouragement.[26–28] Although important, knowledge of pathways to care does not tell us about the preceding journey into, and through, addiction, nor the subsequent, often long and convoluted, path to recovery. Moreover, there is little or no information on the pathways of women or young people with SUD, or in India outside New Delhi.

In a very different nonwestern context, Fatayir[29] (cited in Ali et al.[30]) offers a model of the trajectory through addiction from an Arabian perspective. Five stages are proposed: *Discovery* of pleasure; *Honeymoon* of consolidated consumption; *Early Addiction* where the substance is prioritized; *Elevation of Addiction* of personal and social deterioration; and finally *Zenith of Addiction* in which everything is destroyed except focus on the substance. There are interesting parallels with our model suggesting some potential transferability to wider contexts. *Discovery* may be similar to *Recreational Use*; *Honeymoon* to the transition to, and sojourn in, *Relaxed Addiction; Elevation of Addiction* may echo *Chaotic Addiction;* and *Zenith of Addiction* echo *Strategic Addiction*. However, unlike our model, Fatayir[29] does not include recovery, implies a linear trajectory based on the intensity of focus on the substance, and may have been developed from men's experiences only. Moreover, our model allows for individualized and cycling trajectories around a small number of behaviourally recognizable phases with differential intervention needs.

In a western context, the Transtheoretical Model of Change (TTM) is a popular addiction recovery framework.[31] First of five stages is *Precontemplation* in which no problem is acknowledged. Second is *Contemplation* where some thought is given to the problem and what to do. Third, in the *Preparation* stage, there is an intention to change. The fourth stage is *Action* in which change is initiated. Finally, in the *Maintenance* stage, efforts are made to consolidate progress and avoid relapse. Comparing TTM with our own model, youth in *Recreational Use* and *Relaxed Addiction* are likely *Precontemplators*. Interestingly, those in *Chaotic* and in *Strategic Addiction* are already in *Action*, presumably having undergone some *Contemplation* and *Preparation*. However, they have initiated action that undermines recovery, inadvertently in *Chaotic Addiction* and deliberately in *Strategic Addiction*. For the most part, it is only those in *Supported Recovery* that have taken effective *Action* and have a chance of reaching *Maintenance*. Research is mixed on the efficacy of TTM for understanding addiction and recovery, and a major criticism is that the stages are arbitrary and overlap. However, as is the case with our own model, DiClemente[31] does not claim that TTM posits a determined order or single linear pathway but is, instead, a developmental model of recovery.[32] On the other hand, TTM is a generic model of change and does not appear to capture well the cycling, cynical and self-destructive processes of addiction, particularly those we identify as *Chaotic* and *Strategic*. Moreover, TTM may posit an overly rational and individualistic change processes contrary to the often distorted and socially situated experiences informing our model. However, models like TTM, with a strong theory of change may complement inductively derived, contextually situated models such as ours and contribute to the transferability of our work to wider contexts (see also Hansen et al.[33]).

In Phase 1 of our model, *Recreational Use*, the risk of engaging with addictive substances is experienced as psychosocial pushes and/or pulls. Pushes consist of attempts to escape boring and/or difficult life circumstances. Pulls consist of personal curiosity and the need for social prestige and integration with peers. Similarly, a study of nursing students in Karnataka,[34] found the main causes of substance abuse to be, in order of frequency, peer pressure, enjoyment and family problems. In Assam, itself, Katoki et al.[5] report slum-living adolescents to be primarily influenced to take illicit substances by friends and peers and, secondarily, out of enjoyment or curiosity. Arguably, difficult life circumstances are endemic to this group, and fights, vandalism and criminal activities were reported by Katoki et al.[5] alongside SUD. These psychosocial pushes and pulls can be extremely powerful and we can extrapolate with support from the literature that resilience to drug initiation will include the presence of positive peer influences and role models, and strong family and community support,[35] particularly parental monitoring.[36] To this we add the directing of curiosity in constructive directions.

In Phase 2 *Addiction*, a major risk is remaining in an extended period of *Relaxed Addiction* because substances are not perceived to be a problem and/or are viewed as a remedy to pressing psychosocial issues. Although opinion is somewhat divided in the literature,[37] our participants were clear that, in their experience, having entered *Addiction*, it

is impossible to return to a 'take-it-or-leave-it' *Recreational Use* pattern. This may be due to biological changes[38] and/or the troubled contexts in which substances are secured and consumed.[39] Illicit drug use is often attempted self-medication, particularly in relation to psychological disorders. However, as Temmingh[40] points out, there is little research on this in LMIC. Psychosocial skills education, and early identification and treatment of young people with psychological disorders, could prevent many cases of SUD.[41]

Although research indicates that it is possible to recover from SUD unassisted, for example, through 'maturing out',[42] our work demonstrates that a risk for young Assamese people attempting to quit substances alone is entering an extended period of *Chaotic Addiction* characterized by a cycle of *abstinence* and relapse. A key aspect of this phase is captured by the slogan 'abstinence is not recovery', in which recovery is considered to be a 'voluntarily maintained lifestyle characterized by sobriety, personal health and citizenship'.[35],p.259 In the context of another nonwestern county—South Africa—Stokes et al.[43] report also the importance of a psychological mind-set for sustaining recovery, avoiding situations associated with substance use, and keeping otherwise meaningfully engaged. The skills and stamina to transform abstinence into recovery are unlikely without a strong, nonjudgemental support system of people and organizations that know what it takes.[44] However, as evidenced by one of our female participants, support does not need to be via official services.

Finally, within *Addiction*, there is a risk of settling into *Strategic Addiction* in which there is no real or sustained intention to quit but only to mollify other people and/or to deal on a known temporary basis with negative impacts and interrupted supply. In fact, a key finding of our study is that many parents and service providers are unaware of, or that a young person is in, *Strategic Addiction* and so waste scarce resources supporting interventions that actually sustain addiction. Specifically, our study suggests that, in Assam, there may be a particular risk of inadvertently colluding with *Strategic Addiction* through placing too much emphasis, and hope, on stand-alone detoxification treatment.

Important to understanding Phase 3 *Supported Recovery* is that it comprises, indefinitely, a complex constellation of de-addiction support, meaning that recovery must be viewed as a continual work-in-progress. Stokes et al.[43] report the importance of social support while adding that a transition into recovery is often sparked by a crisis turning-point. In Assam, people with SUD and associated mental health challenges are often stigmatized and assumed to be criminals.[45] As an educational tool, our model has the potential to challenge myths about SUD and increase the possibility of constructive community support for young people and their families. The transformation of community 'gossip' from negative to positive over an individual's recovery journey is documented in rural America, supporting the conclusion that community education is an important route to help addicts into long-term recovery.[46] Although our participants were predominantly from urban settings, there are strong resonances with Krentzman and Glass's[46] study given tendency to the interweaving of lives in Assamese neighbourhoods. There is also potential to explore longer-term recovery as Webb et al.[47] did with a British sample to understand the transferability of their findings that gratitude and reliance on support groups transformed into greater self-determination and independent decision-making.

It is exceptionally important to recognize the phase and stage in which a young person is so that the most appropriate intervention can be made. The two transitions associated with Phase 1 *Recreational Use* offer particularly fruitful points for *prevention* interventions: (i) prevention of starting casual drug use and (ii) prevention of SUD through quitting casual drug use before addiction occurs. The risk of slipping into addiction unawares cannot be overestimated and is noted also in Hansen et al.'s[33] study of nine American addicts in long-term recovery. However, prevention interventions are not appropriate if the young person has, even unwittingly, entered the phase of *Addiction* and that this has happened is often hidden deliberately from others.

Phase 2 *Addiction* interventions are best geared towards *accepting support to quit* and the importance of admitting to needing help is highlighted also by Hansen et al.[33] The process of recovery is difficult to sustain on one's own and *Chaotic Addiction*, in which this is attempted, can be physically, psychologically and socially destructive and lead to a dangerous level of hopelessness. Medicalised interventions with no long-term psychosocial follow-up are particularly problematic and can be used cynically in *Strategic Addiction*. Although it is a major success to enter

Phase 3 *Supported Recovery* there is always a risk that commitment to recovery flounders, and relapse can occur before or after a period of being in recovery. Support from others is vital to develop a real and sustained commitment to the personal and lifestyle changes required, particularly in peer recovery-oriented communities.[35]

We incorporated a purposive sampling strategy, selecting for diversity to generate rich and relevant material for an in-depth study. However, the relatively small sample size and situational specificity could be viewed as limitations. For example, to recruit 15 participants, 27 people attended an initial meeting to discuss the study and, although Hope Foundation Rehabilitation Centre identified five candidates, only one took part. We do not know why individuals did not join the study and the information sheet stated that we would not ask. Informally, we understand that several candidates relapsed just before reaching a substance-free year. All but one of our participants had attended residential rehabilitation services and, hence, are distinguished by having access to some financial resources. Moreover, 9 of our 15 participants were working as service providers in the recovery sector. However, many rehabilitation facilities in Assam provide the opportunity to give service as part of on-going recovery and Stokes et al.[43] note that helping others and working in a recovery environment is common for addicts in sustained recovery.

In terms of strengths, our participants were reasonably representative in terms of the type of addictive substances engaged, including those most commonly abused by young people in India, that is, alcohol (N = 14), cannabis (N = 11) and opioids (N = 7)[7] and, in Assam, solvent abuse (N = 5).[20] Alcohol was the main addiction of three of the four women, but this is commensurate with national figures that 26.3% of women aged 15–49 years in Assam consume alcohol, the highest by far of the 36 states surveyed.[48] Finally, our model was confirmed in expert and participant credibility checks including both male and female addicts-in-recovery and the participant who did not use professional rehabilitation services.

The key implications of this research for policy development in Assam are as follows. In addiction, interventions are best geared towards encouraging a young person to accept support to quit. Effective interventions, including medical treatment, require also long-term psychosocial support to have the best chance of sustaining sobriety. Investment in women's rehabilitation is needed due to the immense stigma women experience, even when in recovery, and will have the additional potential benefit of contributing to the well-being of their current and future children. Finally, investment in family and community education and peer-to-peer support is likely an economical and effective strategy for preventing youth SUD and enabling rehabilitation.

In support of these potential policy initiatives, we have cocreated a visually informed community education package around our model.[49] Early piloting has been conducted in Assam with high school students, the general public, postgraduate mental health trainees and women in rural and semirural districts. This demonstrated that the education package was successful in promoting young people's voice with regard to SUD prevention and recovery, increasing awareness of their needs and has the potential for stigma reduction. We are currently demonstrating our educational package to schools, colleges, rehabilitation services and health providers in Assam to encourage uptake, collect feedback and cocreate further ideas for incorporating the model in activities such as group lesson plans, personal recovery journaling and peer-to-peer mentoring. Future research includes a trial of the effectiveness of the education package and extending our understanding of the pathways to long-term recovery from SUD in Assam.

AUTHOR CONTRIBUTIONS

Anna Madill: Conceptualization (lead); formal analysis (lead); funding acquisition (lead); methodology (lead); project administration (lead); resources (equal); supervision (lead); visualization (equal); writing –original draft preparation (lead). **Raginie Duara**: Conceptualization (supporting); data curation (lead); formal analysis (supporting); investigation (lead); methodology (supporting); project administration (supporting); visualization (equal); validation (lead); writing –original draft preparation (supporting). **Sangeeta Goswami**: Conceptualization (supporting); formal analysis (supporting); investigation (equal); methodology (supporting); project administration (supporting); resources (equal); validation (supporting); writing –review &editing (equal). **Rebecca Graber**: Conceptualization (supporting); formal analysis (supporting); funding acquisition (supporting); methodology (supporting); writing –review &editing

(equal). **Siobhan Hugh-Jones**: Conceptualization (supporting); formal analysis (supporting); funding acquisition (supporting); methodology (supporting); writing –review &editing (equal).

ACKNOWLEDGEMENTS

We thank our participants for giving so generously of their time and for sharing their experiences with us in such depth. Our gratitude is extended also to Mr. Ratul Dey, and colleagues at Nirmaan Rehabilitation Facility Guwahati, and Dr. Diptarup Chowdhury Lokopriya Gopinath Bordoloi Regional Institute of Mental Health for their insights as clinical practitioners. Finally, we thank our many transcriber-translators, and Anjani Hathi and Charlotte Butler for undertaking literature searches. More information can be found on the project website https://projectresilience.co.uk/ . This research was funded by the UK Research and Innovation Global Research Challenges Fund jointly via the Economic and Social Research Council and the Arts and Humanities Research Council grant number ES/S00047X/1.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are openly available in ReShare at https://reshare.ukdataservice.ac.uk/855418/.

ETHICS STATEMENT

Approval was obtained from the Ethics Committee of the Lokopriya Gopinath Bordoloi Regional Institute of Mental Health, Assam and from the Ethics Committee of the School of Psychology, University of Leeds, UK. The study conforms to the Declaration of Helsinki guidelines. All persons gave their free informed consent before their inclusion in the study.

DETAILS

Subject:	Risk reduction; Mental health; Risk management; Bicycles; Public health; Relapse; Drug abuse; Recovery; Health surveys; Recreational use; Gender; Mental disorders; Rehabilitation; Prevention; Addicts; Narcotics; Marijuana; Substance use disorder; Heroin; Substance use; Addictions; Interviews; Drug use; Cocaine; Health professional-Patient communication; Alcohol; Tobacco; Transferability; Children &youth; Psychiatrists; Males; Teenagers; Substance abuse treatment; Youth
Location:	India
Identifier / keyword:	addiction recovery; LMIC; mental health; substance abuse disorder; visual methods
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	318-328
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES

Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-09
Milestone dates:	2022-10-26 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-06-09 (manuscriptReceived); 2022-11-09 (publishedOnlineEarlyUnpaginated); 2022-10-27 (manuscriptAccepted)
Publication history :	
First posting date:	09 Nov 2022
DOI:	https://doi.org/10.1111/hex.13658
ProQuest document ID:	2767101983
Document URL:	https://www.proquest.com/scholarly-journals/pathways-recovery-model-youth-substance-misuse/docview/2767101983/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the “License”). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-09-12
Database:	Public Health Database,Publicly Available Content Database

‘I don't know what to do or where to go’. Experiences of accessing healthcare support from the

perspectives of people living with Long Covid and healthcare professionals: A qualitative study in Bradford, UK

Baz, Sarah A [1]
; Chao, Fang [2]
; Carpentieri, J D [2]
; Sheard, Laura [1]
[1] Department of Health Sciences, University of York, York, UK [2] Institute of Education, University College London, London, UK

ABSTRACT (ENGLISH)

Background
In October 2022, it was estimated 2.3 million people in the United Kingdom have self-reported Long Covid (LC). Many people have reported not receiving adequate healthcare support. There is a lack of research which provides an in-depth exploration of the barriers faced by people with LC in accessing healthcare support. It is important to understand these barriers to provide better support, care and advice for those experiencing LC.
Objective
To understand the barriers faced in accessing primary, secondary and specialist healthcare support for people with LC.
Design and Participation
40 interviews were conducted with people living with LC in Bradford alongside 12 interviews with healthcare professionals (HCPs) providing LC support in Bradford healthcare settings. Interviews were analysed using reflexive thematic analysis.
Results
People living with LC had a large degree of difficulty in accessing healthcare services for LC support. We categorized the healthcare access experiences of participants into five main types: (1) being unable to access primary care, (2) accessing primary care but receiving (perceived) inadequate support, (3) extreme persistence, (4) alternatives to mainstream health care and (5) positive experiences. There was a severe lack of access to specialist LC services. Ethnic minority participants faced a further barrier of mistrust and fear of services deterring them from accessing support. HCPs discussed systemic barriers to delivering services. Experiences were embedded in macrostructural issues further exacerbated by the pandemic.
Conclusion
To better support people with LC, the barriers faced in accessing healthcare support must be addressed. Of significance, improvements to general practitioner access are required; especially as GPs are the first line of support for people living with LC.
Patient and Public Involvement
A patient and public involvement group is engaged at regular intervals in the project.

FULL TEXT

INTRODUCTION

Long Covid (LC) is a rapidly emerging medical condition that first drew headlines nationally and internationally in 2020.[1] In the early stages of the pandemic, many medical professionals and patients reported being neglected or disbelieved about their persisting COVID-19 symptoms.[2,3] Thus, they mobilized online via social media to create

awareness of their condition. As such, LC is believed to be the first illness constructed by patients.[2,4] Despite the increasing prevalence of LC, its definitions remain vague and are continuously evolving. Adopting the WHO definition, NICE states that the term LC 'is commonly used to describe signs and symptoms that continue to develop after acute COVID-19. It includes both ongoing symptomatic COVID-19 (from 4 to 12 weeks) and post-COVID-19 syndrome (12 weeks or more)'.[5,p.5] Post-COVID-19 syndrome is described as presenting with a cluster of often overlapping symptoms which fluctuate, change over time, affect any system in the body[5] and impact 'everyday functioning'.[6,p.674] Symptoms of LC include breathlessness, fatigue, cough, fever, neurological symptoms (such as loss of taste and smell and brain fog), skin rash and chest pain.[7] There has been an increasing emergence of academic studies exploring LC and the medical and social impacts it has on people's lives.[7–11]

1BoxPatient and Public Contribution

Designing an interview schedule for people with LC: The wider CONVALESCENCE research project has a patient and public involvement (PPI) group involved in various work packages. The PPI group is hosted by researchers at the University of West of England who have expertise in patient and public involvement. Members of the PPI group all have or had LC. After an extensive literature review,[4] a draft of the interview schedule was presented to the group via a workshop. Feedback suggestions included simplifying the language of questions and approaching questions sensitively. The interview schedule was then revised and piloted by the research team for further refinement (see Appendix A).

Data interpretation workshop: Following the advice of the UWE researchers, the PPI group were presented ahead of time with four interview transcripts from the data set and provided their interpretation of the interviews via a workshop. The theme of barriers to accessing health care was also highlighted by attendees, for example, they discussed patients being disbelieved (particularly young people) and fragmented services as some points of interest within the transcripts.

The United Kingdom has universal healthcare provision, which is free for most at the point of delivery.[12] However, barriers to access are impacted by a healthcare system which has faced years of austerity, budget caps, increasing waiting times, pressurized services, backlogs and workforce shortages.[12,13] This has been further exacerbated by the pandemic, consequently impacting people's ability to access health care. COVID-19 has been said to have created a 'perfect storm' 'interacting with and exacerbated by social, economic and health inequalities'.[12,p.3] The pandemic has further intensified health inequalities, and existing chronic health and social conditions.[12] Healthcare services are fragmented, with patients transitioning between multiple care pathways; often, patients consult with GPs who act as gatekeepers to other specialist services.[14,15] Given the complexities and uncertainties surrounding the diagnosis, treatments and impacts of LC, it is expected that it may become a burden upon the healthcare system.[16] Although some studies and commentary pieces have touched upon LC patients not being believed by healthcare professionals (HCPs) leading to them managing symptoms alone,[7,8] and the importance of relationship-based care,[17] there is less critical analysis of nonhospitalized people's experiences of not being able to access adequate healthcare support.[7]

Moreover, there is a lack of interpretative studies that embed ethnic minority and/or socioeconomically deprived LC patients' experiences of health care within the wider structural impact the pandemic has had on the National Health Service (NHS), health inequalities and consequently, how this shapes access to healthcare services. This paper will present findings from the Bradford sample of a national qualitative study. The aim of this paper is to understand healthcare access for people living with LC.

METHODSStudy design and setting

This paper is based on a qualitative interview study with 40 self-identified nonhospitalized people who are living with LC. Participants were drawn from the Born in Bradford (BiB) cohort, and further sampling via community connections in Bradford. The BiB cohort tracks the health and well-being of over 13,500 children, and their parents over time. The second component is three interviews overtime with 12–15 HCPs and those working in/with public health supporting people with LC in Bradford. In-depth semistructured interviews allowed people to share the lived experiences and challenges of having LC and for HCPs to share reflections on delivering care. A PPI group is

engaged at regular intervals in the project (Box 1).

Bradford is a city in the North of England with high levels of deprivation, poverty and health inequalities.[18] As such, we engaged with a socially and ethnically diverse sample. Bradford experienced a high number of COVID-19 cases compared to the rest of the United Kingdom. This was cited as 'likely to be due to greater deprivation, high population density and a higher-than-average number of multi-generational households'.[19,p.1160] Furthermore, it has been found in racial disparities report that ethnic minorities have been overexposed to and underprotected against COVID-19.[20,21] People from deprived localities are also more vulnerable to COVID-19 infections, both groups could disproportionately experience LC.[8,12]

Sample and data collectionPeople with LC

Interviews were conducted with 40 people living with LC in Bradford. Sampling purposively, we aimed to oversample ethnic minorities and those living in medium to high deprivation, using postcode and IMD score as a proxy for deprivation status. We approached people with a range of engagement with healthcare services and considered the severity of LC (mild to severe self-defined symptoms). BiB cohort participants were largely in their 30s and 40s. Twenty-one participants were drawn from the BiB cohort identified via a recurrent cohort survey. From February to August 2021, survey respondents had been asked if they had COVID-19 and how long their symptoms lasted, with four options to choose from. Reflecting the literature at the time, although there was no firm definition of LC, it was understood to be defined as having persistent symptoms for over 4 weeks.[9] Consequently, the main exclusion criteria were having symptoms of COVID-19 for 4 weeks or less. We sampled respondents who stated that their symptoms were either 5–12 weeks or over 12 weeks in duration. Once the list of potential 50 participants was generated by BiB, a research assistant called respondents, inviting them to take part. Twenty-eight were interested in taking part, the remaining 22 were either unreachable or not interested. Information sheets and consent forms were then sent out. The first author arranged the interviews. Out of the 28, 7 did not participate either because they were no longer interested or there were already enough female participants in the study, prompting us to recruit more men outside the cohort. 19 people were recruited outside the cohort through community workers and snowball sampling. Another three were approached but were not interested in participating or did not reply to correspondences. Those embedded in community settings had established trust and rapport with local people, which allowed for a diverse range of respondents to be approached.[19] Snowball sampling was used to take a more targeted recruitment approach and engage with underserved groups, for example, those whose first language is not English and men.

Participants were predominantly female, reflecting there being more mothers registered in the BiB cohort than fathers and females being cited as having a higher risk of developing LC.[22] Participants came from 10 different postcodes dispersed across Bradford. They worked in a range of occupations, from low-paid/low-skilled jobs, like warehouse workers, and professional occupations like nurses. The timeframe of when participants had their initial COVID-19 infection was broad. Four participants had COVID-19 at the very start of the pandemic when testing was not available. The rest of the participants had a confirmed infection via a PCR or lateral flow test as and when testing was available. Participants were at different stages of their LC illness; some had recovered, and most were still experiencing symptoms. At the time of the interview, participants had LC for a range of durations, from 6 weeks to around 20 months. There was a range of persistent symptoms reported, with a loss of taste and smell, fatigue and breathlessness being common. Table 1 provides further participant demographics.

Table 1 Participant demographics

Sex	
Female	29
Male	11

Ethnicity	
White British	7
Pakistani or Kashmiri	25
Indian	3
Filipino	2
White Other/Eastern European	3
Age group	
18–29	3
30–39	16
40–49	18
50–59	2
60–69	1

The interviews took place between November 2021 and March 2022. All interviews were conducted remotely by the first author either over the phone or via video call. Interviews in Urdu or Mirpuri were conducted with three people. The interviews ranged from a duration of 16 min to almost 2 h (average length of 38 min). All participants gave informed consent via a verbal audio recording at the start of the interviews. Interviews were recorded digitally and transcribed by a professional transcriber with identifiable information removed. Interviews in Urdu or Mirpuri were transcribed by the first author, with data used in outputs translated over to English.

HCPs

HCPs were recruited starting off by contacting existing HCP contacts of the last author, followed by further snowball sampling and identification of HCPs via already recruited participants. Emails/letters were sent with an information sheet and details of what was involved. Sixteen NHS HCPs and people working with/in public health running and supporting LC services in Bradford, a key criterion for recruitment, were identified. Four did not take part as they did not meet this criterion or were unresponsive. Overall, from the 12 interviewees included, there were 3 lead clinical practitioners from Bradford LC clinics, 1 occupational therapist, 2 physiotherapists, 2 GPs, 2 service managers, 1 public health official and 1 charity CEO working with local health services. Remote interviews via video call took place from December 2021 to April 2022. Again, these were recorded digitally and transcribed professionally.

Analysis

A reflexive thematic analysis approach was taken.[23] Regular analysis sessions were held by the research team (all authors) to develop themes. Healthcare access arose as a striking finding during our initial analysis sessions, with most participants discussing substantive, lengthy content about their experiences of accessing—or failing to access—various elements of the healthcare system. This participant-driven content about healthcare access continued to dominate interviews as fieldwork proceeded. After a close reading of transcripts and analytic discussion amongst the research team, we developed a coding framework which focused on healthcare access. The first author

then coded the transcripts, sense-checking with the other authors. Reflexive thematic analysis encourages the researcher to be explicit about their subjectivities, which are considered a resource rather than a threat.[23] During the interpretation phase, the first author drew on her expertise in ethnicity and health, and the last author drew on her expertise of macrolevel healthcare systems, to situate the findings within both the Bradford and national context. The first author conducted further reflexive and interpretative work to analyse and write up the paper, producing a coherent narrative about healthcare access for LC rather than reporting basic 'facts' about the topic.[24] HCPs perspectives were analysed in relation to healthcare access, integrating the two data sets. The interviews were analysed in Microsoft Word without any software package.

FINDINGS

We were immediately struck by the difficulty the majority of participants faced when accessing healthcare support for LC. This was a vociferous and very clear overarching narrative which proceeded throughout data collection. We applied the following research question to the data to help us make sense of what participants were telling us: 'What happens when patients try to access care and support for their LC symptoms?' We found this could be delineated into three themes. First, the differing experiences that participants had of healthcare access, which we break down into five main types. Second, experiences of mistrust and fear among ethnic minorities in our sample. Third, systematic barriers to service delivery which was an issue discussed predominantly within the HCP interviews.

Experiences of accessing healthcare

We found five main types of experience that participants discussed when accessing or trying to access healthcare support for LC. First, some people with LC were not able to get through to primary care and were not able to secure a general practitioner (GP) appointment. Second, many were able to access primary care but did not receive (perceived) adequate support from either their GP or secondary care. Third, a small group of participants who were extremely persistent in their interaction with health care sought LC support. Fourth, a group used alternatives to accessing mainstream health care for various reasons. Fifth, a small number of people had positive experiences. We also discovered a severe lack of access to specialist LC clinics.

Not getting through to primary care

Most notably, some participants were falling at the first hurdle when trying to access support and advice for LC symptoms from GP practices, often the first point of contact for patients. A common barrier was not being able to get through to practices via the phone, often facing a prolonged wait for someone to pick up, as this extract illustrates:

I will ring them and then I'm waiting on my break for like 10 minutes and nobody is answering, so I'll wait another 20 minutes. When I'm at home and I've got a day off, I don't know where to start. So I don't want to ring my doctor waiting, you know, 2 hours on the phone because I've got no time for it and I'm trying to manage my symptoms with ginger or garlic. (Interviewee 11, 40–49, female, Eastern European)

One participant was already aware that her GP was 'extraordinarily difficult' to get through to and instead went to her local pharmacist for advice:

I spoke to the chemist because our GP is extraordinarily difficult to get through and it's very difficult to talk to anybody other than the receptionist. So I thought I'd just go and talk to our local pharmacist and see if they can suggest anything and they just said that I've just got to let the symptoms come out naturally or take paracetamol for my headaches … relax…. there was nothing else offered if they could offer anything else I don't know…. (Interviewee 29, 30–39, female, White British)

HCP interviewees also acknowledged prolonged waiting time to get through to services as a key barrier. Resultantly, people could end up self-managing, potentially risking further health complications:

I think there are going to be a lot of people who we're not touching. I mean it's how do you get hold of your GP? Last time it took 50 phone calls, 50 tries on my mobile. How do you do that if you're exhausted? (HCP1, physiotherapist)

A further barrier for this group was having to justify their need for an appointment with the receptionist, often facing pushback. For example, when interviewee 34, a British Pakistani male in his early 30s, contacted his GP, he felt that he was not a priority. He mentioned the LC clinic to the receptionist, however, had to face a long waiting time of 3 weeks for an initial appointment with the GP before referral, leaving him to state: 'So in my mind at that time it was

just kind of that natural response to when you're being pushed back to say, “okay I'll leave it then” and that was that’. Thus, being able to get a GPs appointment in the first instance is one major barrier many people with LC are facing. But those who were able to eventually get through also faced hurdles within the healthcare system.

Accessing primary care but receiving (perceived) inadequate support

Most participants were able to get through to their GP and received an appointment but felt they had received (perceived) inadequate support from primary care. An interview with a couple with LC, living in a deprived area of Bradford, provides one account of such experiences. The difficulty of being able to access their local GP was further exacerbated during the pandemic. When they finally got through, they were not happy with the advice given:

Wife: …They said just take paracetamol.

Husband: This is normal. This is common in here. Take paracetamol and ring two months. (Couple interview, 40–49, Pakistani)

Some participants described a sense of disappointment in primary care. One participant stated that he felt ‘hopeless’ and ‘neglected’ (Couple interview, 40–49, male, Pakistani). Participants wanted to receive more advice and support from their GPs. Another participant felt that LC was not taken seriously compared to other medical conditions, a common finding reported in previous studies[8,25]:

…when I spoke to the doctor's about feeling rubbish ‘oh well it will be just Long Covid but we don't do anything about it’. If I'd said ‘oh it's anaemia’, then they do all these tests and you can progress. But if it's Long Covid it's just ‘well that's what [it] is’. (Interviewee 20, 40–49, female, White British)

Two participants stated that they had been referred to secondary care services by their GP but were still on a waiting list after many months. Interviewee 38 had been referred to E.N.T. and was on the neurology service waiting list for 6 months but still had no answer regarding why she was experiencing persistent head and ear pains for over a year. From the interview, there was a clear sense of frustration about how long it was taking to get an appointment and navigate a fragmented healthcare service. She wanted to find answers about the cause of symptoms experienced since her COVID-19 inflexion and had not been diagnosed with LC. The participant contemplated a private healthcare check-up as an alternative when visiting India to see family. There she felt she would be able to get all the tests needed in one hospital visit and find some answers:

GP is also waiting for investigations and they're just giving the medications but at the end of the day I mean I'm anxious, I don't know what's happening … Wherever I go, whatever they do they're saying everything is fine. My chest x-ray is fine … So nobody knows. They've not diagnosed it … Maybe I will go … back home in India … if I get a chance when I go back that maybe I'll go for proper treatment…. So think how many months I'm just waiting for this, you know, they could have done that CT head[scan] when I was actually there. They said no, E.N.T is only doing certain parts … I don't know what to do or where to go, to whom to ask and nothing is easy access. It takes forever … I'm really fed up with this … it's really hopeless. I'm trying to live with it now. (Interviewee 38, 40–49, female, Indian)

Extreme persistence

We found that a high level of persistence and familiarity with approaches to get through to the right person was required to gain access to primary care. A few participants were persistent in navigating their way through services to gain medical support. These were those working in professional occupations, for example, public health, or those who had extensive previous experiences of navigating GP services because of other long-term illnesses. Thus, they had high health literacy and access to resources. They too acknowledged the difficulties of getting through to GPs over the phone and illustrated the importance of making sure that people get through to a GP who knows their medical history and that they access continued care over time from the same practitioner,[17] as this extract shows:

First of all they put you on a triage list and get someone to call you back and I've had to insist and say, I need to be put on my doctor's list for her to ring me. There's no point in anybody else ringing me because they don't know me. I think that's the bugbear isn't it … sometimes I've had to speak to other doctors, but they've not really known and you get mixed messaging … I just need to speak to my own because it's having that trust in somebody as well isn't it. But on the whole, I don't have any quibble … they've genuinely been supportive. (Interviewee 10, 40–49, female,

White British)

Furthermore, interviewee 20, who has rheumatoid arthritis, discussed the resources that she drew upon to access primary care and be listened to:

…I think I probably I've got a bit more access than most people because of my rheumatology team. They do listen you see and because of my medication, you know, they have to listen to me. Whereas if I hadn't have had that communication and opened to me, I'm not sure it would have continued. (Interviewee 20, 40–49, female, White British)

Positive experiences of healthcare

A few participants described having an overall positive experience of engaging with primary care for LC support. This included GPs listening, providing reassurance, practical and emotional support, receiving continuous care and follow-up phone calls:

What I did appreciate was that my telephone call with the GP was probably slightly extended to the other ones that I have had in the past and the fact that it was the same person that I spoke to … There's an element of continuity of care that really helps. (Interviewee 1, mid-30s, female, Pakistani)

A participant who was initially hospitalized for COVID-19 described receiving follow-up support from her GP, who provided practical advice on breathing exercises to help with continued experiences of breathlessness:

For my breathing, I spoke to my GP and he recommended me to like get balloons and kind of blow into them. Breathing exercises. So I used to do breathing exercises…. (Interviewee 5, 18–29, female, Pakistani)

Overall, such approaches were cited as being helpful in managing the illness and can be learnt from to provide better support to people with LC.

Using alternatives to accessing mainstream healthcare

Every participant described a degree of self-management or 'burden of illness',[26] for example, prioritizing rest, reducing physical activity or using home remedies. However, some participants self-managed symptoms from the offset and chose not to engage with healthcare services. This was due to several reasons, including, not preferring to approach healthcare services unless necessary, not liking medicine, preferring self-management, not wanting to burden an already overwhelmed NHS, not knowing what help was available, mistrust, fear and past negative experiences which deterred healthcare access (see Section 3.2) and learning to live with symptoms with the hope that they would see change over time. One HCP stated that some may be 'accepting that this is how life is for them' (HCP5, GP).

Limited access to LC clinics

A startling finding was that only 1 out of 40 interviewees in Bradford had engaged with an LC clinic service. This one person was an NHS staff member and had accessed an LC clinic through their workplace that was designed to help NHS staff recover and progress back to work. A very small proportion was beginning to discuss the possibility of a referral with their GP if symptoms worsened, many had not even heard of an LC clinic.

Patients have to go through a prolonged process with their GP to gain a referral to the LC clinic. A clinician interviewee stated that to gain access to the clinic, symptoms must last for 12 weeks or more. Patients must go through an initial assessment with their GP to eliminate other health risks. It was argued that this timeframe can be reduced so people can receive earlier interventions:

I think probably identifying the right people, you know, so making sure that people don't miss out. I think probably not being necessarily so strict about this 12-week cut off, you know, because even at the moment the GPs are not allowed to refer to the community team until it's 12 weeks. But why not refer at 7 weeks, you know? Why wait? (HCP2, Physician)

Evidently, participants had to do the 'hard and heavy work'[8] to receive healthcare support for their symptoms. This depicts the different barriers and inequalities in accessing services among participants. The next section will further focus on mistrust and fear as an extra layer of barriers to accessing services for ethnic minorities.

Mistrust and fear

Mistrust and fear were pertinent issues amongst some ethnic minority participants. This has previously been cited as

a barrier in relation to COVID-19 vaccine uptake amongst ethnic minorities[19] and creates an additional bottleneck for people with LC. A few participants expressed fear of going to the hospital for treatment of their LC symptoms. In one participant's case, this reflected 'fake news' stories and rumours going around the Pakistani community in Bradford at the height of the pandemic that hospitalization could lead to death.[19] Interviewee 4 emphasized a lack of trust in doctors and a need to increase trust in healthcare services to tackle such rumours. This can be embedded in both experiences of historical and contemporary structural racism, which leads to mistrust in the healthcare system and may be further exacerbated in the case of COVID-19, as there has been a disproportionate number of deaths amongst ethnic minority people.[27]

…you should have that full trust in him [GP] … this 'negativity' that is spread this this this should not happen because I understand because I I didn't go to the hospital I didn't go because of this that I heard that that's it if you go to the hospital then a person does not come back alive…. (translated from Urdu) (Interviewee 4, 30–39, male, Pakistani)

The participant stated that he attained medical advice informally from a GP, which a family member put him in touch with. The GP spoke his preferred language, and provided reassurance, advice and 'emotional support'.

Other participants also expressed fear of being hospitalized, put on a ventilator and dying. This also reflects some participants knowing of people who have died from COVID-19, making it more 'real'.[19] This was deterring interviewee 36 from seeking medical support when experiencing frightening pains in his chest due to his LC. He was yet to take the first step to engage with services:

I felt so wheezy and I felt like my chest was tightening up around me and I was really close a few times to making the call to 911[111] to say, you know, this is happening, what should I do and I couldn't do it because I was too scared to make the call.

Interviewer: 'In what ways were you scared?'

I think like I try not to listen to people but I heard a lot of stories at the time that people were going to the hospital and not coming back out and were put on ventilators and stuff. That essentially was really scaring me and I did have one of my close friends, his brother passed away … Now that didn't scare me but it kind of puts that thing in your mind. He wasn't vaccinated. I'm vaccinated. But yeah just things like that really. I don't want to put myself in that position. (Interviewee 36, 30–39, male, Pakistani)

There were also accounts of participants lacking confidence and trust in HCPs. This was embedded in previous encounters with GP surgeries where they were misbelieved or not taken seriously. These past encounters played a part in deterring them from seeking medical advice for LC. These experiences occurred at the intersection of aged, gendered and racialized discrimination. For example, a young Pakistani woman described not being taken seriously by her GP:

I mean my doctors aren't really that good in that sense anyway, so I wouldn't even go to them for help … I went to the doctors once because I had a lump on my breast and he told me to lose weight. They didn't even check. In that kind of sense I don't go to the GP anymore because of them not being really practical about anything … it stops me…. (Interviewee 30, 18–29, female, Pakistani)

Another Pakistani man in his early 30s described feeling that as a 'youngish man' he was not prioritized or taken seriously and was previously 'denied' being given antibiotics for a medical condition, with his practice stating: 'you're a young fit guy, you'll be fine'. Therefore, mistrust ran in both directions as patients have been mistrusted by HCPs which consequently shaped their mistrust of the system.

Often in relation to ethnic minority experiences of healthcare access, language is cited as the key barrier. However, the three Urdu/Mirpuri-speaking participants in this study stated that their families, husband or support networks supported them in seeking medical advice. This raises the importance of shifting the conversation beyond a narrow focus on solely language to other barriers, namely mistrust and fear. This creates an additional barrier to access for ethnic minority people despite their language abilities. Past encounters of being disbelieved, having mistrust and fear can lead to a lack of confidence that adequate healthcare support will be provided, consequently impacting people's decisions on seeking support for LC symptoms, resulting in people self-managing symptoms.

Systemic barriers to service delivery and access: HCPs perspectives

HCPs in Bradford shared their perceptions about the barriers people with LC face when accessing health care. There was a mix of both praise and criticism of services. However, a salient finding was the systemic healthcare access issues that HCPs had to work around.

First, there was a lack of training for GPs about LC, particularly during the onset of the illness. HCPs were overstretched and often had to figure out themselves what LC was and how to support patients, drawing on knowledge of other illnesses like chronic fatigue syndrome, and in one GPs case via her own experience of LC:

…there was nothing to offer so we were kind of winging it … making sure we weren't missing our you know bread and butter stuff erm but it just kind of felt like there was something happening to these patients that we didn't know what was happening … it was something I was reading a lot about…. (HCP12, GP)

As discussed in Section 3.1, there was limited access to specialist LC services, with an emphasis being placed on access for NHS staff with LC and patients who had been hospitalized. A physiotherapist was informed to use existing services in her own practice to support people with LC, despite already being overstretched and with increasing workloads. Nevertheless, she continued to support her LC patients:

we do more than we should and erm we work more and more and later and later and then we cannot fill all our workload … we all know that in reality it means services are overstretched ones that are already overstretched…. (HCP1, physiotherapist)

At the time of fieldwork, a newly set up LC clinic aimed to provide holistic care to Bradford patients taking a multidisciplinary approach, which is particularly key as LC is often seen as a primary respiratory phenomenon. The main barrier to accessing this clinic was the long waiting list. HCP10 (a lead clinician) stated that the clinic was 'lagging behind' due to the time it had taken to set up and allocate funding. Additionally, there has been a struggle to recruit staff due to shortages of specialist staff, a wider system issue which is also impacting this service.[13]

Although HCPs felt that GPs were best equipped to support LC patients, as they had knowledge of the whole body, a CEO of a third-sector organization working with health services raised the concern of accessing the clinic via GPs, the primary route of referral. This can create an immediate 'bottleneck' as many face barriers to accessing GP appointments, particularly marginalized groups. This further illustrates the high importance of improving access to primary care but also using other methods to signpost patients to specialist services, including more engagement in grassroots community settings that are connected to the most underserved:

if there was a way for a wide range of groups to be able to refer, connect, signpost people to that service without having to jump through hoops for a GP then I think that will be more effective. (HCP8, charity CEO)

One GP working in a deprived locality stated that some of her patients were now able to access the clinic and shared positive experiences. However, she was yet to receive any information from the clinic on the progress made and instead had to ask patients. This reflects a fragmented healthcare system where patient record systems are not linked together between services, creating barriers to better-supporting patients.[28]

Importantly, these findings illustrate the impact of systemic issues on service delivery and the access and support people with LC get from HCPs.

DISCUSSIONExisting literature

This study presents similar broader findings to existing studies into LC, particularly the experiences of being disbelieved, trying to find answers, barriers getting through to GPs, having to navigate fragmented services and self-managing symptoms.[7,8,11] However, previous findings are largely considered in the context of broader descriptive findings of the multifaceted impacts LC has on people's lives and there has been a lack of in-depth critical exploration of the barriers to accessing health care, particularly for disadvantaged groups with LC. Previous studies have not fully captured the voices of ethnic minorities, with participants predominantly being White British.[7] This study particularly addresses this gap, with the sample being 75% ethnic minority and mostly living in deprived areas of Bradford, which allowed us to understand how the experience of healthcare access is shaped for this group of people and capture insights about mistrust. We found five different types of experience when accessing health care alongside a lack of access to LC specialist services. Overall, it is evident that people faced worrying difficulties in

accessing the healthcare system at all, with a high degree of persistence required just to access primary care.[29] As found in previous studies, there were some positive experiences of primary care, such as GPs following up and listening,[11] but many participants felt that their symptoms were not taken seriously.[28] People who were referred to secondary care had to wait many months to access services. Only 1/40 of the interviewees had accessed a multidisciplinary LC specialist service, with a few people discussing the possibilities of future referrals with GPs.

It is important to embed these experiences in literature from an inequality and structural lens, given COVID-19 and LC being both a health and socioeconomic crisis (often termed a 'syndemic pandemic') and experiences of access being shaped by inequalities and structural factors.[12,21] As previously argued,[7] the sociological theory of candidacy, which describes how eligibility for care is jointly negotiated between individuals and health services, is useful here in contextualizing experiences.[29] It is acknowledged that access requires considerable work by users and is argued that a number of factors, such as those at the material, structural, cultural, professional and individual levels, can shape the views of the most disadvantaged as to whether they are eligible for care.[29,30] Access to health care is lower in disadvantaged and deprived communities, with the number of patients per GP higher in the most deprived areas than in the least-deprived.[12] This results in reduced access to health care, further creating a bottleneck for people with LC. This can further contribute to inequalities and lead to worse health outcomes from LC for the most disadvantaged.[12] Similar to previous studies on LC, participants in this study were having to still do the 'hard and heavy' work of both understanding and managing a new illness and navigating fragmented healthcare services.[8,11] Moreso systemic barriers, including backlogs, a decimated and underfunded healthcare system and workforce shortages, mean people with LC experience barriers to access.[7,15,31,32]

Furthermore, this research adds further to emerging literature surrounding COVID-19, ethnicity and mistrust amongst ethnic minorities.[16,19,33] In relation to accessing LC support, experiences of mistrust and fear were rooted in the disproportionate impact of COVID-19 on ethnic minorities, intersectional accounts of discrimination and previous negative encounters with the healthcare system.[32,34] Shahid and Dogra[32] conceptualize this as 'medical mistrust'. This results in fear and reduced trust in HCPs and disparities and inequalities in the utilization of healthcare services.[32] This creates an additional barrier to accessing healthcare support for LC amongst ethnic minorities.

Implications for practice

Evidently, GPs are often the first point of contact for patients and play a crucial role as gatekeepers in facilitating access to secondary care and LC clinics and assessing patients.[15,25] Therefore, it is essential to improve access to primary care so people with LC are provided with better support and referral. Our study shows that this is a major barrier for LC sufferers, this was emphasized by both people with LC and HCPs. A backdrop of mistrust exists, this must also be addressed when looking at access to and engagement with healthcare services, particularly as Bradford has a diverse ethnic minority population and socioeconomic inequalities, which have led to greater risks of contracting COVID-19. Although progress has been made in setting up an LC clinic, HCPs cited the structural barriers in the healthcare system which impacted their ability to provide support to LC sufferers. As previously cited,[14,28,35] better communication between fragmented services is required so GPs can provide better follow-up support, alongside more training and education for HCPs about LC. Wider systemic issues routed in years of austerity are evidently also impacting access and service delivery. There is concern that not everyone is able to seek help in an overwhelmed system.

Strengths

Earlier studies into LC focused predominately on White British populations, and HCPs with LC, and recruited participants from online platforms.[3,7,25,36] A key strength of this study is that it accounts for the experiences of ethnic minorities, underresearched populations (such as those with English as a second language) and people living in deprived areas, allowing us to capture their experiences of healthcare access. Another key strength of this study is that it explores HCPs' perspective of LC service delivery and access, addressing a significant gap in the literature. Future research into LC needs to explore the perspectives of HCPs in different UK settings.

Limitations

We do not have 'evidence' of COVID-19 infection for some participants, particularly those who were infected during

Spring 2020 when testing was largely unavailable. The severity of LC was also self-defined by participants. These could be viewed as limitations, but, rather, we see the self-identification of our sample as a positive move echoing Alwan's[37,p.201] assertion that 'the burden of proof should not be on ill people'. Furthermore, the data reported in this study only focuses on one city and one time point. There is a lack of longitudinal follow-up research involving people with LC, and exploring their experiences over time.

Further research

Our study is the first sweep of data collection of a three-sweep longitudinal interview study, where we will follow participants both in Bradford and across the United Kingdom over three time points over an 18-month period. As this paper only presents the Bradford sample, we do not situate our current data as longitudinal or nationally representative. However, it is worth noting that in future publications, we will explore varying topics of importance to participants (such as LC, identity and existential loss), changes over time and whether participants have engaged with and accessed further healthcare support.

CONCLUSION

This paper has contributed to providing a more nuanced and in-depth understanding of the barriers and 'hard and heavy work'[8] people with LC face in accessing healthcare support, drawing on the perspectives of people living with LC and HCPs. These subjective experiences are embedded within deep-seated structural and systemic barriers, discrimination and health inequalities which create healthcare access barriers for people with LC.

AUTHOR CONTRIBUTIONS

J. D. Carpentieri and Laura Sheard designed the wider qualitative longitudinal study and obtained funding as part of the CONVALESCENCE grant. Sarah A. Baz collected all the data discussed in this paper and was the primary analyst for the healthcare access focus. All authors fed into analytic discussions. Sarah A. Baz and Laura Sheard wrote the first drafts of the paper. All authors read, commented and provided feedback on the paper, providing approval for the final manuscript.

ACKNOWLEDGEMENTS

The authors would like to thank the Born in Bradford cohort, researchers at Bradford Institute for Health Research who supported the study and recruitment process (particularly Dagmar Waiblinger, Zenam Bi and Shahid Islam), the PPI group at the University of West England and Andy Gibson, John Kellas and Sophie Taysom who facilitated PPI workshops. A special thank you to all of the participants who took part in this study and shared their experiences. Characterization, determinants, mechanisms and consequences of the long-term effects of COVID-19: providing the evidence base for healthcare services (CONVALESCENCE) was funded by NIHR (COV-LT-0009). This research team is working on the Qualitative component of the work package, reported here are findings from the Bradford sample of a nationally representative longitudinal qualitative study.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

This study has been ethically reviewed and given a favourable opinion by the University of York Health Sciences Research Governance Committee on 01/10/21 (ref: HSRGC/2021/466/B).

AAPPENDIXINTERVIEW SCHEDULE

- 1.

Introduction

We are interested in talking to you about your experience of living with symptoms of Covid. We know from the Born in Bradford survey you stated you had Covid symptoms for more than 4/5/12 weeks. Thanks very much for answering questions about Covid on the survey. Today, I'd like to have a more in-depth conversation about your

experiences of living with Covid.

We are aware that some people experience longer Covid symptoms than others. You may have heard of the phrase ‘Long Covid’ as it is an increasingly popular term (although we are aware that not everyone with Covid symptoms for more than a month would identify as having ‘Long Covid’). We are carrying out this study to understand more about the experiences of living with Covid symptoms for about 5–12 weeks or more. We aim to provide evidence to improve practice and policy.

This interview will explore the impact that Covid has on your everyday life and your experiences of accessing healthcare support for Covid symptoms. The interview will last between no more than 1 h. Importantly, you do not have to answer any questions you are not comfortable with. You can also stop or pause the interview at any time. You have the right to withdraw during and after the interview—any data collected will be destroyed if you decide to withdraw. If you would like me to repeat any question or provide further explanation, please feel free to ask. You can also ask questions at any time during the interview.

•2.

Opening question/ice breaker

(i) Tell me a bit about yourself (e.g., are you working/studying/retired)

•2.1.

Initial experience of Covid

•(I)

Can you remember when you first had COVID-19 symptoms?

•(II)

Tell me about your initial COVID-19 experiences.

•(III)

Did you get a test? (lateral flow or PCR?) What happened?

•(IV)

Did you have any contact with your GP, the hospital or other healthcare services? Can you tell me more about that?

•(V)

What support did you seek when you got Covid-19?

DETAILS

Subject:	Pandemics; Polls &surveys; Barriers; Minority &ethnic groups; Citizen participation; Health care policy; Health services; Health care; Medical personnel; Ethnic groups; Health care access; Health disparities; Primary care; Interviews; COVID-19; Patients; Public involvement; Minority groups; Medical research; Family physicians; Professionals; Females; Occupations; Qualitative research; Coronaviruses; Social services delivery; Access

Business indexing term:	Subject: Professionals
Location:	United Kingdom--UK; England
Identifier / keyword:	access; ethnic minority; healthcare support; ineq uality; Long Covid; qualitative
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	542-554
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Documenttype:	Journal Article
Publication history :	
Online publication date:	2022-12-13
Milestone dates:	2022-11-29 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-08-02 (manuscriptReceived); 2022-12-13 (publishedOnlineEarlyUnpaginated); 2022-12-01 (manuscriptAccepted)
Publication history :	
First posting date:	13 Dec 2022
DOI:	https://doi.org/10.1111/hex.13687

COVID-19 community assessment hubs in Ireland: A study of staff and patient perceptions of their value

McAuliffe, Eilish [1]
; Sophie Mulcahy Symmons [1]
; Conlon, Ciara [2]
; Rogers, Lisa [1] ; De Brún, Aoife [1]
; Mannion, Marese [3] ; Keane, Niamh [4] ; Glynn, Liam [5] ; Ryan, Joseph [6] ; Quinlan, Diarmuid [7] [1] IRIS Centre, School of Nursing, Midwifery &Health Systems, University College Dublin, Dublin, Ireland [2] Academic Affairs, Trinity College Dublin, Dublin, Ireland [3] Burren Medical Centre, Clare, Ireland [4] Midwest Community Healthcare Organisation (CHO3), Limerick, Ireland [5] School of Medicine, University of Limerick &HRB Prmary Care Clinical Trials Network Ireland, Galawy, Ireland [6] The Weir Clinic, Bandon, Cork, Ireland [7] Irish College of General Practitioners, Dublin, Ireland

ABSTRACT (ENGLISH)

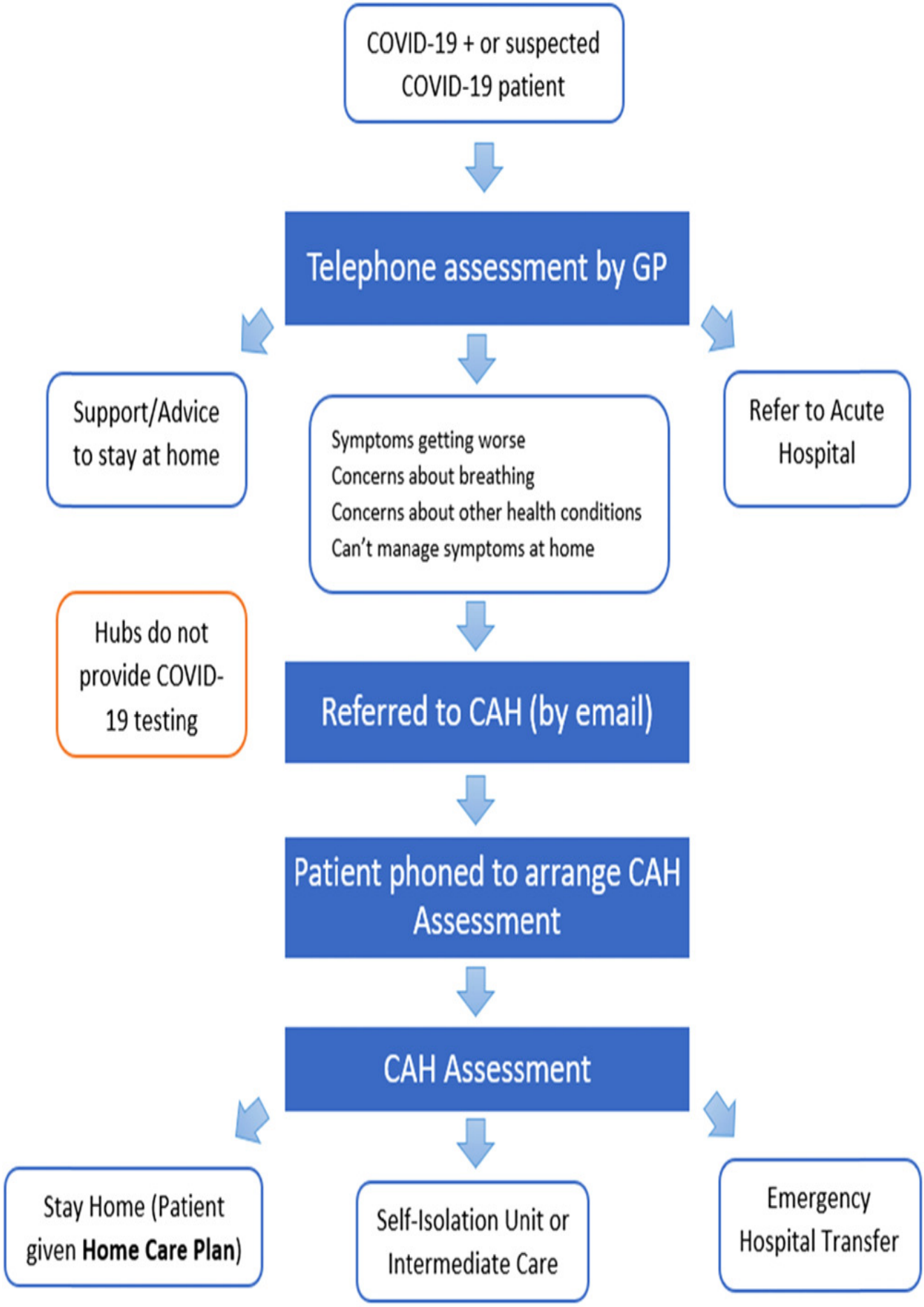
Background
Critical care bed capacity per capita in Ireland is among the lowest in Europe. The COVID-19 pandemic has put additional strain on an over-stretched healthcare system. COVID-19 community assessment hubs (CAHs) were established to prevent unnecessary admission to acute hospitals and to reduce infection spread.
Objective
The aim of this study was to assess the effectiveness and acceptability of CAHs and identify how the service might be improved or adapted for possible future use.
Design
This was a mixed methods study, incorporating co-design with clinical stakeholders. Data collection was via an online survey and semistructured telephone interviews with staff and patients conducted between January and May 2021.
Setting and participants
Thirty-one patients completed the survey and nine were interviewed. Twenty interviews were conducted with staff.

Results
The findings suggest that the CAH model was successful in providing a dedicated pathway for assessing patients with COVID-19 symptoms, whilst mitigating the risk of infection. Patients were particularly positive about the timely, comprehensive and holistic care they received, as well as the accessibility of the clinics and the friendly attitudes of the staff. Staff welcomed the training and clinical protocols which contributed to their feelings of safety and competency in delivering care to this cohort of patients. They also highlighted the benefits of working in a multidisciplinary environment. Both staff and patients felt that the hubs could be repurposed for alternative use, including the treatment of chronic diseases.
Discussion
This study describes staff and patients' experiences of these hubs. An unexpected outcome of this study is its demonstration of the true value of effective multidisciplinary working, not only for the staff who were deployed to this service but also for the patients in receipt of care in these hubs.
Conclusion
This multidisciplinary patient-centred service may provide a useful model for the delivery of other services currently delivered in hospital settings.
Patient or Public Contribution
An earlier phase of this study involved interviews with COVID-19-positive patients on a remote monitoring programme. The data informed this phase. Several of the authors had worked in the CAHs and provided valuable input into the design of the staff and patient interviews.

FULL TEXT

INTRODUCTION

The Irish healthcare system was considered poorly equipped to manage a global infectious disease pandemic. Irish hospitals operate at near full capacity on a regular basis. The low ratio of intensive care unit (ICU) beds to population size compared to other countries was of concern, given a late presentation to hospitals and subsequent rapid deterioration, resulting in more patients requiring admission to the ICU.[1,2] High levels of COVID-19 amongst healthcare staff added to the existing staff shortages and demand on the system. In addition, Ireland's low general practitioner (GP) to population ratio[3] resulted in primary care experiencing significant challenges managing the surges in COVID-19-positive or suspected positive patients. In summary, Ireland's healthcare system was not well-equipped to manage an escalating number of people presenting with COVID-19 symptoms, mild or severe. Thus, Ireland faced many challenges in the early stages of managing the COVID-19 crisis. In response to these challenges, the Health Service Executive rapidly developed a novel community assessment service in April 2020, namely the COVID-19 community assessment hubs (CAHs) to reduce infection of staff and patients in primary care services, avoid unnecessary emergency department (ED) attendance and provide timely specialized care for patients.[4] The World Health Organization recommended that countries set up services to assess, test and treat COVID-19 patients to combat the pandemic and the strain placed on health systems.[5] Service models with varying levels of integration with hospitals and level of care provided, including ‘fever clinics’, have been used effectively across the globe in other infectious epidemics,[6–10] but there is limited information on experiences of these services. The models vary across countries, the Irish model provides an assessment, triage and either treatment or referral. It is most similar to the primary care assessment services set up in the United Kingdom to alleviate the high demand for primary care services during the pandemic. These CAHs acted to triage patients who were COVID-19-positive or suspected positive. Figure 1 describes the CAH patient pathway where patients were referred by their GPs, assessed at the CAHs and then provided supportive information to isolate at home or at an isolation facility or referred to the ED if needed.[11] CAHs consisted of a multidisciplinary team of volunteer GPs, and redeployed nurses, physiotherapists and administration staff. Approximately, 50 CAHs were established around Ireland, operating 12 h a day, 7 days a week initially, although this decreased significantly as demand reduced. The CAHs were closed in March 2021.



Enlarge this image.

The COVID-19 pandemic provoked fear and uncertainty globally. The public was fearful of the disease, falling ill, spreading the disease to their loved ones and were hesitant to attend health services.[12] These concerns were

paralleled in healthcare staff. For healthcare staff, the pandemic provoked experiences that were both positive and negative.[13,14] Some staff felt unprotected, with limited personal protective equipment (PPE) and training, yet there was an atmosphere of solidarity and learning.[13,14] It is critical to understand these experiences and learn from them to inform service reconfiguration and redeployment of staff in response to any future pandemics or similar healthcare crises.[15]

Literature emerging from the COVID-19 pandemic has shown widespread strain on health services and staff and vulnerabilities of health systems. It highlights the need for more robust systems, developed protocols and adequate resourcing and capacity to manage in emergencies.

New services set up in emergency situations require evaluation just as services that are established over time. Despite this, they receive limited attention and therefore valuable learning may be lost. As the CAHs were a novel service, rapidly set up with little evaluation to date, it is critical to explore their effectiveness and acceptability from both the service user and staff perspective. Gaining experiential information through unveiling insight into patients' preferred care pathways and through understanding the benefits and challenges of delivering care at the CAHs is critical to this evaluation.

This paper explores the effectiveness, acceptability and experience of CAH-delivered services from staff and patients' perspectives in two regions in Ireland. The study time period is from April 2020 through to March 2021, when the CAHs closed.

Aim

The aim of this study is to assess the effectiveness and acceptability of CAHs and identify how the service might be improved or adapted for possible future waves of COVID-19 and other public health emergencies.

MATERIALS AND METHODSDesign

The study design chosen was a retrospective mixed methods study. The survey method was selected to gather data from as many patients as possible on acceptability, with follow-up telephone interviews being utilized to explore patient experiences in more depth. As this was a new experience for staff, we believed it was important to allow them space to expand on their experiences through interviews rather than constraining their responses within a survey format. The method was particularly useful for exploring possible future use of the hubs. Full details of the methods are reported elsewhere.[11] The online survey consisted of questions relating to the patient's demographic characteristics, COVID-19 symptoms and their experience with the CAH service (including the information they received to isolate at home [four questions rated on a scale of 1 (not at all) to 5 (greatly)], access to care [rated as very easy, easy, neither easy nor difficult, difficult, very difficult], quality of care [using the Patient–Professional Interaction Questionnaire (PPIQ) scale (16 questions rated on a scale of 1–5)]).[16] The interview guides for both patient and staff participants were designed to gain an in-depth understanding of the acceptability and experiences they had been assessed or working at the CAHs respectively. The guides included questions pertaining to their experience including changes in clinical practice, management of patients, communication, perceptions of care and timeliness at the CAHs. The interview guide was refined during data collection based on topics of importance to the participants.[11]

Ethics

The study was approved by the COVID-19 National Research Ethics Committee (Ref: 20-NREC-COV-093).

Participants and setting

Four CAHs across two regions in Ireland were the setting for this study. There were three CAHs in region A and one in region B. All staff who worked in a CAH (114 from region A and 60 from region B) for at least a week were invited to participate in the study via email with the information leaflet and consent forms emailed through gatekeepers.

All patients who were referred to and assessed in a CAH were aged over 18 and had the capacity to consent (all 194 patients from April to June 2020 in Region A and 200 patients assessed in January 2021 in Region B) were eligible to participate in the study. Those who met these criteria and had been discharged were therefore sent, via posted letter, an invitation to participate, an information leaflet and a link to a survey through gatekeepers. The survey was hosted on Qualtrics.com, which is GDPR (general data protection regulation) compliant. Additionally, 10

patients in Region B, assessed in January 2021, who were not contacted about the survey were sent an invitation to take part in the interview only. This CAH though not included in the initial study design, indicated an interest in being included. The timeframe for completion of the project did not allow for the survey to be completed in this CAH, but patients were contacted for interviews in an attempt to increase the number of patients interviewed.

Data collection

Telephone interviews were conducted with all participants due to the COVID-19 pandemic and the facilitation of social distancing guidelines and a national lockdown. Interviews took place between January and May 2021. Staff who returned the consent forms were contacted and a 20–30 min telephone interview was arranged with them at a time of their convenience. The aim was to purposively sample staff to ensure a range of views from each discipline (GP, nurse, physiotherapist, admin) were captured. Due to the low response rate, staff was convenience sampled and a representative sample of 20 staff participants from across all disciplines was obtained. Patients who completed the online survey and provided their contact details for a 20-min follow-up interview were contacted. Demographic data were collected from the survey. There was a low response rate for the online survey, hence convenience sampling was necessary. One researcher conducted all interviews remotely by phone, which were recorded and transcribed verbatim.

Analysis

Thematic analysis was conducted to explore all participants' perceptions of the CAHs and identify common themes.[18] Coding was conducted on NVivo 12 software.[19] Themes were drawn out inductively and reviewed and discussed with the research team. One researcher developed the initial codes from transcripts of the staff interviews. Initial themes were discussed with two other researchers and refined iteratively as more data were collected until consensus was achieved. A fourth researcher independently coded and developed themes from transcripts from patient participants, discussed with researcher 1 and integrated them with themes from the staff data. A fifth researcher independently coded a subset of transcripts to ensure the quality of the research.

Simple descriptive statistics were conducted on the 31 survey responses. Basic percentage summaries were calculated to determine the acceptability and experiences of patients attending the CAHs. The means and standard deviations (SDs) were calculated for the overall scores of the PPIQ and for the information on symptom management.

RESULTS

In total, 31 patients completed the online survey, of which, 21 indicated interest to participate in a follow-up interview. Fourteen were subsequently contacted for interviews and nine interviews took place, five in region A and five in region B.

Twenty-seven staff indicated an interest in participating by returning consent forms, however, 7 later declined, therefore 20 interviews with staff were conducted in total, 14 in region A and 6 in region B.

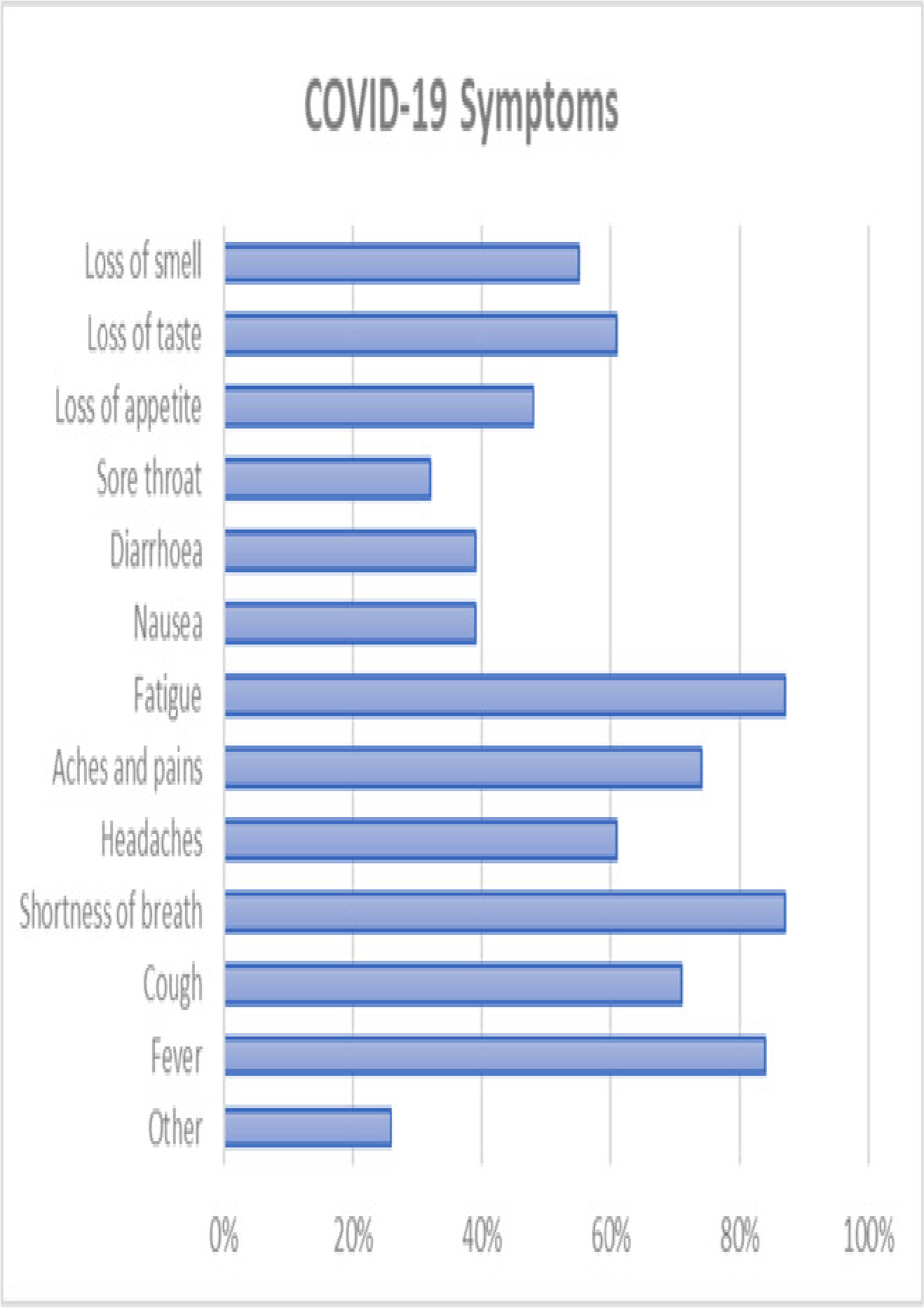
The demographic characteristics of respondents who completed the survey are displayed in Table 1. The sample was distributed across the age ranges from 30 to 60+ years with only 6% of the sample being under 30 years. The majority were female (61%), White Irish (83%) and had a university degree or above (58%). Of these patients, 27 (87%) tested positive for COVID-19, 2 tested negative and 2 were not tested (likely due to contracting COVID-19 early on in the pandemic when testing criteria were stricter).

Table 1 Characteristics of respondents who completed the survey

Age	18–29	30–39	40–49	50–59	60+
	6%	23%	26%	19%	26%
Gender	Male	Female			

	39%	61%			
Ethnicity	White Irish	White other	Asian		
	84%	3%	13%		
Education level	Lower secondary	Upper secondary	Postsecondary certificate/vocational	Degree/third level	
	10%	16%	16%	58%	

Figure 2 displays symptoms as reported by patients. The most common symptoms patients experienced over the course of their illness were shortness of breath (87%), fatigue (87%) and fever (84%). Some patients reported still experiencing symptoms at the time of completing the survey. Patients were also asked about underlying conditions with the most commonly reported being high blood pressure (26%), asthma (23%), followed by obesity (19%), and diabetes (19%).



Enlarge this image.

Table 2 provides a summary of how patients rated their experience of care. Most patients rated their experience of accessing care at the CAHs as ‘easy’ or ‘very easy’. In relation to the patient–healthcare staff interaction, patients

rated the quality of care highly (the mean PPIQ rating was 72.8 [(SD = 10.7]) where the maximum score was 80. The majority of patients (71%) rated their care as better than usual, with 26% rating the service as 'about the same' as their usual experience of healthcare services. Most patients received information about how to manage their symptoms at home, but 29% were referred to the hospital for further investigation/treatment. Patients rated the information they received with a mean rating received 18.9 (SD = 6.9), where the maximum score was 25.

Table 2 Patients' experience of care received as measured through the survey (*N* = 31 respondents)

Variable	*N* = 31, *n* (%)
Ease of access	
Getting through to my GP/family doctor on the telephone	
Very easy	11 (35%)
Easy	12 (39%)
Neither easy/nor difficult	4 (13%)
Difficult	2 (6%)
Very difficult	2 (6%)
Does not apply	0
Getting an appointment at the COVID-19 CAH	
Very easy	19 (61%)
Easy	9 (29%)
Neither easy/nor difficult	3 (10%)
Difficult	0
Very difficult	0
Does not apply	0
Getting the results of your assessment	
Very easy	18 (58%)
Easy	9 (29%)

Neither easy/nor difficult	2 (6%)
Difficult	1 (3%)
Very difficult	0
Does not apply	1 (3%)
Getting access to further care (hospital/isolation facilities)	
Very easy	10 (32%)
Easy	3 (10%)
Neither easy/nor difficult	4 (13%)
Difficult	3 (10%)
Very Difficult	3 (10%)
Does not apply	8 (26%)
Received information to isolate at home	
Yes	21 (68%)
No-referred to hospital	9 (29%)
No	1 (3%)
	Mean (standard deviation)
Patient–Professional Interaction Questionnaire	
Total sum	72.8 (10.7)
He/she provided me with clear information	4.6 (0.87)
He/she was interested in what I feel about my current health status	4.6 (0.8)
He/she turned to me in a calm and quiet tone	4.7 (0.63)
He/she understood my emotions	4.6 (0.85)
He/she was interested in what I know about my disease/prognosis	4.5 (0.85)

He/she respected me as a person	4.9 (0.43)
He/she was interested in what I want from care	4.5 (0.85)
He/she was able to listen	4.8 (0.56)
He/she paid attention to what I was saying	4.7 (0.69)
He/she was able to put him/herself in 'my shoes'	4.2 (0.99)
He/she gave me time to ask and to talk about the disease	4.5 (0.93)
He/she inspired confidence and security when touching me and being nearby	4.7 (0.59)
He/she asked questions that allowed me to express my view	4.4 (0.99)
He/she was interested in what I expect from care	4.4 (0.99)
He/she gave me encouragement and transmitted optimism	4.4 (1.05)
He/she offered me the opportunity to discuss and decide together the 'things to do'	4.3 (1.25)
Information on symptom management	
Total sum	18.9 (6.9)
Increased my knowledge about COVID-19	3.9 (1.3)
Was useful to help me manage my symptoms at home	3.6 (1.6)
Reassured me that I could manage my symptoms at home	3.6 (1.6)
Reduced my anxiety about COVID-19	3.6 (1.6)
Reassured me that I would receive appropriate care if my symptoms worsened	4.2 (1.5)

Abbreviations: CAH, community assessment hub; GP, general practitioner.

Of the CAH staff interviewed, there were six nurses, six GPs, two physiotherapists and six admin staff. There were 15 females and 5 males, reflective of the overall gender balance in healthcare staff.[20] Of the patients interviewed six were female and three were male. The age range most common among the patients interviewed was 30–39 (n = 4), while three were between 40 and 49 and the rest 50 or above. The majority of patient participants were White Irish (n = 6) and 8 (90%) indicated they held a degree or above. Four main themes were identified in the staff and patient interviews: (1) Efficiency of protocols and referral pathways in mitigating risk, (2) Patients' positive experience of care, (3) Positive working environment and (4) Potential for an expanded role for hubs.

Efficiency of protocols and referral pathways in mitigating risk

This major theme consisted of a number of subthemes namely: Mitigated exposure to risk; Specialized training;

Dedicated pathways of care for COVID; and Referral systems.

Mitigated exposure to risk

Safety was referred to many times in staff narratives as it is critical for staff when delivering healthcare, particularly in the context of a progressing pandemic. Feeling safe in accessing care was also crucial for patients, as hesitancy to access healthcare has been reported during COVID-19, as well as in previous pandemics. Both staff and patients were satisfied with the processes related to the delivery of care, such as patient pathways and infection control protocols put in place in the hub. One staff member described being prioritized in terms of access to PPE, and many referred to the thorough clinical training around COVID and PPE they received. Efficient organization and responsive management also contributed to a sense of safety, which alleviated the worry of contracting COVID-19:

They were very well organized. We were trained extremely well. I felt very comfortable at work and working with the team (Staff 12, Region A)

A couple of patients also described feeling reassured by the protocols in place, with one stating they felt staff were taking ‘all precautions for me and themselves’ (Patient 8).

Specialized training

Although many staff referred to the specialized training provided on COVID-19 and PPE and how beneficial this was in helping build their confidence in dealing with COVID patients and refreshing skills, there was variation in the standard and amount of training received across the different hubs. One participant felt a standardized training would be more appropriate to ensure staff was providing consistent standards of care across all hubs:

there was a variety of different levels of training going on. And even when we as a staff got together, and we compared the different training we had got, there was a lot of difference. Which you might think is surprising, but that was the case that was. Depending on where you got your training, you were told different things. So, that isn't a great idea. So, much more, the training needs to be made much more standardised, across everybody involved in the hubs (Staff 2, Region A)

An example of this variation ‘we did sit down in the hub one day and we had a respiratory nurse specialist speak to us which was a kind of an added-on training if you like. It was not part of what the national guidelines was, that it was recommended training’ (Staff 7, Region A).

Others spoke of this standardized national training programme ‘we did a lot of that beforehand, that was for the month of March really, we were doing it on HSELanD (Health Services Executive training platform) constantly’ (Staff 6, Region A).

Dedicated pathways of care for COVID

Both staff and patients felt that the service served to bridge the gap between the community and acute services, relieving the pressure on the health service. Staff felt that the hubs mitigated the risk that would have arisen had symptomatic or COVID-positive patients been seen in the usual health facilities. Seventy-one percent of patients reported that the dedicated CAH service was better than their usual health service. There was also a positive sentiment expressed from both patients and staff regarding the specific pathway of care the hubs provided to COVID patients, enabling staff to provide appropriate and precise care for specific symptoms:

it's very specific, the care is very specific, and the equipment is there that's needed and there's a process, you know there's no diversion away from it and it's very clear (Staff 4, Region B)

Patients were reassured by the sense that the hubs were a dedicated service, a ‘one stop shop’, where staff were trained specifically and had the expertise required to treat COVID:

…somewhere that was specialised for Covid was reassuring … to go and know that everybody there had the best knowledge that they could of this you know (Patient 6)

Patients that were advised to isolate themselves at home were provided information about how to manage their symptoms, and these patients rated the specific information they received highly (mean = 18.9, SD = 6.9).

There was widespread agreement across all participants that this specialized model of care could be repurposed for other diseases, something we will return to later in this paper.

Referral systems

Staff felt the referral pathways with GPs and EDs were effective, and particularly expressed satisfaction with the utilization of the iNEWS (iNEWS Irish National Early Warning Score—uses a recognized scoring system to determine the degree of illness of a patient and prompts critical care intervention) score, which (because of its widespread use in EDs) improved communication with EDs and fast-tracked patients through the triage process in EDs, reducing the wait time for patients. Patients who were referred to the ED also mentioned this as an additional benefit of attending hubs. The use of an electronic referral system was generally thought to be efficient, with clear and direct communication between services enabling good service integration, although there were some initial technological glitches and some GPs who were less proficient with technology may have found it difficult to navigate at times.

Patients' positive experience of care

Patients were overwhelmingly positive about their experience of receiving care in these hubs, speaking specifically about the timely access and good geographical accessibility of the hubs, as well as the high quality of care and the benefits of being attended to by several healthcare disciplines in one visit.

Timely access and accessibility

Patients expressed high levels of satisfaction with their experience in the hubs and staff also perceived a high level of patient acceptability of care received. The majority of surveyed patients rated the ease of access to CAH services highly, with 90% of responding patients considering it easy or very easy to obtain an appointment Patients found the hubs to be very accessible, both in terms of securing appointments and physical proximity/transport to the hub. Most patients were given an appointment ‘within a few hours’ of referral, and some were provided with transportation to the hubs. Many also felt the hubs were conveniently located. Patients also appreciated how staff anticipated their arrival and dealt with them promptly, as compared to often long waiting times in EDs.

High quality of care

Patients described pleasant interactions with staff, who were described as ‘kind’ (Patient 8) and ‘approachable’ (Patient 7) and providing a high level of reassurance. This perception of patient-centred care was evident in the survey results, with high PPIQ ratings observed (mean = 72.8, SD = 10.7). Patients felt heard, reassured and informed, while staff observed how patients' anxiety reduced during the assessment.

I just felt that they all really listened to what I was saying and how I felt and what my experience was. (Patient 6)

There was a sense that staff was able to dedicate more time to patients and they did not feel rushed, as opposed to other services where they often feel staff are more time pressured:

There was no rush on the assessment hub staff or there didn't seem to be anyway. It wasn't to get you in and out as fast as possible, it was to get you in and have a look at you and do a thorough examination of you (Patient 5)

Overall, patients felt the staff held a high level of expertise and provided a thorough assessment of their condition, although one participant expressed uncertainty as to the seniority of the clinicians, and another was concerned about the lack of continuity of care. Staff credited the positive patient experience to the calm environment of the hub, and the low numbers of patients, in contrast to busy and overcrowded EDs, which allowed them to dedicate more time to each patient:

I think they preferred it to going to A&E where it would be a lot more crowded, and the waiting times are longer (Staff 5, Region B)

Multidisciplinary nature of service

Both patients and staff highlighted the benefits of the multidisciplinary nature of the team providing care, which staff felt helped them provide a more holistic, patient-centred approach to care. Staff felt patients benefited from having a multidisciplinary team that could provide comprehensive care for all of their symptoms:

having those core disciplines there, that was excellent for patients. When else would a patient see all those disciplines together unless if they were in the acute hospital? (Staff 3, Region A)

Patients echoed these sentiments, with one pointing to the availability of a physiotherapist to perform breathing exercises with them, concluding ‘I couldn't have asked for better help’ (Patient 9).

Hubs as a positive working environment

Both staff and patients commented on the positive nature of the hub environment, agreeing that they were well-organized, safe spaces that fostered a high degree of collaboration and learning.

Safe organized spaces

Staff spoke about the anxiety and fear of so many ‘unknowns’ when they were redeployed to work carefully in the hubs. Some staff described an initial discontent and apprehensiveness about the redeployment, which was challenging to deal with from a management perspective.

A lot of them were very disgruntled and unhappy and nervous about coming in. Because we were walking into the unknown (Staff 2, Region A)

However, once training was received and the hubs were operational, staff felt they provided a safe place to work. Staff expressed positive sentiments about their involvement with an innovative health service, that was shifting pressure from acute services. They referred to strong leadership and responsive management who listened to their concerns and kept the staff ‘in the loop’ (Staff 3, Region A). The overall consensus was that the hubs were organized and well managed. This was also reflected in patient narratives who commented on how ‘it was really well planned out and well organized’ (Patient 7).

Fostering collaboration and interdisciplinary learning

It was evident that the staff felt the hubs cultivated collaborative ways of working, interdisciplinary learning and a reduction of hierarchies in the team. Despite some initial reluctance, staff described working in the hubs positively; as an interdisciplinary environment that created a shared learning culture: ‘shared learning between the different disciplines’ (Staff 3, Region A). They described gaining renewed confidence in their own skills and ability to be flexible and adaptable.

Staff felt being co-located fostered collaboration, particularly those who typically worked independently. Most described ‘a strong sense of collegiality’ (Staff 2, Region A) and ‘team spirit’ (Staff 8, Region A), assisting colleagues with PPE. Participants felt this facilitated them in building rapport and establishing new working relationships which their role would not usually facilitate. Some participants felt the hub environment lent itself to reduced hierarchies among the disciplines, and a sense of shared decision-making with more information sharing and consultation between different disciplines:

I was included in the conversation and probably that would never happen in my previous role, but it was yeah I found it inclusive […] we were all linked together, there was that sort of camaraderie feeling about it and everyone supported each other because it was the unknown you know (Staff 10, Region A)

Participants also felt the experience of working together in the hub has facilitated the building of both professional and personal relationships with other healthcare workers in the community, which has proved to be beneficial in community patient management since the hubs closed, for example when referring patients to other services:

Sometimes in the community, doctors, and nurses… you know it's very cut and dry whereas now, you've that relationship with them (Staff 6, Region A)

Potential for an expanded role for hubs

Participants (both patients and staff) were specifically asked if the hub model might be useful for use in the future and for purposes other than COVID-19 assessment. Several commented on the demand and supply imbalance and some frustration was expressed at what was perceived as an underutilization of the hubs. Despite this, many staff and patients felt the hubs could be adapted for use in the management of chronic diseases, to reduce unnecessary reliance on EDs and for other situations where infection control might be paramount. Staff emphasized the need for additional resourcing of the hubs to render them suitable for the expansion of their use into these other areas.

Underutilization of hubs

The peaks and dips in COVID-19 infection rates meant predicting the numbers of patients that might attend the hubs and resourcing the hubs accordingly was a difficult task for management. One positive result from this was that low demand provided a better care delivery experience for patients, with staff having more time to attend to individual patient needs.

the level of care was excellent, the expertise was spot on, they were courteous, there wasn't a feeling of being

processed, it was a matter of people were listening and they were interested and they were, it wasn't a matter of here's your script and off you go, it was a totally different attitude altogether (Patient 9)

However, some staff expressed frustration at the low volume of patients, particularly staff who had been redeployed from services that had increasing waiting lists. Some thought the hubs were underutilized and sometimes inappropriately used by GPs for several reasons including GPs not having enough awareness about the hubs and their role, the financial loss GPs would incur by referring a patient, or a desire to treat their patients on their own practice rather than refer to the hubs. Staff raised questions about the financial viability of the hubs due to low demand:

when you divide that [cost of the service] by the number of patients seen, fairly expensive assessments (Staff 5, Region B)

Providing more holistic care for chronic conditions

Both the patient group and the hub staff remarked on the opportunity to provide more holistic care with several disciplines in one site and how this could be a good model for future services.

and a small team you know of you know GP, physio, and specialist nurses you know you could certainly handle a lot of stuff. You know and then keep it in the community (Staff 4, Region A)

Several staff mentioned diabetes as a condition that could benefit from this holistic multidisciplinary care model.

diabetic patients. If they were able to come and have their outpatient care, like if they were attached to a particular hub… I mean if they had to come for say wound care, dressings, you know skin care, you know you'd obviously have tissue viability Nurses, or you know Public Health Nurses there who might be able to look at that, and then if there's issues around general health and physical fitness and, you know a lot of diabetic patients, for example, end up with particular things like neuropathy, frozen shoulders—things that Physiotherapists might help with—and the same an Occupational Therapist might have a role in terms of helping them with particular equipment or activities that they need to do, and then the Doctor would look at managing of their blood sugars and all of that, and I suppose it would be just nice to have a whole team collaborative approach, rather than having to go to the acute hospital. (Staff 8, Region A)

The management of other chronic respiratory or cardiac conditions through community hubs was also mentioned by several staff.

people with chest problems could go there, especially for the physios and that you know and even just dedicated clinics really, that's what it would be very good for, even leg ulcer clinics, just different clinics you could bring people back to one place with an IT system that works well (Staff 6, Region A)

I think there's definitely a big gap in the community for not just respiratory, and I definitely think you know, a lot of the patients that are in A&E don't need to be there. They need to be assessed in somewhere like a Hub (Staff 5, Region A)

Reducing utilization of EDs

This perception that there are many patients who attend EDs that do not need to be there, was echoed in the patients' comments. They were particularly positive about the time saved:

the nature of the expertise that was there locally without the endless ED waiting time because that's probably where my GP would have sent me next (Patient 9)

Patients were also concerned about not utilizing the scarce resources in hospitals when they could be effectively treated elsewhere and felt that the hubs facilitated this.

I definitely think those units are an amazing idea because if the GPs can't see you, you are very sick like that, you don't want to be going to the hospital where there are people sicker than you are, do you know what I mean, obviously I didn't want to be taking up a bed that I didn't need. (Patient 4)

There was therefore strong consensus between patients and staff that these hubs could play a vital role in reducing the number of patients that may be attending EDs when their condition could be more efficiently treated in a community setting. Some staff did, however, draw comparisons to the Medical Assessment Units in acute hospitals and were concerned that utilizing the hubs to reduce the burden on EDs may just result in a duplication of the work

that these established units are currently undertaking.

Infection control and expert knowledge

Staff repeatedly referred to the quality of infection control measures and the training in the management of COVID that they received when deployed to work in the hubs. As already highlighted, many staff commented on a feeling of being protected and being in a safe working environment. Patients were also reassured by the expert knowledge of the staff and the measures that were being taken to ensure the risk of infection was minimized.

it was very, very reassuring to go and know that everybody there had the best knowledge that they could of this you know. (Patient 6)

Given the high-quality infection control and expertise of staff, it is not surprising that many of those interviewed were in agreement that this model of service delivery should be replicated for any future pandemic or outbreak of infectious disease.

Further resourcing of hubs

Many participants commented on the absence of diagnostic tools, such as X-ray machines and blood testing requests, and felt that without these the hubs were not being used to the maximum potential for COVID patients, and the absence of such tools would limit their utility for other conditions.

It's called the respiratory hub and that should have all the facilities for what you are being assessed for (Patient 1)

Patients thought these resources would have streamlined their care experience and prevented them from being referred to EDs. Some staff also felt limited in what they could do for the patients.

Many participants felt the hub model, if better resourced, could be applied to other conditions to create more specialized holistic and patient-centred care using a multidisciplinary approach.

some of them just might need bloods and chest X-ray to see you know and check their oxygenation and see are they safe at home you know, can they be managed at home and could the community intervention team then give IV-antibiotics at home you know, save a bed you know (Staff 4, Region B)

There were also suggestions about staffing resources that would be required should the hubs be used as specialized clinics:

GPs who have a special interest in a certain thing could be brought in to run a clinic or day hospital you know … and you could have a consultant in there maybe for the afternoon and you could have lots of day cases to do you know and—so I think that model you know is a good model (Staff 10, Region A)

One staff member felt that there was a specific gap in paediatric care that CAHs could usefully fill.

We have the medical assessment unit and the surgical assessment unit and the local injury clinic for adults, but something, yeah, like a paediatric assessment unit would be helpful and run in probably a similar way … Yeah. Well not necessarily a GP, I suppose, but maybe a paediatric doctor or a GP that specialises in paediatrics (Staff 11, Region A)

DISCUSSION

This study adds to the limited evidence on the acceptability of COVID-19 primary care services from both the staff and patient perspectives. Only one study has assessed the CAHs in Ireland to date,[17] which found high compliance with infection control procedures at the hubs, as well as reported positive experiences of staff working at these hubs, corresponding to the findings of this study. In the United Kingdom, where similar primary assessment centres were established, staff reported improved knowledge of COVID-19 and confidence in assessing patients.[10]

The findings of this study suggest that the CAH model was successful in providing a dedicated pathway for the assessment of patients with COVID symptoms and in mitigating the risk of infection associated with being assessed within the established health facilities, that is, general practice clinics or EDs. Patients and staff articulated the many positive aspects of the model, with particular emphasis on feelings of safety and being afforded the time to provide/receive good quality care. The shared sense of safety highlights the success of the clinical protocols in overcoming the initial fear associated with COVID-19. The overwhelmingly positive experiences of staff who participated in this study do not correspond with previous research which found a negative psychological impact of the COVID-19 pandemic on frontline workers and healthcare professionals.[13,21] This may be due to specialized

training, efficient protocols in place and a sense of physical safety maintained by the teams' diligence in complying with all recommended infection control measures.

To make sense of the success of the CAH model, it is useful to draw on integrated theories of urgent care. Turnbull et al.[22] in their conceptual model of urgent care sense-making and care seeking, outline three distinct types of 'work' or thinking processes that patients typically navigate before accessing urgent care: (i) 'illness work'—When people make sense of illness by interpreting the severity of symptoms, assessing risks and making decisions about accessing services; (ii) 'moral work'—Work undertaken to present as an appropriate, legitimate or responsible user of healthcare—'a credible patient'; and (iii) 'Navigation work'—Identifying and making sense of the range of services on offer and how to access healthcare services. Illness work is, to a large extent, influenced by the level of health literacy the patient has and their knowledge about patterns of symptomatology and indicators of risk. During the COVID-19 pandemic in Ireland, extensive efforts were undertaken through public campaigns to ensure that the general population was fully informed of the signs and symptoms of COVID-19 and clear instructions were provided on the level of severity at which one should seek healthcare and from where one such seek such care. It could therefore be argued that patients had to perform less illness, moral and navigation work to access the CAHs than would have been the case to access EDs. Thus resulting in more efficient utilization of the CAH services.

This efficiency is evident in patients' positive comments about the comprehensive and holistic care they received in a timely manner, as well as the easy accessibility of the clinics and the friendly attitudes of staff. For many, the experience contrasted sharply with the busy, overcrowded nature of EDs (often serving as a point of access for patients that could be more effectively treated in other settings), where staff has little time to spend with individual patients due to workload pressures, and where patients may endure long waiting times in less than ideal conditions. Whilst there is a clear association between staff stress, burnout and poor quality of care measured through patient safety errors,[23] the impact of relieving stress on staff and the potential impact on patient care is not as well researched. What this study demonstrates is that when staff is working in a well-managed environment, where they feel adequately prepared for their role and are afforded the time to deliver care, the result is patients who feel listened to and respected, and whose experience is one of receiving high quality, holistic care. In addition, the findings make a strong argument for providing more information to the general public on what, when, and how to access ED services.

The multidisciplinary nature of the hubs also elicited positive sentiments towards the working environment. The pandemic has shown that, despite the pressure, there has been great resilience and solidarity in healthcare staff[13] working in new environments with new colleagues. A surprisingly positive finding from this study was how novel and well-received multidisciplinary working was by hub staff. The hierarchical culture that is typical in many healthcare settings, seems to have been suspended in these settings with many staff noting the opportunity to contribute to problem-solving and decision-making in a manner that would not be typical in their usual roles. A study of healthcare staff narratives of implementing change during the COVID-19 pandemic found that the focus was on completing the task, and the position in the hierarchy was less relevant as all team members relied on each other for clinical and emotional support.[15] This absence of hierarchy and camaraderie in the teams led to staff feeling supported, and encouraged to work and learn together as well as building professional relationships along the way. Another study where healthcare staff was surveyed on their experiences during the COVID-19 pandemic, reported that the interprofessional collaboration enabled an atmosphere of psychological safety and creativity, where ideas and innovations were actively sought and developed collectively.[24] This sense of psychological safety is also evident in the CAHs as the staff was able to express opinions and there was a levelling of traditional hierarchies. These unsolicited positive comments on collaboration, multidisciplinary working and levelling of hierarchies do, however, raise the question as to why this way of working is not by now the norm in healthcare, particularly given the strong emphasis on integration of care and primary care teams and networks in Ireland's health strategy.[25]

Despite the positive outlook of the CAHs, several issues were brought to light, primarily around matching resources with the demand. Nearly all staff highlighted that the CAHs were over-resourced, particularly at the beginning of the pandemic where COVID-19 cases were lower than expected. Staff numbers were then reduced but the staff was

sometimes left with little work due to low patient throughput and the limited role CAHs played in COVID patient care, that is, they had limited ability to conduct diagnostic work other than assessing COVID symptoms. For further development of the CAHs to be a viable option, they would need to be given more powers to do more detailed assessments or provide care or treat patients. The staff view generally was that their skills and time could have been better utilized. Greater awareness of the existence and function of the CAHs amongst GPs would also assist in expanding their use. The scope of practice and the cost-effectiveness of this model of service is something that requires further research to understand if it does have a place in the healthcare system.

Staff and patients were in agreement that this model could be used to treat patients with minor conditions or chronic illnesses who may need to be seen by an array of professionals or those who need more specialized care than a GP practice can give, but who are not so unwell as to require emergency care. The complexity of COVID-19 has highlighted that multidisciplinary teams may be required to provide care to patients with long-COVID symptoms. Participants identified the lack of follow-up care for these patients. Statistics from the UK Office for National Statistics (ONS) suggest that 13.7% continued to experience symptoms for at least 12 weeks.[26] However, a retrospective study of 273,618 patients with COVID-19 found that in the 6 months after a COVID-19 diagnosis, 57% had at least one feature of long-COVID recorded.[25] In the 90- to 180-day ‘long’ phase postdiagnosis, over 36% had a long-COVID feature recorded. Additionally, the study found that the incidence of ‘any’ long-COVID feature varied from 46% in the 10- to 21-year age group, to 61% in the over 65s.[27] The CAHs may therefore provide the basis for a model for long-COVID care. One such integrated multidisciplinary model of care for post-COVID pneumonia hospitalized patients outlines the patient's physical and psychological support needs.[28] Another study co-designed a potential long-COVID pathway with healthcare professionals and long-COVID patients demonstrating that this patient group's complex care needs require a holistic ‘one-stop-shop’ with multiple disciplines' expertise.[29] There is a need for continuation of care for these patients, particularly those who were not hospitalized, to ensure they do not fall through the service gaps.

Limitations

There were a number of limitations to this study. Engaging with busy healthcare professionals throughout peaks and dips in the pandemic proved difficult and flexible remote meeting arrangements and sustained open dialogue across sites were used to facilitate collaboration and reduce the burden on the team. This inevitably caused delays in the data collection. Our ethics permissions required that recruitment of patient participants to the study needed to be conducted through the healthcare facilities to avoid sharing patient contact information with the research team. Communicating with patients about the study placed an additional burden on staff and because of this in Region B, the decision was taken to reduce the sample size to 100 patients. An additional limitation was that patients were contacted via posted letters, as email addresses were not recorded for all patients. These limitations may have negatively impacted the response rate. The research team was not allowed to send reminders because of the aforementioned ethics stipulation regarding patient contact. An additional constraint on the research team's ability to expand the sample size was the short time frame for the study, initially 7 months but subsequently extended by 2 months. The resultant small sample size for the survey results is a significant limitation of the study. For some patients, there was several months time gap between attending the clinic and completing the study, so responses may be influenced by potential recall bias. It must also be noted that this study was conducted on a sample of CAHs and does not represent the national picture. Most of these CAHs were relatively quiet throughout the pandemic, other CAHs may have different experiences based on demand and variations in training as this was not standardized nationally.

CONCLUSION

This study provides an understanding of the challenges of delivering care to COVID-19 patients, mitigating the risk of cross-infection whilst providing a service without access to diagnostic capabilities. It demonstrates what is important to patients who contract an infectious disease about which they have limited knowledge. It also highlights the importance for staff of working in a safe environment and having the knowledge to deliver quality care to their patient cohort. An unexpected outcome of this study is its demonstration of the true value of effective

multidisciplinary working, not only for the staff who were deployed to this service but also for the patients in receipt of care in these hubs. This multidisciplinary patient-centred service provides evidence of the benefits of such models of care, and important learnings for their implementation. This has relevance to proposed healthcare programmes pertaining to long-covid, chronic disease and integrated care in a community setting. However, further research is needed to assess the cost-effectiveness of this model of service.

AUTHOR CONTRIBUTIONS

Eilish McAuliffe contributed to the conceptualization of study, funding acquisition, data collection, data analysis and drafting and reviewing the paper. Sophie Mulcahy Symmons collected the data and contributed to the data analysis and drafting of the paper. Lisa Rogers contributed to the data analysis. Ciara Conlon contributed to the data analysis and reviewed the paper. Aoife De Brún contributed to funding acquisition, reviewed the paper. Marese Mannion assisted with recruitment of participants and reviewed the paper. Niamh Keane assisted with recruitment of participants and reviewed the paper. Liam Glynn contributed to the study design, funding acquisition and reviewed the paper. Joseph Ryan assisted with recruitment of participants and reviewed the paper. Diarmuid Quinlan contributed to the data collection and reviewed the paper.

ACKNOWLEDGEMENTS

This project was funded by Science Foundation Ireland, Enterprise Ireland, and IDA Ireland's COVID-19 Rapid Response Funding Call [SFI20-0221].

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Participant consent was obtained to utilize their data for this study's purpose only. The data therefore cannot be made available to other researchers for use.

DETAILS

Subject:	Pandemics; Health care access; Emergency medical care; Patient-centered care; Clinical training; Professional attitudes; Signs and symptoms; Patients; Hospitals; Remote monitoring; Value; COVID-19; Interviews; Data collection; Access; Holistic approach; Infections; Consent; Hubs; Co-design; Health care; Chronic illnesses; Patient satisfaction; Medical research; Response rates; Primary care; Surveys; Interdisciplinary aspects; Polls &surveys; Critical care; Coronaviruses; Hospitalization; Health services; Health risks; Disease transmission
Location:	Ireland
Identifier / keyword:	community assessment hubs; coronavirus; COVID-19; patient experience; staff experience
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	119-131
Publication year:	2023

Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-05
Milestone dates:	2022-06-14 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2021-12-21 (manuscriptReceived); 2022-11-05 (publishedOnlineEarlyUnpaginated); 2022-09-06 (manuscriptAccepted)
Publication history :	
First posting date:	05 Nov 2022
DOI:	https://doi.org/10.1111/hex.13603
ProQuest document ID:	2767101976
Document URL:	https://www.proquest.com/scholarly-journals/covid-19-community-assessment-hubs-ireland-study/docview/2767101976/se-2?accountid=211160
Copyright:	
Last updated:	2023-08-01
Database:	Public Health Database,Publicly Available Content Database

‘It feels like my metabolism has shut down’. Negotiating interactional roles and epistemic positions in a primary care consultation

Lian, Olaug S [1]
; Nettleton, Sarah [2] ; Grange, Huw [1] ; Dowrick, Christopher [3] [1] Department of Community Medicine, Faculty of Health Sciences, UiT—The Arctic University of Norway, Tromsø, Norway [2] Department of Sociology, University of York, York, UK [3] Department of Primary Care and Mental Health, University of Liverpool, Liverpool, UK

ABSTRACT (ENGLISH)

Introduction
Our aim is to explore the ways in which a patient and a general practitioner (GP) negotiate knowledge claims stemming from different epistemic domains while dealing with a mismatch between experiential and biomedical knowledge during the clinical consultation. We interpret their interaction in relation to the sociocultural context in which their negotiation is embedded and identify factors facilitating their successful negotiation (a medical error is avoided).
Methods
Based on a narrative analysis of a verbatim transcript of a complete naturally occurring primary care consultation, we explore the moment-to-moment unfolding of talk between the patient and the GP (two women).
Findings
The patient experiences symptoms of what she interprets as a thyroid condition, and indirectly asks for medication. She presents her case by drawing on experiential knowledge (‘it feels like my metabolism has shut down’) and biomedical knowledge (while suggesting a diagnosis and a diagnostic test). The GP informs her that her thyroid blood tests are normal and uses biomedical knowledge to explain why she turns down the patient's request. This stages a potential conflict between the patient's embodied experiential knowledge and the doctor's biomedical knowledge. However, during their encounter, the patient and the GP manage to co-construct the patient's illness story and make shared decisions about further actions.
Conclusion
The transition from potential conflict to consensus is a result of the mutual efforts of two parties: a patient who persistently claims experiential as well as biomedical knowledge while at the same time deferring to the GP's professional knowledge, and a GP who maintains her epistemic authority while also acknowledging the patient's experiential and biomedical knowledge.
Patient and Public Contribution
Our empirical data are sourced from a data archive and patients were not involved in the design or conduct of the study, but our study is based on a naturally occurring clinical consultation with a patient.

FULL TEXT

INTRODUCTION

What furnishes a person's status as a knower? Personal experience for one. There are events, activities and sensations ‘to which the experiencer has primary, sole and definitive epistemic access’.[1] When we communicate these experiences to others, it is a testimony based on first-hand knowledge that testifies to the truth of the matter. This means that when patients communicate their illness experiences to doctors in a clinical setting, doctors gain

access to ‘testimonially based knowledge’.[2] Together with knowledge derived from systematic research and clinical experience, patients' testimonial experiential knowledge constitutes a key component in clinical practice.

In the wake of the increasing emphasis on patient-centred care and shared decision-making (SDM), it has become increasingly important for doctors to be attentive to patients' experience-based knowledge. The SDM model, which is founded on a collaborative doctor-patient relationship and a two-way exchange of knowledge, means providing patients with decision-making influence.[3] To give patients meaningful decision-making influence, doctors need to be attentive to patients' knowledge and normative stances, and supply sufficient information for patients to be able to make decisions about their healthcare.[4] Acknowledging patients' expertise, whether experiential or biomedical, is a key prerequisite.

SDM is constrained by the different *institutional* roles and knowledge positions that patients and doctors occupy in clinical consultation.[5] The medical encounter brings together two ‘territories of knowledge’[1]: the patient's embodied experience and the doctor's biomedical knowledge and technical expertise. The former is subordinated to the latter. It means that while interacting in institutional settings, they do so within a context of epistemic asymmetry.[6–9] For each epistemic domain, actors occupy a position on a gradient from knowing to not-knowing, which they implicitly mark by pointing to ‘presupposed access to knowledge or the rights to knowledge’.[10] Their institutional epistemic *positions* must be distinguished from their epistemic *stances*, which concerns ‘the moment-by-moment expression of these relationships, as managed through the design of turns at talk’.[11] Positions are fixed, stances are not. People often align their epistemic stance to their epistemic position, but such congruence is not inevitable.[11] The epistemic primacy of biomedical over experiential knowledge in the medical system severely constrains patients' ability to exercise choice,[12] and ‘patients' testimonies are often dismissed as irrelevant, confused, too emotional, unhelpful, or time-consuming’.[6]

Our aim in this study is to explore the ways in which a patient and a GP negotiate knowledge claims stemming from different epistemic domains while dealing with a mismatch between experiential and biomedical knowledge through a case study of one complete naturally occurring clinical consultation. Our analysis involves capturing the ways in which the two parties mark their epistemic positions and stances, and interpreting their knowledge claims in relation to the sociocultural context in which their interaction is embedded. After narratively exploring the moment-to-moment unfolding of the consultation, from beginning to end, we reflect on how their interaction might have contributed to solving the potential conflict it entails.

Reducing the epistemic divide between patients and healthcare providers is widely advocated, but SDM is difficult to achieve.[13] Previous research drawing on naturally occurring consultations points to various ways in which both patients and health professionals are invested in maintaining differences in epistemic authority.[14] Doctors may limit patients' epistemic access to medical knowledge, for example by providing only interpretations of test results rather than the results themselves,[15,16] or disguise power by generating *perceptions* of choice.[17] Patients have been found to deny and downgrade their own knowledge during medical encounters, for example, through the use of epistemic disclaimers like ‘I don't know’,[10,18] nonconstraining expressions of caution and uncertainty like ‘I was wondering’ and ‘I'm not sure’,[9,19] or attributions to third parties like ‘my husband thought it could be…’.[9,20] Particularly in the final decision-making phases of consultations, patients typically defer to doctors' expertise regardless of their own level of understanding.[18,21–23] Patients who display knowledge in ways that disrupt or resist epistemic asymmetry may be treated as problematic.[24]

Contrary to previous research, we are not focusing on the negative (i.e., epistemic asymmetry) but on the positive, in the sense that we explore how constructive negotiations of knowledge claims across epistemic domains might be successfully achieved in a clinical consultation. Because the onus of achieving SDM is usually placed on healthcare professionals,[4] patients' role is easily overlooked, and their engagement remains underinvestigated.[25] We, therefore, emphasize the interactional aspect, and the role of the patient in the decision-making process.

METHODS

This is a case study based on a verbatim transcription of a complete naturally occurring primary care consultation, sourced from the *One in a Million: Primary Care Consultations Archive* (Table 1). We chose the case-study design

because of its potential for generating detailed knowledge about complex processes as they occur in their natural setting.[28]

Table 1 One in a Million: Primary Care Consultations Archive

Data archive (*n* = 300)	A prospective observational study containing an initial data set archived at the data repository of the University of Bristol, UK. The data set includes 327 film- or audio-recorded and verbatim transcribed naturally occurring GP consultations collected between 2014 and 2015 in 12 National Health Service (NHS) practices in and around the City of Bristol. A total of 300 patients gave informed written consent for their data to be accessed and reused by other researchers, subject to specific ethical approval. The data set also includes patient records; longitudinal patient pre- and postconsultation survey data; sociodemographic data of patients and GPs and GP practice data. The *One in a Million* study was funded by the National Institute for Health Research (NIHR) School for Primary Care Research (208) and the South West GP Trust, and received ethics approval from South West—Central Bristol Research Ethics Committee (ref.: 14/SW/0112).[26,27]
Our sample (*n* = 212)	All consultations classified as endocrine/metabolic, neurological, musculoskeletal, psychological, digestive, cardiovascular and general. *Patients*: 135 women and 77 men aged 18–96 (average = 51 years). *GPs*: 13 women and 10 men aged 32–62 (average = 46 years), divided between 12 different practices, who conducted 7–14 consultations each. *Consultations*: 101 consultations were performed with what patients defined as their 'usual' GP, 122 were conducted by women GPs and 86 were woman-to-woman consultations. All 212 consultations were systematically coded in NVivo (version 12.4) based on a codebook with data-grounded themes (master-themes and subthemes) and semantic codes, which means we stayed close to the language of participants and coded what was overtly and explicitly expressed. The codebook, which was generated after coding 25 randomly chosen transcripts (by the first author), was developed collaboratively by the research team, and the final coding was done by the first author and a researcher. We have previously published studies based on larger samples of the data set.[15,18,19]

Abbreviation: GP, general practitioner.

Data material

To identify a consultation where the patient was actively engaged in decision-making processes, we identified all patients who proposed both interpretations of their condition *and* treatment options (Table 2). After reading all 16 identified consultations, we chose to proceed with a case where the two parties negotiated epistemic positions throughout the whole consultation (Table 3), and therefore was most likely to maximize 'what we can learn'.[30] Our patient is averagely engaged in terms of *describing* symptoms and action taken to manage her illness, but above averagely engaged when it comes to making *suggestions* about diagnosis and treatment (Table 2). This is consistent with the case study methodology, where it is common to study an 'unusual' case[31] 'because of its uniqueness'.[28] We used the patient record and the patient's responses in the pre- and postconsultation surveys as supporting data.

Table 2 Patient utterances: Our case compared to the complete data set (selected variables)

Patients	Number of utterancesa

	Our case (n = 1)	Average (n = 212)	Median (n = 212)	Range (n = 212)
(1) Describe symptoms	14	14	13	0–54
(2) Describe actions taken to manage their illness	9	9	7	0–49
(3) Suggest interpretations of their condition	1	0.3	0	0–10b
(4) Suggest treatment initiatives	4	0.6	0	0–5c
Total	28	24.5	21	3–99d

a
Utterances, or speech acts, implying a speaker temporary ‘taking the floor’, are housed in turns at talk, coupled into an alternation of takers.[29]

b
Scores identified in 45 (21%) of the 212 consultations.

c
Scores identified in 76 (36%) of the 212 consultations.

d
In 16 of the 212 consultations, we identified scores on both numbers 3 and 4.

Table 3 The consultation

Duration	17 min
Contact reason	Endocrine/metabolic
Patient	Woman in her early 50s, who is ‘Unable to work due to illness’. Education: ‘O-levels/CSEs/GCSEs or equivalent’ (lowest level).
Patient survey information	The patient answered ‘Strongly agree’ on the questions ‘I know this doctor very well’ and ‘This doctor knows me as a person’.
GP	Woman in her mid-40s. She is a salaried GP who has worked in her current practice for nearly 9 years. The patient classified her as her ‘usual’ GP.

Abbreviation: GP, general practitioner.

Data analysis

Through this case study, we aim to capture the complexity of a single case and relate its particularities to the institutional setting in which the interaction unfolds. Our first obligation is to understand the presented case. However, because the case represents a social practice in a social institution in which culture is enacted, the case might teach us something about the institution itself. Our study has both an intrinsic and an instrumental approach.[30] During our analysis, we treated the consultation as a narrative[32] and explored the complete transcript in relation to *what* was uttered (content), *how* it was uttered (form) and *by whom* (speaker). Our empirical data consist of a dialogue, where meanings emerge through reciprocal exchange. Every utterance is ‘either a statement establishing

the next speaker's words as a reply, or a reply to what the prior speaker has just established'.[33] To preserve context and meaning, while also capturing the ongoing dynamics of the interactional flow, we only worked with dialogue sequences. Our focus on the interactional dynamics is in keeping with Riessman's[32] performative narrative analysis. By quoting long extracts, analysing components in light of the whole, and attending to sequentiality, we respect the integrity of the narrative.

FINDINGS

In our selected case, a woman who has experienced a wide range of symptoms for the last 4–5 years meets her 'usual' general practitioner (GP). During the consultation, they discuss the patient's symptoms, diagnostic alternatives, causal explanations and options for medical examinations and treatments. Here, we present all key dialogue sequences from these negotiations, in chronological order (Table 4). Each dialogue extract is introduced by a quote from the patient. In the final part, we quote from the GPs entry in the patient record.

Table 4 Key content of dialogue extracts

1	The beginning: 'Yes, I'm fine'
2	The patient moves their conversation to menopause-related hormone replacement treatment (HRT): 'The hot flushes are driving me nuts'
3	The patient elaborates on her symptoms: 'I had a look at the Thyroid UK website thing again'
4	The patient indirectly suggests starting a low dose of thyroxine: 'A friend of mine did say, "Ask if you can be put on a low dose of thyroxine"'
5	The patient indicates that she is searching for an explanation of her symptoms: 'I still don't know what the hell is going on'
6	The end: 'It feels like my metabolism has shut down'
7	Information from patient record 10 days after the visit: 'Lab results!'

'Yes, I'm fine'

The GP starts the consultation with a very common introductory question:

GP: How are you doing?

P: Yes.

GP: Alright? […]

P: Yes, I'm fine. First of all was the result for the [jobbie] yes, which I kept forgetting to phone in about. I'm still old-fashioned and expect people to phone me back.

GP: Sorry, yes.

P: No, it's not you, I'm saying when they get the results in, if that is what they used to do.

GP: Yes.

P: About thirty years ago I think they did that still. (Laughs)

GP: So that was normal, it was 2.7.

P: Right.

After first refraining from answering the 'How are you doing?', the patient adds a brief 'I'm fine', before requesting information about a blood-test result. Though she seems keen to get down to business, she side-tracks for a moment by indirectly complaining about not being phoned up and informed about these results (which she downplays by labelling herself as old-fashioned to think like that). Through her clear agenda-setting at the start, she

takes control of the conversation. However, she also intimates lack of biomedical and systems knowledge through the colloquial placeholder ‘jobbie’ (i.e., ‘thing’), and ‘forgetting to phone in’ for the test-result. At this stage, neither GP nor the patient mentions which test(s) was taken, but later on we learn that it was thyroid-stimulating hormone (TSH). Too high or too low TSH-levels indicate that the thyroid is not working correctly. The GP informs the patient that the test result was ‘normal’ and adds the exact figure (2.7), with no further explanation about reference-ranges for ‘normal’ (which is 0.4 to 4.2 micro-units per litre).

‘The hot flushes are driving me nuts’

The patient then abruptly moves their conversation to menopause-related hormone replacement treatment (HRT):

P: Yes, the thyroid results. Oh yes, I think I need to go back onto the HRT, but I didn't want to do the Premarin. When I ticked the Premarin box, he wouldn't let me have them, which I kind of understood, because it's been about two or three months since I have taken any, and the hot flushes are driving me nuts. But I was remembering what you were saying about—and I thought, because this is so bad now.

GP: I mean, with HRT it is weighing up pros and cons, isn't it? The reality is officially now you're five years you should be taking it, it is five years after the average of the menopause. […] For you, of course, there are many benefits with the osteoporosis [component] side of things. So, I think that's a good idea to go back on it.

P: Yes.

While drawing on her own experiences (‘hot flushes are driving me nuts’) as well as the GP's knowledge and authority (‘I was remembering what you were saying’), the patient proposes resuming HRT. She formulates her request indirectly and modifies it through a subjectifying clause (‘I think I need’). After reminding her that there are pros and cons of this treatment, the GP complements the patient's indirect proposal: ‘I think that's a good idea’ (aligning with the patient's ‘I think’).

‘I had a look at the Thyroid UK website thing again’

The patient then continues to describe her symptoms, before moving their conversation back to the thyroid issue:

P: I had a friend from [name of American city] come over that I haven't see for eleven years in August. She is my actress friend. For two days of that—she is only here for a week and for two days of it I just got this killer migraine because I'd done a little bit of walking with her. Oh my God, and she is a yoga instructor, so-

GP: It's frustrating, isn't it?

P: She was trying to help me and stuff, but I can't lift my legs or do anything because of that muscle weakness thing.

GP: Yes.

P: I had a look at the Thyroid UK website thing again because it keeps coming up, and I know that that is coming up normal. So, I thought, ‘Right’, and I saw that they had—I think it's like 45 different symptoms, so I thought, ‘I'm going to write down the ones I've got’, so I did. I've got 32 of them so I thought I'd give them to you so that you've always got them. All of those are still standing.

GP: Yes.

Through the detailed mini-narrative about the visit from her actress and yoga-instructor friend, the patient conveys the impact of her symptoms: she spent two of these days with a ‘killer migraine’ just because she had done ‘a little bit’ of walking with her. The GP responds empathically by acknowledging how frustrating that must have been. Then, the patient downplays her own knowledge position by using placeholders (‘thing’ twice) to refer to muscle weakness and a website (Thyroid UK is a charity). Although she signals not knowing official technical terms, she appears to know very well what she is talking about. By collecting online information, she has learned that thyroid conditions might be associated with about ‘45 different symptoms’, of which she experiences 32. She has written down these 32 symptoms so that her GP can ‘always’ refer to them. She confirms she knows that her test ‘is coming up normal’, but a possible thyroid condition is still something she would like to consider because her symptoms tell her something else, which she seems to rely on more. By mentioning the ‘normal’ test results, she pre-empts a potential objection.

‘A friend of mine did say, “Ask if you can be put on a low dose of thyroxine”’

The patient then moves their conversation to the issue about further actions:

P: The back thing is just getting worse and worse and worse. A friend of mine did say, ‘Ask if you can be put on a low dose of thyroxine just to see if it does make any difference’. I was wondering if that was going to be at all possible, even though I know that is coming back normal.
GP: Yes, I think that's a difficult one actually.
P: Yes, I know.
Here, the patient proposes to be ‘put on’ a low dose of thyroxine, even though her blood-tests are normal. She begins indirectly, attributing the suggestion to a friend and using the downtoner ‘just’. Through this ‘displaced authorship’,[20] she bypasses a direct me-to-you challenge of the GPs role by displacing the responsibility of her requests to a third-party. After quoting her friend, she reformulates the suggestion in her own words, but still expresses it tentatively (‘I was wondering if’). When the GP effectively declines the patient's request, the patient acknowledges the difficulties of her proposal (‘Yes, I know’), but she is not giving up yet.

‘I still don't know what the hell is going on’

The patient continues her line of argument by giving more details about her symptoms, and reflecting on why they occur:
P: Because I'm getting really weird pains now in this area here and here.
GP: Okay.
P: I don't know if that is a problem of an internal thing, or if that is just the pain radiating out even more because it's getting even worse. So, I'm still not—I still don't know what the hell is going on […]
GP: I guess the other thing is that a lot of these symptoms are also associated with lack of oestrogen. So, like, joint stiffness, the muscles.
P: So, all of these have been going on for what? The last four or five years.
GP: You take oestrogen, I know.
P: But I was taking oestrogen.
GP: I know, yes.
P: I was taking it so that's why I know that a lot of these can be—what's the word I'm looking for? Attributed to other illnesses and God knows what else, but it's when you read it.
GP: Shall we just check it again?
P: What?
GP: Your T4.
P: Oh, right, T4. Is that what I had done?
GP: No, TSH.
P: So, I looked at this one that you can do—I know there are the normal ones that you always do.
GP: Then there is one extra. So T4 and TSH we always do. T3 is an extra. We can try and request a T3. Sometimes—I can successfully now request it, the lab don't process it, but if I phone them up, usually they will then go through. Why don't we try that?
P: Can we?
After describing the pain she is experiencing, and stating baldly that she does not know what is causing it (‘what the hell is going on?’), the GP links it to a lack of oestrogen, albeit tentatively (‘I guess’). The patient objects to the GP attributing her symptoms to lack of oestrogen and reminds her that she has already tried it (‘But I was taking oestrogen’; the ‘but’ probably links to the GPs statement about ‘associated with lack of oestrogen’). Based on her experiences of the oestrogen treatment, she claims to know that these symptoms might be related to other illnesses (‘that's why I *know*’). The GP picks up on this and suggests taking further thyroid tests, through a collaborative doctor-patient ‘we’ (‘Why don't we try that?’). This appears to be what the patient wants, most of all. When the GP mentions which tests she wants to do (T4 and TSH), the patient takes a more biomedical stance and says she knows that these tests are ‘the normal ones that you always do’, but she also knows about an additional test (‘this one’) that the GP ‘can do’. The GP, who apparently interprets this as a proposal to include other thyroid tests as well, confirms that ‘there is one extra’ they can do, and adds it to the requisition.

In this vital part of the consultation, the patient manages to plead her case effectively, although she repeatedly expresses that she lacks the proper expertise regarding explanations (‘what the hell is going on’ and ‘God knows’), terminology (‘what's the word I am looking for?’) and blood-tests (‘Is that what I had done?’). However, what she knows and what she claims to know might be two different things. When the patient mentions the test ‘that you can do’, she leaves the GP with one of two options: either offer the patient the extra test or explain why she would not. The GP responds by suggesting that they (again, via the collaborative doctor-patient ‘we’) request additional thyroid tests, which the patient agrees to (although it was effectively her suggestion).

‘It feels like my metabolism has shut down'

The GP then moves to explaining why she would not offer the patient the medication she indirectly asked for:

GP: The issue is if we give you thyroxine, it then can have a knock-on effect and put you into heart failure if you are taking thyroxine when you don't need it.

P: I see, right. I can understand that.

GP: That is the issue, really, because it slightly increases your output so potentially can cause that.

P: So how does it stop the muscle fatigue? How does it help with that? […]

GP: I mean, it's just that your body slows down, so when you become low in thyroxine you come really slow and heavy, you gain weight. […] So as soon as that is too low, the basal metabolic rate goes down, so all your cellular processes are just slowing down.

P: Sure, because that is how I feel.

GP: So, I think that is the theory as to how it causes the fatigue, in the same way as if you take too much, you become very hyper.

P: Yes, yes. Absolutely.

GP: So, I mean, that's the worry, I don't want to give you thyroxine.

P: No, I understand, I understand that.

GP: Unless there is a definite need for it. I mean, but let's check again and see all those three.

P: Yes, of course. I'm also aware that we have only got a short time, but I also did look up—because five years ago when all this started off as well, I had just spent that two years with no sleep. I mean, serious, serious sleep deprivation for two years. So, I did look up online several different sites to see if severe sleep deprivation can trigger hypothyroidism. It says it can do that because it fucks about with your—sorry, with your metabolism. I know that is what has shut down on me. It feels like my metabolism has shut down.

GP: Let's do that, just put on here, ‘Query thyroid disease. T4, TSH and T3 please’.

Throughout the GP's explanations, the patient repeatedly aligns with the GP's stance (‘I can understand that’ and ‘Yes, yes. Absolutely’) and asks for further information (‘How does it help with that?’). While responding to the GP's explanations about ‘metabolic’ and ‘cellular’ processes, the patient claims knowledge by referring to personal experiences (‘that is how I feel’ and ‘it feels like my metabolism has shut down’), and—again—online sources (‘several different sites’). The patient then introduces the medical term ‘hypothyroidism’, which is a diagnosis (meaning the thyroid fails to produce enough thyroid hormone) that the GP has not mentioned, before immediately switching to more informal language (‘it fucks about with your—sorry, with your metabolism’). While drawing on the online information, she presents a possible explanation for why she might have developed this disease (‘severe sleep deprivation can trigger hypothyroidism’). After first concluding that she ‘know[s]’ that her metabolism has ‘shut down’, she quickly reformulates from knowledge to feelings: ‘It feels like my metabolism has shut down’.

‘Lab results!’

The consultation ends with a prescription for a hormone patch to ease menopause symptoms, and a referral for four different blood-tests (T4, TSH and T3 plus an antibody test). Ten days later, the entry in the patient record reads: ‘Lab results! Thyroid autoantibodies [AB], agreed to T4 as AB raised—but very unusual with normal TSH […] make nonurgent appt with GP’, which means that the patient has an autoimmune thyroid condition in need of thyroxine treatment.

DISCUSSION

In the presented consultation, the patient describes experiences of symptoms that she interprets as a thyroid condition, but her interpretation is not supported by biomedical findings (her blood-tests are normal). The mismatch between the patient's embodied experiential knowledge and the doctor's biomedical knowledge stages a potential conflict. The absence of a diagnosis means that the patient is not receiving appropriate medical treatment, which she now seeks professional help to get. For the GP, the mismatch complicates her dual obligation: to acknowledge patients' experiential knowledge, and to make decisions based on the most up-to-date reliable scientific evidence. It is easy to imagine a negative outcome here (thyroid condition going undiagnosed for years) because the experiential is still so often subordinated to the biomedical.

Instead, a medical error is avoided. So, what works in this case?

Balancing experiential and biomedical knowledge

The answer lies in the interaction between the patient and the GP. The contribution from the patient is essential. She states the purpose of her visit in their very first exchange, and she continues to control the agenda-setting until the very end by providing information, asking questions, presenting her views and proposing actions. While building her case, she draws on a range knowledge sources. While describing how she experiences her symptoms and their implications, she treats herself as entitled to experiential knowledge. This is how it *is*. When she claims to be entitled to know because of what she experiences, she marks her epistemic stance. Building on her own symptom descriptions, she presents a candidate diagnosis (hypothyroidism), a possible explanation (long-term sleep deprivation) and a possible course of actions (a supplementary diagnostic test and medical treatments). In addition to her embodied experiences, she draws on several knowledge-sources: (a) what the GP previously said, (b) what other people have said and (c) various online sources.

By claiming biomedical knowledge, the patient shows she has done her research. However, she adapts her knowledge-claims to the recipient[34] by continually talking as if she speaks to a person with epistemic primacy: (1) she downplays her own biomedical knowledge, and marks it as the GP's domain (waiting for the GP to fill in correct terms and correcting herself from ‘know’ to ‘feel’); (2) she modifies her proposals with lexical downtoners[35] (‘just’); (3) she marks her proposals as tentative or subjective by embedding them within other clauses (‘I think’) and (4) she asks for permission (‘I was wondering if that was going to be at all possible’). By doing so, she aligns her epistemic stance to her epistemic position, and acts within an implicit framework in which the decisive decision-making power is placed with the GP. In downplaying her own knowledge, she talks as though she is not allowed epistemic access to biomedical understandings of illness, and not allowed to share decision-making with the GP. This is a remnant of the pre-SDM era that indicates a cultural lag, where new institutional ideals about patient-centred care are not yet internalized by the patient. By not challenging the GP's authoritative medical position, she avoids being confrontational.[10,18,20] This might have been significant for the outcome: doctors do not consider it helpful, and may become annoyed, if patients insist on their preferences and doubt their doctors' recommendations.[36]

Although the patient is careful not to challenge the GP's expertise by aligning her epistemic stance to her subordinate epistemic position, her outright claim to biomedical knowledge is of vital importance for the outcome of the consultation. The most forceful of these statements is seen in extract 3.5, when the patient refers to a blood-test not yet taken that the GP ‘can do’. By making clear that she knows about this test, the GP either has to offer her the test or explain why she would not. Given the patient's experiential testimony about her symptoms and their implications, it would be difficult for the GP not to offer her the test, which eventually leads to a correct diagnosis and appropriate medical treatment.

The GP contributes to the positive result (a medical error is avoided) by allowing the patient to talk and listening attentively to what she says, while also expressing understanding and sympathy. It is potentially difficult to spot the toned down, understated and hedged utterances that the patient makes, but this GP detects them and responds with respect. The importance of understanding that patients are constrained by their institutional position, and detecting and attending to patients' downtoners, is a key lesson of this study. The GP maintains her epistemic authority throughout (e.g., initially rejecting the patient's indirect treatment proposal), but she remains open to the patient's contribution, engages with her proposals despite their indirectness and reconsiders the available evidence based on

the patient's knowledge-claims. When she provides professional opinions based on biomedical knowledge, it is always with an openness to complexity and uncertainty (repeatedly ‘think’ but also ‘guess’; ‘weighing up pros and cons’ and ‘that's a difficult one’). By not closing their debates before the topics are thoroughly discussed, she facilitates patient engagement.

All these communicative aspects are likely to facilitate patient engagement, patient-centred care and SDM. To further enhance our knowledge about such interactional factors, we need more research on positive interactions in naturally occurring consultations, more studies with ‘naturalistic’ designs (in contrast to experimental and hypothetical), and more case study research.

The collaborative and consensus-orientated interaction that we see in the presented consultation is of course not only a result of what happened in the consultation room that particular day. Previous research indicates that when patients meet their ‘usual’ GPs, as our patient does, there may be more opportunities for them to resist epistemic asymmetry.[14] Their apparently open, honest and respectful dialogue indicates mutual trust, which clearly contributes to the positive outcome of their interaction: By combining experiential and biomedical knowledge, the patient and the GP manage to co-construct the patient's illness-story and make mutual decisions about further actions.

Strengths and limitations

Our empirical data give us a unique opportunity to explore doctor–patient interaction in situ. By doing an in-depth analysis of a single case, we are able to explore in detail the moment-to-moment unfolding of a complete consultation as it occurs in its natural setting. Working with observation-data, however, prevents us from asking participants to elaborate their utterances, and our only information about what happens outside the consultation room comes from the patient record and her responses to the pre- and postconsultation surveys. Possible biases in the data relate to recruitment of GPs, who self-selected to take part in the study,[27] and participants might have been influenced by their awareness of being filmed.

CONCLUSION

The presented consultation is indicative of how a patient and a GP who face a mismatch between experiential and biomedical knowledge manage to use a mix of knowledge-sources to co-construct the patient's illness story and share decision-making responsibility. Although both parties largely align their epistemic stance to their epistemic position (one speaking the language of ‘knowing’, the other of ‘feeling’), they manage to merge them: the patient finds her symptoms in the GP's description of hypothyroidism (‘Sure, because that is how I feel’), and the GP takes the patient's experiential and biomedical knowledge seriously enough to consider that the test results received so far may not be telling the whole story (‘let's check again’). The transition from potential conflict to consensus is a result of the mutual efforts of two parties: a patient who persistently claims experiential as well as biomedical knowledge without dismissing the expertise and authority of the GP, and a GP who acknowledges not only the patient's experience-based knowledge but also her biomedical knowledge.

ACKNOWLEDGEMENTS

The *One in a Million* study was funded by the National Institute for Health Research (NIHR) School for Primary Care Research (208) and the South West GP Trust. The research behind this paper did not receive any specific grant from funding agencies in the public, commercial or not-for-profit sectors.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no new data were created or analyzed in this study. The data used in this study are sourced from the One in a Million: Primary care consultations archive (https://www.bristol.ac.uk/primaryhealthcare/researchthemes/one-in-a-million/one-in-a-million/). Restrictions apply to the availability of these data, which were used under license for this study.

ETHICS STATEMENT

The One in a million study received ethics approval from South West—Central Bristol Research Ethics Committee (ref.: 14/SW/0112). All participants (patients and GPs) gave informed written consent for their data to be accessed

and reused by other researchers, subject to specific ethical approval. Our study received ethics approvals from the National Health Service (Research Ethics Committee reference 18/WM/0008; Integrated Research Application System project ID 232578), and Bristol Data Repository clearance from the Data Access Committee. All data were anonymized upon receipt, and there was no contact with study participants.

DETAILS

Subject:	Negotiation; Negotiations; Datasets; Epistemology; Medical diagnosis; Metabolism; Women; Sociocultural factors; Diagnostic tests; Thyroid; Empirical analysis; Primary care; Professional ethics; Asymmetry; Patients; Drugs; Professional knowledge; Health care; Knowledge; Decision making; Shutdowns; Blood tests; Medical research; Archives &records; Family physicians; Critical incidents; Physicians
Business indexing term:	Subject: Professional ethics Physicians
Identifier / keyword:	epistemic position; epistemic stance; general practice; narrative analysis; patient experience; shared decision-making
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	366-375
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article

Publication history :	
Online publication date:	2022-11-16
Milestone dates:	2022-10-07 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-07-20 (manuscriptReceived); 2022-11-16 (publishedOnlineEarlyUnpaginated); 2022-11-06 (manuscriptAccepted)
Publication history :	
First posting date:	16 Nov 2022
DOI:	https://doi.org/10.1111/hex.13666
ProQuest document ID:	2767101972
Document URL:	https://www.proquest.com/scholarly-journals/feels-like-my-metabolism-has-shut-down/docview/2767101972/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the “License”). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-11-08
Database:	Public Health Database,Publicly Available Content Database

Bereaved parents involvement in maternity hospital perinatal death review processes: ‘Nobody even thought to ask us anything’

Helps, Änne [1]
; O'Donoghue, Keelin [2] ; O'Connell, Orla [3] ; Leitao, Sara [4] [1] Department of Obstetrics and Gynaecology, Pregnancy Loss Research Group, University College Cork, Cork, Ireland; Department of Obstetrics and Gynaecology, INFANT Research Centre, University College Cork, Cork, Ireland; Department of Obstetrics and Gynaecology, National Perinatal Epidemiology Centre (NPEC), University College Cork, Cork, Ireland [2] Department of Obstetrics and Gynaecology, Pregnancy Loss Research Group, University College Cork, Cork, Ireland; Department of Obstetrics and Gynaecology, INFANT Research Centre, University College Cork, Cork, Ireland [3] Department of Obstetrics and Gynaecology, Pregnancy Loss Research Group, University College Cork, Cork, Ireland [4] Department of Obstetrics and Gynaecology, Pregnancy Loss Research Group, University College Cork, Cork, Ireland; Department of Obstetrics and Gynaecology, National Perinatal Epidemiology Centre (NPEC), University College Cork, Cork, Ireland

ABSTRACT (ENGLISH)

Introduction

The death of a baby is devastating for parents, families and staff involved. Involving bereaved parents in their baby's care and in the maternity hospital perinatal death review can help parents manage their bereavement and plan for the future. In Ireland, bereaved parents generally have not been involved in this review process. The aim of our study was to assess parents' perception of how they may be appropriately involved in the maternity hospital perinatal death review in ways that benefit them and the review process itself.

Methods

Bereaved parents ($n=20$) in Ireland were invited to take part in semistructured interviews. Thematic analysis was carried out on the interview transcripts.

Results

Four main themes were identified based on the participants' views and opinions on how they experienced the review process and how they feel this process may be improved. The themes reflect the journey of the parents through the different stages of the review process: Throughout process; On leaving the hospital; Interaction with the hospital ‘waiting in limbo’; Review itself. Identified subthemes highlighted essential aspects of this process and care provided to parents. For the parents, open, honest communication with staff, as well as having a key hospital contact was essential. Parents wished to provide feedback on their experience and wanted to be included in the review of their baby's death, in a way that was sensitive to their needs and the hospital's schedule.

Conclusion

A respectful, flexible system that allows bereaved parents' involvement in their baby's perinatal death review and is tailored to their needs is essential. A collaborative process between staff and parents can highlight clinical areas in need of change, enhance lessons learned, improve bereavement services and may prevent future perinatal deaths.

Public Contribution

Bereaved parents were interviewed for this study.

FULL TEXT

INTRODUCTION

The death of a baby during pregnancy or shortly after birth is devastating for parents and families and can deeply affect the healthcare staff involved. Unfortunately, some deaths are inevitable (e.g., due to a fatal foetal abnormality) but others may be preventable if significant risk factors are recognized antenatally (i.e., during pregnancy) or intrapartum (i.e., during labour). After a perinatal loss (stillbirth or death within 4 weeks after birth, i.e., neonatal death), parents commonly experience negative psychological symptoms which can persist into subsequent pregnancies.[1] Acknowledging the importance of the deceased baby as an individual and involving the bereaved parents in all aspects of the baby's care (such as washing, dressing and examinations if appropriate) can help the parents process their bereavement and plan for the future.[2–4]

The purpose of local child death reviews, like the ones carried out after a perinatal death (stillbirth or neonatal death) in maternity hospitals, is to gather all the information on events relevant to the death, identify contributory factors and cause of death, and to recommend changes to prevent future deaths in maternity hospitals by identifying and addressing modifiable risk factors.[5] The bereaved families should be treated with compassion and be offered the opportunity to be part of the review process.[5,6] A study examining parental involvement in perinatal mortality review processes in six high-income countries found procedures were not established, and only 1 in 20 of the 1104 participating healthcare professionals described a detailed approach to parental engagement in reviews.[7]

In the United Kingdom, the PARENTS 1 and PARENTS 2 studies examined how bereaved parents want to be involved in the local perinatal review process and how this can be achieved.[2,8–10] The PARENTS 1 study showed that bereaved parents want to be part of the perinatal review process in a way that is ‘open and transparent, and emphasised the need for an inclusive and positive approach to both medical and emotional aspects of care’.[2,10]

Many benefits of involving parents in reviews were identified, such as the parents providing additional, relevant information to the process; helping the parents to understand events around their baby's death, and encouraging the hospital to learn lessons and change practices accordingly.[8,10] Barriers to parental involvement in reviews mentioned in previous studies included the cost involved and fear of litigation,[7] a language barrier between some bereaved parents and professionals[11] and variations in bereavement care service provision across maternity units.[10]

The Perinatal Mortality Review Tool (UK-PMRT) was launched in 2018 to standardize perinatal mortality reviews across the United Kingdom and to ensure bereaved parents are always involved in the review of their baby's death.[12,13] Specific material is readily available to facilitate parental engagement in reviews using the PMRT.[14]

In Ireland, bereaved parents generally have not been invited to be involved in the perinatal death review, as the current process in place does not facilitate their involvement.[15] Instead, the final results and findings are usually discussed at the parents' follow-up visit with their consultant.[16] Of note, the National Incident Management Framework published in 2018 by the Irish health service stated that families must be informed if a review is going to be carried out and should be given the opportunity to give their perspective of events.[17] However, in Ireland, there is no specific guidance on involving bereaved parents in review processes specifically.

A study from 2019 showed that just over half (58%) of Irish maternity units regularly informed bereaved parents of the local perinatal death review taking place.[18] Furthermore, only 17% of Irish maternity units stated that the final review report was provided to the bereaved parents.[18] A study on 10 inquiry reports relating to perinatal deaths and pregnancy loss services in Irish maternity services stated that only 40% of the inquiries involved all of the affected families.[19]

This study aimed to learn from and with bereaved parents, how they may be appropriately involved in the local maternity hospital perinatal death review process in Ireland in a way that is beneficial to both them and the review process itself.

METHODSRecruitment

Bereaved parents from all regions in the Republic of Ireland were invited to participate in the study. Purposeful sampling was implemented to recruit bereaved parents in collaboration with Clinical Midwife Specialists in Bereavement Care and parent representatives working within Voluntary Organizations supporting bereaved parents. These acted as a liaison to bereaved parents who had experienced a perinatal death (stillbirth or neonatal death), informing these potential participants about the study over the phone or through emails. Inclusion criteria included parents who were over 18 years of age, spoke fluent English, were at least 6 months postperinatal bereavement (stillbirth or neonatal death) and had no more than 6 years since completion of their child's death review. Previous research with bereaved parents showed that 6 months after their bereavement was an acceptable timeframe for parents to be approached about research participation.[20]

Once a parent gave consent to be contacted, they were contacted by email or phone by one of the researchers with a personal invitation to participate in a semistructured interview. Each participating parent was invited to extend the invitation to participate to their partner. Recruitment occurred between October 2020 and March 2021.

Setting

There are 19 maternity units in the Republic of Ireland, which are funded through the Department of Health.[21] The maternity units vary significantly in size and activity; with between 1000 and 9000 babies being born per annum.[21] The majority of births (>90%) in Ireland occur in the hospital setting, under consultant-led care.[21] To maintain anonymity the parents were not asked which hospital their baby was born and/or died in. To ensure representation from all regions in Ireland, the parents were asked which province they lived in.

Interviews

Semistructured interviews were carried out at a time convenient for the parent(s). A topic guide was used with open exploratory questions to encourage a conversational flow and allow participants to express their experiences, views and opinions on how, when and where parents would like to be and can be involved in the local review process in their and their baby's care.

Before recruitment began, a pilot interview was carried out with a parent representative from the local pregnancy

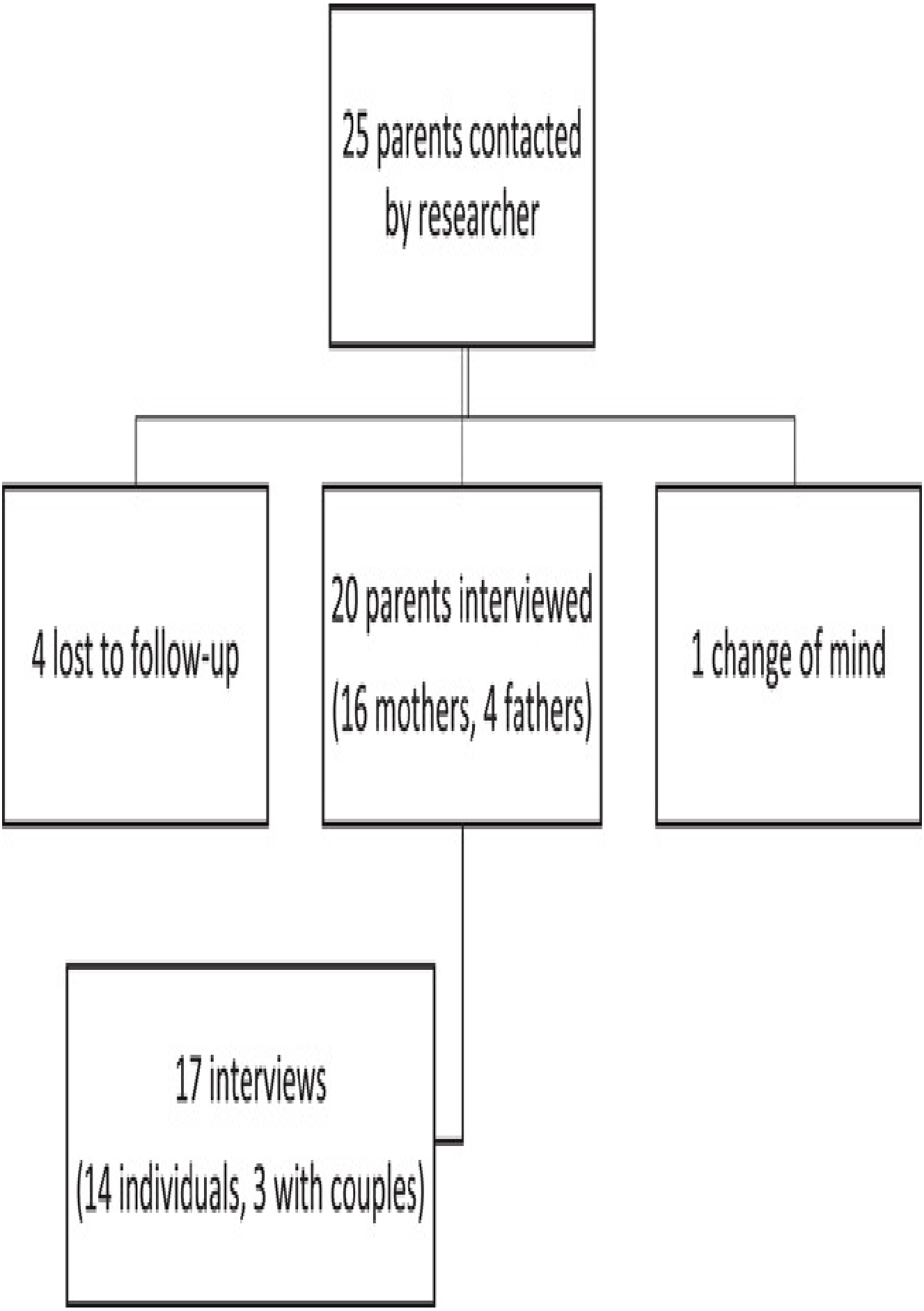
loss research group to check the topic guide for clarity. All terminology was confirmed to be sensitive to the parents' bereavement during the pilot interview. This interview was not recorded and was not included in the analysis.
All parents were offered to have interviews carried out individually or with their partner present (sometimes for support) according to their preferences. The interviews took place between November 2020 and March 2021. Due to the COVID-19 pandemic and national public health guidance, all interviews were carried out remotely using a virtual meeting platform. Specific interview protocols were established to ensure security. The interviews were semistructured, lasted between 27 and 107 min (median, 58 min), were audio-recorded using a Dictaphone and transcribed verbatim.

Analysis

Data collection and analysis were conducted simultaneously. A qualitative research design was used to identify and report patterns in the data and to describe them in rich and meaningful detail.[22] The data analysis methodology was based on the principles of reflexive thematic analysis as described by Braun and Clarke and followed their six-phase process.[22,23] First, all transcripts were anonymized, read and reread by the first author to become familiarized with the data and identify initial codes. Second, open, systematic coding facilitated the researcher to identify codes (and quotes) related to the research objective. Six of the interviews were read and coded independently by two of the other authors (three each). The three researchers with the aid of thematic maps discussed, reviewed and grouped the initial codes to reach a consensus and actively generate the main themes and related subthemes. The transcripts were then re-examined by the first author to ensure all relevant and poignant data extracts were included and fitted within the generated themes and subthemes. Two of the authors discussed, further developed and refined the themes and subthemes to generate clear definitions and names for each, as well as clarify the overall flow of the analysis of bereaved parents' involvement in hospital reviews. Finally, these themes and subthemes were reviewed and agreed on by all authors. The four final themes are united by a central concept (i.e., the bereaved parents' journey through their review process) and the subthemes share patterns of meaning within each theme.[23] An audit trail of the phases of continuous analysis was kept.

RESULTSParticipants

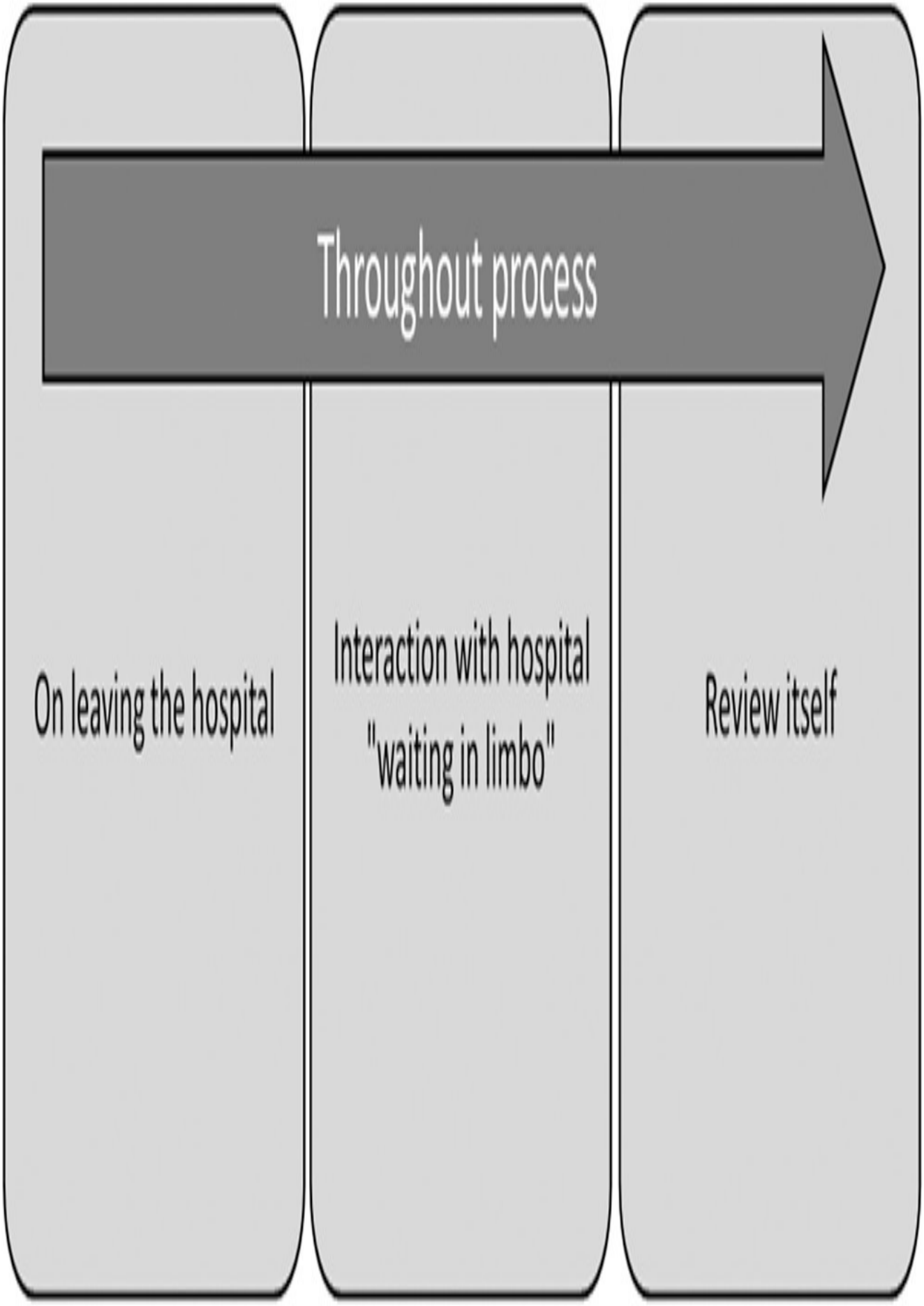
Twenty-five parents were contacted by the researcher, 20 of whom participated in 17 semistructured interviews (Figure 1). In total, 16 mothers and 4 fathers were interviewed. Ten of their babies were stillborn and six died in the neonatal period. It was at least 6 months since their bereavement for all parents (median 3.5 years). There was representation from three of the four provinces in Ireland, as well as from regional and tertiary Irish maternity units.



Enlarge this image.

Results are reported on the lived experiences of the parents and their views on how meaningful engagement by parents in review processes may be achieved, as well as the reasons why this is important.

Four overarching themes were identified from the data (Figure 2). Three of the themes represented different (though at times overlapping) stages of the bereaved parents' journey through the hospital review process, and the fourth theme ‘throughout the process’ contains subthemes that were important and relevant throughout the whole journey (Figure 2).



Enlarge this image.

The 13 subthemes stemming from the four themes are presented in Table 1. Direct quotes from the interviews (indicated by '*Interview*' and the interview number) are used to highlight each theme. Short quotes are present within

the main text (and subheadings); further quotes are presented in Tables 2–5.

Table 1 Themes and subthemes identified from the interviews

Themes	Subthemes
Throughout process ‘an informed approach is a fair approach’	Impact of grief on parents
	A just, compassionate culture with honesty
	Importance of communicating with parents with regular updates
	Support for parents
On leaving the hospital ‘you're just given so much information inside the hospital’	Information given to parents (verbal and written)
	Having a point of contact/key contact
Interaction with hospital ‘waiting in limbo’	The follow-up meeting for parents
	Parents providing feedback to the hospital
Review itself ‘a way to get answers’	Aims of reviews
	Parents' contribution to reviews
	Delivering information to parents
	Inconsistencies for parents with reviews
	Outcomes of the reviews for the hospital and the parents

Table 2 Theme ‘Throughout process’

Subtheme	Quotes
Impact of grief on parents	1. …, when you're leaving the hospital, you're in a kind of a (pause) eh haze. …you're not able to take things in and you have questions afterwards, you know, after, you know, maybe a couple of weeks after that you you, you're kind of you have further questions that you are like, oh I should have asked that …I can't remember, you're told things as well, but you can't remember them because you're, …you're totally (pause) consumed with grief. (*Interview 8*)

	2. You're going up for facts and you come away with em (pause) …kind of worry and you know you're sent down a different, a different road. Thinking I wasn't that before but now I am, you know, and that's grief and trauma too, eh grieving process, it's a very up and down road. So you need the medical side to be consistent. (*Interview 2*)
A just, compassionate culture with honesty	3. But even now every day if I have time, if I go to the hospital or if I am passing the hospital I always have a warm feeling about it because of the way the staff were up there. And even down to the explaining the process of what the pathologist does and the coroner was all done very naturally and there was warmth in it, there was no talking about clinical things and all that kind of stuff. (*Interview 14*)
	4. …she was fantastic I have to say, I got very lucky with the lead on our review, just with her compassionate empathic approach. She was fair to all sides, …she made sure that all sides were appropriately met with fairness and justice. (*Interview 13*)
	5. We had a list of questions we wanted to ask, they weren't hard, they were basic questions about my care, about the systems that failed us. We just wanted the simple answers to those. And we firmly believe if staff could engage, if there was the culture in place for this to go together …where it is protected, where we can all sit in a room, for once we could get the answers that we were looking for. (*Interview 16*)
	6. And you know, obviously if there's, if there was an issue, if they were able to tell us the truth and, and you know from the very beginning obviously that would have been better and then we could have gone away I suppose, and, and try and absorb that. (*Interview 11*)
	7. I suppose my big issue with the whole situation was the lack of information, literally we had to keep asking and asking. It was like trying to get blood from a stone. They wanted to be open and transparent but they wanted to be open and transparent if it suited them. I found the amount of information that was hidden, that was underhanded was an absolute joke. (*Interview 17*)
Importance of communicating with parents	8. So I was always kept informed. It is so important. An informed approach is a fair approach and that needs to be taken with bereaved parents …Like this is the death of a child, it is an ongoing process and all you are doing is sitting at home waiting for some sort of information. And the information that is received and the information that is given to bereaved parents, it is not enough. It is not okay to just send a one sentence email to say, yes they are still working on it. (*Interview 13*)

	9. So I suppose we wanted to know why and how, some people don't want to know that and that is entirely up to them. …all parents should be given the information whether they want to act on that information I suppose is entirely up to them, but they should definitely be given the option. (*Interview 17*)
	10. She rang a few times and then she was texting and stuff just to see how we were getting on. And I suppose, only because I kind of a good rapport built up with (name). I felt I could text her and ask her, you know, questions …And I mean, she would be great, like you'd never be, you would never be waiting long for her or anything like that. (*Interview 7*)
	11. …for me it just seemed like (pause) there hasn't been and there's still not any kind of appetite to hear my views. And like, I kind of feel like even though I've only sent a few emails, but I still kind of feel like I'm almost pestering to try to get things reviewed properly, and like our baby died! You know, I, I don't think I should be the one to have to keep following up, to try to make things be done properly …Like you know, obviously, if I wasn't trying, there'd be zero interaction. And even when I am really really trying, it's slow and it doesn't feel like that people really want to engage or listen really. (*Interview 5*)
Support for parents	12. I think the only time that I really felt I could have used more support was when I was discharged. Because you're going from a circumstance whereby you have midwives around you the whole time looking after you. You know, and you're getting fed at certain times. …you have this really good support bubble, you're wrapped in cotton wool, and then suddenly you're sent out to the real world and you have to stand on your own two feet and you're grieving. (*Interview 4*)
	13. I think the hospital should make contact with the parents and be like, you know, look, the support is here if you need it, like, you can contact us when you're ready like …D'you know, eh would you like to talk about this? Would you, like, give them time, but also give them the option that there is always someone there to talk to. Like when they're ready. (*Interview 6*)
	14. And like, she wasn't pushy. She rang all right. …But after that, then she'd text. So, like, if you didn't feel like talking, that was fine. No, she wasn't em, she wasn't pushy at all, at all. You know, you appreciated the phone call the first week because everything was so new and she was, but …Some people might be more private. I don't know. But I definitely liked (name of bereavement midwife) checking in. (*Interview 7*)
	15. So we didn't really know what to expect or what to do. But in hindsight, only for our bereavement midwife at the time. She guided us through all of that. Our consultant didn't ask to meet with us. (*Interview 3*)

	16. But that support *(from patient advocate)* continued all the way along throughout the years. She would always pop in every now and again and say how are things. Or if there was something I needed to ask about that I was very unsure about myself it was only a matter of picking up the phone. (*Interview 13*)

Table 3 Theme 'On leaving the hospital'

Subtheme	Quotes
Information given to parents (verbal and written)	1. …because sometimes you're just given so much information inside in the hospital. I think maybe even a follow up, a call …two or three weeks later, just to kind of nearly check in and see, 'do you want to have the follow up, do you want to have …these are the numbers available, do you want to come and have a meeting?' Here, you know, I think you're just given so much information in the hospital, sometimes you kind half forget, you know, there's so much coming at you. (*Interview 8*)
	2. I think it should be arranged before you leave the hospital, just to say that this is coming down the line. The results will come back. They may show something or they may not show something. Em would you, I mean, would you like to meet with us? (*Interview 3*)
	3. There must be some two or three steps that could be, could be time-lined and person specific and explain, given to the parents on paper to go this is what the hospital will come to you with. (*Interview 2*)
	4. I think there needs to be a booklet developed and that for information, that they are given to families, the coroner process, inquest, investigation, the parents' rights, advocacy support services. But it needs to explain the whole lot, even the terms of reference, as simple as that, I didn't understand enough back then and I know from listening to others that they didn't either. (*Interview 15*)
	5. So I think, you know, have their doctor sit down with them the day they're being discharged and say, look, these are the, you know, supports available to you. And here are the numbers and someone will be in contact with you from these supports, you know, to know if you want to talk or when you're ready, you can reach out and talk yourself. (*Interview 6*)

	6. I would have appreciated that before I left the hospital and I would have appreciated if they had already decided they were going to do a review, a hand out about what this process is. And that as parents you can contact this person at the patient advocacy service and they are there only to support you …And had we had that we would have known what to expect. (*Interview 16*)
Point of contact/key contact	7. And just to, you know, be introduced to each other and say this is your, you know, if you feel that you want to have this contact in the hospital, then, yeah, I think that's important. That's very important I think. (*Interview 11*)
	8. So I suppose like, she was kind of, like if there was anything, I'd probably go to her before somebody else, because (pause) I don't know, I suppose like, you don't really know otherwise who to contact. Em so, yeah, it is important to have a key, you know, probably a single point of contact who maybe could follow up on some things. (*Interview 5*)
	9. Em to have someone, you know, just to be able to, like a key worker or something like that, just so that you're able to talk to someone about the situation and be like, 'OK, what's what's happening with this' or 'how is this gonna go…' (*Interview 6*)
	10. …I suppose, like there are the bereavement midwifes up there. Em so it's, it probably just needs to be a little clearer to the parents though, like em, who is my contact person if I want to follow up on anything that's happened? (*Interview 2*)

Table 4 Theme 'Interaction with hospital "waiting in limbo"'

Subtheme	Quotes
The follow-up meeting for parents	1. I would have liked to have had a meeting sooner after we lost (name of daughter), because as I said, like from the minute she died, I was in overdrive. …Em And I would have loved to have sat down with (name of consultant obstetrician) sooner and just been able to just converse with her about it. (*Interview 4*)
	2. And it was now time to start slowly picking ourselves up a little bit. And moving forward with her. …the time was right for us. And we were ready to meet him …So I think a time frame of maybe 6 to 12 weeks, or definitely 12 weeks post, was a good time for us. (*Interview 3*)

	3. Definitely, em I'm not sure if 6 weeks is long enough. I think em parents need longer, longer to try and process everything …And this just on top of having all the normal hormones that you'd have after having a baby. Em I think maybe a longer space of time before, before that discussion is maybe had. Even if it was another month added, you know. (*Interview 11*)
	4. Em I felt like maybe they should have done it in another ward or another floor …it just, it was just horrible, like I was shaking, my whole body was shaking …Like, it just brought back so many memories. Maybe if they are having their meetings and stuff. Maybe they should be on a different floor or over in (name of hospital) in another room like, you know, rather than going back into the maternity hospital, where you know your baby, you had your baby there…. (*Interview 9*)
	5. Well, I know she said that she felt initially coming back into (name of maternity hospital) we'll be upset you know, that she could set up something outside of the hospital …I'm so glad she set it up in the hospital because the day we went in, we met a nurse that looked after me, we met another midwife. And it really grounded us again, to say that (name of daughter) was real and that it did happen and we did deliver her here in the hospital. So initially put my foot inside the door, I did get upset. …But after that it was quite a safe, comfortable place and it was a safe place to go. (*Interview 3*)
	6. So when I met with him, he checked with me. …So just really lovely. You know, he had offered his time. He was very respectful about me as the grieving parent. So, so he, he basically checked in saying, 'what way would you like to do this? Would you like me to, would you like to ask me questions? Would you like me to run through what happened?' He's like, 'just tell me what you need'. (*Interview 2*)
	7. If you are feeling that in the moment you can, you have someone there to support you, like your partner can feel like that as well, so you kind of need someone, either a family member or someone neutral like a patient advocate or someone there with you, I would think is a good idea. We never went to a meeting in the hospital on our own, ever. (*Interview 14*)
Providing feedback to hospital	8. But like you'd love them to know exactly how good their staff were, you know. Em yeah no, I suppose like there isn't really the opportunity to, to say any of that. Like when you go for, when you're getting the results even like it's, it's, it's very medical you know, you're only talking about results, future pregnancies. Like the last thing you'd be thinking about is being like, oh, 'by the way, I had a great experience, thank you'. (*Interview 7*)

	9. I see that to give feedback would be great. If anything I can do to help other parents going through this and to prevent, I suppose, certain things that happened for us, not that we were met with much negativity to be honest. …Em yes, we would have loved that. Em and I suppose not just a letter but to be met, (pause) and to, to give our side of things or what we were unhappy with, or happy with …Most definitely, I think for moving forward and closure and for grieving, it would be very important on both sides, to get both sides of the story. (*Interview 3*)
	10. They were listening to us. Like we spoke to them for two hours …Em but kind of highlighting all of the things that we felt, you know (pause) possibly could have made a difference. And we haven't really got response on some things and, you know, kind of highlighting the things, the areas where we thought there might have been kind of gaps, em and not just for us. Like just in general, you know, like we were kinda saying, 'look we're not experts, we're not trying to tell anybody what to do, but, this is kind of our experience'. (*Interview 5*)
	11. Like maybe a couple of months down the line, not really straight after, because, you know, like especially if a mother is angry, they're going to say, 'I hate this, I hate that', you know what I'm saying? So, like, give it a couple of months and then, like, you know, phone them up and ask them. Or if they're meeting up with someone on the bereavement team, you know, get the person on the bereavement team to be like, 'look were you happy with the level of care you received and your baby received at this time. And if, if not, how could we change that in the future?'…. (*Interview 6*)
	12. Again I would think around the 12 weeks, you know, let you process everything again, let the hormones settle down …So I think definitely let all that settle after the couple of weeks and then you would be able to speak up. And you would have time to process what has happened as well and speaking with your partner and stuff, he would have picked up things that you mightn't have picked up on. (*Interview 10*)
	13. Maybe even a questionnaire or something because maybe people would be more confident to say things on a questionnaire or an email or something than they would face to face. People might shy away. I would have no problem speaking up for myself but not everybody would …There could be a comments section at the end then if people did want to put in their own little, because obviously everyone's journey was a bit different and their experience. So whether they wanted to express their anger. (*Interview 10*)
	14. I don't know how, like I suppose the, if there was a kind of a follow up meeting that it would, that would be part of that follow-up meeting, you know. Em if the bereavement midwife, whoever it is or whoever meets to go through, to meet to see if you are, you know, how you are doing, to talk to as part of that process, get feedback there, d'you know em. It, through that, that way I think would probably be a good idea. (*Interview 8*)

	15. Because, like, OK, not everybody might be used to doing emails or, you know, sometimes talking on the phone isn't, well some people might find it easier to do it on the phone, other people might do it face to face. So, yeah, I think an option. I think there should be whatever option a parent wants, really. Like I don't think it should be restricted to just, you know, contact this number between these times or something like that. I think em yeah, just an option of different ways to contact somebody would be good. (*Interview 5*)

Table 5 Theme 'Review itself "a way to get answers"'

Subtheme	Quotes
Aims of reviews	1. As a parent I suppose you want to be your child's voice and I think the review process for a parent, as I said, is a way to get answers to something that (a) they don't understand because it is all medical, and (b) it is giving them closure. (*Interview 14*)
	2. And at every single meeting we kept saying that this is about a systems failure, systemic systems failure where improvements could be made, where this was not to happen to another family. And that is what was most important. (*Interview 16*)
	3. I suppose how it came across to us, was almost they were covering themselves. Unfortunately. But obviously, I know it's to assess what has taken place…. (*Interview 2*)
Parents' contribution to reviews	4. Well to get our side of the story, first of all, because it was a very one sided review, they only got what the doctors and nurses involved. So there was no statement from us at all. …and, you know, for the doctors and nurses involved, em you know, obviously they're not going to try and and, em you know, say anything bad about themselves. There are, so they didn't get the full picture …I think mainly just to write down from our side of things what exactly happened and just explain exactly from our side of things. (*Interview 11*)
	5. I know it's not going to be, in most cases medical information, but it's relevant …And like I was trying to (pause) kind of complement my notes as opposed to contradict them, like I was trying to give more information for it to be reviewed properly …And it's like, like I know most parents probably aren't doctors, but I mean, it's not just the kind of soft, emotional side of it that we can give, like a lot of the time it's actual proper information as well…. (*Interview 5*)

	6. But, you know, just ask parents, like, would you like to provide any further information, you know. I don't know …giving a form would work or, you know, give people just an opportunity, at least ask them at some point, you know, would you like to give any more information? Do you have any other information that you'd like to have included in anything? (*Interview 5*)
	7. …I find it easier for writing down information, personally. I know everybody wouldn't be the same, em (pause) maybe to write it down and then, you know, when they have seen that written statement or, and then maybe set up a meeting with everybody involved, then. (*Interview 11*)
	8. And then it's up to the parents, obviously, whether they decide to be part of it or not. And again, that it needs to be very clear and honest, because the parents need to know what they're getting into obviously. …So as long as the parents, whatever the parents are told they're going to be involved in, is what they're involved in. They're very clear what they're agreeing to. (*Interview 2*)
	9. It was months really trying to get all it sorted, so I think as a rule the system should be changed …because it is a traumatic time of your life and not many people want to go over this. Because every time we went over it, it was heart-breaking and it was very hard in the days afterwards we found because it is constantly on your mind. You give your account and then a few days later you weren't right really, we both found it, it was very hard. (*Interview 17*)
Delivering information to parents	10. There was no warning you will get it next week, nothing, there you go, in your in-box. So to say the least that was extremely hurtful and extremely disrespectful to a family. (*Interview 16*)
	11. I think to get the results in the post, if it was me on my own, hormones raging, no baby here at home and to open a letter with the results. I think my (pause) the ground would just opened beneath me again. And it would have just added to my extra grief. The fact that (name of bereavement midwife) phoned me and said, 'would you like to come and collect them and bring somebody with you?' And to meet her personally and just the touch of her hand and just be able to get the results into my hand, helped. It really did help. (*Interview 3*)

	12. Em so for us, initially getting our post-mortem result, we still came home with a lot of weight on our shoulders, whereas the second time round meeting with (name of consultant obstetrician), it was totally different. We came home with our bag was empty. We didn't feel that burden. So I think delivery of the information and how we're met as parents, grieving parents and that our child is acknowledged throughout the meeting. (*Interview 3*)
	13. They gave it to us in the meeting. We had a patient advocate with us and that is when she kinda said, 'one second now we need the room to go through this'. If we didn't have that I don't know would we have got anywhere, would we have got half of what we needed to get out of it. (*Interview 17*)
	14. But I definitely do think that if parents provide feedback, then it should be, you know, reviewed properly and noted and maybe give parents an opportunity to em review the report. And for that not to be a kind of a final report, maybe provide something to feed back into it and then finalize the report or something like that. (*Interview 5*)
	15. …we had to constantly write, after three or four months, guys what stage is this at now? When it is supposed to take the 120 days, to take the length of time it took is just insane for the actual report that we got in the end. (*Interview 16*)
	16. And I will never know the answer to that. That was one thing that really upset me, I really thought that by getting a review I would have all the answers …That was the hardest part of it all I think because when you get a review as a parent you expect all your questions to be answered. Because they tell you that is what it is going to be. (*Interview 14*)
	17. So like these people, they must think that you're never going to look for freedom of information. You're never going to get all your files. You're never going to read all these emails. Like, yes, there's an awful lot of emails but, oh, my baby's dead. So I had time to read them. And as difficult as that was, I've read them. (*Interview 12*)
Inconsistencies for parents with reviews	18. You don't know what's what like, and I was asking his opinion. He basically told me 'oh reviews are not really worthwhile'. And I was like, really? Because I have a lot of unanswered questions here like, and you're here telling me this. (*Interview 12*)

	19. Em it did say patient concerns were noted, but there was no (pause) eh no detail as to what my concern were. Em or how they had been noted or how they had been reviewed at all …So, you know, I'd kind of really tried (pause) to give as much information as we could. And the only reason I was trying is because I wanted …a full review of (name of son) dying. Like, it's nothing to do with anything, it was literally just, I had information to give. I tried my best to give that information. And then even when I was kind of, you know, going out of my way to follow up and provide all the information that I could. That then was just completely ignored, you know, and it's just, it's just really annoying …I just kind of felt like we were just completely taken out of it, even though it was us and our baby who died. …it's like nobody even thought to ask us anything. (*Interview 5*)
Outcomes of the review for the hospital and the parents	20. …it wasn't like that for us in terms of, we know when staff get up that morning it wasn't their intention to not look after you, it was never their intention. They are human, they make mistakes but the biggest thing from mistakes is learning from them. (*Interview 14*)
	21. But then you are looking at where these recommendations go, who is in charge of overseeing that these recommendations will be actually carried through? So it makes an absolute …What is the point in doing these investigations when they go into a drawer basically? (*Interview 16*)
	22. And that was really amazing that, like, I felt like that was because of our little boy, that he inspired change and that would have been a really lovely thing to hear. I can understand how you have to be careful how you say things to parents, because there are people who sue for anything or anything. But I just thought that was very lovely to hear. (*Interview 2*)
	23. And you don't want the same thing to happen to somebody else, so I think if a parent can, you know, knows themselves that they can give a bit more information and know or think at least that maybe that information might help somebody else. Or help, you know, so that another situation of the same or a similar kind of occurrence …that they might be able to stop that happening. So I think a lot of it is down to trying to (pause), do something for their baby because, like our babies have died. There's nothing really that we can do for them now. But I think for most parents, you know, you probably want to do something kind of for them so that it's not gonna happen to somebody else. (*Interview 5*)

Throughout process ‘an informed approach is a fair approach’Impact of grief on parents

Grief and its impact was a core experience that was mentioned by the bereaved parents. The parents expressed how grief has an enormous impact on them (‘grief is a killer’, *Interview 1*) and how it affected them both physically

('we weren't eating properly, we weren't sleeping', *Interview 11*) and mentally ('we were very raw', *Interview 3*). Some described their state of mind like a 'haze' or 'being in a dream' and 'in total shock', 'totally consumed with grief'. Grief further impacted the way the parents were able to absorb the information given to them and communicate with professionals (Table 2, Quotes 1 and 2). The participants felt that this needs to be taken into consideration when interacting and communicating with bereaved parents throughout the review process.

A just, compassionate culture with honesty

A compassionate culture within the hospitals and the supportive manner of professionals helped parents cope with their bereavement. The hospital culture affected how the parents were able to manage their grief and process the events around the birth and death of their baby, as well as navigate the investigations and reviews that followed (Table 2, Quotes 3 and 4). Parents expressed their gratitude when they met kind, supportive staff: 'they were just amazing and I can't put it in words how good they were to us' (*Interview 3*). Whereas those parents that encountered a cold, uncaring environment described the detrimental consequences this had on them: 'I can tell you the disappointment through the whole thing was their care and like, those words. I regularly get night terrors. I relive that whole experience' (*Interview 12*).

Being able to ask questions and get answers ('why did it happen', *Interview 4*) was essential to the parents (Table 2 , Quote 5). The majority of parents expressed how important honesty and openness from professionals were to them from the beginning ('…if there was an issue, if they were able to tell us the truth and, and you know from the very beginning obviously that would have been better…', Table 2, Quote 6) and throughout their bereavement journey. Those that were confronted with dishonesty or were 'kind of pushed aside' said it made them feel 'very confused', 'suspicious' and 'paranoid'. Two parents explained that they felt that openness was on the professionals' terms only ('…literally we had to keep asking and asking. It was like trying to get blood from a stone…', Table 2, Quote 7). Some felt that the current culture in maternity services after a baby dies is to 'deny and defend' and explained that all they wanted was to 'feel safe to sit down and just say the truth' with the staff involved.

Importance of communicating with parents with regular updates

All the parents agreed that communication with regular updates from professionals was extremely important throughout the review process (Table 2, Quote 8). However, they did feel this should be optional and adapted to each parent's needs, as some parents might prefer not to have regular contact (Table 2, Quote 9). A number of parents described how they had an open two-way channel of communication with staff in the hospital and could ask questions any time, an aspect they said was particularly valuable to them (Table 2, Quote 10). A few parents stated that they had difficulties in establishing contact with professionals, describing how they had to take the initiative, 'pursue' contact and meetings. They had to insist on being heard 'at every turn' (Table 2, Quote 11). One mother raised a concern about the parents that may not be adept at navigating these communication challenges: 'what about the parent that isn't willing to do that? Or doesn't even know that that's an option for them?' (*Interview 2*).

Support for parents

The participants also talked about their experiences of support throughout their bereavement journey. Some felt supported throughout this difficult time and through the hospital review process. Others mentioned how this support was lacking and how they felt they 'were forgotten' and felt like 'just another number'. Most mentioned the concept and the need for someone 'checking in' with them, especially after leaving the hospital with the abrupt change from 24 h care to 'nothing' and feeling 'very alone' (Table 2, Quote 12). Those that had a bereavement midwife 'check in' really appreciated this support. A few parents mentioned how this contact should be cautious, and not be insistent ('…She rang all right …But after that, then she'd text. So, like, if you didn't feel like talking, that was fine. No, she wasn't em, she wasn't pushy at all, at all…', Table 2, Quotes 13 and 14). A number of parents experienced and valued this support to include guidance through follow-up meetings and the review process ('…But in hindsight, only for our bereavement midwife at the time. She guided us through all of that…', Table 2, Quotes 15 and 16).

On leaving the hospital 'you're just given so much information inside in the hospital'Information given to parents (verbal and written)

The first stage of parental involvement in the maternity hospital perinatal death review process began on leaving (or

just before) the hospital. The parents spoke about the hospital stay after the birth being about spending time with their baby, making precious memories and their own/their partner's physical recovery. It was on leaving the hospital that they felt the information given to parents regarding what to expect next was ‘so much’ and ‘there's a lot to process already’, for example, the funeral arrangements. A few parents explained that being told a review would take place was welcomed, however, they did not understand at the time what this would entail. Some parents described that providing written material about future meetings and review processes could help avoid this information overload. In addition, they recommended that making a follow-up call to ensure parents had received all relevant information accurately would be useful (Table 3, Quote 1).

The parents were very clear about what type of information was important for bereaved parents to receive as they were leaving the hospital. They felt it would be important for parents to know what happens after they leave the hospital, who will be in touch and what supports are available to them. Further, the participants thought it would be beneficial to receive clear information and timelines on follow-up visits, investigations and reviews, and the options they have regarding these (‘I think it should be arranged before you leave the hospital, just to say that this is coming down the line. The results will come back…’, Table 3, Quotes 2 and 3). Seven parents suggested that, ideally, information should be provided in writing in the form of a booklet or information pack (Table 3, Quote 4). They said this would allow bereaved parents to process the information in their own time and ‘soak things in better’ during the initial period after leaving the hospital. Furthermore, the parents wanted to know what local and national support services (i.e., bereavement support, counselling, voluntary support organizations, patient advocacy) are available to them and not to have ‘go looking’ for these supports (‘…have their doctor sit down with them the day they're being discharged and say, look, these are the, you know, supports available to you. And here are the numbers…’, Table 3 , Quotes 5 and 6).

Having a point of contact/key contact

Having a point of contact in the hospital (and/or a liaison for the review process) was important to the parents, to have ‘a go-to person, just that one link person, just one name’ (Table 3, Quotes 7 and 8). They recommended for this contact keep parents updated on investigations/reviews, be available to answer questions and liaise with other professionals (Table 3, Quote 9). The local bereavement midwife fulfilled this role for many of the parents, but this was not always made clear to them when they were leaving the hospital (‘…it probably just needs to be a little clearer to the parents though, like em, who is my contact person if I want to follow up on anything that's happened?…’, Table 3, Quote 10). Some parents thought it was valuable to meet their key contact in person before leaving the hospital so they would ‘have a face to the person’ and that they would have a familiar person who would establish contact with them (‘that it's not some random person that rings you after’, *Interview 7*). One mother indicated that she believed that the workload was too arduous for a single individual: ‘I'm not saying questions weren't answered or people weren't contacted. They were (pause), but they're counselling, they're liaising, they're contacting. That's too much for one person’ (*Interview 1*).

Interaction with hospital ‘waiting in limbo’The follow-up meeting for parents

Once the parents had left the hospital and their baby's funeral had taken place, they said they were at home ‘wanting to know what happened to your child’. At this stage many felt the follow-up meeting for parents with the consultant would be essential to get some answers (‘waiting in limbo for weeks and weeks and not knowing is terrible’, *Interview 16*) and to dispel some misperceptions (‘I had this tightness in my chest all along because you would feel blame, but after that meeting, I felt a lot better’, *Interview 10*) (Table 4, Quote 1). The timing for this follow-up meeting, according to the parents, needed to be flexible and suit each individual couple, though somewhere between 6 and 12 weeks after the birth was recommended (‘…So I think a time frame of maybe 6 to 12 weeks, or definitely 12 weeks post, was a good time for us…’, Table 4, Quotes 2 and 3). The parents were divided in their opinions regarding the location for the follow-up meeting, some thought it should be away from the maternity hospital, and others felt going back into the maternity hospital was an opportunity to meet the staff that had cared for them and their baby (‘…the day we went in, we met a nurse that looked after me, we met another midwife. And it really grounded us again, to say that [name of daughter] was real…’, Table 4, Quotes 4 and 5). Ultimately, the

consensus was that it should be the parents' choice where they want to attend for their follow-up visit.
The conduct of this visit and the manner of the consultant can have a huge impact on the parents, either positively (‘it was very reassuring’, *Interview 4*) or negatively (‘that meeting with that man did my mental health no good’, *Interview 12*). Those that had a positive experience were grateful, especially when the meeting had been conducted according to their preferences (Table 4, Quote 6). In contrast, one mother described this visit as an ‘opportunity missed from the hospital to keep the relationship going’ (*Interview 16*). Options regarding the follow-up visit that participants felt should be offered to parents included meeting a team of professionals rather than one individual, multiple appointments (‘and leave it up to the individual person then to choose to pursue the appointments or not’, *Interview 4*) and having ‘someone neutral’ present (Table 4, Quote 7).

Parents providing feedback to the hospital

When asking the bereaved parents if they would have liked to provide feedback to the hospital on their own and their baby's care, the majority of the parents said they would have liked to, but very few were given the opportunity to do so (Table 4, Quote 8). Parents felt by giving feedback, both positive and negative, they would be able to give their side of events, highlight gaps and/or excellence in care and ultimately help other parents (‘…to give our side of things or what we were unhappy with, or happy with…’, Table 4, Quotes 9 and 10). Again, the consensus was that the timeframe for providing this feedback should be flexible, but around 6–12 weeks after the birth was deemed appropriate (Table 4, Quotes 11 and 12). Parents thought the invitation to provide feedback to the hospital should be both verbally and in writing, with a follow-up letter or phone-call to say ‘if you want to opt-in, if you want to have a chat, we're more than happy to do that’ (*Interview 1*). Many different ways of giving feedback were mentioned. Some thought writing using a questionnaire/feedback form, via email or a letter was appropriate (Table 4, Quote 13). Others thought verbally, over the phone or face-to-face, as part of the follow-up meeting or a separate meeting, was best (Table 4, Quote 14). Many parents felt different options should be offered, so the bereaved parents themselves may choose how to provide feedback (‘…I think there should be whatever option a parent wants, really. Like I don't think it should be restricted to just, you know, contact this number between these times…’, Table 4, Quote 15).

Review itself ‘a way to get answers’

Eight of the 20 parents were informed of a formal review into their care and their baby's death as it was taking place, one further mother learned of the internal review after it had been completed. The other 11 parents were not aware of any formal review taking place. However, all parents had investigations and/or meetings with professionals to identify the cause of death for their babies and any potential contributory factors. In this section, we discuss all these processes together under the term ‘review’ as for the parents the aim and desired outcomes are the same: ‘to try and piece together what exactly did happen’ and ‘to prevent this happening to anybody else’ (if possible).

Aim of reviews

For the parents, the aim of reviews was to get answers, identify errors, prevent events recurring and possibly give them some closure (Table 5, Quotes 1 and 2). Unfortunately, overall the parents felt that what they experienced in the review process was not consistent with this perception (‘…how it came across to us, was almost they were covering themselves…’, Table 5, Quote 3) but rather that it was done to satisfy a predetermined requirement: ‘I really don't see how what's being done at the moment is in any way useful or meaningful, other than just to say that it's been done’ (*Interview 5*).

Parents' contribution to the review

This was also the case in relation to the parents' contribution to the review process. As one mother put it: ‘I think as a parent the review process will mean very little until a parent's voice is heard a bit louder’ (*Interview 14*). All the parents agreed that their contribution to the review process was ‘relevant’, ‘important’ and ‘has to be treated with credibility’. The parents' reasons for contributing to the review were ‘to get the full picture’, ensure all sides of events are recorded, ‘the chronology’, and ultimately so that lessons can be learned (Table 5, Quotes 4 and 5). Suggestions for involving parents appropriately included an invitation to all (‘input at the start and again before it's finalized, so that you can actually see what's been discussed’, *Interview 5*) and a written statement and/or a meeting (‘…when they have seen that written statement, and then maybe set up a meeting with everybody involved…’, Table 5,

Quotes 6 and 7). The parents were clear that it should be up to the parents themselves to decide whether to contribute to the review or not, and that 'the invite should be there anyway'. And what is offered to the parents is followed through (Table 5, Quote 8). Several participants felt that the current process of involving parents is protracted, one father described how 'the process was so long' and the effect this had on them ('…Because every time we went over it, it was heart-breaking and it was very hard in the days afterwards we found because it is constantly on your mind…', Table 5, Quote 9). For the parents it was essential that the information they provided was 'taken with honesty and listened to' and not 'dismissed' or 'treated as unreliable, uncredible, hearsay'. Some were asked to provide a written statement, as well as attend a meeting for an interview, at times the experience of the review meeting was described as 'traumatic', it was particularly distressing if in the end, they realized their input 'had no impact, it meant nothing'.

Delivering information to parents

The manner of delivering information to parents needs to be clear and compassionate. The parents requested clarity from the beginning regarding when and how results and review reports would be delivered to them. Getting results/reports without prior notice ('out of the blue', *Interview 16*) at home was described as 'disrespectful' ('…There was no warning you will get it next week, nothing, there you go, in your in-box. So to say the least that was extremely hurtful…', Table 5, Quotes 10 and 11). Many parents preferred receiving reports in person, with professionals facilitating the time and space to process the findings in their own time and ask questions, while also acknowledging their child as a person and their grief ('…how we're met as parents, grieving parents and that our child is acknowledged throughout the meeting…', Table 5, Quotes 12 and 13). Some parents stated that bereaved parents need to be offered input to a preliminary review report, rather than being presented with the final version (Table 5, Quote 14). Again, the length of the review process until receiving results/reports were described as 'long' and 'very slow' by several parents ('having that hanging over you for months is torturous', *Interview 7*), especially if there was uncertainty regarding the date of completion (Table 5, Quote 15). A few of the parents did not receive answers to their questions through the review process, and in some instances, freedom of information requests was experienced as necessary to receive missing information (Table 5, Quotes 16 and 17).

Inconsistencies for parents with reviews

The bereaved parents illustrated many inconsistencies with reviews. One mother felt that most parents are not made aware of reviews being started ('most people don't know that there's a hospital review happen[ing]', *Interview 5*), and two sets of parents were actually discouraged from pursuing a review (Table 5, Quote 18). Some of the parents explained how they were not appropriately involved in the review process ('they have to ask you these questions but they don't really want to know the answers', *Interview 17*) and what they said was not included (Table 5, Quote 19). It required significant effort from the parents to have to persistently contact the review team to 'be heard' and for updates ('it takes so much strength and it takes so much energy', *Interview 16*). One mother described how she felt 'shoved off, shoved off' when asking for updates. Another impact of the review process on parents was a 'burden of responsibility' and pressure to ensure recommended changes were implemented in the hospital ('we felt under enormous pressure to make sure that [em] the proper processes were put in place in the hospital to make sure that wasn't gonna happen *(sic)* to anybody else', *Interview 11*). The dread of the same issues with care potentially recurring has led to parents feeling a sense of responsibility to ensure recommendations were progressed, solutions found and changes implemented. This added a significant level of pressure and stress to the anxiety they were already experiencing. They felt this was not fair on them and should not have been their responsibility ('we shouldn't have to do that, we have been through enough, that is not our job', *Interview 16*).

Outcomes of the reviews for the hospital and the parents

Outcomes of the reviews for the hospital and the parents differed but ultimately the one aim for both families and hospitals was to try and prevent further deaths if possible. The outcomes of the reviews for the hospital, the parents specified, should be learning and change ('…They are human, they make mistakes but the biggest thing from mistakes is learning from them…', Table 5, Quote 20). However, several parents experienced their review to be a 'tick-box exercise' and felt that recommendations from the reviews were not implemented (Table 5, Quote 21). The

parents said the review process has the potential to get answers, see positive change and have the acknowledgement that their baby's life mattered, and therefore it can help to bring healing and closure ('…I felt like that was because of our little boy, that he inspired change and that would have been a really lovely thing to hear…', Table 5, Quotes 22 and 23). One mother summarized the potential outcomes of the review process aptly: 'it brings comfort and healing at the end of the day when it is done right, and when they are not done right you just have repetitive hurt' (*Interview 15*).

DISCUSSION

In Ireland, bereaved parents have been infrequently included in local maternity hospitals' perinatal death review processes.[15] In this study, we learned from and with bereaved parents, how they may be appropriately involved in these reviews to aid the review process and have their views heard. From the interviews, it was apparent that meaningful parental involvement needs to be considered as a process and not a once-off meeting where report findings are divulged. Throughout this process, open and clear communication between professionals and the parents is paramount, including unambiguous verbal and written information, as expressed by parents in this study and in line with previous literature.[2,8] Similarly to what has been reported in previous research, the parents in this study stated that it is essential to have a person as a key contact and support so that they know who to contact with concerns and/or questions once they have left the hospital.[8] The bereaved parents stated that they want to give feedback to the hospital and the review process, both positive and negative, as well as receive results and reports in a supportive timely manner. The overarching expression from parents in our study was that parental inclusion in reviews needs to be flexible with realistic options available that are sensitive to parents' needs and state of mind, acknowledging their child, their role as his or her parents and their grief. This reflects the findings from earlier studies in this field.[2]

When bereaved parents are meaningfully included in maternity hospitals' perinatal death reviews, they feel their concerns are heard and their views are appreciated.[8] As expressed in our study and in line with previous research, for parents it is important to understand the circumstances and cause of death of their baby to help to process their grief and potentially plan for future pregnancies.[5,8] Further, parents in the current study explained how they can provide valuable clinical and nonclinical information to the review process, as well as highlight good or deficient aspects of maternity and/or neonatal care. Previous literature has highlighted the importance of parental contribution to reviews and the value of such participation.[2,8,10] Collaboration between staff and bereaved parents can result in learning from events and improve services for other parents as indicated, and potentially prevent other perinatal deaths in the future.[10]

Our findings in the Irish setting mirrored many of the findings of the PARENTS 1 and PARENTS 2 studies in the United Kingdom, adding to the growing evidence of the benefits of involving bereaved families in hospitals' mortality reviews.[2,7–10,24] Like the participants in the PARENTS 2 study, our cohort of parents emphasized the importance of having a point of contact and the need for personal interaction rather than limited written correspondence.[8] Further, the concept of someone 'checking-in' was felt to be important by the participants in this study. Not having a designated person in the hospital for parents to contact with questions is an ongoing problem in some Irish maternity units.[25]

In Ireland, parents should be informed if a review into their care and their baby's death is taking place and if not, the reason for not reviewing a death. This has been stipulated in the 2018 national Incident Management Framework.[17] However, in this study parents were not consistently informed about these reviews. Further, the Incident Framework states that questions from affected persons should be considered as part of the review.[17] Despite this, many of the parents felt their queries or opinions were not considered appropriately. In the PARENTS 2 study, there were mixed reactions to the feedback form developed for parents to complete after the death of their baby.[8] Similarly in our study, the parents felt there should be different options available to provide feedback to the hospital in writing or in person, depending on their preferences.

The UK-PMRT was developed and put into practice to improve the quality of local reviews by incorporating the parents' perspectives and standardizing the review process.[13] Further research to examine the potential of

implementing an electronic review tool like the UK's PMRT in Ireland is warranted, along with identifying barriers and facilitators to its use and uptake in practice. The PARENTS studies highlighted potential challenges to the engagement of bereaved parents in the reviews including the need for endorsement by local management, as well as the provision of extra human resources and support.[10] Further, it may be difficult to balance the timing of the parental engagement to be sensitive to the parents' grief and fit with the hospital's schedule.[8] Many of the parents in this study were upset by the protracted nature of reviews in Ireland currently, with it taking many months or even years to receive postmortem and/or review reports. A previous study by the authors showed that 4 of the 19 Irish maternity units released review reports more than 6 months after the event occurred.[18] Furthermore, recent research examining the timelines in the investigations of 122 stillbirths in Ireland reported the median time from stillbirth to the follow-up meeting with the consultant with the final report was 140 days (ranging from 54 to 579 days).[26] The current system with long delays and/or difficulties for parents to get answers and resolution, does not appear to put bereaved parents at the centre of the review process and may be contributing to delayed or complicated grief reactions of parents and families.[16,27]

Since the publication of the National Standards for Bereavement Care Following Pregnancy Loss and Perinatal Death in 2016, the emphasis has been on providing sensitive and individualized bereavement care in Irish maternity hospitals.[28] Promoting a culture of compassion and honesty is key to this.[28,29] This culture of openness and compassionate bereavement care needs to continue throughout the review process and is not limited to the bereaved parents' stay in the hospital. It is not acceptable to be unaware and insensitive to parents' emotional and bereavement needs, as every interaction with a member of the hospital staff has the potential to have lasting positive or negative effects.[4,16,27,30] Regular multidisciplinary, interactive education on communicating and interacting with bereaved parents for all hospital staff would help to foster a compassionate culture.[30] A workshop (called TEARDROP [Teaching, Excellent, pArent, peRinatal, Deaths-related, inteRactions, tO, Professionals]) has been developed and evaluated in Ireland, and the aim is to expand this training programme nationally.[30] TEARDROP consists of six interactive, multidisciplinary workstations covering areas of the National Bereavement Standards to equip staff with skills to provide optimal bereavement care for parents.[28,30]

Practice and policy implications

The current study provides relevant insight and information which can have relevance to practice and the care provided to bereaved parents. Numerous important pieces of information (e.g., contact details, bereavement supports, information on the coronial system) are currently provided to bereaved parents on discharge from the maternity hospital after the birth/death of their baby. Seven of the parents suggested the development of an information booklet explaining the different aspects of the review process (i.e., key contact, supports available, ways to provide feedback, timelines and possible outcomes including results and reports) to complement existing information given to parents. This would be a simple intervention with a potentially large impact on parents. Further, this information booklet should clearly outline the voluntary support organizations and services available to parents. Realistic timelines for follow-up meetings and review processes, as well as options for receiving information, results and reports, should be clearly stated to parents and adhered to. Standardization of the local perinatal death review processes at the national level (based on the existing Incident Management Framework), including ways of incorporating parents' views and questions, would be helpful and alleviate discrepancies occurring in reviews and experienced by parents. The development of a review tool like the UK-PMRT and adaptation of the available associated support material[14] for the Irish setting may unify review practices further. Regular training sessions for all staff would form part of these standardized practices in Irish maternity hospitals.

A regular system of feedback is important to ensure the meaningful involvement of bereaved parents in review processes is taking place, practices that are sensitive to parents' needs and identify areas in need of further development. Parent experience surveys or a regular audit of parental involvement in maternity hospital perinatal death reviews could provide this feedback.

The findings and implications from this study are transferable to other countries and other mortality reviews, especially child death reviews. Factors affecting the transferability of findings include the availability of specially

trained staff, for example, bereavement specialists and resources, that is, time and facilities.

Strength and limitations

Purposeful sampling was implemented to invite bereaved parents in this study with some potential selection bias as participants who were willing to participate in this research likely being those already engaged with maternity services and/or who had raised a previous concern about the review process to parent representatives working within Voluntary Organizations. Efforts were made to include a representative sample across Ireland and invaluable and relevant views on this matter were provided. Due to the need to maintain the anonymity of participants, it was not possible to analyse the findings taking into consideration or adjusting for sociodemographic or obstetric history. Further research with a wider (and perhaps international) sample would be valuable to understand how different individual and family characteristics can impact bereaved parents' experiences.

Both individuals and couples were interviewed. Similar to other research studies,[31] we found it challenging to recruit fathers to this study with only four taking part, even though the invitation to participate was extended to all partners of those that agreed to be interviewed. Limiting participation to parents who spoke fluent English may have excluded parents from some ethnic minority groups with broader cultural preferences. Due to the COVID-19 pandemic and public health guidance, all interviews were carried out virtually, which facilitated the geographical widespread representation of participants but hindered the personal rapport between interviewer and interviewee. Further, it excluded participants not comfortable with carrying out an interview virtually.

CONCLUSIONS

In this study, parents clearly voiced their concerns with and desire to be included in perinatal mortality reviews. A respectful, compassionate and flexible system, tailored to the needs of parents is essential, however, this is not yet consistently present for all bereaved parents in Ireland during their baby's review process. The involvement of parents in reviews needs to be carefully considered and resourced, as poorly managed engagement has the potential to cause more hurt. Causing upset and emotional harm through disrespectful or dismissive comments or practices at any stage of the review process must discontinue. Hearing parents' voices in open transparent collaboration with the hospital staff respects their baby and their grief. It has the potential to both support their healing process and make real differences for parents and babies in the future.

ACKNOWLEDGEMENTS

We would like to thank all the parents for participating in this study and for sharing their child's story, views and opinions so openly and honestly with us, without them this research would not have been possible. We would also like to thank the parent representative for participating in the pilot interview and all the individuals involved in the recruitment process. Open access funding provided by IReL.

CONFLICT OF INTEREST

The authors declare that there is no conflict of interest.

DATA AVAILABILITY STATEMENT

Data are available on request from the authors.

ETHICS STATEMENT

Ethical approval was received from the Clinical Research Ethics Committee of the Cork Teaching Hospitals, Ref. No. ECM 4 (d) 08/09/2020.

DETAILS

Subject:	Parents &parenting; Bereavement; Death &dying; Infants; Interviews; Mortality; Hospitals; Stillbirth; Families &family life; Pregnancy; Death; Qualitative research; Reviews; Parental death; Babies; Perinatal mortality
Location:	Ireland; United Kingdom--UK

Identifier / keyword:	bereaved parents; maternity hospital death reviews; parents involvement; perinatal death; stillbirth
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	183-198
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-06
Milestone dates:	2022-09-26 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2021-08-28 (manuscriptReceived); 2022-11-06 (publishedOnlineEarlyUnpaginated); 2022-10-19 (manuscriptAccepted)
Publication history :	
First posting date:	06 Nov 2022
DOI:	https://doi.org/10.1111/hex.13645
ProQuest document ID:	2767101967

Experiences of antibiotic use among Brazilian healthcare users: An exploratory study

Zago, Luiz F [1] ; Correa, Juliana S [1] ; da Silva-Brandão, Roberto R [1] ; Fracolli, Lislaine A [1] ; Padoveze, Maria Clara [1] ; de Oliveira, Sandi Michele [2] ; Gloria C. Corboda Currea [3] [1] Department of Nursing in Collective Health, School of Nursing, University of São Paulo, São Paulo, Brazil [2] Section of General Practice, Institute of Public Health, Faculty of Health and Medical Sciences, University of Copenhagen, Copenhagen, Denmark [3] Section of General Practice, Institute of Public Health, Faculty of Health and Medical Sciences, University of Copenhagen, Copenhagen, Denmark; Antimicrobial Research Unit, School of Health Sciences, University of Kwazulu-Natal, Durban, South Africa

ABSTRACT (ENGLISH)

Introduction

This article analyzes experiences of antibiotic use and bacterial infections among Primary Health Care users of the Brazilian Unified Health System (SUS) and the possible implications for antimicrobial resistance (AMR). The aim is to map aspects that shape users' lay knowledge regarding antibiotics use and AMR.

Methods

This is an exploratory study, which consists primarily of individual in-depth interviews with 19 respondents. Recurrent interview topics were coded and analysed according to thematic content analysis.

Results

Our findings show users' lived experiences constitute three dimensions related to users' previous antibiotic use: (1) lay knowledge about medicines; (2) previous bacterial infections and (3) communication during the consultation. Lay knowledge encompasses the users' understanding of how antibiotics work in comparison to other drugs and experimentations they make with medication. Users' narratives about bacterial infections are divided into situations of urinary tract infections and antibiotic treatments for other conditions. Communication during the consultation is mainly characterized by a lack of shared knowledge and trust in the doctor–patient relationship.

Discussion

Users bring together knowledge learned from their own experiences to create the rationale, which shapes how they understand antibiotic use, bacterial infections and medical advice. These experiences are interwoven with information received from healthcare professionals (HPs) on these topics, creating a scenario that goes beyond professional information about antibiotic use. Users have knowledge about medication, antibiotics use and bacterial

infection but do not have room to share it with HP, allowing lived experiences to take precedence over professional information.

Conclusion

Users ascribe symbolic meanings to antibiotics creating a lay knowledge frame, even if this knowledge is not scientifically correct. The personal experiences of bacterial infections and their treatment are also an important source of knowledge about antibiotic use and AMR among users. Users demand from their HPs both trust and willingness to listen to their health narratives and experiences. By considering lay knowledge as part of the assessment of a user's health condition, rather than dismissing it as erroneous and therefore unworthy of attention, HPs may enhance the compliance of users.

Patient or Public Contribution

Patients or community members did not participate in the design stage of the study. Primary Care patients were invited to participate as respondents of in-depth interviews, which were carried out by the first author at a Primary Care Unit (PCU) in the suburb of Campo Limpo, Southern region of São Paulo, Brazil. Patients were interviewed after reading and signing a Free and Informed Consent Form, holding with them a copy of the Form. Among the final activities of the project, a feedback session at the same PCU is planned to report on the results of the study. All respondents will have the opportunity to contribute further information regarding their antibiotic use and exchange knowledge and experiences on antimicrobial resistance.

FULL TEXT

INTRODUCTION

Antibiotic (mis)use is one of the contributing factors to the increase in antimicrobial resistance (AMR) worldwide, a global health threat.[1,2] A major approach to defining context-specific actions to curb the inappropriate use of antibiotics involves knowledge, attitudes and practices (KAP) surveys.[3–6] Biomedical information is a criterion to assess which KAP are ‘appropriate’, reinforcing the prevalent notion that healthcare users have ‘knowledge gaps’ regarding antibiotic use and AMR.[7] The way KAP research is generally developed in analysing AMR and antibiotic use may underestimate the narratives from which the individual's practices and behaviours come and gain meaning. Taking an approach different from the traditional KAP perspective, this article analyzes experiences of antibiotic use and bacterial infections among Primary Health Care users (hereinafter ‘users’) of the Brazilian Unified Health System (SUS) and the possible implications for AMR. The aim is to map aspects that shape lay knowledge regarding antibiotics and AMR at the community level. We rely on Haenssgen et al.[8] by defining ‘lay knowledge’ as the local, nonbiomedical notions of health formed within a specific cultural background. Our approach stresses how lived experiences are intertwined with one's cultural background. This intertwining serves as the context in which attitudes and practices related to antibiotic use and AMR understanding emerge.[9–11] Patients' perspectives of past treatments should be acknowledged to enhance the success of actions to tackle AMR in health services.[12] Analysis from this perspective is crucial to design and implement tailored actions of antimicrobial stewardship planned with community engagement[13] and a social science approach.[14] This exploratory analysis is part of a long-term process which aims to support the implementation of the Brazilian National Action Plan (PAN-BR)[15] to tackle AMR. The article may also broaden the conceptual discussion regarding AMR by raising awareness about community members' perspectives.

To our knowledge, no previous qualitative study focusing on antibiotic use among users of Primary Care in Brazil has been conducted. This study was carried out at a Primary Care Unit (PCU) of the SUS in the area of Campo Limpo, a suburb of São Paulo. The PCU delivers free, public health services to the local population. It has 10 Family Health Strategy (FHS) teams, each comprising one physician (a general practitioner or an FH specialist), one nurse, one nurse technician and five community health workers. Community health workers are responsible for facilitating the users' access to health services and accompanying their health conditions. Each FHS team covers different territories within the surrounding region where the PCU offers health services.

METHODOLOGY

This study is part of a broader, qualitative project using the One Health approach to explore the perspectives of

those involved in the demand and supply of antibiotics ($N = 76$). It examines aspects of antibiotic use at the community level among users; prescription practices among antibiotic prescribers, dispensers and other healthcare professionals (HP) in Primary Care; and the development of AMR policy in Brazil among policymakers, researchers and other stakeholders.[16] The perspectives of the three groups of participants are integrated to provide a holistic view of the social dimensions for tackling AMR in the country. In this article, we focus on the perspectives of 19 users of the SUS, as they are generally underrepresented in biomedical or governance research. The number of participants was defined primarily by the saturation of responses related to the main domains of the study,[17] which was discussed with the research team; also, the COVID-19 pandemic in Brazil imposed restrictions on the face-to-face interactions between the interviewer and the participants. The project included observation of the local environment (general field notes by the first author) combined with in-depth interviews. The aim was to explore closely the societal reality of the community covered by the healthcare service of the PCU.

Selection and invitation of participants

All interviews were conducted in Portuguese by the first author, who self-identified as a cis-male. He holds Master's and PhD degrees in Education and has skills in conducting journalistic and ethnographic interviews as well as experience in supervising qualitative research projects.

Purposeful sampling was used to select the participants, and users covered by the FHS implemented at the PCU. We presented the research scope and methodology to the PCU management team and community health workers of five different FHS teams. The community health workers proposed the names of users filling the criteria described below. The interviewer presented himself as a research assistant of a Brazilian University and invited participants by phone. Those who accepted were interviewed individually and privately at the PCU at a secure and discrete location. Interviews were conducted between August and October 2021. All hygiene rules adopted by the local Secretary of Health were followed to preserve the safety of both the interviewees and the interviewer. Seeking diversity in the participant sample, we sought users from different regions within the territory covered by the PCU, as well as a variety of gender and age. Additional inclusion criteria included (a) age over 18; (b) active registration at the PCU; (c) having attended the PCU at least once in the last 2 years. Due to the last criterion, most respondents were female, as women represent the majority of users within SUS.[18]

Data collection and analysis

This study consists primarily of individual in-depth interviews. We developed a comprehensive interview guide for in-depth interviews before starting data collection. It covered three broad domains: (1) how users understand their health conditions and how they deal with medication (including the features and use of antibiotics); (2) users' relationships with HPs at the PCU and (3) users' understanding of the risk of AMR, and how and by whom that information is communicated, as Appendix show. The interview guide was developed from both questions from previous qualitative studies and from an informational needs analysis of the project's broader international research team. It was not tested before the first interview, but questions were refined throughout the data collection process, as needed. Each interview lasted approximately 1 h. Three participants did not show up at the scheduled time for the interview, ceasing communication with the interviewer without reporting their reasons. There were no repeated interviews with users. The digitally recorded audio was transcribed verbatim in Portuguese by the interviewer. All transcriptions were reviewed by the first author and by the sixth author, who is the scientific coordinator of the project. Excerpts used in this article were translated into English by the first and sixth authors of this article; the latter was a native speaker. Quotes were coded according to thematic content analysis.[19] The first author conducted the line-by-line analyses and original coding of themes derived from the data. The second and third authors collaborated in the creation and refinement of codes. Ongoing discussions on the data and its coding were carried out with the research group throughout the project, ensuring consistency throughout the different areas of the study. Interviews were then reviewed to understand the ways interviewees framed their practices and AMR to develop key themes. These themes were structured separately and then grouped into major clusters, as explored in the results and discussion sections that follow. Microsoft Word was used to highlight relevant excerpts of each theme and subtheme. All relevant excerpts were then extracted to Microsoft Excel, and identified by the corresponding codes.

Participants were coded by number, gender and age (e.g., R14, female, 58). Table 1 illustrates the themes and subthemes from which the interview quotes here presented were selected and categorized.

Table 1 Themes and subthemes derived from the data

Health conditions and antibiotic use	Antibiotics	Understanding of the features of antibiotics Understanding of potential harms of antibiotic use Following medical orientations to take antibiotics
	Other medications	Understanding of the differences between antibiotics and other medications Understanding of the features of other medications Frequency of use of other medications
Relationship with HP	Demand for consultation	Painful symptoms Accident Periodic healthcare consultation Healthcare level of access
	Valuable characteristics of HP during consultation	Educational skill Professional experience Ability to listen
	Information exchange during consultation	Prescription orientations Medication features Potential harms of general medication use
Understanding of the risk of AMR	Information exchanged with HP	Length of treatment and dosage of antibiotic Potential harms of antibiotics Understanding of HP language
	Knowledge gathered through personal experiences	Previous bacterial infections Children's caregiving and administration of antibiotics Shared narratives within the household and the community

	Information gathered on media	Health campaigns TV or radio Social media, internet

Abbreviation: HP, healthcare professional.

Source: Main author.

RESULTS

Respondents self-identified their race in the following way: two as White, three as Black and 14 as mixed-race Brown. Their education level was diverse, with seven having attended elementary school (completed or not), nine reaching high school (completed or not) and three reaching university level (completed or not). Participants were aged 19–62, and most were women (n = 17).

Our findings show the important role that users' experiences play in three dimensions related to antibiotic use: (1) lay knowledge about medicines; (2) previous bacterial infections and (3) communication during the consultation.

Dimension 1: Lay knowledge about medicines

Lay knowledge encompasses the respondents' understanding of how antibiotics work in comparison to other drugs, and the experimentation they do with medication.

In response to our general question about what they know about antibiotics and medication, users affirm an awareness of classes of medication, such as antibiotics, analgesics and anti-inflammatory drugs. Their explanations about differences and applications vary. Some affirm that antibiotics are a stronger type of anti-inflammatory (‘antibiotics, I think [it] is a little bit stronger than anti-inflammatory but with the same properties’, R14, female, 58), or that antibiotics function as anti-inflammatories but are more intense (‘anti-inflammatory works for a deeper inflammation, antibiotics I also believe that should be more or less the same thing’, R17, female, 35). Others view the difference between anti-inflammatories and antibiotics as a dichotomy between topical and oral medication: ‘anti-inflammatory works for what? For inflammation. Antibiotics work for a chronic inflammation which is not external, it is something that should be taken care of from the inside out’ (R10, male, 61).

Additionally, most respondents operate their own symbolic categories of ‘strong’ and ‘weak’ medication according to prior experiences with pain, illness and its symptoms. The faster a medication cures painful symptoms, the stronger they consider it. If they do not perceive any positive effect from a medication, they consider it as ‘weak’. Antibiotics are considered ‘strong’ by most respondents, not only because they experience rapid healing (R15), but also because they must follow specific rules regarding the length of treatment and dosage (R16).

R: Amoxicillin is very strong, amoxicillin, diclofenac, and Dorflex.

I: And why do you consider amoxicillin strong?

R: Because I myself, in my opinion, when I take it, when I have a problem with my body, I quickly get better.

(R15, female, 50)

R: I consider amoxicillin strong because my mother … says we can't use it for [just] anything, right? […] amoxicillin for example, you have to take it for an exact number of days, it is not the same as Doril [a brand of analgesic] or something that we take once and that's it, it has to be scheduled […] it's a rule, so as it has more restrictions, we already give it more importance, so I think it's stronger.

(R16, female, 19)

We asked respondents about their understanding of relevant terms related to AMR. Respondents were not uniform in their understanding of ‘resistance’. For instance, when R19 was first asked about ‘bacterial resistance’, she did not recognize the term. However, ‘antibiotic resistance’ was familiar to her. Her understanding of antibiotic ineffectiveness comes from a deduction made from a conversation within the household on the functioning of dipyrone (anti-inflammatory) as compared to antibiotics.

I: And have you heard about bacterial resistance?

R: No, no.

I: Antibiotic resistance?
R: I think it's when you've taken the same medicine many times and then the medicine no longer has the effect it would […]
I: Do you remember from where you heard about this antibiotic resistance?
R: Once, at home, everyone talking as a family, then I have an aunt, who is crazy for dipyrone, right, then my cousin said to her, 'Mom, stop taking dipyrone, soon it won't have effect on you', then later I deduced that with all medicines it is the same thing.
(R19, female, 20)

Respondents assemble an array of lived experiences to deal with their health problems. As a result, they may not seek professional care if they feel they have resources at home that may treat their condition. Some respondents state they have used medication and chemical products to treat specific diseases based on perceived properties that, in their understanding, have healing potential. Those practices arise from their experiences in using medication and through shared narratives about healing within their network and serve as the basis for the rationale they present in choosing these nonconventional solutions. The first excerpt involves using vaginal cream for an inflamed inner ear, and in the second the respondent uses creolin (an environmental disinfectant) to combat gastritis:

R: I feel like I'm going to get the flu, my ear starts to itch, it gets inflamed, it turns red, […] Then, do you know what I put on? I take the swab, I get vaginal cream, I put it on the swab, I put it inside my ear, can you believe it?
I: But why the vaginal cream?
R: Because the vaginal cream is good, because if we have a wound, if we have an infection, we put it on, right? It eliminates the pain and everything from us. And in the ear it's the same thing, it's an internal place, a place where you can put the medicine, it inflames, we can't see it, it is the same thing when we have a problem in the vagina, we put the medicine, the ointment, to relieve it.
I: How did you discover this?
R: By myself. […] I didn't have to go to the doctor.
(R6, female, 62)

R: Gastritis, you've heard of gastritis, right? So, I know a perfect remedy, good for gastritis […] do you know what creolin is? … a friend of mine … told me that he had gastritis that was turning into ulcers […] then someone told him that he could take creolin […] well, it makes sense because we use creolin here in the big city more as a disinfectant, […] my father used [creolin] to kill infection in animals […] and soon the animals' wounds would heal, so I said 'if it was good, if it would heal those wounds and also ulcers, gastritis is a wound, so if you take it, it will definitely heal'.
(R8, male, 61)

Dimension 2: Previous bacterial infections

Respondents' narratives about bacterial infections can be divided into situations of urinary tract infections (UTIs) and antibiotic treatment for other conditions.

Respondents' knowledge about bacterial diseases comes primarily from UTIs, which may reflect the predominance of female participants in our study. Patterns of antibiotic use are associated with narratives of UTIs that encompass the search for healthcare assistance in hospitals, gender positionality, ageing and difficulties during treatment. Moreover, respondents also highlight memories of frustration when the treatment is ineffective. R7 says she became frustrated because her grandmother was ill, and 'they [HPs] couldn't find the right medicine to fight the bacteria she had. […] the three courses of medication didn't fight the bacteria' (R7, female, 41). In a second case, R11 recounts the story of the mother of her mother's ex-employer, who:

had bacteria in her urine […], and then the doctor couldn't treat it, she did several, and several, and several surgeries and [they] didn't cure it, she was in a wheelchair and that was not cured yet, then I heard that the urinary tract infection bacteria is very strong. […] people talk about the urinary tract infection bacteria.
(R11, female, 35)

In the following excerpt, R14 reveals her frustration with the prolonged need for care and notes that the HPs may

lose track of the treatment history to date. R14's experience also reveals tensions that may arise between users and professionals, which leads to frustration:

R: My mother had a urinary tract infection for a long time, […] every Saturday I would go to the healthcare service with her, and the doctors would prescribe medication, or give medicine, it would be resolved for 2, 3 days and then come back, then one day I arrived at the hospital and said 'look, young man, we need to know what is the bacteria that is making my mother like this, let's make a more complex test to be able to treat the bacteria, right' […] I said 'here, look at the prescriptions, she already took them, and I don't want to come to the doctor with my mother every week, so I want you to ask for an exam, a urine culture for her to be able to treat the bacteria that is leaving her with this pain'.

(R14, female, 58)

Previous antibiotic experiences provide a relevant set of narratives that may influence the way respondents use that medication in their present time. Despite rapid healing, discussed in Dimension 1, harms from antibiotics are also considered:

R: […] the memory I have is when you usually take antibiotics, depending on the antibiotic you are taking, the doctor even says 'don't take them on an empty stomach', the excessive use of antibiotics also harms the teeth, you know, of the child […] from what I have already experienced, being close to people that this happened to, children who were very sick in childhood, the color of their teeth began to change because the drug is very strong.

(R17, female, 35)

R3 mentions that her information about that topic came from her mother but was confirmed by her own experience in taking care of her daughter, who had bronchiolitis as a child.

I: And has a doctor ever told you about the consequences of using antibiotics or not?

R: No. Never. I always knew it causes some harm, right, in children. My daughter, she took a lot of antibiotics when she was young, because she had bronchiolitis, all that stuff, right? […] My mother always said that it ruined my teeth, weakened them, that sort of thing. But not because the doctor told me so.

I: It was from your mother's comments?

R: Yes.

(R3, female, 52)

In presenting experiences of female family members with UTIs, respondents also acknowledge their having witnessed treatment difficulties, harm associated with antibiotics and tensions with HPs.

Dimension 3: Communication during the consultation

Communication during consultation is characterized by a lack of shared knowledge and trust in the doctor-user relationship.

As elements shown in Dimension 2 above suggest, respondents have their own knowledge about medication use, antibiotics and bacterial infections. However, they feel there is no opportunity to share their knowledge during the consultation. HPs communicate basic information when prescribing antibiotics, such as treatment length and dosage, but users report that HPs seldom provide guidance about possible harm from the medication, nor do they ask users what they already know about AMR. It creates a communication gap:

I: What about the consequences of eventually taking [antibiotics?]

R: No, never, he [the doctor] just told me that it was the deadline I had to use until the seventh day. And if necessary, to continue the treatment, it had to be guided by them. That's all.

I: So, information on bacterial resistance, antimicrobial resistance?

R: No, no, never, never spoke about and I never asked either, no.

(R4, female, 43)

One explanation provided for the low quality of information exchange between the users and the HPs is the short time allocated to the consultation (15 min). Another aspect hindering good communication is the shame some users feel when asking questions:

R: Because there are moments when the consultation is very fast, you know? And there are moments when the

doctor also doesn't give us the opportunity to ask, then we ask something, then we feel ashamed to ask the next question.
(R3, female, 52)
Additionally, some respondents reveal their awareness of the high demand for health services and do not want to overstay their allotted time:
R: I know that sometimes the doctor, when he is not so thoughtful, […] when he does not have so much time to talk to the patient, it is not because he doesn't want to, it is because he has to respect the demand for scheduled appointments [at the PCU] because after you there can be someone with a more serious problem, you have to understand that.
(R17, female, 35)
Aspects of the doctor–patient relationship are evoked as relevant to determine whether there will be trust in the HPs' advice. Respondents' perception of the doctor's attitude towards them also plays a decisive role in following (or not) the doctor's advice (R17).
Also, being able to express feelings is important for the respondents, as they feel they ‘know their own bodies’ better than the HP (R10).
I: And do you always follow all the advice the doctors give?
R: So, […] when I feel it's true, yes […] because you know your own body, nobody better than you to know if you're okay or not […] if the doctor doesn't care about you, there are doctors who don't even look at your face, […] he doesn't know what is really going on with you.
(R10, male, 61)
R: Look, I follow [the doctor's orientations], but there are things I don't follow so strictly, it's not that I don't trust in what the doctor is saying, he studied for that […] but sometimes the doctor says some things that are not part of what you're feeling, […] sometimes you can come to a PCU like this one […] and suddenly the doctor is so rude, so gross, [she/he comes with] the prescription in hand and you'll say ‘I'm not going to take this [medication] that he prescribed because I don't know if it is really part of what I'm feeling’.
(R17, female, 35)

DISCUSSION

Our findings illustrate how users build their knowledge from their own experiences, which shape their understanding of antibiotic use, bacterial infections and AMR. We suggest that these experiences are interwoven with the information received from HPs on these topics, indicating that professional information about antibiotic use and its implications shared during the consultation is not the only source of users' ‘lay knowledge’. In line with other scholars,[8–10,12,20–23] our analysis shows that respondents rely on a set of experiences and values embedded in their cultural settings that shape both antibiotic use and knowledge about AMR, and users develop an important sense of autonomy about medication and their own bodies in the intertwining context of health experiences and information gathered within their community network.
Respondents integrate their antibiotic experiences into their knowledge about other drugs, such as analgesics and anti-inflammatories, as other studies have shown.[3,24] They claim that antibiotics are ‘strong’, setting them apart from other drugs, based on the duration, dosage and frequency of treatment.[23] Respondents show they know antibiotics have specific features, as they believe that antibiotics work to cure ‘chronic internal inflammation’ in the body and aid in rapid recovery. These results parallel those of other studies in that users perceive that antibiotics are a special type of drug.[23,25,26]
Despite viewing antibiotics as ‘strong’, some respondents forgo their use, choosing instead alternative practices completely outside standard medical care (e.g., off-label use of vaginal cream, creolin). If these alternative practices seem to work, these experiences reinforce the users' sense of autonomy in dealing with their own health and reliance on lived experiences and shared narratives of healing. These practices indicate a mixed knowledge frame between lay knowledge and biomedical information and suggest cultural entanglements in which antibiotics are intertwined for the respondents.[9] Our study shows that some of these entanglements come from comparisons with

other drugs, previous use of antibiotics and experiences of illness that are shared within households and the community-level network. Because of these practices, respondents' current ideas about the ‘appropriate’ use of medication, like antibiotics, can be resignified through the negotiation between lay knowledge and professional information.[11,20,23]

The issue of gender is important to our article in three ways: first, studies investigating gender, antibiotic use and AMR as articulated themes are scarce.[10,11,27] Second, antibiotics are prescribed in Primary Care more often to women,[11] who are portrayed as having acquired better knowledge about antibiotic use and AMR.[27] Third, the role of women as family caretakers appeared to be relevant in our study. This central role, combined with the high incidence of bacterial infections (particularly to treat UTIs[28]) among women, suggests that a focus on female users provides a relevant dimension to better map the sociocultural context that shapes antibiotic use.[10,11]

Lived experiences with bacterial infections which are not properly treated can lead to tensions in the relationship with HPs. As repeated diagnostic procedures and the use of inadequate antibiotics do not cure a given health problem, there is a feeling of failure and confusion among users (cf. Boiko et al.[29]).

There may be a ‘grey area’[28] in the communication between HPs and users, as neither seems to address differences between medications or their potential harm.[30] This ‘grey area’ may be a result of structural constraints, such as the limited time for sharing information between HPs and users[31] and the high demand for consultations at PCU, but it is also related to the feeling of trust users have regarding the quality of their relationship with HP during the consultation. The users' assessments will influence how their consideration of and degree of compliance with doctors' instructions. In fact, users affirm they often let their experiences take precedence over professional information, because they trust their knowledge about medication, antibiotic use and bacterial infections, and their personal narratives have not been heard by the FHS team. In contrast to other studies,[32] our findings show that users expect the HP to listen to their experiences during consultation rather than providing pieces of information which are disconnected from their day-by-day life.

Our results align with those of Haenssgen et al.,[8] whose approach to the use of antibiotics and AMR explores the lay knowledge and the ‘tales of treatment’ that are shared at the community level. The medical consultation is one moment, among others, in which information on medication use can be exchanged, sometimes competing with narratives, experiences and previous ‘tales of treatment’ of the users. Thus, even if doctor–patient communication is relevant, the information shared is not necessarily the most important for the respondents: information is assembled with other health experiences and can be relativized, ignored or contested.

The respondents' expectation is that the consultation is a moment of proximity with HPs, as other studies also pointed out.[23,33] Once the respondents effectively build a repertoire of experiences about antibiotics use and health issues, their expectation is that HPs will listen to and respond to these experiences.[12,34] Compliance with medical orientation, as the respondents' state, is linked to the trust they have in the doctor, paralleling recent research.[30] Thus, active listening to users' experiences can help HPs identify the specific contexts in which they make decisions on following or not medical instructions.[10]

Trust is renegotiated at every consultation and requires that the HPs acknowledge the users' assessments of ‘their own bodies’. Also, even if the evaluation of medical care is positive, the idea that everyone ‘knows their own body’ prevails among users. ‘Knowing one's own body’, in this sense, is something different from the apprehension of biomedical knowledge (e.g., diagnosis and appropriate treatment); it is about the possibility of expressing what one feels through the body, and how these feelings refer to previous experiences of medication use and illness. For the respondents, shared narratives in the community and lived experiences constitute what they call ‘knowing one's own body’. In this sense, they merge experiences to build a knowledge frame related to medication, antibiotic use and bacterial infections, through which they come to understand AMR. Eventually, they also apply this knowledge frame to negotiate compliance with medical orientations and to ascribe their own meanings to the potential harms of antibiotic use.

One strength of our study is fostering the appreciation of qualitative aspects of antibiotic use in Brazil by continuously exploring three complementary perspectives: users, antibiotic prescribers and dispensers and policy

stakeholders, to comprehend the views of all actors involved in antibiotic use in Brazil from a holistic perspective. It also adds information in qualitative research of themes concerning AMR in the country, where there are scarce studies of this type. Regarding the Brazilian NAP,[15] our findings support the view that acknowledging the role of community members is fundamental to the success of its implementation, as opposed to taking a purely top-down approach to policy development. We further suggest that the resulting interventions (e.g., local health promotion campaigns and tailored, specific educational training for HP of the FHS team) would be more impactful within the community if they incorporate the values of the communities they represent. Finally, our findings may not be generalized to the national level or abroad, although parallels can be found in other contexts.

CONCLUSION

Our study underlines how lived experiences are intertwined with professional information about antibiotics for users in Brazil. We adopt an approach different from most of traditional KAP research, as we investigate narratives of how these attitudes and behaviours are constructed instead of portraying them as single actions, disconnected from the users' contexts. Their sets of experience play an important role in healthcare, sometimes determining noncompliance of medical orientations and allowing unexpected uses of medication. The experiences of bacterial infections and their treatment, shared within the household and the community, are an important source of knowledge about antibiotic use and AMR among users. Users demand from their HPs both trust and willingness to listen to their health narratives and experiences. Still, users recognize the structural constraints that limit their communication with HPs, like the time allotted for each consultation and the burden of the public health system, both of which affect the quality of the information exchange.

Recognition that users have autonomy in deciding whether to follow medical advice or not and acknowledgement that this autonomy is based on users' sets of experiences may contribute to HPs' overall comprehension of users' attitudes and practices regarding their own health and bodies. Ongoing learning of users' experiences and understanding of antibiotic use is a shared responsibility among all HPs and is not restricted to information-sharing in a single consultation. Users ascribe symbolic meanings to antibiotics and learn about AMR through lived experiences of bacterial infections, creating a lay knowledge frame, even if this knowledge is not scientifically correct. By considering lay knowledge as part of the assessment of a user's health condition, rather than dismissing it as erroneous and therefore unworthy of attention, HPs may enhance compliance of users in efforts to tackle AMR.

AUTHOR CONTRIBUTIONS

Luiz F. Zago has conducted all interviews and has them transcribed. Luiz F. Zago, Juliana S. Correa, Roberto R. da Silva-Brandão and Sandi Michele de Oliveira created the interview guide, interpreted and analysed data. Gloria C. Corboda Currea and Sandi Michele de Oliveira have created the research project and methodological design. Maria Clara Padoveze and Lislaine A. Fracolli have contributed to the research programme and worked as local academic liaison for the development of fieldwork. Roberto R. da Silva-Brandão and Juliana S. Correa have contributed by summarizing the main contributions of the study. Luiz F. Zago and Sandi Michele de Oliveira have written the first draft of the manuscript. All authors drafted the interview guides at different stages of the research. All authors commented on subsequent versions of the manuscript. All authors have read and approved the final manuscript.

ACKNOWLEDGEMENTS

We thank the Israeli Institute Albert Einstein, which manage the Primary Care Unit where the study was conducted, for facilitating the contact with potential respondents and allocating the place where the interviews happened. This work was funded by the Ministry of Foreign Affairs of Denmark and administered by Danida Fellowship Centre, grant number 19-M04-KU.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

The research project was submitted to the Research Ethics Committee of the School of Nursing, University of São Paulo and to the Brazilian National Research Ethics Committee (CONEP) under the number CAAE 42442921.7.0000.5392. All participants were instructed about the objectives of the research project and were informed about the findings, following recommendations of the local research ethical committee. Participants are anonymous and the interviews were conducted upon their consent, registered on the Free and Informed Consent Form.

APPENDIXLIST OF DOMAINS &QUESTIONS

This document presents the domains to be covered in the interviews associated with each of the outcomes laid out in the project proposal.

This is not the interview guide itself.

WP 1: The individual, the household and the community

Preliminary questioning—warm-up, general information

•Household and the community

•■

Number of years in the community

•■

Number of years as patient at the health center in Campo Limpo

•■

Number of people in the household

•■

Number of pets in the household (what are they?)—Additional questions on where the pet sleep, if the pet wander outdoors, what the pet eat, etc.

•■

Meaning of the relationship between humans and animals (psychological health and wellbeing by providing companionship, emotional and social support, a sense of safety and security, entertainent, happiness and relaxation)

DETAILS

Subject:	Narratives; Bacteria; Drug resistance; Bacterial infection; Suburban areas; Patients; Health care; Antimicrobial resistance; Antibiotics; Primary care; Recurrent; Ascription; Bacterial diseases; Community involvement; Infections; Knowledge; Informed consent; Urinary tract infections; Urinary tract; Content analysis; Respondents; Personal experiences; Community health care; Interviews; Drugs; Teams; Bacterial infections; Antimicrobial agents; Health professional-Patient communication; Data collection; Medical personnel; Qualitative research; Health services; Physician patient relationships
Location:	Brazil
Identifier / keyword:	antibiotic use; antimicrobial resistance; exploratory research; patient experience; primary care

Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	343-354
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-24
Milestone dates:	2022-10-26 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-08-17 (manuscriptReceived); 2022-11-24 (publishedOnlineEarlyUnpaginated); 2022-10-31 (manuscriptAccepted)
Publication history :	
First posting date:	24 Nov 2022
DOI:	https://doi.org/10.1111/hex.13664
ProQuest document ID:	2767101966
Document URL:	https://www.proquest.com/scholarly-journals/experiences-antibiotic-use-among-brazilian/docview/2767101966/se-2?accountid=211160

Compliant citizens, defiant rebels or neither? Exploring change and complexity in COVID-19 vaccine attitudes and decisions in Bradford, UK: Findings from a follow-up qualitative study

Lockyer, Bridget [1]
; Moss, Rachael H [1]
; Endacott, Charlotte [1] ; Islam, Shahid [1] ; Sheard, Laura [2]
[1] Bradford Institute for Health Research, Bradford Teaching Hospitals NHS Foundation Trust, Bradford, UK [2] Department of Health Sciences, University of York, York, UK

ABSTRACT (ENGLISH)

Background
COVID-19 vaccines have been the central pillar of the public health response to the pandemic, intended to enable us to 'live with Covid'. It is important to understand change and complexity of COVID-19 vaccines attitudes and decisions to maximize uptake through an empathetic lens.
Objective
To explore the factors that influenced people's COVID-19 vaccines decisions and how their complex attitudes towards the vaccines had changed in an eventful year.
Design and Participants
This is a follow-up study that took place in Bradford, UK between October 2021 and January 2022, 1 year after the original study. In-depth phone interviews were conducted with 12 (of the 20 originally interviewed) people from different ethnic groups and areas of Bradford. Reflexive thematic analysis was conducted.
Results
Eleven of the 12 participants interviewed had received both doses of the COVID-19 vaccine and most intended to have a booster dose. Participants described a variety of reasons why they had decided to have the vaccines, including the following: feeling at increased risk at work; protecting family and others in their communities; unrestricted travel and being influenced by the vaccine decisions of family, friends and colleagues. All participants discussed ongoing interaction with COVID-19 misinformation and for some, this meant they were uneasy about their decision to have the vaccine. They described feeling overloaded by and disengaged from COVID-19 information, which they often found contradictory and some felt mistrustful of the UK Government's motives and decisions during the pandemic.

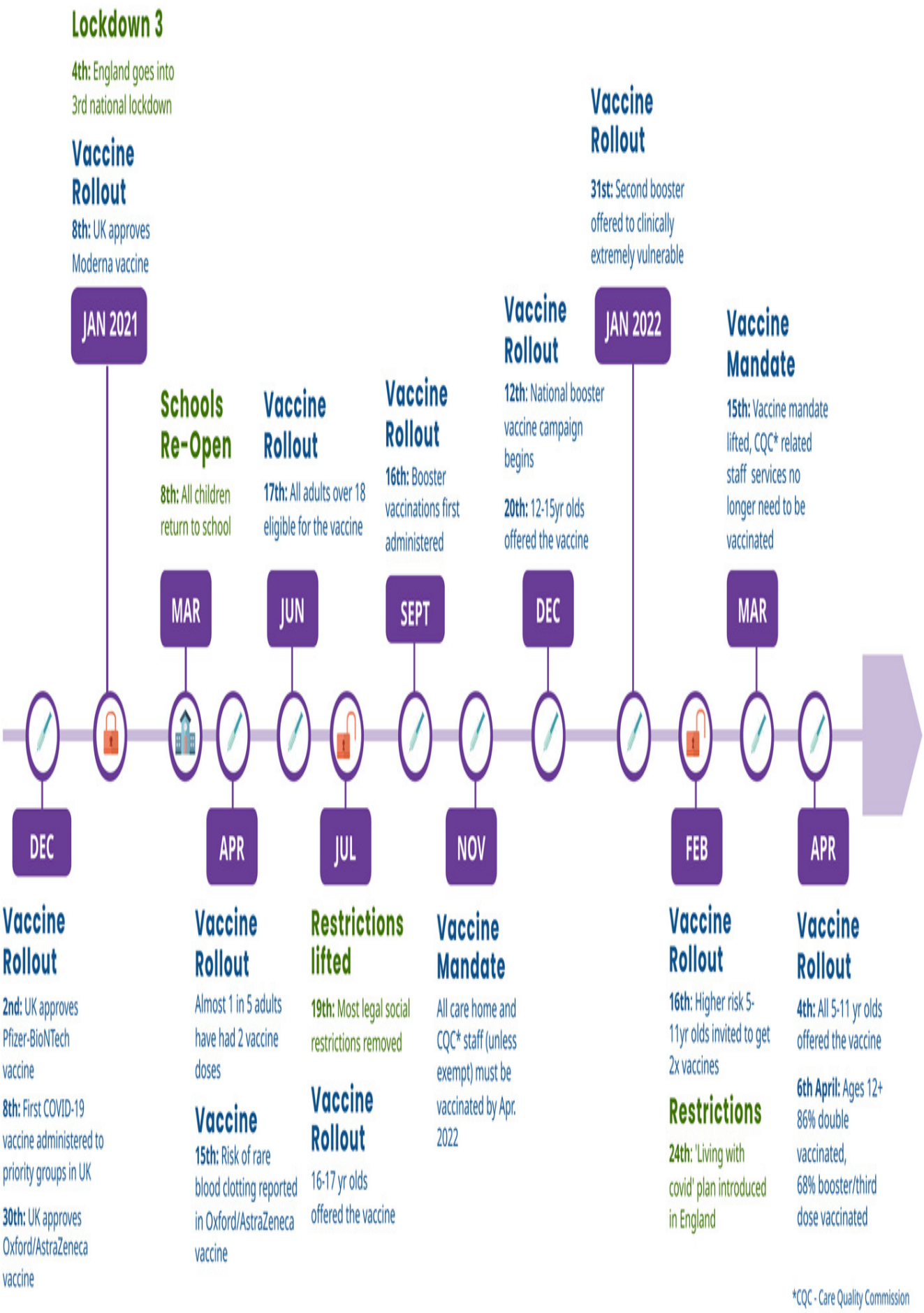
Conclusions
The majority of participants had managed to navigate an overwhelming amount of circulating COVID-19 misinformation and chosen to have two or more COVID-19 vaccines, even if they had been previously said they were unsure. However, these decisions were complicated, demonstrating the continuum of vaccine hesitancy and acceptance. This follow-up study underlines that vaccine attitudes are changeable and contextual.
Patient or Public Contribution
The original study was developed through a rapid community and stakeholder engagement process in 2020. Discussion with the Bradford Council Public Health team and the public through the Bradford COVID-19 Community Insights Group was undertaken in 2021 to identify important priorities for this follow-up study.

FULL TEXT

INTRODUCTION

Worldwide, the COVID-19 pandemic has brought a multitude of immediate challenges, including severe illness and deaths, periods of social restriction and isolation, national lockdowns, travel restrictions and extreme economic disruption. However, since the identification of several successful COVID-19 vaccines at the end of 2020, countries across the globe have attempted mass vaccination programmes to reduce transmission and disease severity, bringing the COVID-19 pandemic ‘under control’.[1]

In the United Kingdom, and specifically in England, the Government has considered vaccination to be the central pillar of the public health response to the pandemic, intended to enable us to ‘live with Covid’.[2] Since the Pfizer/BioNTech vaccine was approved for use in the United Kingdom on 2nd December 2020, a large-scale COVID-19 vaccination programme was introduced and rolled out at a rapid pace. By 17th June 2021, all adults over the age of 18 had been given the opportunity to receive their first (of two) COVID-19 vaccine(s). Several months later, a booster (third dose) was introduced, and all adults in England were given the opportunity to book an appointment to receive a COVID-19 booster vaccine by the end of 2021. By 12th June 2022, 90.9% of over 12s in the United Kingdom had received their first dose, 86.3% had received their second and 71.9% had received their booster or third dose[3] (See Figure 1 for key COVID-19 milestones in England). Studies focused on COVID-19 vaccine acceptance found that people were motivated by wanting to protect themselves and others, by trust in science and the evidence of vaccines' effectiveness, by trust in their GP and by wanting to get back to normal life after periods of social restrictions.[4–6]



Enlarge this image.

Before the introduction of COVID-19 vaccines, there were concerns about vaccine hesitancy and unequal uptake, due to historical patterns of low vaccine uptake.[7] In the United Kingdom, there were early indications that some

population groups were more hesitant than others. Higher vaccine hesitancy was associated with women, people from younger age groups, lower education levels and being from certain minority ethnic groups.[8–10] This hesitancy appears to have been somewhat borne out in the uptake figures. Vaccine uptake was found to be lowest in some of the communities for whom COVID-19 has the biggest risk, including the Pakistani, Black Caribbean, Black African and Bangladeshi communities, undocumented migrants, and studies have found a strong negative association between socioeconomic deprivation and the rate of declining COVID-19 vaccinations.[11–15] Conversely, higher vaccine acceptance was associated with being White British, older and more educated.[14] Since the COVID-19 vaccines were first rolled out, there have been serious concerns about widening health inequalities as a result of uneven uptake and there is emerging evidence to suggest that is the case.[16,17]

Much of the recent public discourse around COVID-19 vaccines has been divisive and has often strayed into racist and classist territory to explain patterns of uptake amongst different population groups.[18,19] Vaccine hesitancy, which refers to a delay in acceptance or refusal of vaccination despite its availability,[20] is in itself a contested term and considered by some to place blame on certain population groups or individuals, when wider structural forces are at play.[21,22] These include access to and relationship with health services, long-standing mistrust in institutions, poor or inappropriate methods of health communication and socioeconomic factors such as being unable to travel to vaccination centres or get time off work to attend a vaccination appointment. However, it is our understanding that using the term ‘vaccine hesitancy’ can and should take these factors into account. In 2014, the SAGE Working Group on Vaccine Hesitancy developed the confidence, complacency, convenience model of vaccine hesitancy, the 3C model.[20] This model highlights (1) confidence and trust barriers, (2) complacency and perception of risk barriers and (3) convenience, structural and socioeconomic barriers. Conversely, greater vaccine acceptance is associated with greater trust in health authorities,[23,24] greater availability of accurate and accessible information for those considering vaccines[25,26] and confidence in vaccine effectiveness and length of disease protection.[27]

Where we think the term ‘vaccine hesitancy’ can be useful is that it does not suggest a binary; people exist on a hesitancy and acceptance spectrum or continuum.[20] Whilst the spectrum ranges from full acceptance to total refusal, there are a lot of people in the middle and their beliefs, situations and decisions can alter and shift, moving them up and/or down the spectrum.

Our previous study explored the impact of the COVID-19 ‘infodemic’ and ‘misinfodemic’[28,29] through interviews with 20 citizens in Bradford, UK.[30] We found the deluge of conflicting, alarming and often inaccurate health information intensified feelings of confusion, distress and mistrust, leading to greater uncertainty about whether to have the vaccine. This study was conducted in Autumn 2020 before any COVID-19 vaccines were approved for use, so questions about vaccination intention were hypothetical. Subsequent studies have enabled us to further understand that exposure to misinformation, particularly online, increases vaccine hesitancy.[31,32] However, this process did not take place in a vacuum and existing levels of trust in governments, media, science and the health service were found to be an important influence, and lower levels of trust were found in ethnic minority groups due to long-standing institutional racism.[14,33–36] Attempts to counter misinformation have had mixed; providing clear communications on the risks and benefits of the vaccine alone was found not to be sufficient in increasing vaccine uptake nor was correcting.[37,38] Many studies have recommended local approaches, offering accessible information for different groups in the community and leveraging trusted in-group messengers.[13,29,39] On a structural level, increasing trust in health organizations, science and information via reputable sources is key but remains challenging.[29,40]

This study follows up our 2020 study, returning to conduct interviews with the same participants.[30] Unlike other recent studies, which have largely and understandably only focused on the reasons why some people choose to get vaccinated and why some people choose not to, we explore vaccine motivations, hesitancy and acceptance on a spectrum. As we had interviewed the participants before, we had a good understanding of their COVID-19 beliefs, experiences and vaccine intentions, and therefore could consider if, how and why they have changed. This also allows us to start to explore the implications of sustained exposure to COVID-19 information and misinformation on both those that have chosen to have the vaccine and those who have not.

METHODSStudy design and PPI

This follow-up descriptive, inductive qualitative study was completed as part of a larger mixed-method, longitudinal research study to provide actionable intelligence to local decision makers, developed in response to community and stakeholder consultation processes described in our previous article.[30] Our earlier findings were shared and discussed with Bradford public health teams and with the public through a COVID-19 Community Insights Group. This group was established in March 2021, and met every 6 weeks to engage with residents from the district to understand how they and their communities are coping during the COVID-19 pandemic. The group consisted of 14 (well-connected and diverse) community members. During the development of a follow-up study, we went back to both the public health team and the community group to ask them what their priorities were at that time and the topics they thought we should include on the interview schedule so these could be incorporated. We used in-depth interviews to explore the same individuals' health experiences and beliefs during COVID-19, one year on. University ethical approval for the follow-up study was secured in October 2021.

Study setting

Our follow-up study was conducted in Bradford, a city in the North of England. Bradford and its surrounding district is the fifth largest metropolitan district in England and is an area of high deprivation and ethnic diversity, with established Pakistani, Bangladeshi and Eastern European communities. Since March 2020, Bradford has experienced a relatively high number of COVID-19 cases, and stricter lockdown measures from July 2020 which remained in place until the introduction of the tier system in October 2020.[30] Initially higher rates of COVID-19 in areas like Bradford were attributed to greater deprivation, high population density and a higher than average number of multigenerational households.[30]

In late 2020, multiple UK surveys indicated that between 54% and 64% of respondents would definitely or are very likely to accept a COVID-19 vaccine and between 4% and 9% reported that they would definitely not or were unlikely to accept it, with the rest unsure.[31,41,42] Results from a survey of 535 Bradford parents at the same point of time indicated that vaccine hesitancy was higher in Bradford compared to the United Kingdom as a whole (29% would accept, 10% would not, 29% had not considered it and 32% were not sure yet).[43] The follow-up interviews took place a year on from this survey between October 2021 and February 2022, after COVID-19 vaccines had become universally available to UK adults and when COVID-19 rates in the United Kingdom were rising, mostly linked to the Omicron variant. During this time the number of daily cases in England varied between 24,134 in October 2021 and 78,512 in February 2022, with a peak of 150,786 in January 2022.[44] The number of weekly deaths in England varied between 715 in October 2021 and 1162 in February 2022, with a peak of 1378 in January 2022.[44] At the time of writing in October 2022, the total number of Bradford deaths within 28 days of a positive Covid test was 1690, a rate of 311 per 100,000 people. This is slightly higher than the England average of 297 per 100,000 people.[44]

Sampling and data collection

In our initial study, we conducted in-depth interviews with 20 people in different communities and geographical areas of Bradford using a maximum variation sample. Our key sampling motivation was diversity of ethnicity and age. Nine ‘community influencers’ were identified by S. I., a community-based researcher with significant local knowledge. These nine (three people from each major ethnic group in Bradford—South Asian, White British and Eastern European) were contacted via phone and email and invited to take part in an interview or identify others who could be interested. This method was favoured because community influencers, people embedded in community settings through their paid or voluntary work (e.g., advice worker, school and nursery community liaison, community councillor), are more likely to be trusted by their peers and people with whom they engage. Snowball sampling was used to recruit further participants. When 15 interviews had been completed, demographic and geographical gaps were identified, and additional participants were recruited via contact with volunteers at a city centre community organization.

We returned to the same participants from our previous study, contacting them via email and phone. We attempted to contact all 20 previous participants, two declined to be interviewed on a first approach, four agreed to be interviewed but were repeatedly unavailable (which we took to be a subtle decline) and we were unable to reach

two.

Fieldwork took place between October 2021 and February 2022. Six women and six men participated; their ages ranged from 25 to 86 years old, but two thirds were aged between 35 and 54. In terms of ethnicity, participants identified as Asian or Asian British (Pakistani, Indian and Bangladeshi) (n = 5), White British (n = 4) and White Other (Eastern European) (n = 3) (see Table 1 for participant demographics). The participants lived in five different Bradford postcodes, representing some degree of variation in geography and deprivation status. Many of the participants were in paid or volunteer community roles; other jobs included teacher, supermarket worker and childminder.

Table 1 Participant demographics

Pseudonym	Age	Sex	Ethnicity
Angela	45–54	F	White British
Bilal	45–54	M	Asian or Asian British
Bina	45–54	F	Asian or Asian British
Robert	Over 65	M	White British
Hasan	45–54	M	Asian or Asian British
Jackie	45–54	F	White British
Louise	45–54	F	White British
Masood	25–34	M	Asian or Asian British
Monika	55–64	F	White Other-Eastern European
Sofija	35–44	F	White Other-Eastern European
Tariq	35–44	M	Asian or Asian British
Tomasz	35–44	M	White Other-Eastern European

All interviews were conducted in English over the phone by B. L. The interviews ranged in duration from 25 to 120 min, with the average length being around 50 min. All participants gave written, informed consent through one of the following methods: (a) emailing a completed consent form or (b) emailing/texting stating that they had read the information sheet and consent form and fully consented to taking part in the study. In addition, all participants confirmed consent verbally at the start of each interview. All interviews were digitally recorded and transcribed by a professional transcriber with identifying information removed and participants' names pseudonymized.

Interview questioning

Headline topic guide questioning was adapted from the previous guide, updated to take into account the current COVID-19 vaccine programme. For example, in this guide, we included questions about participants' intentions to have the COVID-19 booster vaccine. Questions were added through consultation with local public health teams and

the community group described above. The format of the topic guide and interview questioning was flexible to allow participants to voice what they considered important.

Analysis

We undertook the analysis using the principles of reflexive thematic analysis.[45] All transcripts and interview field notes were coded independently by B. L., R. M. and C. E. We held two analysis sessions to identify commonalities and differences in the interview narratives and worked towards ordering the data into loose themes. These themes were then refined by B. L., R. H. M. and C. E. with example quotes. B. L. subsequently analysed all interviews and conducted further interpretive work to write up the findings and sense checking with the other authors as necessary. The analysis was conducted manually without the use of a software package. The analysis was wholly inductive, and, as such, we did not structure it on any existing theoretical frameworks and it was not based on the themes developed in our previous study.

FINDINGS

Out of the 12 people interviewed for this follow-up study, 11 had had at least two COVID-19 vaccines, and most were intending to get their booster. In the original study, four people in this follow-up group (Jackie, Louise, Tariq and Sofija) had been unsure and hesitant about whether they would have the vaccine when it became available to them. The findings are presented in two sections. The first explores reasons why the participants and their friends and family had the vaccine, which included travel, protecting family and friends, influence of others and personal experience of COVID-19. The second section ‘consequences of the (mis) infodemic’ builds on our previous work which contended that misinformation had stoked confusion, mistrust and distress during the pandemic, increasing participants' hesitancy about a COVID-19 vaccine. In this follow-up study, we found that although most participants had chosen to be vaccinated, continued interactions with misinformation and feeling overwhelmed by conflicting information about COVID-19 generally, had led to feelings of uneasiness about the vaccines' effects, overload and disengagement with health information and a sustained sense of mistrust in government.

Why did people get vaccinated?

Participants described various reasons why they and their friends and family had chosen to have COVID-19 vaccine(s). People were largely concerned about their health and the health of those they were closest to. Participants who had jobs which put them and their family at greater risk of virus transmission, such as Masood who worked as a security guard, Louise who worked in a supermarket and Angela who worked as a childminder, said that this prompted them to get a COVID-19 vaccine as soon as it became available to them:

I work like face-to-face like with the customers so I can understand because my family is at home, so I just …straight away I get it [the vaccine]. (Masood)

I mean I did it for my son basically, you know, who's been quite poorly as a baby with asthma and I didn't want, I work, you know, I won't say I work on the frontline but I am a key worker and I, I work on a checkout in a supermarket so I could potentially be bringing Covid home from my workplace without knowing about it. (Louise)

My daughter (aged 17) didn't want it. She weren't keen on it. I don't know, she didn't give an excuse but I didn't give her a choice. I told her she had to because we work with children and children are the carriers of a lot of germs and that. (Angela)

For some, this sense of protecting others extended to their wider community and society:

I did it for the greater good, I went and had mine done. (Jackie)

A factor which appeared to influence participants' vaccine decisions was that they now had had more personal experience of COVID-19. In the original study conducted in Autumn 2020, none of the participants had knowingly had COVID-19; this time, four had been ill with COVID-19 and the majority had a close family member or friend who had had the virus. Angela discussed her husband's experience of the virus, which was particularly worrying as he had existing respiratory issues:

Me husband did, he got really poorly. Well, he nearly died. He couldn't breathe but he has a breathing machine so if I sent him into hospital they'd only put him on that so I got him on his breathing machine. (Angela)

Sofija discussed her step-son who also had existing health conditions and was ill for months:

[He was] very bad, and very difficult to fight with this, and I think he's still got like a loss of taste and smell, so it took a long time, he recovered. (Sofija)

Another of the main motivations for having COVID-19 vaccines appeared to be practicality, as the vaccines enabled individuals to travel to different countries more freely:

I had a battle with my own children. We had to talk it through. They were talking nonsense but they are so fickle as young people as soon as they said ‘you can't go on holiday until you've had your vaccines’, they all had their vaccines done. (Bina)

One friend who was sceptical about it, after talking to us has now had the vaccine. But I think many people in the Polish community are taking it now because they want to travel. (Tomasz)

Some people described having the vaccine as common sense and ‘an easy decision’ (Monika). A few participants suggested that their previous life experience and having had many vaccines in the past made them less concerned about having a COVID-19 vaccine. This included Robert who had been in the armed forces and as such, was required to have a lot of vaccinations as part of his work:

No, as I say I've had so many vaccines and injections and inoculations over the years from everything from cholera, yellow fever, you name it, I've had it. (Robert)

Bina, although significantly younger than Robert, described coming from an age group which valued vaccines, and how this caused her to be more ready, informed and willing to get a COVID-19 vaccine than subsequent generations:

I am a believer, I'm a generation of vaccines, I was vaccinated from an early age…. So I believe in vaccine, I understand the purpose of vaccines…. (Bina)

Interestingly, despite their very willing acceptance of the COVID-19 vaccine, Robert and Bina had declined their annual influenza vaccine invitations, believing that it was not necessary for them because they were healthy. Whilst they discussed why they had the COVID-19 through the lens of social responsibility, they appeared to view the flu vaccine as a more personal health choice.

Some participants did suggest that they were initially worried about the immediate side effects and perceived long-term health effects of the vaccine, but felt reassured when they saw and heard about people they knew having it:

…when you see more people getting the vaccine then it kind of like gives you a bit more faith …it's okay to get it done. (Louise)

As seen in some of the responses above, the influence and active encouragement of friends, family and peers to get the vaccine was evident. Sofija described how her husband, who was initially worried about the vaccines' side effects, came to the decision to have it four months after it became available to him:

[Seeing] work colleagues they have been, they're all vaccinated and so I think this pushed him as well to see that they're fine, yeah. (Sofija)

She also described appealing to his emotions, and asking him what would happen to their children if they were both ill or died from COVID-19. At the time of interview, she said her husband was trying to persuade his brother to get the vaccine.

There were some discernible differences between participants' reasons (and the reasons of those close to them) for accepting COVID-19 vaccines. There appeared to be more of a push to get the vaccines for travel purposes amongst younger people and those with family who lived in other countries (such as Poland or Pakistan). This acceptance appeared to hinge on practicalities, rather than a belief in the vaccines' efficacy and safety. Social responsibility was highlighted by the slightly older participants, but as in Jackie's case, this did not mean an automatic trust in the safety of the vaccines. Those who felt more vulnerable to COVID-19 because of their jobs or because of health vulnerabilities put more emphasis on protecting themselves and their families by having the vaccines. The positive influence of talking to family and friends and observing their vaccine decisions appeared to be strong amongst those who had been hesitant a year before.

Consequences of the (mis)infodemicUneasiness about vaccine safety

Despite their decision to have the vaccine, participants still conveyed some uneasiness about the long-term impacts

of COVID-19 vaccines:

I don't know what it'll do to my body in the future, I don't know if there'll be any […] I've no idea what could go wrong. I weighed it up and it might, it might not …and no, I just needed to get it done, needed to do my bit. (Jackie)

Jackie's account suggested she had struggled to weigh up what was best for her and what was best for society. She indicated that she thought that her long-term health had been put at risk by having these vaccines. Similarly, Louise said that although she had been persuaded to have the COVID-19 vaccines by seeing her friends who were nurses have it, she still harboured thoughts that the vaccine she received could have unknown long-term health consequences:

So, you know, and I think when you see your nurse friends and family, you know, having the vaccine it gives you a bit more confidence that, you know, if, well if they're going to get wiped out I'll go with them [laughs]. You know? I mean if, if we've all had this vaccine and we're going to die in the next couple of years we'll all go together, won't we? (Louise)

The possibility that the COVID-19 vaccines could have lasting health implications appeared to be rooted in misinformation that the participants had encountered. All participants showed an awareness of misinformation about COVID-19 vaccines within the interviews. As in the previous study, most were keen to make clear that they did not believe any 'conspiracy' stories about the vaccines, yet common tropes about the vaccines' safety, such as them changing your DNA or causing infertility, were part of their narratives:

They did say that it changed DNA, one of them, didn't they and stuff? (Angela)

I've read there is a special code and the code will change your DNA and you will die. And younger people wouldn't be able to have children. (Sofija)

My daughter said she didn't want to take a vaccine because what if they affected the time when she wanted to have children …When the story came out that the Pfizer vaccine was affecting the foetal part of the body when they're pregnant and so on she was then reading up on how people had been affected. (Bina)

Overload and disengagement

Whilst the spread of misinformation may have begun online and been facilitated by social media, it was evident that it was being discussed amongst friends and families and had influenced people's beliefs and decisions. Tomasz discussed why a friend of his had refused to have the vaccine:

I think it's because he's got you know, other friends around yeah, and maybe there's a lot more people who are sceptical about it in his circles. (Tomasz)

Tariq, the only participant not yet vaccinated, had been heavily influenced by his friends:

Some friends, that I've made over last couple of years they've got quite close to me and they're like 'oh you'd better not take it, you'd better not take it, you don't know what's in it …you're not well already and who's going to look after you if something happens to you?' (Tariq)

Tariq was saturated in COVID-19 misinformation. His friends had invited him to join a very active WhatsApp group which was inundating him with negative videos and pictures about the safety and inefficacy of the vaccine. He discussed some of this content:

Like some people have said 'oh this person's had the jab and then they've got, they've been paralysed or something's happened to them or they've got ringing in their ears, you know, they've had a heart attack or they've had to go and have more surgery or kidney's failed or they've had a blood clot', stuff like that. So I don't know if it's true. Did the British Airways pilots die of blood clots or was that just a hoax? (Tariq)

Other stories he discussed were the building of a camp in the United Kingdom to imprison people who refused to get the vaccine and the local hospital hiring actors to make it look full of COVID-19 patients. He was obviously conflicted by the information he saw and was not sure what to believe:

I'm trying to stay away from it now because it's just like, it's proper confusing and I'm trying my best to stay away from it but it's quite hard because I've not left the group and I'm thinking 'shall I leave the group? Shall I leave the group or not?' and I've not done and then I'm still listening to what's coming through and I'm like thinking 'I should have left the group, I should have left the group'. But then you're thinking like 'hang on, are they making sense?'

(Tariq)

Tariq's sense of confusion and feeling of being overwhelmed was very apparent. Although he was the only one who had not yet had the vaccine, other participants described being overwhelmed and confused about COVID-19 information and misinformation and indicated these feelings had led them to take less notice of the news:

I turn off the TV when I see people arguing about vaccinations. (Jackie)

I get fed up of hearing it, I turn the telly off. (Angela)

Making sense of the deluge of information about COVID-19 since March 2020 appeared to have had an emotional and mental toll, which led to people purposefully choosing to disengage:

I've avoided the news and social media, I made a decision to because of my health anxiety. (Monika)

I'm just keeping abreast of information but not letting it take over my life like it did at the beginning…. (Bina)

I feel better without this information …because I'm very emotional. (Sofija)

The distress Sofija experienced from seeing both information and misinformation during the early part of the pandemic was still clearly influencing her ability to engage:

They scared me so much that yeah [laughs], that you know, that yeah, depression and, yeah, it was horrible. I don't want to be in that state anymore, that's why I'm like, I don't want to hear anything because it affected me so much. (Sofija)

There was some evidence that this disengagement had led to people being less informed about what the current official guidance on vaccines was. When Tomasz was asked about whether he was planning to get his booster jab he did not appear to have considered it much, although he was soon to be eligible and it was being encouraged amongst his age group:

If we need to take a booster vaccine then I'll take it. And the virus has, you know, mutated and stuff like that, so I think it's worth, you know, taking but I will read into that. Actually, that one I might check-out you know, how it works and…. (Tomasz)

The information people appeared to take most account of was local information, provided by the council and local health centres:

So I signed up to Bradford Council email, so I get emails, updates quite regularly from Bradford Council and they're quite good …I think that it's been really good. I work with a community centre and they get information obviously from the Council and they've found the information really, really good. (Bilal)

Mistrust

One of the reasons why people described feeling overwhelmed and confused was because they were not sure who to trust. Official health information felt contradictory, and at the time of the interviews, there had been several reports about those in the Government not abiding by lockdown rules. Sofija said she had found information from the Government:

…very mixed. The politicians not following their own messages and you know, it's not like trustworthy. (Sofija)

There was a lot of scepticism about the Government's response to the pandemic, with people thinking that they have been scaremongering and lying about the numbers of deaths related to COVID-19:

I've seen them on telly and you don't know if they're telling truth or making it worse. Saying thousands and thousands have died and it's going here and going there. No, you don't know if they're just doing it to scaremonger the whole of the world. (Angela)

Tomasz felt the Government had allowed private companies to ‘monetise the pandemic’ and said a lot of his friends felt more strongly than he did:

I think lots of my friends don't trust yeah, the Government yeah. And then people are generally fed-up with all the information. I see lots of comment about oh stop scaremongering blah, blah blah, something like that, you know. So, people I think are fed-up, they don't want to hear about it. (Tomasz)

For Hasan, despite believing in COVID-19, laughing at ‘conspiracy theories’ and having had two vaccines and booster when offered as it is better to be ‘safe than sorry’, he was incredibly cynical about the Government's response and felt they had exaggerated COVID-19's impact for political and financial gain:

I'm probably the wrong person but for me Covid has just been an excuse and a reason for government in order to do certain things and be able to do things, so it's been a God send and that's all it is at the end of the day. (Hasan)

Ultimately, mistrust had not prevented our participants from getting the vaccine but it was easy to understand from their responses the ways in which it was contributing to hesitancy.

Eleven of the 12 participants in this follow-up had chosen to have at least two COVID-19 vaccines and most had or were planning to get their boosters. However, their decisions and beliefs were complex and there was still uneasiness, disengagement and mistrust. Some of those within the 2020 study that had been most hesitant (such as Jackie and Louise) remained so, even though they had opted to have two vaccines. Sofija was relatively hesitant in 2020, anxious about the vaccine's safety, but was now much more accepting, even encouraging family and friends to take it. Participants like Angela and Hasan who said they were going to have the vaccine in 2020 and did, still suggested that they believed some wider conspiracy theories about COVID-19. Bina and Robert were very positive about the vaccine in 2020 and 2021/2022 but this acceptance did not lead to an overarching acceptance of all vaccines, as they were firm that they did not want a seasonal flu vaccine. Tariq, one of our participant still unvaccinated, appeared to be even more conflicted and overwhelmed than he had been in 2020.

DISCUSSION

This follow-up study aimed to understand people's COVID-19 vaccine decisions and explore their health beliefs and experiences around COVID-19 and COVID-19 vaccines. We found that most participants had chosen to have two or more COVID-19 vaccines. Their reasons and the reasons of those close to them included the following: protecting their health and the health of loved ones; travelling without restrictions; closer experience of COVID-19 and the positive influence of others around them having the vaccine. These reasons corresponded to the findings of other studies which explored vaccine acceptance, although the influence of others was more pronounced and there was more ambivalence about the vaccines' safety and effectiveness.[4–6]

In our previous study, we found that exposure to COVID-19 misinformation had led to confusion, distress and mistrust and was contributing to uncertainty in vaccine intentions. Amongst these participants, exposure to misinformation appeared to be higher than the UK average, as one survey conducted regularly between March 2020 and September 2021 estimated that around half of people in the United Kingdom had come across some form of misinformation every month.[46] Multiple studies have shown a correlation between exposure to misinformation and vaccine intent.[31,32,47] Our follow-up study found that the majority of participants and those close to them had managed to overcome the tidal wave of misinformation and get the vaccine. However, their accounts still suggested exposure to and engagement with misinformation, particularly around vaccine safety and long-term effects, which was causing some to be uneasy about their decision.

However, after over 18 months of interacting with distressing, confusing and often contradictory COVID-19 information and misinformation in news, social media and within their social circles, they were feeling overwhelmed. As a result, several participants described actively disengaging with anything related to COVID-19 to feel more in control and less anxious. Many were distrustful and suspicious about the Government's response to the pandemic, some were even doubting what they had been told about the severity of the COVID-19 virus. This is an important finding, as it suggests that living through this tumultuous period could have lasting impact on people's trust in government, future vaccination programmes and perhaps public health interventions in general. Indeed, the latest vaccination statistics for children up to 5 years of age in the United Kingdom show coverage decreased for 13 out of the 14 routine vaccination programmes measured in 2021 to 2022.[48]

Our findings illustrate the continuum of vaccine hesitancy and acceptance and recognize that those who have chosen to have the vaccine may still have doubts and concerns. This is important to understand because it means that we cannot take their current vaccine acceptance for granted, it is based on current social and political conditions and patterns which can change.[49] This aligns with the study of expected vaccine uptake,[50] which found that both conditional acceptance and rejection of COVID-19 vaccines were dynamic and volatile. By following up with the same participants, we were able to see that people's location along the hesitancy/acceptance spectrum is changeable. Sofija and her husband were good examples of that, moving from vaccine hesitancy and delay to

encouraging others to get their vaccines. This is further evidence that people who appear 'vaccine hesitant' can indeed be convinced.[50]

This method has also allowed us to understand the different and compounding reasons why people may move further along the spectrum towards acceptance despite reservations, including health concerns peer/family encouragement and practicalities. Our study has also highlighted that people's levels of vaccine acceptance can differ for different vaccines. For example, some participants were happy to have the COVID-19 vaccine and were not willing to have a seasonal flu vaccine. Being vaccinated for COVID-19 was deemed to be socially responsible, but being vaccinated for flu was a personal health choice, suggesting that people did not fully understand the social and human costs of a widespread flu outbreak. This reminds us that vaccine hesitancy and acceptance should be looked at in the historical, political and socio-cultural context in which vaccination occurs.[51] We had less insight into the beliefs and motivations of those still refusing to be vaccinated as this two-wave study depended on participants agreeing to be re-interviewed. Yet the internal struggles of Tariq and his willingness to discuss them demonstrated that even those who appear to be staunchly resistant could have the potential to move towards acceptance within a less confusing and distressing environment.

Often local and national media write-ups of studies which report and discuss COVID-19 vaccine uptake amongst different population groups (including our own work) have been met with social media comments which contain a 'them and us' narrative, sometimes with implicit and explicit racist and classist tropes. Lower than average vaccine uptake in Bradford has been blamed by online commentators on the Pakistani and Eastern European populations, the poor and the less educated.[52] Public discourse on both 'sides' have called one another stupid and uneducated, for either their perceived compliance or defiance.[52] Yet our participants' views and decisions were nuanced and complex, and largely understanding of others' doubts and fears because they shared some of them too. This study adds further weight to a need to move away from the binary of vaccine hesitancy and acceptance, not only because it can contribute to stigmatizing certain demographic groups but also because it is incorrect and unhelpful.

Implications for policy and practice

We found misinformation, whether it was regarded as misinformation or not, to still be present and sometimes influential in the experiences and narrative of our participants. Numerous studies have advocated for local, targeted, community-driven and accessible health information,[13,29,39] and we think this study gives further weight to persisting with this, particularly because trust in the national government was low. The quality of local government–public relationships is positively associated with provaccine outcomes, including more frequent risk information seeking, pro-vaccine attitudes and greater vaccination intention[53] and our study found that participants' most trusted health source was the local council. Although the misinformation machine is global, continuing to foster and develop strong and trusting relationships locally can help erode some of its impacts.

This study also found that encouragement and positive discussion about COVID-19 vaccines with family and friends was persuasive. Seeing vaccine acceptance normalized amongst friends, colleagues and acquaintances was also influential, as people trusted the decisions of those close to them. This further underlines the importance of health messaging that leverages personal relationships and positive emotions.[54] Our findings were able to capture how overwhelmed people were by the COVID-19 (mis)infodemic, and the potential for disengagement with future COVID-19 booster(s) or other vaccine campaigns as a result. Again, clear and positive public health communication should be prioritized over messaging that is likely to engender further anxiety and distress.

Limitations

As this was a follow-up study, our sample size was necessarily restrained. In addition, we found that some of those who said they were very unlikely to have a COVID-19 vaccine, were unwilling to talk to us this time or were unreachable. This is perhaps understandable, given, how politicized and fraught vaccine decisions have become. As a result, we have less insight into the uptake and beliefs of some of those who appeared most hesitant in 2020.

CONCLUSIONS

The public discourse around COVID-19, and vaccines in particular, has often felt polarized. There has been an assumption that people have either been compliant citizens or defiant rebels, assumptions which often have classist

and racist undertones. The findings of this study ask us to consider the feelings and behaviours of the vast majority of the population who are neither. By following up with the same group of people from a largely deprived and multi-ethnic city, we could appreciate, in context, how and why they made their decisions and more deeply explore the complex influences of family and peers, health (mis)information and (mis)trust in institutions. The majority of the participants had chosen to be vaccinated, but this was not without some uneasiness and their narratives still contained threads of misinformation and mistrust. As well as underlining the persistent effects of misinformation, this study re-emphasizes vaccine hesitancy/acceptance as a continuum, rather than as a binary concept. In doing so, we hope to contribute to a greater and more empathetic understanding of what shapes the health beliefs and behaviours of all of us on the continuum.

AUTHOR CONTRIBUTIONS

This research has been designed and developed by the first and last authors (Bridget Lockyer and Laura Sheard) in collaboration with Shahid Islam and the wider Bradford Covid-19 Scientific Advisory Group. Bridget Lockyer collected the data and Bridget Lockyer, Charlotte Endacott and Rachael H. Moss carried out the analysis. Results have been interpreted and discussed by Bridget Lockyer, Charlotte Endacott and Rachael H. Moss and the wider Bradford Covid-19 Scientific Advisory Group. Bridget Lockyer, Charlotte Endacott and Rachael H. Moss wrote the first version of the paper and then Laura Sheard and Shahid Islam read, commented on, made edits and approved the final manuscript.

ACKNOWLEDGEMENTS

We are grateful for all the support and feedback given to us by people within the Bradford Council Public Health team, the Bradford Covid-19 Community Insights Group and the members of the Bradford Covid-19 Scientific Advisory Group. Finally, we would like to thank all our interview participants who took the time to be part of this follow-up study. This study was funded by The Health Foundation Covid-19 Award (2301201); A Wellcome Trust infrastructure grant (WT101597MA); The National Institute for Health Research under its Applied Research Collaboration Yorkshire and Humber (NIHR200166); ActEarly UK Prevention Research Partnership Consortium (MR/S037527/1).

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data that support the findings of this study are available on reasonable request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

DETAILS

Subject:	Pandemics; COVID-19 vaccines; Socioeconomic factors; Minority &ethnic groups; Public health; Vaccines; Community; Ethnic groups; Teams; Immunization; Attitudes; Change agents; Misinformation; COVID-19; Ethnic factors; Health professional-Patient communication; Uptake; False information; Complexity; Qualitative research; Coronaviruses; Decisions; Disease transmission; Dosage; Friendship
Location:	United Kingdom--UK; England
Identifier / keyword:	Bradford; COVID-19; qualitative; vaccine hesitancy; vaccine acceptance; misinformation
Publication title:	Health Expectations; Oxford
Volume:	26

Issue:	1
Pages:	376-387
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-12-01
Milestone dates:	2022-10-24 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-06-21 (manuscriptReceived); 2022-12-01 (publishedOnlineEarlyUnpaginated); 2022-11-06 (manuscriptAccepte d)
Publication history :	
First posting date:	01 Dec 2022
DOI:	https://doi.org/10.1111/hex.13667
ProQuest document ID:	2767101965
Document URL:	https://www.proquest.com/scholarly-journals/compliant-citizens-defiant-rebels-neither/docview/2767101965/se-2?accountid=211160

| Last updated: | 2023-10-04 |

Influences on indoor environmental trigger remediation uptake for children and young people with asthma: A scoping review

Lewis, Grace [1]
; Milnes, Linda [1] ; Adams, Alexandra [2] ; Schwarze, Jürgen [3] ; Duff, Alistair [4] [1] School of Healthcare, Faculty of Medicine and Health, University of Leeds, Leeds, UK; Asthma UK Centre for Applied Research, USHER Institute, University of Edinburgh, Edinburgh, UK [2] Asthma UK Centre for Applied Research, USHER Institute, University of Edinburgh, Edinburgh, UK; Paediatric Respiratory Unit, Leeds Children's Hospital, Leeds, UK [3] Asthma UK Centre for Applied Research, USHER Institute, University of Edinburgh, Edinburgh, UK; Child Life and Health, Centre for Inflammation Research, The University of Edinburgh, Edinburgh, United Kingdom [4] School of Healthcare, Faculty of Medicine and Health, University of Leeds, Leeds, UK; Asthma UK Centre for Applied Research, USHER Institute, University of Edinburgh, Edinburgh, UK; Paediatric Respiratory Unit, Leeds Children's Hospital, Leeds, UK

ABSTRACT (ENGLISH)

Introduction
Children and young people (CYP) with asthma can benefit from reduced exposure to indoor environmental allergens and triggers but may not consistently have avoidance strategies implemented. To inform future interventions to increase trigger and allergen avoidance and enhance asthma control, a greater understanding of the influences on avoidance behaviours is necessary.

Methods
A systematic scoping review was selected to summarize evidence on what influences family uptake of indoor environmental asthma trigger avoidance strategies for CYP with asthma and identify research gaps. Primary studies of any design, including CYP (≤18 years) with asthma, and/or parent-carers, available in English and conducted since 1993, were eligible. Searches included nine databases, hand-searching reference lists and citation searching.

Findings
Thirty-three articles were included and are summarized narratively due to heterogeneity. Influences appear complex and multifactorial and include barriers to strategy uptake, health beliefs and personal motivation. Research specifically related to family understanding of allergic sensitisation status and exposure risks, and how these may inform avoidance implementation is required. Patient and public involvement (PPI) was not reported in included articles, although two studies used participatory methods.

Conclusion
There is limited research on family asthma trigger management, particularly what influences current management behaviours. Variation in families' ability to identify important triggers, understand exposure risk and consistently reduce exposures warrants further exploratory research to explain how families reach avoidance decisions, and what future interventions should aim to address. Further PPI-informed research to address such gaps, could enable theory-based, person-centred interventions to improve the uptake of asthma trigger remediation.

Patient or Public Contribution

An asthma-specific PPI group contributed to the decision-making for the funding for the wider project this review sits within. The findings of this scoping review have informed the subsequent phases of the project, and this was discussed with PPI groups (both adult and CYP groups) when proposing the next phases of the project.

FULL TEXT

INTRODUCTION

Asthma is a complex, heterogeneous, chronic airway condition, affecting more than one million children and young people (CYP) in the United Kingdom, and contributes to substantial economic and emotional burdens.[1] Attempts to support CYP and families include self-management programmes, which are multifaceted with medicating, monitoring and managing asthma triggers seen as core components.[2] Physical asthma triggers can be broadly grouped as allergic, and irritant, and can be further subdivided into indoor and outdoor exposures. The focus of this review will be indoor environmental triggers including irritants and allergens.

There are potentially multiple indoor environmental triggers and exposure has been associated with increased asthma severity, exacerbations and reduced quality of life in CYP.[3] Whilst intervention trials aim to reduce allergen presence in homes, including house-dust mites (HDM), and pet allergens, many methods are not recommended for all by clinical guidance, in the United Kingdom. This is due to limited evidence for HDM exposure reduction methods, [2] the complexity and heterogeneity of trials of reduction methods and subsequent challenges of aggregating data for systematic reviews or meta-analyses for HDM and furry pet allergen reduction.[4] Thus, trigger-management advice is often to remove or avoid trigger sources, such as pets. However, longitudinal epidemiological evidence suggests that having a family member with asthma (without co-existing rhino conjunctivitis) is not associated with pet withdrawal and does not deter pet acquisition.[5] Moreover, a multicentre study conducted in 22 countries, demonstrated that adults with asthma and/or allergy, who owned pets and subsequently had children with an asthma diagnosis continued to keep pets, although with greater avoidance of cats than dogs or birds.[6] Potential recall and selection bias were acknowledged in both studies.[5,6]

Systematic reviews of asthma-trigger education programmes have shown some promising outcomes. However, these are limited due to bias in included studies,[7] a scarcity of eligible studies, and heterogeneous outcome measures further limited conclusions regarding intervention effectiveness.[8] A more recent systematic review of educational interventions for CYP with asthma in underserved communities or minority groups, noted a lack of theory use and consideration of health literacy in intervention trials. Authors suggested greater attention be given to the beliefs and attitudes of those whose behaviour the interventions are designed to change.[9]

Despite healthcare providers giving avoidance advice, clinicians anecdotally note that families report continued exposures, particularly regarding pets they are emotionally attached to,[10] and reluctance to rehome pets can lead to reluctance to suggest this.[11] Multiple HDM reduction methods exist with varying levels of evidence to support their promotion for use in the homes of people with asthma.[4] Given the aforementioned complexities surrounding HDM reduction method effectiveness measures, practical patient-specific advice has been advocated, instead of relying on meta-analyses of intervention trials.[4,12] However, little is known about whether families implement these measures, and how they choose between methods or barriers they may encounter, in real-life settings. Avoidance of other indoor environmental exposures, such as environmental tobacco smoke (ETS) is advocated for general health, [13] asthma control and primary prevention.[2] How families actually manage indoor environmental asthma triggers outside of trial settings is not well described and long-term intervention effectiveness also depends upon adherence to such health advice. Understanding adherence to supported self-management plans by CYP and their families is complex since these include monitoring asthma, taking medications and managing asthma triggers with health-provider support and to date, the literature focuses heavily on asthma medication adherence challenges. To enable the development of future interventions to address the apparent gap between clinical advice and environmental trigger avoidance uptake, a clearer understanding of the influences on avoidance and nonavoidance behaviours is needed. Furthermore, there is consensus that interventions should build from an evidence-based understanding of

the target problem or behaviours and context, in addition to careful selection and use of theory from early stages and iteratively throughout intervention development.[14]

Objectives and justification for selecting a scoping review

The objective of this scoping review is to describe what is known about CYP and/or parent-carer beliefs, motivations and other influences involved in the uptake of avoidance of indoor environmental asthma triggers, in homes with a CYP with an asthma diagnosis. Additionally, the review aimed to discover evidence gaps. The overarching objective of the scoping review was to ascertain whether there is sufficient evidence to inform the development, or adaptation of a behavioural intervention to address continued exposures in CYP with moderate-severe asthma and co-existing allergic sensitisation* particularly to pets and/or HDM (*the presence of a positive reaction to allergens on testing, showing that there is an immune response mediated by exposure to the specific allergen. The immune response leads to airway inflammation and asthma symptoms and/or suboptimal control of asthma). Early literature searching to clarify the ideal type of literature review for these purposes suggested there is scant research into influences on asthma trigger avoidance behaviours. This led to the decision to select a scoping review to provide a high-level overview of what is known and to identify research gaps.[15]

METHOD

This scoping review was guided by a seminal framework,[16] alongside recent guidance.[15,17–19] These include review question development and study identification through database searching, study selection, charting or synthesizing and disseminating findings.[16] A priori protocol was written as recommended.[15,20]

Scoping review questions

- (1)
  What is known about CYP and parent/carer beliefs regarding indoor environmental asthma triggers in homes?
- (2)
  Do their beliefs inform exposure reduction strategy uptake?
- (3)
  What factors influence avoidance/nonavoidance behaviours or adherence to avoidance advice?
- (4)
  Are CYP/parent-carers motivated to reduce environmental trigger exposures at home, and what may further motivate avoidance?
- (5)
  Are there any relevant research gaps which may require attention before further behavioural intervention development or adaptation?

Search strategy

Search terms were developed according to participants, concepts and contexts of interest[19]: asthma AND/OR allergic sensitisation AND triggers AND children AND/OR parent/carers AND beliefs AND/OR behaviours AND qualitative OR quantitative OR mixed methods. A search string is available in Supporting Information: 1. Table 1 details the inclusion/exclusion criteria, including reasons for the selection of participants, concepts and contexts of interest for the scoping review.

Terms were refined following an initial search using Ovid Medline, and relevant synonyms, mesh terms and headings were used. Final terms were extended to Embase, CINAHL, PsychINFO, Google scholar and Cochrane Database. Grey literature databases searched included Zetoc, OpenGrey and Ethos. Systematic reviews were not included in the scoping review but were read and reference lists were reviewed for relevant primary studies. Citation

searching was conducted using Scopus, Google Scholar and Web of Science, and reference lists of key and included articles were searched. This strategy was developed to capture broader studies of self-management that included data related to influences on asthma trigger avoidance strategy uptake.

Searches were limited to articles available in English and those with primary, empirical data collected and published since 1993. This reflected that British Thoracic Society guidance changed in 1993 to include asthma trigger avoidance advice.[21] Further inclusion/exclusion criteria are detailed in Table 1. These restrictions allowed a balance between relevance and search breadth.[18,22] Initial searches were run from January to March 2020 and updated in August 2021. Database alerts were used throughout to track additions to the literature with matching search terms. Deduplication within databases was conducted where available, and further deduplication was recorded after importations to Mendeley.

Table 1 Inclusion/exclusion criteria

Restriction area	Inclusion	Exclusion	Explanation
Study design	Any primary study design highlighting beliefs and opinions about asthma triggers (concept of interest) in CYP and/or parents/carers of children with asthma and trigger avoidance strategies	Studies designed to evaluate effectiveness of an intervention. However, if baseline measures were taken to establish beliefs before an intervention, these could be included if they could be extracted in isolation	The aim of the scoping review is to understand whether triggers are noted and/or avoided by CYP/parents under usual care, rather than those who have undergone an intervention trial. Incorporating all designs allowed for broad evidence scoping
Studies exploring other triggers	Those including indoor triggers, in any country (context) where findings relating to these can be extracted separately	Studies exploring only beliefs around psychological triggers or outdoor environmental triggers	Numerous studies were noted exploring only psychological or outdoor triggers on developing and piloting search strategies
Participants	CYP (under 18 years) or parents/caregivers of CYP with asthma or asthma and co-existing allergic sensitisation—if reported	Adult only participants with asthma or unclear descriptions of diagnoses (e.g., wheeze rather than asthma). Studies including only those under the age of 5 years/parents of under 5s with asthma, were ineligible	Due to differences in asthma and asthma management between adults and those under 18 years.[23,24] Asthma is difficult to diagnose in under 5s.[2,25]

Abbreviation: CYP, children and young people.

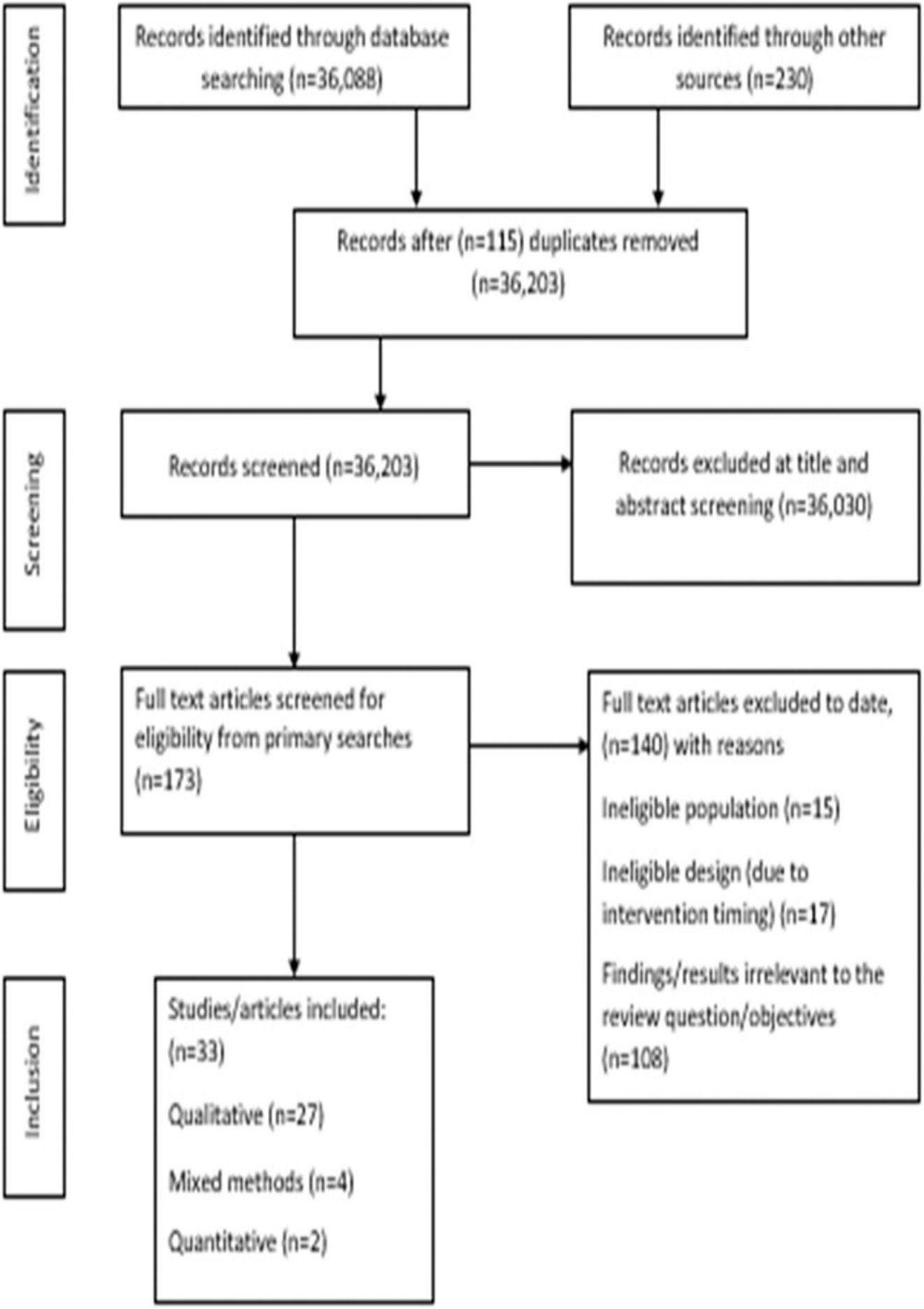
Data extraction

Two reviewers (G. L. and L. M.) conducted article selection and data extraction. Data extraction followed scoping review guidance,[16] and further details included study aims, dates of data collection (where available) and confirmation of ethical approval. A copy of the data extraction table is available via the protocol.[20]

RESULTSArticle retrievals

Electronic database searching retrieved 36,088 articles and a further 230 were retrieved through hand-searching

reference lists and database citation searching. After deduplication, 36,203 were screened and 36,030 were excluded based on title or abstract. Full-text screening was conducted for 173 articles and 33 were included. Reasons for exclusion include ineligible populations/participants (*n* = 15), ineligible study design, due to intervention timing (*n* = 17), or results that were not suited to answer the scoping review questions (*n* = 108). Retrieval results are displayed in Figure 1: PRISMA flow diagram and a PRISMA-ScR checklist[26] is available in Supporting Information: 2.



Enlarge this image.

Study designs

Of the 33 studies, 27 were qualitative, two quantitative and four mixed methods. Methodologies and methods employed are detailed in Supporting Information: Table 2 (Supporting Information 3) alongside study aims, context/setting, participants, and study designs. Due to the heterogeneity of included studies, narrative findings are presented. Due to the broad scoping nature of this review, included studies also reported findings that are outside the scope of this review. For clarity, only the findings or results that are pertinent to this review are reported.

Narrative summary of findingsAbility to identify triggers

A family's ability to identify triggers is an important step preceding decisions about avoidance strategy implementation. Participants were able to identify some potential indoor environmental triggers across the majority of included studies.[27–51] A mixed methods study reported that 77% of 200 parents of 5–12-year-olds with asthma, avoided some indoor environmental triggers suggesting recognition, although most identified ETS and dust.[52] Parents and CYP with asthma were asked to rank triggers by impact in an American study with socioeconomically disadvantaged families; ETS, dust and cockroaches were believed to have the highest impact, followed by pets, mould and dry heat.[49]

Trigger identification was not universal across studies. An English qualitative study of 11–18-year-olds with severe, uncontrolled asthma, reported that participants had very limited trigger knowledge; whilst most were aware of ETS as a trigger, many lived with a pet but were unaware pet allergens could trigger asthma or denied that pet exposure led to symptoms, even where participant descriptions suggested otherwise.[37] Another qualitative study from England described parents' belief that some CYP with asthma were unable to identify triggers themselves.[50] Authors of a grounded theory study conducted in America noted some families were not able to recognise which triggers led to exacerbations and such uncertainty resulted in anxiety.[53]

Trigger information provision and experiences with triggers

Receipt of trigger information was identified as a potential factor influencing avoidance strategy implementation in a mixed methods pilot study that investigated parents' trigger knowledge and strategy uptake[39]: African American parents (n = 4, of n = 12 participants) reported having not received trigger information from healthcare providers, in contrast to the eight White participants. This was reflected in greater awareness of triggers (t = 2.43 p = .017) and higher uptake of avoidance strategies (t = 1.98; p = .04; particularly for HDM reduction t = 3.23; p = .009) in White families with a child with asthma. Although, all were aware of ETS and pets as triggers. Those who had not received trigger avoidance advice were less trusting of healthcare providers and were less likely to report having discussed asthma with others or sought information themselves. However, due to this being a pilot study, the results were deemed provisional.[39]

A qualitative study with White British and British South Asian families with a child with asthma, also highlighted that across ethnicities families reported they had not received an asthma action plan, in which families are invited to note triggers. Furthermore, those who did not speak English as a first language experienced additional barriers where information was not provided in their first language. Although, all families experienced problems with accessing or understanding information, including trigger information.[46]

For fathers in a Canadian qualitative study, trigger advice from other parents of children with asthma was valued and observing an exacerbation following trigger exposure led to trigger recognition.[54] This was echoed in a Norwegian study of 15 children (7–10 years) who learned to recognise triggers through previous exacerbations or allergic reactions and at times endured continued exposures or continued activities that left them feeling exhausted, to maintain social normality.[55]

Findings from two North American qualitative studies reported that participants who were unable to identify triggers described that they did not know the information they needed to enable identification.[44,53] A qualitative study in the

United States described parents being overwhelmed when multiple trigger exposures were possible, and there was uncertainty in 9 of their 10 participants about risks attributable to triggers.[56] Younger children (7–12-year-olds with moderate-severe asthma), in the United States, were able to identify triggers such as a family pet and attribute coughing to exposure, but rarely knew how to avoid triggers.[29]

Two articles mentioned that where trigger exposure also led to noticeable allergic symptoms, such as facial oedema, triggers were more easily recalled,[51] by children as young as 7 years old.[55] In contrast, some parents did not consistently notice signs of allergy or deteriorating asthma control.[44] Parent participants in a study in Taiwan described that they did not know their children's triggering allergens and that their 8–12-year-olds should be aware themselves. This contrasted parental beliefs that they should help CYP with asthma control, in the same study. Parents also experienced difficulty differentiating colds, asthma and allergic rhinitis symptoms.[42]

HDMs were mentioned far less than other triggers. One article reporting a qualitative study with mothers of children with asthma in Australia noted that HDM had to be identified as a trigger and explained by health professionals and that not knowing this sooner led parents to reassess their competence after an exacerbation and led to feelings of guilt.[31]

Myths and misconceptions

Misconceptions and myths recurred in accounts of participants' beliefs. Asthma was believed to be episodic rather than chronic with symptomatic episodes by some participants.[44] There was also confusion between perceived asthma aetiology and asthma symptom triggers. For instance, parents believed asthma only occurred when CYP were exposed to triggers,[46] such as dusty schools.[33] Such misconceptions led parents to believe that asthma could be cured by trigger eradication.[44] Parents also believed that whilst dust should be minimized, children were likely to outgrow asthma and ‘willpower’ could limit the likelihood of chronicity.[43,p.134]

ETS beliefs and experiences

Despite broad recognition of ETS as an asthma trigger, some studies highlighted misconceptions and risk-taking. Some parent-caregivers in an American qualitative study believed ETS exposure could enable tolerance.[41] Some CYP reported experimenting with cigarettes, despite knowing the risks.[37,47] Conversely, in some studies, CYP noted that parent-carers continued to expose them to tobacco smoke.[32,33,40] CYP explained this by noting cultural norms and the unacceptability of requesting guests to smoke outdoors in one study.[33] Parents also reported feeling they lacked control over the presence of pets and smokers,[28] but the underlying reasons for limited control were not clear. Barriers to tackling this appeared related to parental health beliefs, personal and environmental circumstances rather than socioeconomic limitations, such as healthcare access and medical insurance coverage, as often presumed in low-income groups, as sampled in this study.[28] Some teenagers became able to self-advocate ETS avoidance at home either by removing themselves from the area or requesting parents smoke outside.[45] In contrast, CYP sometimes avoided confrontation with others over ETS exposure by moving away or using reliever inhalers.[32]

CYP age

Age has also been identified as a factor in CYP taking responsibility for asthma self-management.[41,42] A qualitative study with parent-carers of teens (14–18 years), with asthma, reported parent-carers believed that age was a suitable measure of when CYP could take responsibility, and 14–18 years was an appropriate age; one exception was a parent of a teenager with learning difficulties.[41] However, whilst teenagers were keen to mitigate their asthma diagnosis as they moved into adulthood, few noted trigger avoidance in their mitigation strategy in an American study.[45]

Avoidance strategies noted by participants and influences on strategy uptake

Whilst beliefs and perceptions likely influence strategy uptake, other issues were apparent that suggested simple

information provision may not lead to uptake. For example, a cross-sectional survey of American parents (n = 638) of CYP (aged 3–16-year-olds with asthma), showed there was no association between previous trigger education (written or discussions in clinic) provision and exposure to triggers in the home. However, dog ownership was associated with lower parental education levels (odds ratio [OR]: 2.3; 95% confidence interval [CI]: 1.2–4.3). Similarly, household smoking was associated with low income (OR: 1.9; 95% CI: 1.0–3.7) and low parental education levels (OR: 4.5; 95% CI: 2.4–8.2). Also, there was no association between exposures and asthma symptoms but the authors did not control for medication use or inhaler technique.[57]

In a mixed methods study of 200 parent-caregivers of 9–12-year-olds with asthma, 77% described avoiding some triggers. However, when questioned about specific triggers, avoidance reports were low for pets (35%), tobacco smoke (29%), HDM (10%) and soft toy removal (14%; undertaken to reduce HDM exposures), and in qualitative interviews, increased cleaning, cleaning when children were not present and smoke-free rules were most frequently reported.[52]

Few articles mentioned parents' use of air purifiers and dehumidifiers,[27] with use, particularly on rainy days.[42] Few articles mentioned HDM-proof bedding,[31,42,51] with one citing parents' uncertainty regarding effectiveness.[42] Although some families reported the use of HDM-proof bedding, they also suggested other strategies that may help (such as carpet removal) but had not yet implemented this,[51] suggesting partial strategy implementation despite knowledge.

Two articles noted CYP knew that pets may trigger asthma and that this led to avoidance but that this was usually where CYP had other (non-asthma-related), allergic symptoms.[38,50] Partial avoidance strategies were also described, with participants disallowing pets into CYP's bedrooms.[44,50] However, how these strategic decisions were reached and their perceived effects on asthma control were not discussed.

An English sociological study of nine families suggested whilst most could identify some triggers, families did not always believe these were applicable. Families with pets either asked children to stay away from pets or made decisions to keep pets (e.g., rabbits) due to their child's emotional attachment.[58] However, it was unclear whether rabbits were kept outdoors. Another family kept cats and dogs despite believing their child may be allergic and felt they mitigated risks by hand washing. A further family timed removal of soft toys for a ‘deep freeze’ (to mitigate HDM exposure) to avoid upsetting their child. Some families noted triggers but did not enforce avoidance as this was considered more unsettling to family life than asthma.[58]

Inhaler use and trigger exposures

The use of reliever inhalers was mentioned in reaction to trigger exposures and for preparedness for potential exposures outside of the home.[45] However, others described that despite knowledge about asthma triggers, CYP did not always carry reliever inhalers,[32] and sometimes took risks related to triggers.[38]

Motivation and trigger avoidance

One study described using ‘structured interviews’ based upon attribution theory, to investigate causal attributions participants applied to explain self-management successes and failures. Both CYP (9–13 years) and parents-carers attributed trigger avoidance success and failure to predominantly internal, and personally controllable reasons. Whilst triggers were referred to broadly, rather than by individual types of trigger or allergen in the article, motivation was discussed. Both intrinsic motivation (including being observant of triggers) and effort for self-management were seen as causally related to self-management successes and failures by participants. Children attributed their trigger avoidance success and failure to mostly internal (85.9%–96.9%) and controllable (73%–93.2%) but unstable (69.2%–79.4%) causes. Parents also believed causes of successful or failed trigger avoidance were internal (79%–68.3%), mostly controllable (85.5%–54%) but unstable (59.7%–73%). However, external issues also impacted participants, for example, some exposures appeared especially challenging to avoid due to their abundance (e.g.,

pollen).[59,p.276]

Whilst no included studies aimed to explore what might motivate increased trigger avoidance, some studies briefly discussed motivation as a barrier to improving asthma self-management in their findings: Teenagers in a Swedish qualitative study were ambivalent about asthma self-care, as they attempted to balance managing asthma with maintaining social norms.[48] In an English qualitative study, some older teenagers described their indifference towards self-care and parents reported teenagers' low motivation and risk-taking behaviours as barriers to successful self-management.[50] However, one Canadian mixed methods study highlighted CYP's wish to learn from other slightly older adolescents with experience in managing allergies and asthma suggesting interaction may enhance self-management.[38]

Other studies of asthma self-management experiences suggested parents and/or CYP are often motivated to improve family management of asthma (including trigger management), but that other social, and familial challenges constrain the implementation of improvement strategies. A grounded theory approach describing the main concerns of 11–16-year-olds with asthma in Ireland, suggested CYP tested boundaries with trigger exposures and attempted to balance trigger management with engagement in activities with peers. However, CYP remained motivated to manage asthma.[47] A further grounded theory study identified that self-management involved families learning about symptoms and associated triggers and that they attempted to ‘catch the asthma before it got out of hand’.[27,p.359] In contrast, older teenagers have acknowledged taking risks with known triggers and needing support to assess risks safely.[38] Younger children (7–10 years) described known triggers but sometimes pushed themselves and ignored triggers to avoid appearing different or being harassed by peers.[55]

Following a qualitative study in England, a parental typology to describe asthma trigger management responses was developed. Parents were grouped as ‘preventors, reactors or compensators’: whilst all were motivated to preserve normality, the strategies and timing of implementation differed depending on whether parents attempted proactive, preventative trigger avoidance or compensated for exposures by implementing some exposure reduction strategies reactively or reacted to triggers only after an asthma exacerbation.[34,p.109]

Other barriers to avoidance

Costs of HDM-proof bedding were noted as a barrier to purchase by parents in one study.[33] Studies reporting recruitment from low-income groups or communities, identified other barriers to trigger avoidance strategy implementation. These included lack of control of overcrowding, financial constraints for pest control,[35] challenges with controlling shared environments and landlord refusal to support tenants with resolving these issues.[36] Parents in disadvantaged settings made as many environmental adaptions as possible (e.g., changing air-conditioning filters). However, CYP identified and prioritised emotional triggers, including the threat of neighbourhood violence, where parent-carers noted physical triggers.[49] Similarly, fear of neighbourhood violence and poor outdoor air quality limited CYP's time spent outdoors and deterred increased ventilation by opening windows.[36]

DISCUSSION

This scoping review was undertaken to outline the extent of current evidence on the influences on indoor environmental trigger avoidance at home, for CYP with asthma. Most of the included articles took a broad view of asthma self-management and explored many aspects beyond trigger management. This limits the extent to which the review questions could be answered in terms of detailed explanations of behaviours, yet this highlights research gaps. Three articles had aims focussed solely upon asthma triggers.[34,39,57] These studies provided insight into parent typological responses to CYP's asthma triggers,[34] the lack of association between advice to avoid triggers and parental uptake of avoidance[57] and reported racial inequity of receipt of avoidance information in an American pilot study of 12 parents.[39] However, all focussed on parent-carer perspectives and did not include CYP as participants.

Inclusion of CYP's perspectives could further understanding of the processes involved in strategy uptake decisions. Moreover, the processes involved in family decision-making regarding trigger avoidance were touched upon in included articles, but detailed explanations of behavioural influences remain unclear. This scarcity of in-depth, explanatory research on the topic is echoed in evidence syntheses of self-management practices and experiences of parent-carers of CYP with asthma,[60] and barriers and facilitators for successful self-management,[61–63] which had a greater focus on medication adherence than trigger avoidance adherence. Whilst this is unsurprising, given the importance of medication in asthma management, it remains challenging to develop evidence-based trigger exposure reduction interventions where current behaviours and behavioural influences remain unclear.

None of the included articles referred to the allergic sensitisation status of included participants (with exception of those with visible signs and symptoms of allergic reaction) or established whether sensitisation was understood and whether this may be related to avoidance strategy uptake. Although there is evidence of good parental recall of positive skin prick test results for allergen sensitivity, parents may not link exposures to aeroallergens (to which their child is sensitised) to acute asthma exacerbations.[64] Some included studies noted that according to parents, children did not recognise symptoms of deteriorating asthma control,[59] which for parents in one study, led to delayed asthma treatment.[42] Suboptimal adherence to asthma monitoring has also been reported.[63] Whilst an intervention to improve symptom and trigger recognition using home monitoring resulted in increased symptom recognition and trigger recognition, these increases were accompanied by a postintervention decrease in quality of life.[65] 'Being on alert' to asthma triggers was noted in a study included in the scoping review,[27,p.361] and others suggested this may increase the emotional burden of asthma management, through increased anxiety.[53] These complexly linked issues warrant further consideration in future intervention development.

Only one study noted the emotional value of pet keeping despite being a suspected trigger. However, the children's sensitisation status had not been confirmed.[58] Whilst evidence suggests few families (4.7%[6]; 8%[5]) rehome pets after advice to do so, greater clarity is needed to explain whether families understand the role of allergic sensitisation and related exposures in asthma control, as this may be a potential factor in pet-keeping decisions and may be considered alongside emotional gains of pet-keeping.

The included articles reporting ETS exposure at home as a trigger,[45,50] and first-hand CYP smoking,[47,50] included some of the most recently published studies. Smoking and ETS are well-established asthma triggers and have well-known causative detrimental effects for CYP.[13] Recent evidence showed an association between reduced asthma-related hospital admissions and Scotland's Take it Right Outside smoke-free home campaign,[66] suggesting a plausible correlation between reduced exposure and reduced exacerbations. However, smoking prevalence remains disproportionately higher in disadvantaged UK homes,[67] potentially placing CYP at risk of exposure. Thus, contemporary data for ETS exposure in homes of CYP with asthma remain important for the development of targeted interventions to reduce exposures. Environmental vapour from electronic cigarettes or similar devices has also emerged in surveys with adults and adolescents with asthma in the United Kingdom as a potential trigger,[68] and maybe an area for further exploration amongst CYP with asthma who may be exposed.

Strengths and limitations

This review sought to provide a high-level overview of what was known about beliefs, and other factors influencing avoidance of indoor environmental asthma triggers. However, there are many asthma triggers and some act in synergy, for example co-existing viral infection and allergen exposure reduce asthma control and increase the risk of hospital admission.[69] Greater understanding of family experiences and perceptions about such synergistic effects may be beneficial for the promotion of trigger avoidance interventions.

The main strengths of this review are the broad search strategy employed to minimise risks of missing relevant

articles and the subsequent identification of research gaps. Although the review focus is indoor environmental triggers, CYP prioritised emotional triggers, where parent-carers appeared to prioritise environmental triggers in one study[49]; whether this relates to strategy prioritisation or uptake is also of interest, particularly if families may not discuss triggers amongst family members, a factor which the included articles did not determine.

The search limitations applied were specific to the broader aims of the project this review sits within and sought to balance breadth with practicality, as guidance recommends.[18] Limiting searches to English language full texts may have introduced language bias.[70] However, where abstracts were available in English, there were none that would have necessitated full-text translation, when assessed against eligibility criteria. Included studies were from high-income countries, which may relate to exclusion of articles in other languages or may reflect other publication biases. Moreover, it may reflect the scarcity of evidence on the topic.

Most scoping reviews do not encompass quality appraisal,[71] despite methodological debates over this and continued critique of appraisal absence.[17,72] The overarching aim was to establish whether there was sufficient evidence to describe factors influencing low uptake of avoidance strategies and had there been sufficient evidence, quality appraisal could have informed whether the evidence was robust enough to begin intervention development. However, due to the scarcity of explanatory evidence for current behaviours, it was concluded more research is needed. Consequently, this review has not included quality appraisal. Yet it is notable that no included studies reported patient and public involvement (PPI) although two used participatory methods.[40,49] Stakeholder consultation [16] for this review was considered but not undertaken due to the project timelines, author expertise and use of a search strategy that sought only published empirical evidence.

CONCLUSIONS

Myths, misconceptions and challenges associated with trigger identification or risk attribution remain for some families and could inform avoidance strategy uptake. Families living with socioeconomic disadvantages often face additional barriers. For those able to identify triggers, and with access to medical advice, strategy uptake appears variable and sometimes partial, which appears to reflect the complexities of balancing other family demands by parent-carers,[27,34,53] and the CYP's desire to live lives close to those of their peers without asthma.[30,47,48,55] Failure to either implement or report the use of behavioural change theory for asthma trigger reduction intervention planning, development and implementation have been acknowledged.[7,9] Future research should aim to elucidate the influences on behaviours to inform the appropriate choice of behavioural theory for interventions. As intervention acceptability and effectiveness are maximised when they are ‘person-based’,[73] such research ahead of intervention development would benefit from PPI and in-depth qualitative study. Further exploratory research focussed on family understanding of allergic sensitisation, indoor environmental asthma triggers related perceived asthma control, and what may motivate increased avoidance, are necessary to inform targeted family-centred interventions applicable to home settings.

AUTHOR CONTRIBUTIONS

Grace Lewis had significant involvement in the review design, extraction, and interpretation of data and the first and subsequent drafts of the review manuscript. Linda Milnes, Alistair Duff, Jürgen Schwarze and Alexandra Adams provided guidance for the design and delivery of the review. Linda Milnes provided guidance for Grace Lewis to conduct searches. Grace Lewis and Linda Milnes conducted article selection and data extraction according to a protocol agreed upon by all authors. Alistair Duff, Jürgen Schwarze and Alexandra Adams were consulted for any disagreements in the article selection or data extraction. All authors provided final approval for publication of the manuscript.

ACKNOWLEDGEMENT

This work is funded by Asthma + Lung UK as part of Asthma UK Centre for Applied Research AUK-AC-2018-01.

The authors wish to acknowledge the support and advice from academic librarians at the Univeristy of Leeds.

CONFLICT OF INTEREST

The author declares no conflict of interest.

DATA AVAILABILITY STATEMENT

No primary data are available to share.

DETAILS

Subject:	Allergens; Parents &parenting; Asthma; Risk reduction; Health beliefs; Intervention; Databases; Families &family life; Allergies; Remediation; Young adults; Exposure; Avoidance behavior; Decision making; Motivation; Heterogeneity; Indoor environments; Children; Citizen participation; Public involvement; Searching; Uptake; Caregivers; Avoidance; Youth
Location:	United Kingdom--UK
Identifier / keyword:	allergic sensitisation; asthma; asthma triggers; behavioural influences; children and young people; parent-carer; scoping review
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	87-97
Publication year:	2023
Publication date:	Feb 2023
Section:	REVIEW ARTICLES
Publisher:	John Wil ey &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English

Developing a Health Literacy Scale for adults in Hong Kong: A modified e-Delphi study with healthcare consumers and providers

Cindy Yue Tian [1]
; Eliza Lai-Yi Wong [2]
; Xu, Richard H [3] ; Cheung, Annie Wai-Ling [2] ; Dong, Dong [2]
; Phoenix K.-H. Mo [2]

[1] JC School of Public Health and Primary Care, Faculty of Medicine, The Chinese University of Hong Kong, Hong Kong, China [2] JC School of Public Health and Primary Care, Faculty of Medicine, The Chinese University of Hong Kong, Hong Kong, China; Center for Health Systems and Policy Research, JC School of Public Health and Primary Care, Centre for Health Systems and Policy Research, The Chinese University of Hong Kong, Hong Kong, China [3] Department of Rehabilitation Sciences, The Hong Kong Polytechnic University, Hong Kong, China

ABSTRACT (ENGLISH)

Introduction

Health literacy (HL) refers to individuals' abilities to process and use health information to promote health. This study aimed to develop the first HL measurement tool for the Chinese Hong Kong population.

Methods

A two-phase methodology was adopted. In *Phase I*, evidence synthesis with a deductive method was conducted to formulate the item list from the literature. In *Phase II*, a modified e-Delphi survey was conducted among stakeholders (i.e., healthcare providers and healthcare consumers) to confirm the content validity of the item list. The stakeholders were invited to rate the relevance of each draft item on a 4-point scale and provide suggestions for revisions, removal or adding new items.

Results

In *Phase I*, a total of 34 items covering functional, interactive and critical HL were generated. In *Phase II*, to obtain a balanced view from experts and laypeople, healthcare professionals ($n=12$) and consumers ($n=12$) were invited to participate in the Delphi panel. The response rates of the three rounds were 100%. After the third round, the consensus was reached for 31 items, and no further comments for adding or revising items were received. All items exhibited excellent content validity (item content validity index: 0.79–1.00; K^*: 0.74–1.00).

Conclusions

A Health Literacy Scale for Hong Kong was developed. Compared with existing HL scales, the scale fully operationalized the skills involved in functional, interactive and critical HL. The Delphi study shows evidence supporting the high content validity of all items in the scale. In future studies, these items should undergo rigorous testing to examine their psychometric properties in our target population groups. By illuminating the details in the development process, this paper provides a deeper understanding of the scale's scope and limitations for others who are interested in using this tool.

Patient or Public Contribution

Public as healthcare consumers, in addition to healthcare providers, were involved in developing a new HL scale for this study. The input from the public contributed to examining the scale's content validity by judging whether all items reflected the skills that they need to find and use health-related information in their daily life.

FULL TEXT

INTRODUCTION

Health literacy (HL) is defined as an individual's capacity to obtain and process health information to promote health.[1] It can contribute to how people interpret symptoms and participate in health-related decision-making. Limited HL has consistently been associated with poorer self-reported health,[2,3] lower health-related quality of life,[4] less use of preventive health services,[5] increased hospitalizations[6] and higher healthcare costs.[7,8] Many national surveys have highlighted high rates of poor HL in populations.[9–12] Previous systematic reviews indicated that the prevalence of low HL in Europe ranged from 27% to 48%,[13] while in Southeast Asia it ranged from 1.6% to 99.5% with a mean of 55.3%,[14] depending on the literacy measurement method applied. The most common factors associated with insufficient HL include educational attainment, age, income and ethnicity.[13,14]

Identifying a relevant measurement is critical for examining HL levels. Early efforts to measure HL primarily focussed on individuals' abilities to read and comprehend health-related materials in a clinical setting.[15,16] With healthcare shifting from a clinical setting to a community setting, more recently developed measurement tools measure a broader understanding of HL, which includes a set of competencies (e.g., information-seeking skills, communication skills and decision-making skills) needed to facilitate health decision-making in both clinical and nonclinical settings.[17–20] Although over 100 HL scales (HLSs) have been developed, no widely adopted measurement tool could reflect our current understanding of HL.[21–23] Taking the most cited HL tools as examples, the Test of Functional Health Literacy,[15] Rapid Estimate of Adult Literacy in Medicine and Newest Vital Sign[16] narrowly measure basic skills and knowledge of health; The Health Literacy Questionnaire[17] did not include the ability to address the broader goal of

promoting health and reducing health disparities among individuals and communities. Moreover, most available HL tools were developed in Western countries.[24] Hence, discussion about HL scale development in Asia is still needed. Scholars have argued that a robust HL scale should allow for discovering new knowledge and testing what we know from previous studies to advance this field.[23,25,26] Therefore, using a testable theory to support the creation of a new scale is vital. The present study is based on Nutbeam's framework of HL, which is widely used in this research area. This framework divides the main skills associated with HL into three levels: functional health literacy (FHL) referring to individuals' basic literacy and numeracy skills (e.g., being able to read and write, basic knowledge of health) to access and act upon health-related materials; interactive health literacy (IHL) referring to individuals' cognitive and social skills to extract information from all kinds of forms of communication and to interact with information providers for achieving better health outcomes (e.g., searching for online health information and requesting clarification during healthcare consulting) and critical health literacy (CHL), which refers to individuals' higher level cognitive and social skills which can be applied to critically analyse information, and to use this information to gain better control over life events that impact health, such as disease management and health promotion.[27] Nutbeam's framework synthesizes HL skills in a comprehensive way compared to other frameworks used in HL research. For instance, the Chinese Resident Health Literacy Scale adopted ‘basic knowledge and skills of people's health’ as the underlying structure, which mainly covered the skills involved in FHL.[28,29] The European Health Literacy Survey Questionnaire used Sørensen et al.s'[19] theoretical model of ‘the competencies needed in the information processing’. The authors of this European scale admitted that the scale could not thoroughly assess an individual's ability to use the information to promote health, which is addressed in CHL.[19]

However, compared with FHL and IHL, CHL is not fully operationalized in current HL scales. As of writing, six scales covering Nutbeam's framework[30–35] and one scale measuring the single domain CHL[36] for adults have been published. These studies[30–36] mainly emphasized the ability involved in critical appraisal of information as the component of CHL. This emphasis, however, was not explicitly linked to the theory of this domain. Nutbeam initially highlighted that CHL includes not only the ability to critically assess the quality of information but also a range of competencies to enable individuals to realize social and structural factors influencing health and take actions to address these factors for better health.[27] Among the above scales, only the All Aspect of Health Literacy Scale[30] made efforts to examine the missing components of CHL: namely, knowledge of and actions to address social determinants of health. But the author admitted that there exist challenges to address this shortage. As such they only adopted three items involved in the capabilities for community empowerment and social engagement for health to indirectly reflect these understandings and actions.[30] The above revealed that continuous discussion on effectively measuring this domain among adults is still needed.

In addition, there is no rigorously validated HL scale for the general population in Hong Kong. Although several studies explored the HL levels in Hong Kong, the scales they used were either condition-specific (i.e., disease-specific and population-specific)[37–41] or directly translated from existing scales without psychometric testing.[42,43] Hong Kong has a dual-track healthcare system encompassing the public and private sectors. The downsides of the two sectors are the long waiting times experienced in public hospitals and high healthcare costs in private hospitals.[44,45] Under such circumstances, patients are expected to actively engage in self-management, which requires a high HL level. It is reasonable to assume that patients with sufficient HL skills are more likely to understand their symptoms and be able to decide when and what healthcare service to utilize in the health system. Therefore, one reliable and valid HL scale is essential to understand residents' HL levels and design research-based strategies to enhance HL in the local health system.

From all these perspectives, we aimed to develop a validated theoretical-based HL scale (HLS-HK) by adopting Nutbeam's framework[21–26] in Hong Kong. Previous studies mainly invited healthcare professionals to design HL scales.[16,33,46–48] Considering HL is a critical component of people-centred health care, which demands participation from the healthcare provider and consumer side,[49,50] we included healthcare providers and consumers in the scale development process. The purpose of this paper is therefore to highlight the development process and the content validity of the HLS-HK via a modified e-Delphi technique.

METHODS

The Delphi technique is a systematic and interactive method to achieve a general agreement or convergence of opinions on a particular topic.[51] It has proven to be a reliable method to develop new concepts[52] and establish consensus across a range of subject areas,[53] including several in the field of HL measurements.[19,54–56] In the present study, two phases were conducted: (a) item development of HLS-HK by evidence synthesis using a deductive method and (b) content validity of HLS-HK employing a modified e-Delphi survey with healthcare consumers and providers.

Phase I: Item development

A deductive method[43] was used to generate items based on our previous two scoping reviews.[57,58]

Theoretical framework

We conducted two scoping reviews[57,58] to ensure the scope and coverage of the scale with the adoption of Nutbeam's framework. The first scoping review synthesized how Nutbeam's framework was operationalized in current HL scales.[57] Given that CHL is the least well-developed domain in Nutbeam's model, we conducted another scoping review to understand the components that need to be measured in this domain. By doing so, the following three subdomains of CHL were identified: CHL-1: ‘critical appraisal of information’ is an individual's ability to evaluate the quality of information; CHL-2: ‘understanding of social determinants of health’ coveys individual's understanding of the relationship between how people experience social determinants and the impact of these determinants on health; CHL-3: ‘actions to address social determinants of health’ focusses on individual's competency to translate knowledge into actions to address the modifiable determinants of health.[58] To sum up, a framework within five content areas (i.e., FHL, IHL and three subdomains of CHL) of this newly developed scale was developed.

Item generation

Then, we turned these five abstract contents into measurable observations. A deductive analysis with the following three steps was performed to generate items: (1) sample: choosing reliable and validated scales with the indicators of interest from the two scoping reviews[57,58]; (2) coding: labelling the content of identified items and then grouping the labels into content categories; (3) results: the final content categories served as the template for the generation of an item pool. The three-step process was conducted by two researchers, and agreement was achieved through discussion with the research team. To ensure the coverage and minimize the cognitive burden, the number of items was expected to be between 30 and 50.

Phase II: Modified e-Delphi study

A modified e-Delphi survey was conducted to assess the content validity[59] of items developed from Phase I.

Participants

In Delphi exercises, 10–18 respondents are suggested as sufficient for ensuring consensus.[60–62] We assembled a panel composed of healthcare providers (Group A) and healthcare consumers (Group B) via nonprobability purposive sampling. Regarding the inclusion criteria, according to Hasson et al.s'[63] suggestion, participants in Group A were required to be healthcare professionals or clinical workers who had been working in the health field for ≥5 years. In Group B, participants were required to be permanent citizens aged ≥18 years and have experience in seeking health-related information. Given that everyone should need healthcare information at some point, we proposed that every citizen could be a participant in Group B. To achieve a representative sample, we selected participants by considering a balance of different professional disciplines in Group A and a balance of gender, age and educational attainment in Group B. To keep the recruitment costs low, for Group A, we invited doctors and nurses from one public hospital and professors with experience in health-related research from one local university. For Group B, we approached citizens who may be interested in joining our study, including staff and students in the local university and people who work outside the university. We expected at least three rounds of exercise to complete the Delphi process. Participants were required to take part in all three rounds. Therefore, if they did not respond to Round 2, they were not invited to participate in Round 3. This study aimed to recruit and complete the process with 20 participants and 10 respondents for each group.

e-Delphi rounds

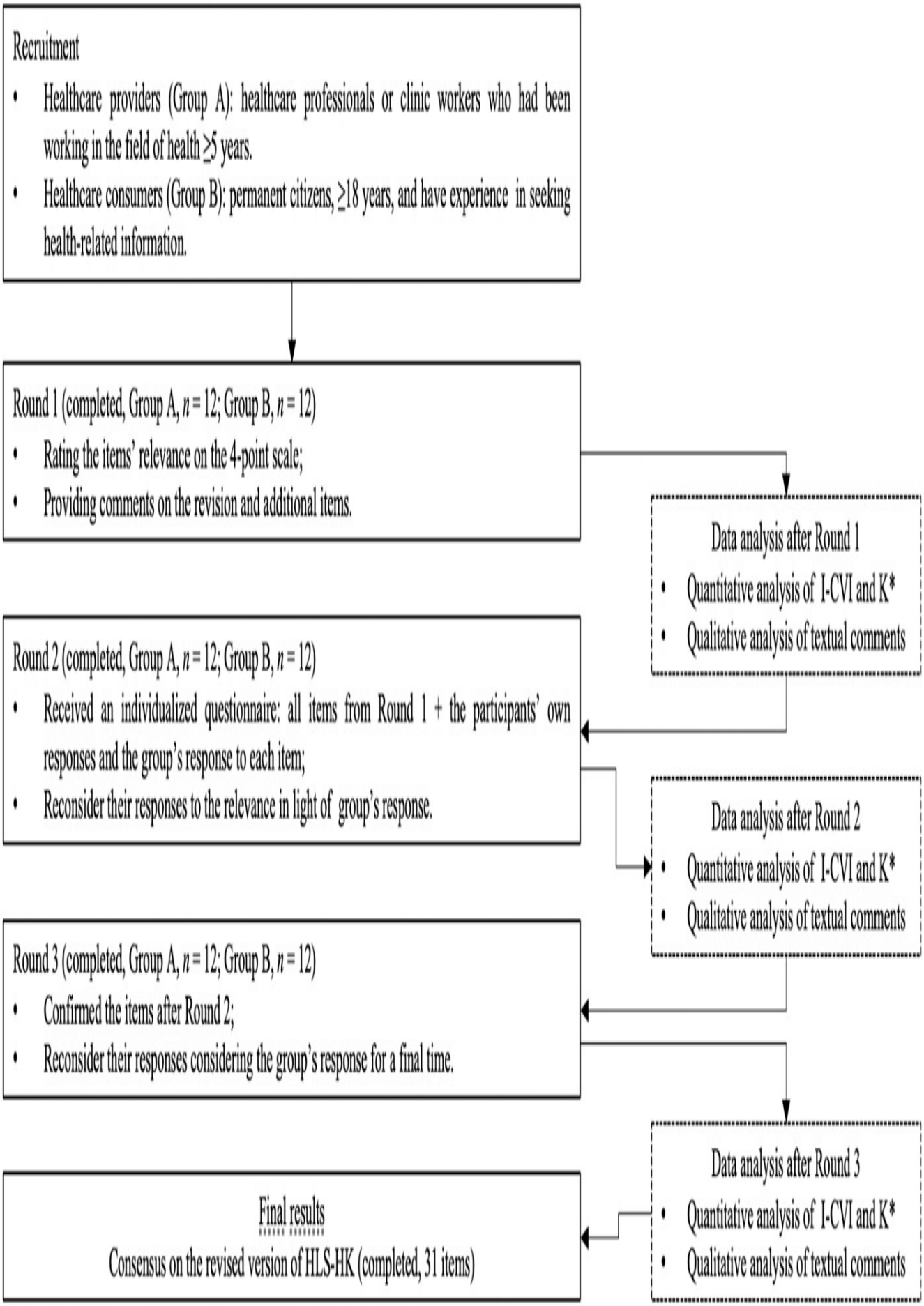
We used Qualtrics software (version August 2021)[64] to develop the online three-round survey and invited potential participants via email or face-to-face. A 4-point Likert-type scale (ranging from 1 = not at all relevant to 4 = extremely relevant) was used to determine raters' agreement on item relevance. Ratings of 1 and 2 were considered 'not relevant', whereas ratings of 3 and 4 were considered 'relevant' as in most studies.[65,66] Additionally, text boxes were provided in the scale for raters to include comments and suggestions.

In Round 1, participants were asked to independently rate each drafted item for relevance on a 4-point scale. They were also encouraged to add free-text comments on the scale's design, clarity and content and suggest additional items that may be used to measure HL skills based on their knowledge and experience. Data on participants' demographics and expertise were also collected in this round. In Round 2, all participants received an individualized questionnaire that included all items from Round 1 which occurred alongside the participants' own responses and all participants' responses to each item. Participants were asked to reconsider their responses in light of the two groups' responses and item modification. Based on the comments we received from the previous round, we revised items and highlighted the changes in the questionnaire for rerating in this round. Additionally, the results on item relevance and a summary of comments of the previous round were provided to the panellists in Supporting Information: Appendix. In Round 3, each participant was asked to confirm the items after the previous round and reconsider their responses, considering the groups' responses for a final time. We also provided a summary of comments and highlighted the item modification from the previous round.

Data analysis

Data analysis was carried out between each round using Microsoft Excel (version 16.63.1).[67] Two approaches were used to calculate content validity. Item content validity index (I-CVI) is the proportion of items that received a rating of 3 or 4 in terms of relevance by panellists. It can be calculated by using this formula: I-CVI = A/N (N is the number of panellists; A is the number of panellists who agree it is relevant). It is recommended that if the I-CVI >0.79, the item is appropriate; if it is between 0.70 and 0.79, the item needs revision; and if the value is below 0.70, the content validity of the item is not acceptable and the item is eliminated.[65,68] Although I-CVI is widely used to estimate content validity, the index does not consider the possibility of chance agreement. The second approach was the Kappa statistic (K^*) which adjusts for chance agreement by examining interrater agreement. To calculate Kappa, the probability of chance agreement was first calculated for each item by the following formula: $p_c = (N!/A!\,[N-A]!) \times 0.5^N$. After calculating I-CVI for all items, Kappa can be computed by using the following formula: $K^* = (\text{I-CVI} - p_c)/(1 - p_c)$. Evaluation criteria for Kappa are as follows: if the values are larger than 0.74, between 0.70 and 0.74, and between 0.40 and 0.69 are considered as excellent, good and fair content validity, respectively.[69] If the K^* is equal to or above 0.70, the content validity of the item is acceptable. After each round, qualitative data were analysed and interpreted to clarify and confirm consensus around the wording.

A consensus was defined as ≥70% of all participants agreeing that one item is relevant in Round 3. We recruited the same number of participants in the two groups. We considered that all participants' responses were weighted equally, as with most studies.[70,71] In this way, the consensus could be achieved while avoiding the impact of dominant individuals and groups. Figure 1 provides a summary of the Delphi process.



Enlarge this image.

RESULTSPhase I

Using a three-step deductive process, we identified seven tools[30,31,35,48,72–74] and consolidated 34 items into the 5

relevant content categories (see Supporting Information: Appendix 1). Given that the items were originally formulated in English, a forward–backward translation was produced by four bilingual translators (two translators for each translation). After that, we performed one review meeting among the research team to determine the primary version of HLS-HK in traditional Chinese.

Phase II

For the modified e-Delphi Survey, a total of 24 experts from Group A and Group B participated in the survey from August to October 2021. All of them completed all three rounds of the survey with response rates of 100%. Table 1 presents the demographic characteristics of the respondents. The experts in Group A (*n* = 12) included six doctors, one nurse, two public health professors, two nursing professors and one social science professor. The participants in Group B (*n* = 12) covered two postdoctoral fellows, two university students and eight workers outside of academia. The three-round survey indicated that the scale has good content validity (see Table 2). The consensus was reached for finalizing 31 items after three rounds (see Table 3). The wording changes and final Chinese version of the HLS-HK can be found in Supporting Information: Appendices 2 and 3, respectively.

Table 1 Demographic characteristics of panellists

	Group A healthcare provider (*n* = 12)	Group B healthcare consumer (*n* = 12)	Total (*n* = 24)
Gender			
Male	9 (75.0%)	5 (42.0%)	14 (58.3%)
Female	3 (25.0%)	7 (58.0%)	10 (41.7%)
Age group			
18–29	0 (0.0%)	4 (33.3%)	4 (16.7%)
30–49	8 (66.7%)	5 (41.7%)	13 (54.2%)
≥50	4 (33.3%)	3 (25.0%)	7 (29.1%)
Education attainment			
Secondary or below	0 (0.0%)	2 (16.7%)	2 (8.3%)
Postsecondary (diploma/certificate course)	0 (0.0%)	1 (8.3%)	1 (4.2%)
Postsecondary (degree course)	12 (100.0%)	9 (75.0%)	21 (87.5%)
Diagnosed chronic disease			
Yes	5 (41.7%)	2 (16.7%)	7 (29.2%)

No	7 (58.3%)	10 (83.3%)	17 (70.8%)
Main work setting			
Academia	5 (41.7%)	2 (16.7%)	7 (29.2%)
Clinic	7 (58.3%)	0 (0.0%)	7 (29.2%)
Industry	0 (0.0%)	8 (67.7%)	8 (33.3%)
Othersa	0 (0.0%)	2 (16.7%)	2 (8.3%)

a

Others refers to students.

Table 2 Content validity of items included in the scale (three-round survey)

	I-CVI	*K**	No. of items					
			FHL	IHL	CHL-1	CHL-2	CHL-3	Total
Round 1	0.75–0.96	0.74–1.00	5	7	7	9	6	34
Round 2	0.79–1.00	0.74–1.00	5	7	6	9	6	33
Round 3	0.79–1.00	0.79–1.00	5	7	6	7	6	31

Abbreviations: FHL, functional health literacy; I-CVI, Item content validity index; IHL, interactive health literacy.

Table 3 Content validity of items included in the scale (Round 3)

Domain	No.	Item	I-CVI	K*	Interpretation
FHL	1	*How often do you*[a]: …need help when you are given information to read by your doctor, nurse or pharmacist	0.88	0.87	Excellent
	2	…need help when you are asked to fill out medical forms by your doctor, nurse or pharmacist	0.88	0.87	Excellent
	3	…find that characters cannot understand when you read instructions or leaflets from hospitals or clinics	0.79	0.79	Excellent
	4	…feel that the content is too difficult to understand when you read instructions or leaflets from hospitals or clinics	0.92	0.92	Excellent

	5	…have problems learning about your medical condition because of difficulty understanding health-related written information	0.96	0.96	Excellent
IHL	6	*How easy would you say it is to*[b]: …find related information when you have questions on disease or health problems	0.96	0.96	Excellent
	7	…find related information when you are not ill but want to do something to further improve your health	0.96	0.96	Excellent
	8	…give all the information a doctor, nurse or pharmacist need when you talk to them	0.88	0.87	Excellent
	9	…ask the questions you want to ask when you talk to a doctor, nurse or pharmacist	0.96	0.96	Excellent
	10	…extract the information you want when you talk to a doctor, nurse or pharmacist	0.96	0.96	Excellent
	11	…ask a doctor, nurse or pharmacist to further explain anything that you do not understand after talking with them	0.92	0.92	Excellent
	12	…understand the obtained information when you talk to a doctor, nurse or pharmacist	1.00	1.00	Excellent
CHL-1	13	*When you get information for health in daily life, how often do you consider the following*[c]: …whether the information source is credible	0.96	0.96	Excellent
	14	…whether the information content is valid and reliable	0.83	0.83	Excellent
	15	…whether the publish time is appropriate	0.79	0.79	Excellent
	16	…whether other reliable sources support the facts or conclusions of this source	0.88	0.87	Excellent
	17	…whether the person or organization that produced the information have a bias	0.83	0.83	Excellent
	18	…whether the information is applicable to you	0.83	0.83	Excellent
CHL-2	19	*How do you agree about the following*[d]: …socioeconomic status affects health	0.92	0.92	ExcellentExcellent

	20	…stress affects health	0.96	0.96	Excellent
	21	…being isolated from the community and workplace impacts health	0.92	0.92	Excellent
	22	…having little control over one's work impacts health	0.92	0.92	Excellent
	23	…poor childhood experience has an impact on one's physical/mental health when he or she becomes an adult	0.92	0.92	Excellent
	24	…good social relations contribute to health	0.96	0.96	Excellent
	25	…transportations impacts health	0.96	0.96	Excellent
CHL-3	26	*How often do you*[e]: …participate in government's programmes about health promotion and disease prevention	0.83	0.83	Excellent
	27	…participate in community's initiatives in health promotion and disease prevention	0.96	0.96	Excellent
	28	…participate in nongovernmental organizations' initiatives in health promotion and disease prevention	0.88	0.87	Excellent
	29	…help your family members or a friend when they had questions concerning health issues	0.96	0.96	Excellent
	30	…seek information from others when you come up with questions concerning a health issue	0.92	0.92	Excellent
	31	…share and communicate your opinion about illness when you talk to a family member or friend	0.92	0.92	Excellent

Note: a, response options range from '1 = always' to '5 = never'; b, response options range from '1 = very difficult' to '5 = very easy'; c, response options range from '1 = never' to '5 = always'; d, response options range from '1 = strongly disagree' to '5 = strongly agree'; d, response options range from '1 = never' to '5 = always'.
Abbreviations: FHL, functional health literacy; IHL, interactive health literacy.

Round 1

In Round 1, all 34 items were content-validated (I-CVI: 0.75–0.96; K^* = 0.74–1.00) (see Table 2) based on the responses of all participants. Only one draft item on 'Whether think about the information is valid' possessed low content validity (I-CVI <0.79, K^* = 0.74). This may have been caused by the difficulty in differentiating it from another draft item on 'Whether think about the information is reliable' in Chinese, as the majority of experts and laypeople highlighted. Thus, we combined these two items into one item (i.e., No. 14) to become 'Whether think about the information is valid and reliable'. In addition, several items (i.e., No. 3, 4, 11, 15) were revised or rephrased since the panel members remarked that their wording remained vague or inappropriate in the text box. For instance, for one item in FHL (i.e., No. 3), one professor in Group A commented: 'The scenario mentioned was not suitable in the local context. Citizens often need to read these instructions or leaflets from hospital and clinic, instead of pharmacy'.

Thus, we changed ‘pharmacy’ to ‘clinic’ for the item. We added several examples to make certain items (i.e., No. 2, 5.8, 9, 23, 25) more specific as suggested by participants. Finally, a total of 33 items from 34 items were retained after Round 1.

Round 2

The 33 items were rerated in Round 2 and content validities improved (I-CVI: 0.79–1.00; *K**: 0.74–1.00). In terms of the clarity on items, we mainly received positive comments. However, three items ‘How do you agree about the lesser the income the greater the tendency to become ill’, ‘How do you agree about socially vulnerable groups more likely turn to alcohol, drugs, and tobacco to relieve the pain of harsh economic and social conditions’ and ‘How do you agree about socially vulnerable groups more likely have no good eating habits and inadequate food supply to promote health and well-being’ were criticized because of the overlapping and different interpretations of ‘socially vulnerable groups’. Therefore, we combined the three items into one item, ‘How do you agree about socioeconomic status affects health’, to make the item content more precise. Thus, the HLS-HK included a total of 31 items from 33 items after Round 2.

Round 3

In Round 3, each participant was asked to confirm the relevance of those items without changes and rerate the relevance with regard to the newly combined item resulting from Round 2. Eventually, for each item, over 70% of all participants agreed that it was relevant in Round 3. Thus, consensus was achieved for individual items and coverage. All 31 times showed excellent content validity (I-CVI: 0.79–1.00; *K**: 0.74–1.00) (see Table 2). We did not receive any further comments for adding or removing or revising items during this round. Thus, the Delphi exercise concluded with three rounds.

DISCUSSION

A validated and theoretically based HL scale, HLS-HK was developed through a rigorous and systematic deductive approach and a modified e-Delphi survey.

Bridging measurement gap

In comparison with the scales[30–35] based on Nutbeam's framework, the HLS-HK fully operationalized the three content areas (i.e., FHL, IHL and CHL) in this framework. In the domain of FHL, we formulated five items to examine individuals' skills to read information, fill out forms and understand health-related materials in healthcare settings. To measure the level of IHL, seven items were built to examine individuals' competencies to search for health-related information and effectively communicate with healthcare workers.

More importantly, this scale bridged the measurement gap in the domain of CHL by providing the following multilevel subdomains. In the subdomain of CHL-1, instead of simply asking the frequency to assess the trustworthiness of information like previous scales,[30,35] we generated seven items to assess subjects' behaviours to critically appraise information in terms of its resources, contents, publication date and publisher. Regarding the CHL-2, as mentioned earlier, the knowledge of how social structural factors affect health was rarely thoroughly measured in HL measurement tools. By learning from one Japanese HL scale,[48] we formulated seven items to directly test participants' knowledge about the impact of several significant social determinants of health. With respect to CHL-3, we found that most of the current HL scales[30,74] only considered an individual's ‘collective action for health’ (i.e., collective efforts to create and preserve public goods, such as a clean environment and herd immunity) as the component of CHL. This might be because the current measurements were mainly developed in Western countries (e.g., the United States and Australia), where people are more open to social action or democratic participation. In this case, only focusing on ‘collective action for health’ cannot fully capture the CHL-3 level of some population groups who have low interest in social movements or limited resources to participate, such as Hong Kong. Thus, we generated three new items (i.e., No. 29–31) to address social determinants of health at the interpersonal level (i.e., creating a supportive social network for health). In fact, the importance of abilities informing interpersonal level actions to address social determinants was addressed in one CHL scale targeting adolescents in Norway.[75] However, those abilities were measured through items related to positive self-beliefs to cope with a variety of situations to promote health in their social network and communities (e.g., ‘I am a person that can share information

on factors that influence health with others'), rather than the real actions as our scale has done. However, we did encounter the challenges to thoroughly measuring CHL, which we have discussed below.

Including opinions of healthcare providers and consumers

Compared with the traditional Delphi method of only recruiting experts into the panel, we included healthcare professionals and the general population, who both play crucial roles in health-related research. To achieve a representative sample, we recruited healthcare providers with diverse professional disciplines and laypeople with a balanced distribution of age and gender. In the progress, we made use of the opinions of all the agents involved and considered them all to be equal in the three-round procedure. The two groups, however, did share different points of view on certain items which may be influenced by their professional or personal experience. Moreover, these differences are mainly reflected in CHL. For example, laymen representatives and healthcare professionals disagreed on item No. 18 (i.e., 'Whether think about the information is applicable to you'). Laypeople mentioned that they usually randomly read the information during their daily life and did not think it is necessary to assess its applicability. By contrast, most experts commented that people should contextualize information for their own good and take actions after fully appraising the information in their own world. This disagreement might be explained by previous studies' findings, that is, even though people might know the strategies to check the quality of information, they do not routinely use these.[76,77] Hence, it is a question of how the scholarly discourse on information appraisal informs people's daily practice and reflects their relevant abilities. Another example is item No. 19 (i.e., 'How do you agree with socioeconomic affects health'). Laypeople acknowledged the impact of socioeconomic factors but tend to feel that individuals' behaviours have a greater impact on health, while experts can thoroughly understand the influence of socioeconomic factors by analysing them from the perspective of health inequities. With relation to this point, Chinn[78] suggested that asking about people's awareness of social determinants of health is methodologically tricky. Individuals who might struggle to link social disadvantage and health, are perhaps more likely to express such ideas through a contextualized narrative description of their own life experience instead of completing a fixed-choice question.[78] However, a narrative interview is a time-consuming procedure that may not be applicable in a busy clinical setting. The above arguments about CHL indicated the complexities in operationalizing of this domain in a real-world setting. We hope our work contributes to further exploring this operationalization from the laypeople and scholars' conceptions.

Implications

Based on the detailed literature review and our rigorous deductive approach, we extended Nutbeams' conceptual framework with 31 items. In the item generation process, we asked stakeholders' opinions to make sure our scale is content-validated and user-friendly. This is critical to build a native measurement and support local researchers, policymakers and practitioners to use this scale for relevant studies and health programmes. These items will undergo further rigorous testing in our target population groups in future studies. Other researchers can use or amend our scale for their research interests and validate the items in various settings and populations. It is thus reasonable to assume that our work can contribute to the further refinement of this conceptual model.

Limitations

Study limitations include the following: First, although we asked experts and laypeople to suggest additional items in the three-round survey, no new items were added by them. This might be insufficient to create a tool that captures all skills related to HL. To enhance the comprehensiveness of a new tool, inductive methods (e.g., in-depth interviews and focus groups) could be used in *Phase I*. Second, the decision to use an agreement index threshold of 0.70 used in this study was arbitrary. Owing to the diversity of topics covered by the Delphi method, there is no standard threshold for determining consensus.[79] This study chose an acceptable threshold, as has been carried out in most studies.[55,80–82] Third, the panel members could not directly discuss any concerns or exchange opinions with other panellists because we conducted the Delphi study online. Although we provided feedback at the conclusion of each round, a structured meeting after the first two rounds may facilitate deeper discussions among the panel members. Fourth, selection biases might exist in the Delphi panellists because we conducted a nonprobability sampling technique. For example, although we intended to achieve a balance of education attainment

in Group B, the actual proportion of the participants who were well-educated was high because sufficient reading levels and cognitive skills were needed to judge the reference of each item. To make sure the scale is suitable to use in the entire population, we will examine its psychometric proprieties among the general population using quota sampling. Additionally, we only included healthcare scholars and clinical workers in Group A because of limited resources. To achieve a deeper understanding of the skills that people need to find and use health-related information in various settings, future studies should consider including a broader range of healthcare providers (e.g., allied health) in Group A. Fifth, the lack of item deduction in this Delphi process highlights the need for future studies such as cognitive interviews and psychometric properties testing to achieve further item reduction.

CONCLUSION

By combining a literature review and a Delphi survey, this study identified a set of content validity items for the HLS-HK. Specifically, the review ensured that all draft items were generated based on scientific evidence. The mixed method approach using a three-round survey provided quantitative and qualitative data which led to item modification and improved content validity. Compared with previous HL scales, this newly developed scale fully operationalized the skills involved in FHL, IHL and CHL. It is useful to examine people's HL levels and identify the barriers that they may encounter in processing health-related information to make appropriate health-related decisions. The next steps in the research will involve testing its face validity for respondents, and psychometric properties to identify its final version and more parsimonious form.

AUTHOR CONTRIBUTIONS

Eliza Lai-Yi Wong, Phoenix K.-H. Mo, Dong Dong and Cindy Yue Tian designed the Delphi study and acquired funding. Cindy Yue Tian performed the literature review and three-round survey, analysed and synthesized the data and wrote the draft manuscript. Eliza Lai-Yi Wong was responsible for data analysis, data curation and project administration. Dong Dong and Phoenix K.-H. Mo were in charge of project administration and supervision. Richard H. Xu provided guidance on the three-round surveys. Annie Wai-Ling Cheung assisted with the funding acquisition and project administration. Eliza Lai-Yi Wong, Phoenix K.-H. Mo, Dong Dong, Richard H. Xu and Annie Wai-Ling Cheung commented and edited the whole draft. Cindy Yue Tian critically revised the manuscript. All authors read and approved the final manuscript.

ACKNOWLEDGEMENTS

The research team would like to acknowledge and sincerely thank all the e-Delphi participants for their contribution. Here are the names of the participants who wished to be acknowledged: Prof. Chi Tim Hung, Prof. Eric Kam-pui Lee, Dr. Fan Ning, Prof. Jojo Cho-lee Wong, Prof. Hung Wong, Prof. Martin Chi-sang Wong, Prof. Nelson Chun-yiu Yeung, Prof. Paul Kwok-ming Poon, Prof. Regina Wing-shan Sit, Prof. Roger Yat-nork Chung, Prof. Ruby Yu, Mr. Chung Ho Chan, Mr. Daniel Lee, Dr. Hoi Lam Charlene Wong, Mr. Shing Fong Chan, Mr. Tang Newton and Dr. Ying Crystal Chan. The authors also extend their thanks to Mr. Clement Cheuk-wai Ng, Dr Janice Ying-en Ho, Ms. Rufina Chan and Ms. Valeria Wing-yu Wong for their support on the forward–backward translation of the identified items. The study was funded by the Direct Grant from The Chinese University of Hong Kong (Reference No. 2020.027).

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data sets generated and/or analysed during the current study are not publicly available to protect the anonymity of participants but are available from the corresponding author on reasonable request.

ETHICS STATEMENT

This study was conducted in accordance with the Declaration of Helsinki and approved by the Survey and Behavioral Research Ethics Committee of the Chinese University of Hong Kong (Reference No. SBRE-20-793). Informed consent for the study participation was obtained before the survey.

DETAILS

Subject:	Health promotion; Health literacy; Literacy; Health care; Skills; Validity; Questionnaires; Content analysis; Consumers; Social skills; Measurement; Stakeholders; Health education; Knowledge; Decision making; Activities of daily living; Medical personnel; Health information; Delphi method; Quantitative psychology
Business indexing term:	Subject: Consumers Delphi method
Location:	Hong Kong China; China
Identifier / keyword:	Delphi survey; Health Literacy Scale; Hong Kong Chinese; mixed methods research
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	245-255
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-08

Milestone dates:	2022-10-12 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-06-07 (manuscriptReceived); 2022-11-08 (publishedOnlineEarlyUnpaginated); 2022-10-23 (manuscriptAccepted)
Publication history :	
First posting date:	08 Nov 2022
DOI:	https://doi.org/10.1111/hex.13651
ProQuest document ID:	2767101962
Document URL:	https://www.proquest.com/scholarly-journals/developing-health-literacy-scale-adults-hong-kong/docview/2767101962/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the "License"). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-07-11
Database:	Public Health Database,Publicly Available Content Database

'What price do you put on your health?': Medical cannabis, financial toxicity and patient perspectives on medication access in advanced cancer

Olson, Rebecca E [1] ; Smith, Alexandra [1] ; Good, Phillip [2] ; Morgan, Dudley [1] ; Gurgenci, Taylan [3] ; Hardy, Janet [4] [1] School of Social Science, University of Queensland, St Lucia, Queensland, Australia [2] Mater Health Services, Mater Research Institute, University of Queensland, Brisbane, Queensland, Australia; Palliative Care, St Vincent's Private Hospital Brisbane, Brisbane, Queensland, Australia [3] Mater Health Services, Mater Research Institute, University of Queensland, Brisbane, Queensland, Australia; School of Medicine, University of Queensland, Herston, Queensland, Australia [4] Mater Health Services, Mater Research Institute, University of Queensland, Brisbane, Queensland, Australia

ProQuest document link

ABSTRACT (ENGLISH)

Introduction

Following 2016 legislation permitting limited access to cannabis for research and medicinal purposes, the number of randomized clinical trials (RCTs) investigating the effectiveness of medicinal cannabis (MC) on symptom burden relief in cancer contexts has increased in Australia. This study aimed to understand the perceptions, hopes and concerns of people with advanced cancer regarding the future availability and regulation of MC in Australia.

Methods
This qualitative study draws on semistructured interviews conducted between February 2019 and October 2020 in Brisbane, Australia, as part of an MC RCT substudy. Interviews were undertaken on 48 patients with advanced cancer in palliative care eligible to participate in an MC trial (n=26 participated in an RCT; n=2 participated in a pilot study; n=20 declined). Interviews included a discussion of patients' decision-making regarding trial participation, concerns about MC and perceptions of future availability, including cost. Transcribed interviews were analysed inductively and abductively, informed by constructivist thematic analysis conventions.
Results
Overall, participants supported making MC legally accessible as a prescription-only medication. Fear of financial toxicity, however, compromised this pathway. Steep posttrial costs of accessing MC prompted several people to decline trial participation, and others to predict—if found effective—that many would either access MC through alternative pathways or reduce their prescribed dosage to enable affordable access.
Conclusions
These findings suggest that—despite a relatively robust universal healthcare system—Australians are potentially vulnerable to and fearful of financial toxicity. Prevalent in the United States, financial toxicity occurs when disadvantaged cancer patients access necessary but expensive medications with lasting consequences: bankruptcy, ongoing anxiety and cancer worry. Interview transcripts indicate that financial fears—and the systems sustaining them—may pose a threat to RCT completion and to equitable access to legal MC. Such findings support calls for embedding qualitative substudies and community partnerships within RCTs, while also suggesting the importance of subsidisation to overcoming injustices.
Patient or Public Contribution
A patient advisory committee informed RCT design. This qualitative substudy foregrounds patients' decision-making, perceptions and experiences.

FULL TEXT

INTRODUCTION

Patients with advanced cancer face numerous symptom burdens: pain, fatigue, nausea and sleep disturbance.[1] Following 2016 legislation permitting limited access to cannabis for research and medicinal purposes in Australia,[2] interest in the potential benefits of medicinal cannabis (MC) as an intervention for relief from symptom burden associated with cancer and advanced cancer has increased substantially.[3–5] Several clinical trials have subsequently been initiated.[6–13] Data from trials, especially randomized controlled trials, however, can be ‘difficult to transfer to real-life experiences’.[14] While randomized clinical trials (RCTs) examine the effectiveness of MC at controlling symptom burden based on an experimental design, little is known about the experiences and concerns of Australians with advanced cancer considering MC.

Understanding patients' concerns, particularly related to access and regulation, is complicated by the history of cannabis as a recreational drug, and the funding, healthcare and regulatory practices specific to each country. Recreational cannabis (RC)—involving smoking or ingesting the cannabis plant which contains over 500 compounds—has been a prohibited substance for most of the 20th century.[15] Recently, cannabis has been progressively remedicalized as a viable treatment for a range of illnesses, conditions and symptoms,[16] typically involving the specific chemical compounds cannabinol (CBD) and tetrahydrocannabinol (THC) in isolation or combination.[17] This remedicalization has occurred on a global scale, with legitimate channels for accessing MC now established in North America, South America, Europe, Israel and Australia.[18] How receptive countries have been to MC, however, varies. Jamaica's legal therapeutic cannabis market, for example, faces diplomatic and marketing challenges, with constraints imposed by agreements with the United Nations and the United States and purchasers conflating RC and MC.[19] Stigma has also been found to be a barrier to accessing MC in the United States,[20] and a contributor to perceptions and experiences of MC use in Thailand[21–23] and Canada,[24] but does not feature as such in the limited Australian-focussed scholarship.[5,25]

In Australia, MC users face two tiers of regulation. Cannabis policy is split between federal and state jurisdictions,

with federal policy progressing slowly, and states devising their own approaches.[26] Despite 2016 legislation changes allowing limited access to MC via prescription from strictly regulated healthcare specialists, survey research suggests Australians still access cannabis through illicit channels and hold concerns about financial and administrative barriers to accessing MC within existing regulatory frameworks.[27] This research suggests that 62.6% of Australians assessed MC as prohibitively expensive, and 87.3% found the existing regulatory framework difficult to negotiate.[27] The number of Australians accessing MC has progressively increased, with a total of 159,665 approvals issued by 31 August 2021, 82.4% of which were issued after January 2020.[28]

Concerns about access must be contextualized with reference to Australia's healthcare system: a mixed public–private system underpinned by Australia's universal coverage known as Medicare.[29] Under Medicare, costs associated with hospital-based and some community-based care (e.g., bulk billing General Practices) are funded through taxation (e.g., Medicare Levy; Medical Levy Surcharge).[30] Australians are, however, incentivized through tax deductions to supplement with private health insurance,[31] and approximately 46% do so, allowing them access to private hospitals and 'extras' coverage, including dental, optical, allied health and other services.[29,32] Within this system, Australians access prescription medication at a reduced cost as most are included on the government's Pharmaceutical Benefits Scheme (PBS). For patients, this scheme dramatically reduces the price of pharmaceuticals filled through a pharmacist, requiring only modest out-of-pocket co-payments. Such co-payments are capped at $42.50 AUD for each PBS medicine dispensed and $6.80 AUD for those with concession cards (e.g., pensioners, students), and cumulatively at $1542.10 AUD or $326.80 AUD annually.[33] In this same environment of government-subsidised medicines, however, authorized MC products cost consumers between $350 and $600 per 100 ml (oil) and around $200 for a 15 ml spray (figures accurate as at August 2022).

Objectives and theoretical framework

Survey research suggests Australians are concerned about MC's financial and administrative burden,[27] but little to no in-depth research has been conducted with Australians with advanced cancer. This is a unique population with incurable, but often long-term disease, who are underrepresented in clinical trials and research more generally—and especially so those with poor performance status and/or high symptom burden.[34–37] This study aims to understand the perceptions, hopes and concerns of people with advanced cancer regarding the future availability and regulation of MC in Australia.

Supporting this objective, we draw on a concept of growing interest in cancer care—financial toxicity—extended by a social constructionist understanding of medication use as situated and agentic. Financial burden has traditionally been understood in terms of the direct financial costs associated with treatment, such as out-of-pocket expenses remaining after government subsidy for certain medications, or the costs associated with attending multiple clinicians across several specialist clinics.[38] Financial toxicity represents growing recognition of the need to broaden conceptualizations of financial burden to account for indirect costs such as the associated emotional burden and the coping strategies patients employ.[38,39]

Financial toxicity occurs when cancer patients—especially those with early and more severe disease—pay out-of-pocket costs (including travel and accommodation) to access necessary but expensive interventions, often while experiencing income loss due to reduced hours or early retirement, with lasting consequences to their finances and mental health, including debt, bankruptcy, emotional well-being (distress, anxiety and worry about a recurrence), quality of life and survival.[39–42] Unsurprisingly, financial toxicity is more common in countries where healthcare is predominantly privately funded; 53.7% of cancer patients surveyed in the United States reported experiencing financial toxicity.[39] It is less prevalent in Australia, with research suggesting its commonality to be near 7% for Australians 12 months postdiagnosis with colorectal cancer, compared to 39% for patients with colorectal cancer in Ireland,[40] and 20% for Australian men with prostate cancer.[43]

Although financial toxicity represents a broader conceptualization of financial burden, with terms like 'cost-related nonadherence' used to describe strategies of coping with financial toxicity,[39] the concept can be critiqued as furthering a clinician-centred understanding of financial burden. Thus, we expand our conceptual framework, drawing on Conrad's[44] classic medical sociology concept of 'medication practice', helping us to shift our focus

towards a patient-centred understanding of MC's financial and regulatory availability for Australians with advanced cancer. Medication practice can be defined as, 'how people manage their medications, focusing on the meaning and use of medications' and viewing 'patients as active agents rather than passive recipients of doctors' orders'.[44] Taking such an approach allowed us to prioritize a patient-centred examination of concerns and hopes regarding MC's future availability, to inform justice-oriented[45] RCT study design and policy.

METHODSStudy design and recruitment

This qualitative substudy examined the perceptions of people with advanced cancer eligible to participate in an MC trial.[25] Semistructured interviews were arranged with recognition of the time and communication needs of people with advanced cancer, taking a pace set by the interviewee to accommodate for any fatigue. Compared to surveys, interviews allowed for the collection of richer, inductive findings into subjective experiences and concerns about MC's future availability.[46] An experienced qualitative researcher oversaw data collection, with interviews facilitated in Brisbane, Australia, between February 2019 and October 2020. The substudy was approved by Human Research Ethics Committees at two hospitals: the Mater Hospital (HREC/17/MHS/97) and St Vincent's Hospital (HREC 17/27). To participate in interviews, participants had to be eligible to consent to one of three MC trials conducted by the research team; the protocols for the two RCTs and results for the pilot study have been published.[6–8] Relevant eligibility criteria for these MC trials included the following: (a) having an advanced (incurable) histology-proven cancer diagnosis as defined by its anatomical components as locally advanced or metastatic; (b) receiving palliative care at the treating hospital; (c) experiencing symptom burden; (d) being aged 25 or older.[6–8] MC was sourced through a registered MC manufacturer and made available to those participating in an MC trial through a hospital pharmacy, dispensed as an oil.[6–8] Recruitment for interviewees, led by the clinical trials coordinator, co-occurred with RCT recruitment. Purposive sampling enabled balanced representation across the two interviewee groups—those who declined and those who consented to MC trial participation—and in terms of gender and age (see Table 1).

Table 1 Demographic characteristics of interview participants

Characteristic	Interview participants		
	Trial participant (*n* = 28)	Declined trial participation (*n* = 20)	Total (*n* = 48)
Gender, *n* (%)			
Male	12 (25)	11 (22.91)	23 (47.91)
Female	16 (33.33)	9 (18.75)	25 (52.08)
Age in years, *n* (%)			
≤49	3 (6.25)		3 (6.25)
50–69	15 (31.25)	9 (20.8)	24 (50)
70–89	10 (20.83)	11 (22.91)	21 (43.75)
Marital status, *n* (%)			
Married/civil partnership	23 (47.91)	11 (22.91)	34 (70.83)

Divorced/separated/widowed	5 (10.42)	8 (16.66)	13 (27.08)
Single		1 (2.08)	1 (2.08)
Ethnicity, *n* (%)			
Anglo-Saxon/English	17 (35.41)	18 (37.5)	35 (72.92)
Australian	3 (6.25)		3 (6.25)
Pacific Islander	3 (6.25)		3 (6.35)
Australasian	1 (2.08)		1 (2.08)
Scottish	1 (2.08)		1 (2.08)
Undisclosed	3 (6.25)	2 (4.16)	5 (10.41)
Education level, *n* (%)			
Did not complete high school	3 (6.25)		3 (6.25)
High school	24 (50)	16 (33.33)	40 (83.33)
Bachelor's degree		1 (2.08)	1 (2.08)
Unknown	1 (2.08)	3 (6.25)	4 (8.33)
Primary cancer diagnosis, *n* (%)			
Breast	6 (12.5)	6 (12.5)	12 (25)
Prostate	3 (6.25)	7 (14.58)	10 (20.83)
Lung	4 (8.33)	3 (6.25)	7 (14.58)
Ovarian	3 (6.25)		3 (6.25)
Endometrial	3 (6.25)		3 (6.25)
Urothelial		2 (4.16)	2 (4.16)
Pancreatic	2 (4.16)		2 (4.16)
Colorectal/rectal	2 (4.16)		2 (4.16)

Bladder	1 (2.08)		1 (2.08)
Bile duct	1 (2.08)		1 (2.08)
Gastrooesophageal	1 (2.08)		1 (2.08)
Glioma	1 (2.08)		1 (2.08)
Kidney		1 (2.08)	1 (2.08)
Mesothelioma	1 (2.08)		1 (2.08)
Unknown primary		1 (2.08)	1 (2.08)

Data collection

Interviews lasted between 20 and 60 min, facilitated by one of two experienced interviewees with backgrounds in sociology and social work. Most interviews (n = 42) were face-to-face, held within a hospital consultation room in a quiet area of the hospital. Following public health measures related to COVID-19, interviewees were given the option—in accordance with an approved ethics amendment—to participate via telephone. Six interviews were subsequently conducted via telephone. Using an interview guide, facilitators prompted participants to reflect on their perspectives on MC and research, their main reasons for participating or not participating in a trial, their perceptions on current and changing MC laws and their opinions on future access. Following an iterative approach to data collection, data generated in earlier interviews refined the focus of the interviews[47]; themes identified in initial analysis informed revisions to the semistructured interview guide. For example, in considering transcripts from initial interviews, several participants described financial barriers, prompting us to add a question about financial concerns to the interview guide. All interviews were audio-recorded, with each interviewee being assigned a numerical pseudonym following verbatim transcription.

Data analysis

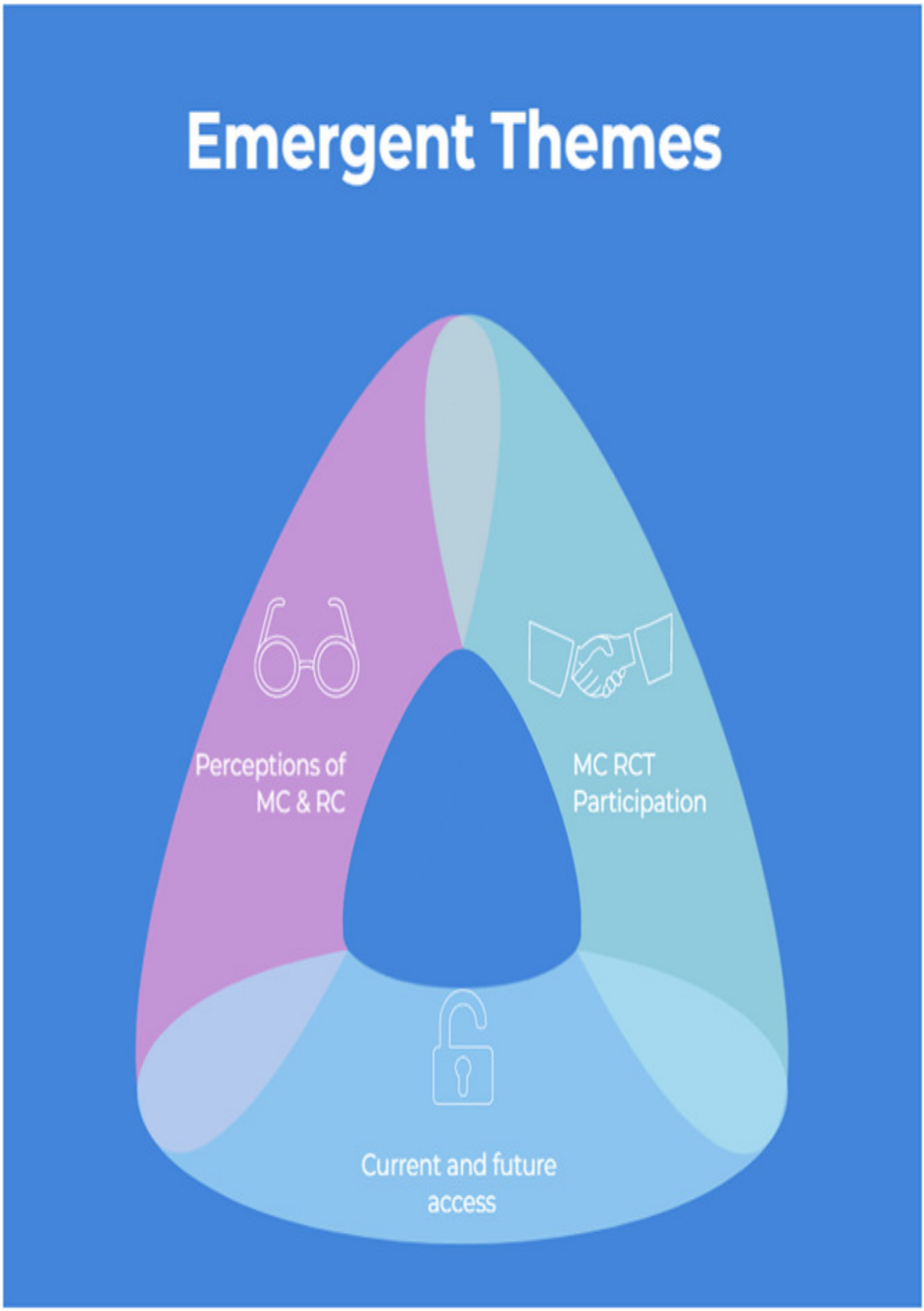
Data analysis was guided by constructivist approaches to thematic analysis.[48–50] Grounded theory informed elements of the study design, such as taking an iterative approach to data collection and analysis.[47] However, in line with the epistemological positioning and appreciation of knowledge as co-constructed[51] that underpins constructivist thematic analysis, the research team's reflexive and theory-informed positioning was foregrounded (rather than bracketed) to prioritize inductive and abductive analysis.

Transcripts were analysed thematically and by case, using NVivo 12 qualitative analysis software to support data management, and to create and organize codes based on the research aims, interview questions and evolving findings. Open coding was undertaken to identify new themes and facilitate comparison. Regular meetings across the team of qualitative researchers and RCT study leads in the early stages of data collection, supported our discussion of findings from selected transcripts. Such meetings prompted us to attend to our differing positions in engaging with transcripts and foreground multiplicity in our theory-informed interpretations, as we come from disciplinary backgrounds in sociology, medicine, social science, social work and anthropology. Following the discussion in our meeting on findings related to cost, themes were mapped against scholarship on financial toxicity, [39] and extended by Conrad's[44] concept of medication practice. Informed by these discussions, two members of the research team progressed data analysis and code development.

RESULTS

A total of 48 people with advanced cancer were recruited: 26 who agreed to participate in an RCT, 2 who agreed to participate in a pilot study and 20 who declined RCT or pilot study participation (see Table 1). Participants were relatively evenly divided in terms of gender (52% female) and age (50% aged 50–69).

Several themes were produced through our analysis(see Figure 1). This section first presents themes on patients' perceptions of how MC should be made accessible in the future, with most supportive of restricting access to a prescription-only medication, dispensed by a pharmacist. We then provide patients' perceptions on current barriers—financial and administrative—to this accessibility, with many expressing concern about the high cost of accessing MC and predicting access outside of pharmacies to manage these costs.



Enlarge this image.

Themes are supported by data displays from patient interviewees, including reference to their participant number (e.g., ‘P5’ for participant 5), gender (e.g., ‘F’ for female), age (e.g., 70s) and trial participation status (e.g., ‘declined’

indicates they did not participate in an MC trial; 'Pilot-CBD' indicates participation on the MC Pilot study[7] receiving the CBD intervention). Information is provided as to which RCT arms participants were assigned (e.g., MC1-CBD refers to the intervention arm of the MC1 study[6]; MC1-Placebo refers to the control group). It is also noted when this information is not yet available (e.g., MC2-Blinded refers to participation on the MC2 study,[8] where information on control and intervention group assignment remains blinded). Where participants described sourcing cannabis for medicinal purposes outside of the trial, this is indicated (e.g., Non-trial MC).

Future access: Who and how

Interviewees were supportive of making MC accessible, to 'people who need it' (P26, F, 50s, MC2-Blinded) and who were 'going to be responsible' in using it (P38, F, 70s, Non-trial MC). One interviewee went so far as to suggest a screening process:

[J]ust as long as …people that actually need it are screened properly and have all the documentation necessary to actually go on it. Because we see too many people who don't really need it, access it and then they screw it up for the rest of us. (P25, F, 30s, MC2-Blinded)

Although a minority said 'it should be open to everyone and anyone' (P14, F, 40s, MC1-CBD), other participants specified that it should be available to populations where other interventions have been shown to be less effective, such as chronic pain and cancer care (P21, F, 30s, MC1-CBD), terminal illness (P18, M, 60s, MC1-CBD) and conditions such as Parkinson's disease:

[It should be made available for] I suppose mainly—there are a lot of cancer sufferers. For anyone really that's suffering, with people that [have] Motor Neurone Disease, if it helps them. People that have multiple sclerosis, if it helps them. Parkinson's, all of these really challenging diseases. If it helps those people, then I believe that they should be able to have access to it. (P23, F, 70s, MC1-Placebo)

In discussing how MC should be made available, one interviewee saw too much regulation as problematic: 'I don't think it should be controlled by state or federal government. I think it's something that they don't need to be involved in' (P18, M, 60s, MC1-CBD). Others (P26, F, 50s, MC2-Blinded; P29, M, 60s, Non-trial MC) suggested RC should be decriminalized and MC made available at pharmacies.

[I think it should be available] freely, over the counter. Yeah …I think it should be decriminalised. There shouldn't be criminal convictions and all the rest because you've got a bit of pot in your pocket. (P29, M, 60s, Non-trial MC)

Most, however, responded by referencing existing mechanisms for regulating, supplying and dispensing medicines in the Australian context. Participants concluded that availability should be 'controlled' (P14, F, 40s, MC1-CBD; P37, M, 66, declined), to prevent MC from being 'abused' (P33, F, 70s, declined) and out of a concern for safety (P13, F, 70s, MC1-Placebo).

If I can't get it on a prescription I wouldn't be having it, no way, no. So, it's got to be something that you've got to get…. through chemists, through your doctor and you must have to have a script for it at all times. (P15, M, 70s, MC1-Placebo)

Accordingly, several participants proposed limiting access to those with a prescription made by a specialist (P13, F, 75, MC1-Placebo) or General Practitioner (P12, M, 60s, MC1-Placebo; P22, M, 70s, MC1-Placebo; P33, F, 70s, declined; P18, M, 60s, MC1-CBD).

Just from your doctor and the same as any other medicine is dispensed …A prescription, yes. I don't think you could just walk in and buy it. It would still have to be controlled. (P37, M, 60s, declined)

Even those participants with a history of sourcing MC through alternative pathways, and who decried the challenges of accessing MC for others, talked about controlling the supply of MC to mitigate perceived abuse:

You've got to put some controls on it otherwise you'll get abuse…. if you just say, 'oh you're feeling depressed and so you can get an [unclear]', well 98 per cent of Australia would be depressed in the morning …it would need to be controlled, there's no doubt about it …possibly the same way as you have the prescription medication. (P30, M, 60s, Non-trial MC)

Overall, most participants saw benefits to limiting access to MC to individuals with a diagnosed condition and a medical prescription, ordered by a physician and dispensed by a pharmacist. Financial and administrative

challenges, however, were said to pose barriers to achieving the perceived safety and hoped-for equity of regulated access to MC.

Barriers to access: Cost

In discussing future access to MC or in response to questions specifically about any financial concerns, 29 interviewees (17/28 on an MC RCT; 12/20 who declined RCT participation) described financial concerns, especially pensioners. Interviewee P13 (F, 70s, MC1-Placebo), for instance, said '$400 is a lot for a little bottle', and Interviewee P39 (M, 70s, declined) described MC as 'cost prohibitive for a pensioner'. Interviewees described their worries regarding the projected cost of accessing MC outside of RCTs, and the ways in which government regulatory control might overcome these barriers.

Five interviewees saw the posttrial cost of accessing MC as prohibitive to RCT participation, among other barriers,[25] directly citing cost as an impetus behind their decision to not participate in an MC RCT. Interviewee P31, for instance, said 'No, the reason I didn't take part in the trial was the fact that [sighs]—in the end I'm not going to benefit from it because it's going to be too expensive' (F, 60s, declined). Interviewee P45 (F, 60s, declined), similarly declined to participate in the RCT, saying, 'I know the trial was free, but the cost, yeah, afterwards. So, I thought, ooh and being on a pension, it would take quite a bit of that'. Interviewee P37 also cited cost in their decision to decline participation: 'I was told, when I first started talking about the trial, that if I felt there was benefit, that I could actually stay on medicinal cannabis. However, the costs were very high' (M, 60s, declined).

Raising questions of justice around equity of access, two participants asked about the merit of conducting the RCTs if the intervention was not going to be accessible.

> I was told that …it was going to be three or four hundred [dollars] a [pop] to get—have it. Just—that's out of the reach for a lot of people. Why are they even doing it if we're not even going to be able to afford to have it? It just seems crazy. (P31, F, 60s, declined)

> [W]hat are we trying to do here? All this study and all this research and trials and everything and people are taking the right drug, but then can't afford to take it. (P14, F, 40s, MC1-CBD)

These interviewees positioned lack of affordable access as a central drawback to the trial: 'the main danger is it would become too expensive for the average person who really needs it' (P38, F, 70s, declined).

Others posed the cost as a concern, but not a deterrent to trial participation. One interviewee, a retired General Practitioner, stated explicitly that cost was not a constraint. 'We are quite comfortable financially…. My wife would …spend whatever it takes, I know that, without the slightest hesitation, to improve the quality of my life' (P44, M, 80s, declined). Rather than cost, symptom-related fatigue posed a barrier to his participation. For others, cost was concerning: 'Once this [trial participation has ended] I don't really know whether I would go further because I've been told that it's very expensive' (P15, M, 70s, MC1-Placebo). The disparity in access prompted Interviewee P8 to lament this financial injustice:

> [I]t should be affordable for everybody. There's no reason why it shouldn't be whether you're rich or poor or whatever …that's where I think the gap is. People when they've got all the money can get it and people that don't are ones that don't even know where to start with it basically. (P8, F, 50s, Pilot-CBD)

Fears of financial difficulty prompted many to deliberate on the cost of the intervention, their need for the intervention, the challenges of financing their posttrial prescription and the consequences of prioritizing MC over other needs. Interviewee P23, for example, positioned health and cost as competing priorities:

> I do have concerns. We are only pensioners. But what price do you put on your health? If I feel that it has been beneficial to me and I'm feeling better on it, then to me, that's priceless. (P23, F, 70s, MC1-Placebo)

Another positioned MC and its cost as like an illicit 'drug habit', jokingly illustrating the impact of the pharmaceutical on their budget:

> After this trial it's going to cost me a fortune and none of it's subsidised, is it? …Apparently, it's around $400 a bottle of what I've got. The little tiny ones. I'd be using that quite a bit so that's a bit scary. Yeah, that's a pretty heavy drug habit [laughs]. (P24, M, 50s, MC1-CBD)

Upon deliberation, these patients concluded that their health should be the priority. Interviewee P18 was less

certain, wary of the risk that MC posed to their budget.

The costs and affordability, yeah …it's one of those things that you've just got to deal with that if—there's ways of finding money to get help, to get funding and that type of thing. Sometimes it's easy, sometimes it's not. Yeah, you've got to consider cost. You have to. (P18, M, 60s, MC1-CBD)

Such deliberations assumed that MC was beneficial. However, not all participants were sure of MC's efficacy. For these interviewees, cost concerns were compounded with worries that they were wasting money—'just throw[ing] it down the drain' (P9, M, 70s, MC1-Placebo)—on a medication unproven to address their symptoms:

Oh, it sounds expensive to me at $100 a week on something that may or may not work. I guess $100 a week wouldn't be a major worry financially, it just seems like a waste for something that I feel doesn't suit my circumstances. (P39, M, 70s, declined)

The quotes presented above relay interviewees' shock and fear—using words like 'ludicrous' (P14, F, 40s, MC1-CBD) 'crazy' (P31, F, 60s, declined) and 'scary' (P24, M, 50s, MC1-CBD)—related to the cost of accessing MC posttrial. Positioning varied, with some having the means to pay, others turning down participation in a trial due to the posttrial cost, and others still—many pensioners—uncertain, caught between prioritizing the cost of attending to their symptom burden through (not yet proven) MC prescriptions and affording other living costs. Without government regulation and subsidy to reign in the cost, interviewees predicted that patients would reduce their intake or access MC via alternative pathways.

Overcoming financial and administrative barriers: Stretching and shifting

To manage the high cost of accessing MC posttrial, and reap the expected benefits, several interviewees described stretching and shifting: making a vial last longer by taking less or changing to alternative pathways. Interviewee P45, for example, described a patient who found MC effective in controlling symptoms, but reduced their MC doses because of financial constraints:

I knew of a friend who did actually buy the medicinal marijuana …because she had really, really bad back pain. It did help her, but it was very expensive. So, she went down to only having half the dose to make it stretch a bit more, but she found it worked amazingly. (P45, F, 60s, declined)

Others considered shifting, accessing MC through other (in some cases, illegal) means, such as via the internet (P42, F, 60s, declined) or black market (P26, F, 50s, MC2-Blinded; P32, M, 60s, declined). Interviewee P11 described this in economic terms:

I'd [access MC] through the legal channels, but if I was sitting here and I didn't have—and the legal channels were $1000 a day, and the illegal channels were $10 a day, and I didn't have the money, then I'd obviously go that way. Do you know what I mean? It all depends on the person's capacity to pay. (P11, M, 60s, MC1-Placebo)

Interviewee P10 emphasized both the financial and administrative 'rigmarole' as deterrents to accessing MC legally:

Because it's too expensive if they buy it. Because they have to go through too much of a rigmarole to get registered to take it. Then they have to pay exorbitant amounts and then they have to renew that every six months. So, it's a lot for people…. I think I probably would know more people who would access it the other way. (P10, F, 60s, MC1-CBD)

Interviewee P40 similarly implied that the current financial and administrative 'speed bumps' associated with legally accessing MC were likely to push many towards unregulated avenues:

Well, I don't think you can ever agree with the black market or even going, buying it online, I don't agree with any of that. But having said that the only way that's ever going to be stopped is for our system to make it available, a lot easier, without putting all the speed bumps in the road. (P40, M, 80s, declined)

Although this interviewee—like many others—didn't 'agree with' shifting to alternative MC access pathways, others showed less reservation. In such interviews, a common thread of pragmatism, and a related lack of stigma, was evident in discussions of accessing MC outside regulated pathways. Interviewee P13, for example, discussed her current experience in obtaining MC where her main concern was about trust in the quality and safety of the MC accessed via an alternative pathway:

I was looking where I could get the oil and I put it on social internet and someone came forward and said yes, I've

been on it …. I can get the oil for you…. The thing that stopped me was the price …I didn't get it from there, but I …rang around all the naturopath people to see if they had it or knew where I could get it. This lady came forward so I just got the cannabis stuff, the marijuana, and she made the oil for me. I trusted her because it helped her …I wouldn't buy it from anybody that I didn't know…. the lady that made it up for me she was okay too. But it's dicey if you just go straight there and don't make inquiries or anything. (P13, F, 70s, MC1-Placebo)

Even interviewee P14, a police officer, viewed accessing MC via alternative pathways pragmatically:

If you can go to [town] and get it, I don't know, $50 or whatever, I don't know, but compared to $200 for every three days. Who would begrudge anyone to do that? As a police officer, yes, I would still charge them, but I can tell you the courts will get sick of it, because that's what we'll be putting in the paperwork. (P14, F, 40s, MC1-CBD)

Despite interviewees' overwhelming support for restricting MC access to a pharmacy prescription, financial and administrative barriers posed a threat to this pathway's viability. Interviewees predicted that—without regulation and subsidisation—patients would likely stretch, altering prescribed dosages to improve affordability, or shift, unashamedly sourcing MC via alternate pathways.

Overcoming financial barriers: Government regulation

To facilitate smoother access—financially and administratively—interviewees, especially those on a pension or reduced income, overwhelmingly suggested government regulation through the PBS.

Because I'm on a pension and I live from week to week, so I can't afford to pay much…. if it gets to be on the PBS …then I really can afford it. (P3, F, 60s, Pilot-CBD)

It should go on the PBS. It'd make it so much easier for so many people…. We're pensioners. We'll find the money, but it's going to be a bit hard. (P13, F, 70s, MC1-Placebo)

Some suggested government regulation through the PBS ‘so that more and more people can get the benefits of it’ (P22, M, 70s, MC1-Placebo): as a matter of equity and justice for patients in need of medication.

Through the PBS. Yeah. Definitely. They do so much for everybody else. Why not for these people that really need it? (P19, F, 50s, MC1-Placebo)

So, the costs for the people who are falling within the categories for its use who want to use it and find benefit from them, it should be made financially within their reach under a PBS-type subsidised scheme and it shouldn't be to their detriment if it's providing them health and pain relief and assistance in coping with their medical health or mental health. (P34, M, 50s, declined)

Others positioned PBS regulation as symbolic of MC's efficacy, if proven, just like any other evidence-based pharmaceutical intervention.

If it's got proven benefits, it should be on the PBS. (P39, M, 70s, declined)

[It should be made available through the PBS …if you've got something that it can help with, like Endone or something like that—that helps—well then you should be able to access that the same sort of way. (P31, F, 60s, declined)

To improve affordability, equity of access for patients and equity in MC's treatment as a pharmaceutical intervention—interviewees supported making MC a prescription-only medication subsided by Australia's PBS.

DISCUSSION

This qualitative study aimed to understand the perceptions, hopes and concerns of people with advanced cancer regarding the future availability and regulation of MC in Australia. Overall findings suggest that patients are supportive of making MC legally accessible as a prescription-only medication. Fear of the financial risks, however, compromised this pathway. The administrative ‘speed bumps’ and steep posttrial cost of accessing MC prompted several people to decline trial participation, and others to predict—if found effective—that many would either reduce their prescribed dosage to enable affordable legal access, or access MC through alternative pathways. Below, we discuss this contribution, theorizing the financial risks of accessing MC posttrial as financial toxicity, and explicating the threat it poses to equitable access to legal MC and RCT participation. We then consider the implications of this finding for policy and RCT design, suggesting subsidisation and qualitative substudies as ways of foregrounding and overcoming possible injustices.

Interviewees were overwhelmingly supportive of making MC legally accessible as a prescription-only medication. As 28 of our interviewees were individuals with advanced cancer consenting to participate in trials—a hypermedicalized context involving a high degree of medical control, including, in these trials, restricting eligibility to those with no cannabis in their system—this sample may seem to be providing a relatively skewed perspective. However, other Australian research examining broader public perceptions of MC use suggests that these patients/participants are not outliers—acceptability of MC is high amongst the general population[27,52] and a majority of general practitioners are also supportive or neutral on MC use.[53] Furthermore, it is important to note that patients were screened for RCT eligibility after consenting to participate. Thus, some interviewees who consented to participate in an MC RCT and interview, may have been found to be ineligible later because of having cannabis in their system. Despite support for it, MC was also perceived as a current or potential source of financial toxicity by patients with advanced cancer interviewed for this study—amongst those who consented MC RCT participation and those declined—with five participants directly citing cost in their decision to not participate in an MC RCT. Said another way, MC was perceived by many as a necessary or potentially necessary intervention, with associated costs that could prompt financial strain.[39] Many reflected on the posttrial cost of accessing MC, using emotional and disparaging language to express their fear and concern. This finding supports research from the United Kingdom[54] and Canada[55] showing significant financial barriers to accessibility for patients, despite MC being available in these countries within regulatory frameworks. However, it may be surprising in the Australian context, given that financial toxicity is less prevalent in this country,[40] especially compared to countries with limited public healthcare systems, such as the United States.[39] This finding may also be surprising considering financial toxicity is often associated with an early-stage diagnosis.[40] Nonetheless, financial toxicity was a concern for interviewees with advanced cancer in this study. Many interviewees were facing chronic symptom burden and were pensioners, with few classified as high socioeconomic status: all factors which have been found to be significant predictors of a financial burden and financial toxicity.[38,56]

In responding to their financial toxicity concerns, interviewees described several mitigation strategies: stretching, shifting and declining. Some participants predicted ‘stretching’ their supply to better weather MC's posttrial financial imposition, taking less than the recommended dose to reduce their weekly MC expenditure. This is a well-known strategy for coping with financial toxicity, referred to as ‘cost-related medical nonadherence’[39] or ‘cost-related medication underuse’[57] within medical scholarship and, less pejoratively, active or agentic ‘medication practices’ within sociological scholarship.[44]

Shifting—to alternative markets—was another financial toxicity coping strategy, but less acknowledged within cancer scholarship and potentially unique to MC. Despite overwhelming support for restricting access to a pharmacy prescription, interviewees described unreservedly sourcing MC via less than legal pathways. Blurring or hybridizing RC and MC, some predicted or actively engaged in abandoning concerns related to safety and control, and sourcing uncompounded cannabis online or via a trusted supplier for a fraction of the cost and without the administrative burden. This finding suggests that pragmatism in the Australian context may override the stigma related to accessing MC found in research from the United States.[20] It also supports research by Mahamad and Hammond[55] pointing to the continued existence, and indeed flourishing, of ‘black market’ sources of medicinally used cannabis in environments of legalized, regulated MC. Within the context of financial toxicity,[39] this study draws attention to the ‘coping’ practice of sourcing medication illegally to treat their conditions—a practice suggested to be widespread but below the ‘public gaze’.[15] Sociologically, this practice is referred to as engaging in ‘covert’ or ‘subaltern’ therapeutics: using interventions deemed outside of medicine, resistant to biomedicine (such as ‘folk medicine’), or, in the case of marijuana, criminalized.[15] For policy, this finding raises important questions about state processes with poor streamlining, potentially posing a threat to MC schemes,[19] and certainly motivating potential MC users' consideration of less legal competitors.

In addition to stretching and shifting, declining was a further strategy for mitigating MC's perceived financial toxicity. Five interviewees declined to participate in an MC trial citing cost as a reason. Despite MC being available at no cost to trial participants, the high posttrial cost prompted these interviewees to circumnavigate financial concerns by

avoiding MC altogether.[25] This barrier to MC RCT participation raises important concerns about equitable access to tested interventions, and the potential impact of these concerns on patient decision-making regarding trial participation. In her research on disparities in RCT participation, Fisher[58] shows marginalized men overrepresented in early-stage pharmaceutical testing, but underrepresented as intervention users. Our study suggests economically disadvantaged participants may be deterred from participating. Such inequities could undermine RCT completion, as well as impact fair and equitable access to tested interventions following trial completion. While recruitment was not an issue for the MC trials supported by this qualitative substudy, it is a common problem. An estimated 50% of RCTs fail to recruit to their targets,[59] a problem amplified within palliative care contexts, where sample attrition is a regular and expected occurrence.[60]

Qualitative substudies—as illustrated through this study—and community partnerships can foreground inequities that threaten to undermine RCT recruitment. Fortuna et al.[45] suggest countering the reductionism that underpins the scientific method—epitomized by RCTs—with humanistic approaches—such as qualitative and participatory methods—that 'prioritize[] the human experience and promote[] the inclusion of disadvantaged populations as partners in research'. As evidenced in this study, 'methodological pluralism'[45]—through a qualitative substudy—can allowed researchers to identify differences in power and resources that could undermine clinical research and clinical outcomes.

While small revisions to study designs and research practice can go some way towards attending to inequities, broader change is also needed. Findings presented here suggest that without subsidisation (e.g., through the PBS), MC poses substantial risks: risk of financial toxicity to patients and their families, and potentially to equitable access to the benefits of RCT participation. Although demonstrated effectiveness is a requirement for pharmaceutical interventions to be listed on the PBS, MC poses a unique scenario where patients are accessing similar interventions covertly through alternative or subaltern therapeutic pathways. There is thus an imperative for commercial entities involved in MC to invest in and support clinical trials to produce high-quality evidence of efficacy and safety, to ensure quality and to embed equity of future access through registration and subsidisation via the PBS.

The strengths of our study included drawing insights from both those who consented, and those who declined, to participate in an MC trial. An iterative and abductive approach[48–51] also foregrounded patients' concerns and critical insights from the study's conceptual framework. The cross-sectional approach, however, limited data to a single timepoint; the exclusive focus on patients' decision-making overshadowed carers' perceptions. Future research will further give insights into perceptions and experiences, by purposively sampling patients and carer participants at different trial stages.

CONCLUSION

The findings and analysis presented here provide novel insights into the perceptions, hopes and concerns of people with advanced cancer regarding the future availability and regulation of MC in Australia. Findings suggest patients are aware and fearful of financial toxicity related to the high cost of accessing MC outside of clinical trials. To improve affordability, equity of access for patients and equity in MC's treatment as a pharmaceutical intervention—interviewees supported making MC a prescription-only medication subsided by Australia's PBS. Qualitative substudies are valued additions to RCTs—shining light on injustices relevant to RCT recruitment and design, but in this context, policy and practice change may be needed to overcome MC's financial toxicity. Put simply, many interviewees assessed legally available MC to be of little use without ensuring commensurate affordability. Future research could examine the prevalence of concerns in Australia related to MC's financial toxicity and establish the commonality of subaltern or covert use of RC/MC.

AUTHOR CONTRIBUTIONS

Rebecca E. Olson designed the qualitative substudy, provided oversight on and contributed to data collection and analysis and drafted the final manuscript. Alexandra Smith led the data analysis and was a major contributor in writing the manuscript. Phillip Good contributed to the study design, data analysis and editing of the final version of the manuscript. Morgan Dudley was a major contributor in writing the manuscript. Taylan Gurgenci was a contributor

in writing the manuscript and was a major contributor in editing the final version of the manuscript. Janet Hardy contributed to the study design, data analysis and a major contributor in editing the final version of the manuscript. All authors read and approved the final manuscript.

ACKNOWLEDGEMENTS

The authors wish to thank the TalkingMedCan participants for their time and insights. We are grateful for the research support provided by Thomas Dinala, Karyn Foster, Georgie Huggett and Zhaoxi Zheng. Funding for this study was provided by a National Health and Medical Research Council (NHMRC) of Australia Medical Research Future Fund (MRFF) Grant (APP1140160, 2018–2022) and a Mater Clinical Research Seeding Grant (2017-2019). None of these funders had a role in the study design, collection, analysis or interpretation of the data, writing the manuscript or the decision to submit the paper for publication.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data generated and analysed for the current study are available to suitably qualified individuals by request, from the corresponding author, subject to HREC approval.

ETHICS STATEMENT

Ethical approval for this study was obtained from the Human Research Ethics Committees at the Mater Hospital (HREC/17/MHS/97) and St Vincent's Hospital (HREC 17/27). All participants provided their written informed consent.

DETAILS

Subject:	Cannabis; Toxicity; Health care policy; Hospitals; Availability; Palliative care; Medicare; Regulation; Cost analysis; Worry; Advisory committees; Cancer; Legislation; Medical marijuana; Patients; Decision making; Conventions; Research; Clinical trials; Medical research; Clinical research; Colorectal cancer; Fear &phobias; Interviews; Access; Drugs; Bankruptcy; Marijuana; Costs; Perceptions; Health professional-Patient communication; Partnerships; Embedding; Dosage
Business indexing term:	Subject: Medicare Costs
Location:	Australia; United States--US
Identifier / keyword:	financial toxicity; medicinal cannabis; qualitative research; sociology
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	160-171
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES

Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-06
Milestone dates:	2022-09-29 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-08-21 (manuscriptReceived); 2022-11-06 (publishedOnlineEarlyUnpaginated); 2022-10-16 (manuscriptAccepted)
Publication history :	
First posting date:	06 Nov 2022
DOI:	https://doi.org/10.1111/hex.13642
ProQuest document ID:	2767101961
Document URL:	https://www.proquest.com/scholarly-journals/what-price-do-you-put-on-your-health-medical/docview/2767101961/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the “License”). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-04-24
Database:	Public Health Database,Publicly Available Content Database

‘it was that … specialist … that finally listened to us … that's probably a weird answer to what you were

expecting': Clinician and carer perspectives on brilliant feeding care

Dadich, Ann [1]
; Kaplun, Simone [1] ; Kaplun, Cathy [2]
; Hopwood, Nick [3]
; Elliot, Christopher [4]
[1] School of Business, Western Sydney University, Parramatta, NSW, Australia [2] Transforming early Education and Child Health (TeEACH) Strategic Research Initiative, Western Sydney University, Westmead, New South Wales, Australia [3] Faculty of Arts and Social Science, University of Technology Sydney, Broadway, New South Wales, Australia [4] Department of Paediatrics, St George Hospital, Kogarah, New South Wales, Australia

ABSTRACT (ENGLISH)

Introduction
To extend research on positive aspects of health care, this article focusses on health care for children who tube-feed—this is because knowledge about tube-feeding for children is limited and fragmented. This is achieved by consulting with clinicians and carers who supported children who tube-feed to clarify their understandings of and experiences with brilliant feeding care.
Methods
Nine clinicians and nine carers who supported children who tube-fed were interviewed. The interview transcripts were analysed thematically.
Results
Findings highlighted several features of brilliant feeding care—namely: practices that go above and beyond; attentiveness; empowerment; being 'on the same page'; hopefulness and normalcy.
Conclusions
These findings show that seemingly trivial or small acts of care can make a significant meaningful difference to carers of children who tube-feed. Such accounts elucidate brilliant care as grounded in feasible, everyday actions, within clinicians' reach. The implications associated with these findings are threefold. First, the findings highlight the need for clinicians to listen, be attuned and committed to the well-being of children who tube-feed and their carers, share decision-making, source resources, and instil hope. Second, the findings suggest that carers should seek out and acknowledge clinicians who listen, involve them in decision-making processes, and continue to source the resources required to optimize child and carer well-being. Third, the findings point to the need for research to clarify the models of care that foster brilliant feeding care, and the conditions required to introduce and sustain these models.
Patient or Public Contribution
All of the carers and clinicians who contributed to this study were invited to participate in a workshop to discuss, critique, and sense-check the findings. Three carers and one clinician accepted this invitation. Collectively, they indicated that the findings resonated with them, and they agreed with the themes, which they indicated were well-substantiated by the data.

FULL TEXT

INTRODUCTION

Many children, worldwide, require a tube to maintain adequate nutrition, orally.[1] Paediatric feeding disorder requiring

tube-feeding (PFD-T)[2] might involve the following: a nasogastric tube, which is inserted into the nose and through to the stomach; an orogastric tube, which is inserted into the mouth and through to the stomach; or a percutaneous endoscopic gastrostomy tube, which is surgically inserted into the stomach. Although it is difficult to estimate the prevalence of PFD-T, it is said to be between 1 and 4 children per 100,000.[3] However, this rate can be as high as 83–92 per 100,000, if not more.[4]

Despite the prevalence of PFD-T, knowledge about it is far from complete. This is largely due to two reasons. First, there are over 350 health conditions that can warrant tube-feeding.[5] These include (but are not limited to) cerebral palsy, neurodevelopmental disabilities, cleft palate, cystic fibrosis, prematurity, recovery postsurgery, and ill health.[6–8] As such, 'There are multiple, complex pathways to paediatric tube-feeding'.[2,p. 1]

Second, different clinicians affiliated with different specialities manage tube-feeding, conceptualizing it differently.[9,10] With few exceptions,[11,12] research on PFD-T tends to focus on particular health conditions.[13–18] Consequently, knowledge about PFD-T remains fragmented.

Regardless of why a child requires tube-feeding or the specialities involved in their care, PFD-T can have personal, social, and economic implications. It can isolate the child and their family from social interactions; compromise the child's well-being; generate carer anxiety, family strain, and relationship issues; as well as warrant greater access to (mental) health services, adding to rising healthcare costs.[4,13,19–23] Furthermore, feeding difficulties and the management of tube-feeding among children are not always readily 'fixed' by health services. This is due to myriad reasons, including poor clinician recognition of carer concerns.[24] For instance, researchers have noted that 'Most of the primary caregivers… found it difficult to coordinate care and obtain support when needed',[25,p.25] and 'parents could benefit not only from sensitive and respectful collaboration but also from anticipatory guidance'.[26,p.212]

This literature, which primarily awards attention to problems and issues, depicts a somewhat bleak portrayal of feeding difficulties and tube-feeding. While it is important to identify problems and issues, a preoccupation with all that is wrong with healthcare can itself be a problem. For instance, for patients and carers, a continued focus on that which is negative can silence their positive experiences with health issues and/or health services—and there are many instances of these[27]; it can also diminish help-seeking behaviours and subsequent access to timely care.[28] For clinicians and service managers, this preoccupation risks unfairly stereotyping them as part of a systemic problem[29,30]—furthermore, it can diminish learning opportunities and innovation.[31] And for policymakers, it might continue to direct their attention (and public funds) to ineffective and/or inefficient healthcare practices—this is because, rather than problematize beliefs and assumptions, the identification of problems and issues is largely based on prevailing beliefs and assumptions, leaving little opportunity for innovation.[32]

Building on the literature that visibilises 'that which is positive, flourishing, and life-giving in [healthcare] organisations',[33,p.731] and redresses the scholarly preoccupation with the problems and issues in feeding care, this article purposely considers what constitutes brilliant feeding care.[34,35] This is achieved by consulting with clinicians and carers who support children who tube-feed. The article commences with a brief overview of brilliant care. After describing the study focus and the research method, the findings on what constitutes brilliant feeding care are presented. The article concludes by explicating the implications associated with these findings for scholars, clinicians, and carers.

Brilliant care

Brilliant care can be conceptualized in ways that are not tied to specific health outcomes. It is a relational experience that exceeds expectations, bringing joy and delight to those who experience or witness it.[36] Brilliant care can be unconventional and serendipitous, and does not necessarily represent business as usual within a service or a sector. Furthermore, brilliant care is interpersonal, uplifting, inspiring, and/or energizing.[37]

Aspiring for brilliance in care goes deeper than meeting or exceeding performance indicators. One aspect of this involves the recognized benefits of positive emotions in diverse contexts, including healthcare. Fredrickson's broaden-and-build theory helps to understand this important feature of brilliance—'Positive emotions… *broaden* people's momentary thought-action repertoires and *build* their enduring personal resources' (p. 147, original italics).[38] The experience of healthcare can benefit from upward spirals as positive emotions and the expanded thinking

they promote become mutually reinforcing.[39]

A second important aspect of brilliance concerns an ethic of care. An ethic of care awards primacy to connections.[40] It recognizes the importance of 'trust and responsibility, protection of individuality, the context in which the relationship takes place, and the quality of the relationship'.[41,p.3] Furthermore, it recognizes listening as a way to fortify trust, strengthen relationships, and diversify voices.

Of particular relevance to brilliant care is the resistance that an ethic of care epitomizes—it counters assumptions and norms that sustain injustice.[42] It recognizes a need to 'negotiate relations between self and other in ways that resist the hierarchies that maintain existing relations of power'.[43,p.13] Correspondingly, brilliant care defies what might be expected to foster connections that enable individuals or collectives to flourish.[34] With this theoretical backdrop, this article considers what constitutes brilliant feeding care according to clinicians and carers who supported children who tube-feed.

METHOD

Following clearance from the relevant human research ethics committee (approval number: H13794), clinicians and carers who supported children who tube-fed were invited to participate in a semi-structured interview. Clinicians were primarily recruited via purposeful sampling. Clinicians aged 18 years or older, who resided in Australia, and had spent most of their working week engaged in feeding care for children aged under 18 years, were invited to participate in this study via email. Carers were recruited via social media platforms (e.g., Facebook, Twitter) and relevant webpages. Carers were invited to contact the researchers to participate in this study if they were aged 18 years or older; resided in Australia; and cared for a child aged under 18 years who required tube-feeding within the last 5 years (to optimize the currency of the findings). Participant recruitment of both cohorts continued until data saturation.[44] Specifically, data analysis occurred in tandem with data collection and when 'no new information, codes or themes …[were] yielded from the data' (p. 202), recruitment efforts ceased.

The researchers devised two interview schedules, one for clinicians and one for carers (see Appendix 1). The schedule for the clinicians pertained to the following: how they became interested in feeding difficulties and/or tube-feeding; what they have found useful when supporting children who tube-feed and/or their carers; their understandings of and experiences with brilliant feeding care; and what they wish they would have known about feeding care, earlier in life. The schedule for the carers pertained to: the lived experiences of tube-feeding; what helped or hindered feeding care; the priorities and considerations that mattered to them; their understandings of and experiences with brilliant feeding care; and what they wish they had known about feeding care, earlier in life. Given the article's focus, only findings pertaining to brilliant feeding care are presented. To ensure the schedules were fit-for-purpose, this study and the schedules were discussed with members of the SUCCEED Child Feeding Alliance. The SUCCEED Child Feeding Alliance represents a unique collaboration between health professionals, academics, artists, and families who are passionate about supporting children with feeding difficulties and their families. Alliance members were invited to consider and critique the study design and inform the development of the schedules.

Following informed, written consent, nine clinician and nine carer interviews were conducted via web conferences for approximately 1 h (see Table 1). The interviews were digitally recorded and transcribed for thematic analysis.[45] One researcher (re)listened to the recordings as well as (re)read and reviewed the transcripts to ascertain patterns within the dataset. They also constructed broad (or higher-order) themes that reflected participant experiences and perceptions. To clarify their understandings of and experiences with brilliant feeding care, particular attention was awarded to experiences that brought joy and delight; '*broaden[ed]* people's momentary thought-action repertoires and *buil[t]* their enduring personal resources'[38,p.147] (original italics); bolstered connections, and exceeded expectation by defying norms.[46] This process was aided by NVivo 12—computer-assisted qualitative data analysis software.[47] To optimize the veracity of the analysis, two other researchers analysed half of the transcripts each. The three researchers conferred about their respective themes and reconciled differences.

Table 1 Participant demographic details and attributes

Cohort	Characteristic	*n* (%)
Clinicians (*n* = 9)	Age (years)	
	20–29	2
	30–39	3
	40–49	3
	50–59	1
	Gender (female)	8 (88.9)
	Geographical location	
	Queensland	9 (100.0)
	Discipline	
	Dietetics	7 (77.8)
	Speech pathology	2 (22.2)
	Experience in child health care (years)	
	1–5	2
	6–10	3
	11–15	1
	16–20	2
	Over 30	1
	Employed in a tertiary health service	3
Carers (*n* = 9)	Age (years)	
	30–39	5
	40–49	2
	50–59	2

	Gender (female)	8 (88.9)
	Geographical location	
	New South Wales	2 (22.2)
	Victoria	2 (22.2)
	Queensland	2 (22.2)
	Unspecified	3 (33.3)
	Employment status	
	Full-time employed	3 (33.3)
	Part-time employed	3 (33.3)
	Unemployed	2 (22.2)
	Retired	1 (11.1)
	Experience in supporting children with feeding disorders (years)	
	0.5–2	4
	3–3.5	4
	15	1
	Age of child with a feeding disorder (years)	
	1–2	4
	3–3.5	4
	15	1
	Gender of child with a feeding disorder (male)	6 (66.7)

RESULTS

Findings from the 18 interviews highlighted six features of brilliant feeding care. Each is addressed in turn.

Going above and beyond

Brilliance was aptly demonstrated when others went above and beyond their substantive role to support a child who tube-fed or their carer. Participants described individuals in their lives who ventured outside their remit to perform acts of care. Sometimes these acts were considerable—they required significant time and effort or placed the

individual in a potentially precarious situation:

[My son] had a really… bad turn… his heartbeat and his breathing just almost stopped… the nurse… called a [medical emergency team]… It wasn't until about two in the morning that I was just standing there watching them… and… his paediatrician just appeared at my shoulder… I was like, 'What are you doing here? You're not oncall'. She said… 'I just came in to make sure everything was okay'… She was there all night until eight in the morning and then did a full day at work. She just came in to make sure that [my son]… was okay. (*carer 14*)

Equally important were small acts of care—deeds that perhaps did not require the individual to invest considerable time and effort or place themselves at risk, but nevertheless made a sizeable impression on others:

there was just these really… small little details that she gave us that made a big difference to make sure that we were… doing the right thing for [our son]. (*carer 15*)

The significant and relatively minor acts of care shared two features. First, the instances typically occurred during times of heightened adversity. For instance, they occurred when a carer experienced considerable strain, distress, or anxiety. During these moments, brilliant care was a helpful antidote:

the one thing that's standing out for me is the parent who said… 'You're the first person who's listened to me and believed that this is a real thing and a real issue, and… told me that it's not my fault or that I'm not being paranoid'… I think listening and really unpacking that with them can have such a big impact. (*clinician 11*)

Second, the acts of brilliant care exceeded expectations. In contrast with the healthcare they were used to, which was often rigid, the carers were moved by displays of care. They were inspired and encouraged by those who acted compassionately, transcending the pedestrian pattern of healthcare that they and their child typically received:

our first paediatrician… told us …[my son]… had silent reflux—'Go home and take this… He will be fine'… we went back… two weeks later and I was like, 'Look, it's getting worse'. So, then he tried us on this… formula. Again, it almost made him worse… then I attempted to see a third paediatrician. They told me the same thing… I had an appointment with our baby health nurse… we weighed him and… she had this look on her face and I said to her… 'What?'… she just said, 'I'm sorry… As a baby health nurse, we can't give recommendations and advice'… I said to her… 'what's the problem right now?' And she said, 'He has just tipped under the third percentile'… I just said… 'If I said to you, I'm going to get a third opinion, would you say that I'm doing the right thing?' And she said, 'Yes'. I said… 'If I said to you that I was going to attempt to… [see Dr A or Dr. B]… what would you say?'… she said, 'You are a fantastic mum… you will know what to do'… I rang [both doctors]… no one picked up, so I left a message… I got a phone call back… from [Dr A's] …rooms… the lady… at the front desk… she said, 'Alright, now just hold on a moment. Just calm down… tell me what's happening'… I just said, 'I need help… I need to save my baby'… she said, 'Look, [Dr A]… is not in today, but I will call him and I will get him to see you on Monday'… I got a call back from [her] …and she said… 'I've just spoken to [Dr A] …and he said if you can be in his room on that Saturday morning at nine o'clock, he will see you'… so we went… and …said to him, 'Look …we have been through the ringer… no one is helping us. If you can't help us today, our car is actually packed and we are going to [the hospital]'… he… said, '…you are not going anywhere; I will be escorting you to hospital'. And so, he actually did and from that point onwards we didn't leave hospital for the ten weeks… we are so incredibly grateful for him and our baby nurse… she subtly gave me the hints of what would be best. And if she ever got into trouble for any of that, I would back her a 1000% because without her and [Dr A]… we actually don't know where we would have been. (*carer 15*)

This account demonstrates the complementary roles of different forms of brilliant care. Mindful of what she was (not) permitted to do, the nurse used praise to gently nudge the carer to source alternative medical advice; while the doctor discernibly and proactively strived to attend to the carer's concerns.

Attentiveness

Several carers described the positive impact of clinician attentiveness. Attentiveness was important because it indicated that the carer and child well being mattered. Rather than prioritize their own interests, like managing limited time or assuming what others needed, the clinicians were thoughtful and they considered what the carer and child might need:

it was that renal specialist in terms of the feeding that finally listened to us… that's probably a weird answer to what you were expecting. (*carer 1*)

Attentiveness was demonstrated directly and indirectly. The former included the following: the respectful questions that others asked and how they deferentially asked them; how they fulfilled promises, such as sourcing supplies or clinical expertise; and unsolicited offers of support. Indirect attentiveness included others' observations—how they noticed the signs that a child's health might be compromised, or that a carer might be struggling with the complexities of feeding care.

Demonstrations of attentiveness were deemed brilliant for two reasons. First, they exceeded expectations. They did not reflect the norms of mediocre or dismissive healthcare that carers were accustomed to. Instead, others' thoughtfulness was serendipitous, positively deviating from what the carers expected:

just the fact that the doctor… was actually asking what I thought. 'What do you think… would help? What about this? Has he tried that?'… they were very open; whereas, I find… with different doctors… it's very much, 'I'm a doctor. I know what… is needed'… not really listening to what your experiences are, what you know that child needs. (*carer 2*)

Second, because managing a feeding disorder can be overwhelming and exhausting, carers were not always able to recognize or articulate their needs. Carers sometimes needed a carer—someone to look out for them, respectfully identify what might be helpful, and support them:

I can just see that parent has had zero sleep, [so I] rework… the plans to make it work… maybe we need to change the overnight [feeds]… to continuous… things like that really help. Sometimes the families aren't in a space to articulate that goal at that particular moment because they're so sleep deprived. But they come back at the next review, and they are glad that we made the change. (*clinician 5*)

Empowerment

According to the participants, brilliant feeding care was demonstrated by empowerment—when they or others experienced improved confidence and were better able to exercise agency. Unfamiliar with and uncertain about feeding difficulties or feeding care, clinicians and carers often struggled to know what to do and how to do it. The associated insecurity and anxiety were sometimes exacerbated by an awareness that, just as a child's failure to thrive can be distressing and dangerous for carers and their child, so too can tube-feeding. Tube (re)insertion can be distressing and uncomfortable for the child—it can also be dangerous if performed incorrectly. A clinician's or carer's feelings of helplessness and hopelessness often subsided when they were encouraged and supported to take greater control over an uncertain or anxiety-provoking situation. Sometimes, this made a world of difference:

what made it really helpful or empowering, was the fact that it was so much about learning to trust your child… ultimately trying to… empower… families and… children… just in terms of knowledge, just in terms of understanding the experiences you're going through, in terms of helping you to find your own way forward with things, that was a really brilliant experience. (*carer 8*)

Empowerment was typically facilitated by clinicians and other carers. Participants described how these individuals reassuringly shared advice, enabling them to manage difficult situations, feel prepared, and gain a greater sense of control:

We… had a buddy system… particularly for those littlies that were going through tube-wean. So, successful tube-weaners would then buddy with families… prior to achieve wean, so they could provide some additional support. I think that worked really well because… hearing it from clinicians is quite different to hearing it from a parent that's had a lived experience. (*clinician 1*)

In the context of empowerment, although the advice was important, so too was the way it was offered. Given that health education was typically offered prescriptively, encouragement and reassurances were welcome juxtapositions:

The [percutaneous endoscopic gastrostomy or] PEG team at the hospital… were awesome… they teach you how to put the PEG in and out by yourself. Just teach you everything about it and make you feel comfortable with it… they… say, 'You're doing a good job, you're doing awesome'… before that, no one ever said stuff like that, ever. (

carer 11)

'We're all on the same page'

Brilliant feeding care involved having a shared understanding with others of what mattered and how to realize aspirations, particularly with those who contributed to the child's care. This was important given that evidence-based child healthcare requires a multidisciplinary approach.[48] As such, several clinicians who represent different disciplines are typically involved in the care of a child who is tube-fed. Despite the potential value of complementary areas of expertise, some participants noted how overwhelming and confusing multidisciplinary care can be—this was largely because different clinicians often espoused different opinions (in different ways) on how to best manage a feeding difficulty. However, when clinicians and carers worked as a team towards shared goals, brilliant care was experienced:

we're all working towards the same goal… we're all on the same page and that's the positive that I take out of all the back and forth with everybody else. (*carer 4*)

Being 'on the same page' was considered brilliant because it surpassed the confusion and inefficiency that many clinicians and carers were used to. When clinicians or carers felt understood, they did not feel obligated to explicate their concerns or experiences at length or repeatedly. The discussion was relatively easier because there was an unstated recognition of what was typically a complex situation, and there was sympathy for those attempting to manage such complexity:

there's no chopping and changing with that department. It still is the same lady… when there's chopping and changing and it's a different person every week, you feel like you're starting from scratch every week and you've got to tell them his… life story to get to the point, every single time… it's always been the same person. That makes a massive difference because she knows his needs. (*carer 11*)

Hopefulness

Participants indicated that brilliant feeding care was demonstrated when they were inspired or offered hope. Depleted by the challenges of caring for a child with a complex health condition, their confidence and their aspirational outlook on life often waned. Yet, this situation and their outlook could be considerably altered when they experienced a semblance of optimism. For instance, when carers felt disheartened, clinicians made a positive impact by working with the carer and child towards feasible goals. Through reassurance and goal achievement, carers felt better equipped to manage their difficult circumstances:

I recently had a little four-month-old bub… she couldn't feed because of her reflux… we did really… well with her because, at the beginning of inserting the tube, we made… three-month goals that… helped guide what we do… that has gone… really well, because the goals that we made were really appropriate for the baby and the family. (*clinician 6*)

According to the participants, the goals need not be feeding-related, but simply a small step that culminated with positive change. This was important because positivity begot positivity[38]—a positive change, even if small, whet a carer's appetite for more change:

I like to think of us [clinicians] as their cheer squad to celebrate those wins with them. (*clinician 9*)

Normalcy

Experiences that exceeded expectations often promoted normalcy. According to the participants, managing a feeding difficulty disrupted the lives that carers had expected for themselves and their children, and sometimes created chaos. The chaos was inflamed by the anxiety and confusion that carers can experience when their child has a complex health condition. And when they felt out of their depth, acts of care that offered a sense of manageability made a considerable difference:

when we'd gone to get his [gastrostomy] button changed with the public system, there was a nurse there and she was really, really good… I was really panicking… and thought it was going to be horrific… She just made everything seem so normal… she was like, 'I'll just take this off and clean it and do that and do that', and we were like, 'Oh, okay, it's quite easy'… It was really, really good. (*carer 12*)

The significance of normalcy was also demonstrated beyond the confines of a health service. The carers and their

children had myriad other relationships, be they with teachers, family members, friends, or community members. Participants noted that their expectations were exceeded when carers and their children felt normal and not shunned. This was important because they often felt stigmatized by others who did not understand feeding difficulties or why tube-feeding was warranted. In contrast to such marginalization, opportunities to feel accepted and part of the collective brought joy. When carers and their children felt welcomed, their extraordinary feeding practices felt somewhat ordinary:

I saw a brilliant school that integrated all the tube kids into the canteen and all the kids had a menu, the same as everybody else. They knew what was going down their tubes… They could choose what they wanted, and they were part of the mealtime. (*clinician 4*)

DISCUSSION

This article clarified clinician and carer perspectives on what constitutes brilliant feeding care—care that exceeds expectations, fostering positive emotion and connections. Participants suggested that brilliant feeding care is bolstered by the following: practices that go above and beyond; attentiveness; empowerment; being 'on the same page'; hopefulness, and normalcy. These ingredients often made a world of difference to carers and their child, particularly during times of heightened adversity.

These themes captured the importance of practices that went beyond the oft-cited delivery of pedestrian or confusing care[21] to provide an unexpected level of support in a sensitive and respectful way.[25] Whether this involved taking extra time to listen to and understand carer concerns,[8,24] offering a kind word or gesture, or sourcing additional support, these unexpected practices demonstrated greater empathy and concern for the child and their carer. In essence, they charged care with brilliance.

Carers who experienced care that exceeded expectations were better able to manage the challenges of tube-feeding. This speaks to an upward spiral,[39] whereby positivity begot positivity—specifically, 'the psychological state influenced the ability to cope with challenges, and challenges impacted the psychological state'.[21,p.8] By stepping in and perhaps stepping up at these key times, those who facilitated brilliant feeding care influenced the child and carer's experiences.

As carer confidence grew with tube-feeding, normalization became an aspiration for many. Normalcy was experienced when they felt: supported to manage their child's feeding difficulty; and accepted, particularly in public spaces. These experiences often incited joy. However, for this to occur, the carer required an understanding and knowledge of tube-feeding. Care could then be integrated into a daily routine, with the hope of eventually forming a 'new' normal. Towards this aim, carers often sought the support of individuals who understood and supported their goals for their child.

The participants cited some of the barriers associated with fragmented care, with some recognizing brilliance in multidisciplinary care in which everyone was 'on the same page'. This reflects research on the value of multidisciplinary child healthcare.[48] Working as a multidisciplinary team also benefitted clinicians who appreciated the reduced burden of care when managing complex health issues. From the carers' perspective, this meant a consistent message from everyone involved in their child's care, reducing the confusion associated with receiving conflicting advice from disparate clinical voices.

Although this study focussed on brilliant feeding care, identifying it often required the participants to recount substandard care. Perhaps necessarily, they narrated the ordinary to personify the extraordinary. This was particularly the case when participants spoke of empowerment. For clinicians, rather than retain control over care, empowerment often involves encouraging others to exercise agency and support each other. For carers, a clinician's brilliant practices often preceded the carer's sense of urgency, heightened concern, or sheer exhaustion. When carers were vulnerable or distraught and shared their situation with a clinician who recognized their plight, a simple yet brilliant act of compassion offered confidence and hope, renewing carer resolve. Participant examples of this align with Fredrickson's[38] broaden-and-build theory, whereby the positive emotions experienced by the carer in those interactions were novel, even unexpected in thought or activity, building their resources and resolve over time; and Gilligan's[46] ethic of care where carers' expectations of what was the norm in care were exceeded and the

relationship with the giver of that care was strengthened. However, this is not to be confused with a paternalistic approach to healthcare, where the agency and autonomy of a patient or carer are undermined. Rather, it serves to highlight the importance of sensitivity and perceptivity to their needs and preferences to foster empowerment. Brilliant healthcare for children with feeding difficulties and their carers can be realized by considering what exceeds expectations and brings joy and delight to those who deliver and receive healthcare.[34]

The findings demonstrate that brilliance happens, sometimes with the smallest gesture or word. Acts of care that might seem trivial can make a world of difference to others, particularly those who experience stress or adversity—like carers of children who tube-feed. This is not to suggest that we should expect more from clinicians or carers, many of whom are time-poor and/or burnout. This offers an opportunity to highlight and reflect on instances of brilliant care that serendipitously occur in the day-to-day care of children who tube-feed. Principally, it is a call to celebrate positive experiences, however small, to acknowledge how people rise above the trials and tribulations often associated with health issues and healthcare, to (re)energise hope and sustain a collective capacity to promote child and carer well being.

Limitations

Despite the value of the findings, three methodological limitations warrant mention. First, the cross-sectional research design limits the lifespan of the findings. Second, there is no claim that the sample was representative of clinicians or carers who have cared for children with feeding difficulties, within or beyond Australia, particularly given the recruitment strategies and the voluntary nature of participation. And third, social desirability bias might have influenced the findings, whereby the participants altered their contributions to this study to present themselves and/or their situation ‘in a way that is perceived to be socially acceptable, but not wholly reflective of one's reality’.[49,p.783] As Nederhof[50] explained:

When the respondent actually believes a statement to be true of him or herself, even though it is inaccurate, ‘self-deception’ occurs… On the other hand, a person might purposely misrepresent the truth as a form of impression management motivated by a desire to avoid evaluation.

Implications

Notwithstanding these limitations, the findings have key implications for clinicians, carers, and scholars. For clinicians, the findings highlight strategies to support children with feeding difficulties and their carers—these include listening, being attuned and committed to their well being, sharing decision-making, sourcing resources, and instilling hope. Incorporating these strategies can capitalize on the interactions with carers without placing additional demands on a clinician's workload. Clinicians should not underestimate the power of a small word or deed as sustenance for carers' resolve the support of their child's health. The findings also point to the importance of normalizing feeding care—this might require education and training, not just for carers, but for anyone with an interest in child well being, including teachers and pastoral care workers. Such efforts are likely to build the skills, knowledge, and confidence required to support children with feeding difficulties and their carers. It is also important to celebrate successes, however small, and commend those who contributed to this success, including the child, their carer, as well as colleagues.

For carers, the findings suggest they should seek out and acknowledge clinicians who listen, involve them in decision-making processes, and continue to source the resources required (including expertise) to optimize child and carer well being. Given the findings, carers might also benefit from a peer support network. Sharing lived experiences can reduce carer stress, partly because of the opportunity to connect with those who are in the ‘same boat’.[51] Peer support can normalize experiences that are difficult and associated with stigma, open opportunities to learn practical strategies to manage challenging situations, build capacity, and boost confidence.

For scholars, this article offers fertile ground for research that builds on these findings. Specifically, scholarship is required to clarify the models of care—that is, ‘the way health services are delivered’,[52,p.3]—that foster brilliant feeding care as well as the conditions required to introduce and sustain these models. These conditions might encompass the leadership styles, the composition of interprofessional teams, the teamwork approaches, and the organizational cultures that enable brilliant feeding care in different contexts, including hospitals, outpatient

clinics, and home-based services, among others. Additionally, given the need to address the negative discourse regarding feeding care, scholarship is required to clarify the methodologies that serve to examine, understand, and promote brilliant healthcare. Given the demonstrated value of participatory methodologies,[53,54] this might involve the use of video-reflexive ethnography and/or co-design approaches with children who tube-feed, their carers, and the clinicians who work with them.

AUTHOR CONTRIBUTIONS

Ann Dadich conceived and managed the study, developed the Introduction and Methods and contributed to all sections. Simone Kaplun analysed the data and developed the Results. Cathy Kaplun developed the Discussion. Nick Hopwood and Christopher Elliot reviewed the article.

ACKNOWLEDGEMENT

This study was funded by the Western Sydney University School of Business.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data are not available due to ethical restrictions.

ETHICS STATEMENT

This study was approved by the Western Sydney University Human Research Ethics Committee (reference number: H13794). All participants provided informed consent.

1APPENDIXInterview schedule for clinicians

- 1.

  How did you become interested in feeding difficulties and/or tube-feeding?

- 2.

  What have you found useful when supporting children who tube-feed and/or their carers, and why?

- 3.

  What are your understandings of and experiences with brilliant feeding care?

- 4.

  What do you wish you would have known about feeding care, earlier in life, and why?

Interview schedule for carers

- 1.

  What is it like to care for a child who is tube-fed?

- 2.

  What helps or hinders feeding care, and why?

- 3.

  What priorities and considerations matter to you, and why?

- 4.

  What are your understandings of and experiences with brilliant feeding care?

- 5.

  What do you wish you would have known about feeding care, earlier in life, and why?

DETAILS

Subject:	Collaboration; Health care; Social networks; Families &family life; Data analysis; Caregivers; Feeding; Decision making; Emotions; Pediatrics; Interviews; Children; Schedules; Well being; Empowerment; Clinical decision making; Ostomy; Children &youth; Health services
Business indexing term:	Subject: Social networks
Location:	Australia
Identifier / keyword:	brilliant care; child health; feeding difficulties; tube-feeding
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	488-497
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-12-08

Milestone dates:	2022-11-19 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-07-26 (manuscriptReceived); 2022-12-08 (publishedOnlineEarlyUnpaginated); 2022-11-23 (manuscriptAccepted)
Publication history :	
First posting date:	08 Dec 2022
DOI:	https://doi.org/10.1111/hex.13683
ProQuest document ID:	2767101960
Document URL:	https://www.proquest.com/scholarly-journals/xa0-was-that-specialist-finally-listened-us-thats/docview/2767101960/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the "License"). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-04-24
Database:	Public Health Database,Publicly Available Content Database

Co-creation of information materials within the assent process: From theory to practice

Fons-Martinez, Jaime [1] ; Ferrer-Albero, Cristina [2] ; Diez-Domingo, Javier [3 1] Vaccine Research Area, Foundation for the Promotion of Health and Biomedical Research of Valencia Region, FISABIO, Valencia, Spain [2] Facultad de Medicina y Ciencias de la Salud, Universidad Católica de Valencia San Vicente Mártir, Valencia, Spain [3] Vaccine Research Area, Foundation for the Promotion of Health and Biomedical Research of Valencia Region, FISABIO, Valencia, Spain; Facultad de Medicina y Ciencias de la Salud, Universidad Católica de Valencia San Vicente Mártir, Valencia, Spain

ABSTRACT (ENGLISH)

Introduction

The informed consent process is key to safeguarding the autonomy of the participant in medical research. For this process to be valid, the information presented to the potential participant should meet their needs and be understood by them. The i-CONSENT project has developed 'Guidelines for adapting the informed consent process in clinical trials' which aim to improve informed consent so that they are easier to understand and better adapted to the needs and preferences of the target population. The best way to tailor information to the characteristics and preferences of the target population is to involve the community itself.

Methods
Following guidelines developed by i-CONSENT, assent materials were co-created for a mock clinical trial of the human papillomavirus vaccine in adolescents. During the process, two design thinking sessions were conducted involving a total of 10 children and 5 parents. The objectives of the sessions were to find out the children's opinion of the informed consent (assent in their case) process in clinical trials, identify the parts that were most difficult to understand and alternatives for their presentation and wording, identify the preferred formats for receiving the information and the main characteristics of these formats, design a video explaining the clinical trial and evaluate a tool for assessing comprehension.
Results
Assent materials were co-created in three formats: a web-based material following a layered approach; a video in story format; a pdf document with an innovative way of presenting information compared to traditional assent documents. In addition, the Comprehension of Assent Questionnaire was co-designed, based on the Quality of Informed Consent questionnaire.
Conclusion
The design thinking methodology has proven to be an easy and useful tool for involving children in designing information tailored to their needs and preferences.
Patient or Public Contribution
A sample of the target population participated in the design and piloting of the materials created using design thinking methodology. In addition, patient representatives participated in the design and evaluation of the guidelines developed by the i-CONSENT project that were followed for the development of the materials in this study.

FULL TEXT

INTRODUCTION

Many people still believe that the term informed consent (IC) is limited solely to obtaining the signature of research participants in the Informed Consent Form (ICF), unaware that this act is part of a much broader Informed Consent Process (ICP).[1]

During the ICP, efforts are made to protect the rights and welfare of participants at all times. The right to health protection is the main objective of legislators, researchers, sponsors, health professionals and the pharmaceutical industry. But the right to justice, freedom and participant autonomy must be ensured in all research involving human subjects.[2]

The ICP, described step-by-step in the 'Guidelines for tailoring the Informed Consent Process in Clinical Studies',[3] focuses on a continuous bidirectional communication process between the participant and the research team. It starts at the first contact of the potential participant with the study and continues until the end of the study and the corresponding dissemination of its results.[4]

There are therefore a series of phases in which relevant information is provided from the first contact with the potential participant. This information is discussed and clarified in an interview with a member of the research team who is trained to perform competently and with integrity.[1] The decision on whether or not to participate in the study should be made after ensuring that the potential participant has understood all relevant information provided and that any doubts that may have arisen have been resolved.

The central axis of the whole process is the relationship that is created between the researcher and the study participants. Knowledge, empathy, active listening, communication skills and respect should not be lacking in this relationship.

But since the interpersonal relationship that is created is not traceable and no record of what is discussed or talked about can be kept, it is necessary to ensure that the relevant information from any research study is presented and available to the potential participant in a clear, concise and patient-friendly manner.

The best way to adapt it to the characteristics and preferences of the target population is to involve the community itself, or a representative group of the community, in the design, development and execution of the ICP monitoring of the research, as well as in the dissemination of the results.[1]

In the same way that lay members are included in Ethics Committees to provide that perspective of potential participants, inviting lay members or patient groups to participate in the development of IC materials and resources will have a positive impact on the end result, as the process will be better understood and more suited to potential participants. Industry and patient organizations are committed to improving collaboration and building trust with all parties involved. The document developed by the European Federation of Pharmaceutical Industries and Association (EFPIA) on how to work with patient groups[5] is a reference point to guide these interactions.

This is the result of a shift from the traditional paternalist paradigm of care, inherited from Hippocratic medicine to a patient- and family-centred paradigm of care.

One of the first initiatives in this direction was the creation of Patient-Focused Medicine Development (PFMD) in 2015,[6] whose mission was to bring together and include all healthcare stakeholders in an open coalition for shared decision-making and to provide healthcare solutions. Among the outcomes of this collaboration, a practical guide was developed[7] for planning, developing and evaluating the quality of patient involvement activities and projects in the development and lifecycle of medicines.

Between 2012 and 2017, the European Patients' Academy on Therapeutic Innovation (EUPATI)[8] project was developed with the aim of increasing patients' involvement in the development and research of new medicines and treatments, improving their health literacy, becoming patient experts and empowering them in the management of their own health.

In the field of rare diseases, the Share4Rare project launched in 2018,[9] and seeks to empower patients by increasing their knowledge through information materials created in collaboration with patients.

With the aim of developing guidelines to help improve the ICP, the i-CONSENT project was launched in 2017.[4] One of the key points of the project is the inclusion of potential participants in the design and review of the information materials in a research setting, to ensure that they are understandable and tailored to the needs and preferences of the target population.[3]

Balik's[10] approach to providing patient- and/or family-centred care envisages three different approaches: ‘doing to’, ‘doing for’ and ‘doing with’. When we apply this to the ICP, we are faced with the challenge of making IC materials with the patient, where potential participants are involved in all phases of the process, especially in the design of information materials. To do this, sponsors and researchers must first understand the target population and then incorporate them into the design, development and review of the information materials to make them more inclusive and tailored to the actual needs of the participants.[3]

Tool V proposed in the guidelines, entitled ‘Methodologies and tools to incorporate the participants' perspective’,[3] proposes design thinking and focus group methodology to identify problem areas in the IPC, define and prioritize these problems and develop joint ideas and prototypes to solve them.

The participant is thus an active part of scientific progress and not a passive research subject. Co-creation in the ICP within any study seeks to encourage fair and open participation and quality input based on the experience and expertise of all stakeholders.

This article describes the process of developing informational materials for a hypothetical clinical trial (CT) with children following the recommendations of the i-CONSENT project. It focuses on the description of strategies for the co-creation of materials based on the characteristics of the target population, their needs and preferences.

METHODS

Taking for granted the social and scientific value that any research must have to be carried out, we worked on the design and co-creation phase of the information materials for a simulated study, following the recommendations of the i-CONSENT guidelines.[3] The steps to be followed in the development of materials are summarized in Table 1.

Table 1 Points to consider when preparing study information

•□ Have information materials been prepared taking into account the target population?

•□ Have you tested your communication materials with representatives of your target population? Have you tested it with men and women (if applicable)?
•□ Is the information clear and concise?
•□ Is the information relevant and complete?
•□ Has it been presented in a neutral/balanced way?
•□ Have you provided references to reliable sources of information?
•□ Does the study include placebo control? Have you informed participants about the details of its use and the placebo effect?
•□ Have you informed participants about incidental findings policy?
•□ Have you considered a range of media channels/platforms/formats?
•□ Have all the information materials been approved by an Independent Ethics Committee?

Source: *Guidelines for Tailoring the Informed Consent Process in Clinical Studies*.[3]

The scenario for the assent materials is that of the human papillomavirus (HPV) vaccine CT in adolescents, taking into account gender differences.

The target population and the scenario were defined according to the i-CONSENT project study protocol,[4] considering healthy children aged 12–13 years old for participation. In the same way and following the same protocol, the result of the co-creation work of information materials was validated in a later phase, measuring their comprehension in Romania, Spain and the United Kingdom. It was therefore necessary to create an information comprehension assessment tool.

The technique chosen to work with the target group was 'design thinking',[11–15] as it is a directly user-centred, action-oriented technique aimed at generating innovative solutions to a given problem. It involves several phases: empathizing, defining, devising, prototyping and validating or testing.

Development of the design thinking sessions

Two face-to-face sessions were scheduled in Valencia, Spain. Recruitment was done through the paediatric network VIVA (Vaccine Institute of Valencia), together with members of the i-CONSENT team. Participants were boys and girls aged 12–13 years, with no previous experience of participating in CTs and in good health. This is a challenge for vaccine CTs, as participants have no experience with the disease and are not aware of the indirect benefit of their participation.

As the aim of the sessions was to prepare materials that could be useful and easy to understand for both those who

have previously participated in CTs and have knowledge of the terminology and processes used in them, and those who have never participated in this type of research, it was decided to include only participants with no previous research experience, since they are the ones who, in principle, are at a disadvantage in understanding and have the greatest need for information. It was also considered that there may be a risk that those who had already participated in CTs could monopolize the conversation and make the rest of the participants uncomfortable because they were unfamiliar with certain terminology or processes. Convenience sampling was used, where three paediatricians from the VIVA network offered participation to parents and children in the consultation. Those who showed interest in participating voluntarily were invited to contact the i-CONSENT research team. All participants were informed of the purpose of the sessions, the benefit to other children, the inconvenience their participation might entail in terms of time and travel, the protection of their data and the right to withdraw at any time without giving any reason. They gave their assent to participate, and the parents gave their consent. A total of 10 children participated in the design sessions.

To create a safe and open space to increase comfort, trust and participation, the following strategy was applied:

•(1)

Sessions began with group dynamics focused on: introducing the participants and the researchers; informing them that other children had participated or were going to participate in similar sessions; highlighting the importance of each participant's role in the research, making them feel that a diversity of opinions among the participants was welcome and that all contributions were important to us.

•(2)

Many of the activities included written expression, with subsequent reading aloud by the researcher. This meant that an idea or answer was not attributed to any specific person, encouraged all opinions to be heard no matter who said it and prevented the exercise from being monopolized by any one participant.

First session

The objectives of the first session were:

•(1)

Create a climate of trust and empathy between children, parents and the research team.

•(2)

Share views on CTs for vaccine development and identify wishes and needs relevant to the group of participants and their parents.

•(3)

Prototype assent materials with preferred formats.

Two members of the research team welcomed the five children and their five parents and acted as facilitators guiding the group through the process. The participants were introduced to each other using a dynamic presentation through a game with a ball to encourage interaction between them. With this playful component, a positive emotional climate was established and the relaxation of those involved was achieved.

As this was a group of healthy children with no previous experience of participating in CTs, and in order for them to understand what a CT is, a 5-min 11-s educational video in Spanish on how a CT is developed and conducted, produced by the European Communication on Research Awareness Needs (ECRAN),[16] was shown. The aim was to understand what would be really relevant for children and parents if they would participate in a CT with vaccines. Subsequently, a role-play was conducted with a vaccine CT scenario, in which both children and parents

participated by assuming a role (participant, parents, researcher or doctor) and following a given script. At the end of the role-play, participants were given a traditional assent form to read and make decisions. They were given the paper-based assent document, based on the ICF used in a real trial (EudraCT no. 2006-000764-85) and were given the time they needed to read it.

Participants expressed their emotions, using balloons on which they drew faces expressing their mood with the information received in the assent and how they would feel if they had to make the decision to participate in the CT at that moment. In this way, it was possible to better understand the problems experienced by the participants and the feelings they have in a situation such as this.

With the information obtained the focus of action could be defined by focusing on the aspects relevant to the participants. The format 'The (user) wants/needs (want/need) because (insight)' was used.

The information collected was clustered into different areas of improvement: information (purpose, risks, benefits, personal data, right to revoke, conditions, procedure), format (web, app, video, comic, text, oral explanation) and decision-making (individual, shared).

Once the focal points for action had been collected and synthesized, the question arose as to how we could devise and design the best solutions to the problems raised.

To this end, through brainstorming, participants reflected on the information presentation formats they would prefer and were asked to design a prototype of assent material (video and infographic).

With all this work (summarized in Table 2), the first session ended and their participation was thanked.

Table 2 Objectives and methodology for the first design thinking session with children and their parents

Objectives	Methodology
Empathize	Presentation dynamic: 'passing the ball'
Identify and define	Viewing video on Clinical Trials
	Vaccine clinical trial role play and decision making with a traditional text-based reporting document
	Clustering to define areas for improvement: information and formatting
Devising	Brainstorming for alternative presentation formats
Designing prototypes	Design of prototypes with different formats (video and infographics)

Second session

The second design thinking session included more detailed tasks involving another five children at the same age. The objectives were different, as the results of the previous session were already being used as a starting point:

•(1)

Detect words that are difficult to understand, and propose a glossary of terms.

•(2)

Read the modified written assent document for the hypothetical HPV vaccine study to identify information that is difficult to understand and propose a plain language explanation.

•(3)
Evaluate the comprehension assessment tool.

•(4)
Assess the understanding of the information provided.

The second session began with a review of the previous session in the form of a narrative story, telling them about when and where the previous session took place, the characteristics of the children who participated, the objectives of the session and the results obtained. The points for improvement identified in session 1 were presented on a whiteboard using a mind map. This allowed to focus the children's attention, introduce them to the topic and the progress of the first session and explain the objectives of the second session.

The mind map graphically represented the main ideas, highlighting the most relevant points and making it easier for the children to focus their attention and follow the story. The first area of improvement detected in the previous session referred to the amount of information included in the initial document. Following the guidelines set out in Fact Sheet IV of the i-CONSENT guide: 'Information to be given to potential participants during the information phase' and taking into account the EU 536/2014 Regulation on CTs,[17] the original information document worked on in the first session was adapted.

The title proposed as a result of the text adaptation was: 'Phase III study on the HPV vaccine in youth from 9 to 14 years of age'. The i-CONSENT guidelines recommend using inclusive language and avoiding gendered roles. We also followed the recommendations on the gender perspective included in the guidelines, which recommends developing a single material for all participants, in the event that there are no exclusion criteria based on gender; and the recommendations to adapt the information to the minor's age and maturity.

As the amount of information in the text document proved to be overwhelming in the first session, the information was presented using a layered approach, maintaining the completeness of the information provided. The first layer was prepared with the relevant information, and the second was left for further information and a glossary of terms difficult to understand.

To test the new assent document prepared for the second session, the participants were asked to mark in colour the words they did not understand. Members of the research team explained the terms they did not understand, and the participants were asked to write an explanation in their own words. The definitions were accompanied by their own illustrations, which provided guidance on the type of drawing and the aspects to be highlighted.

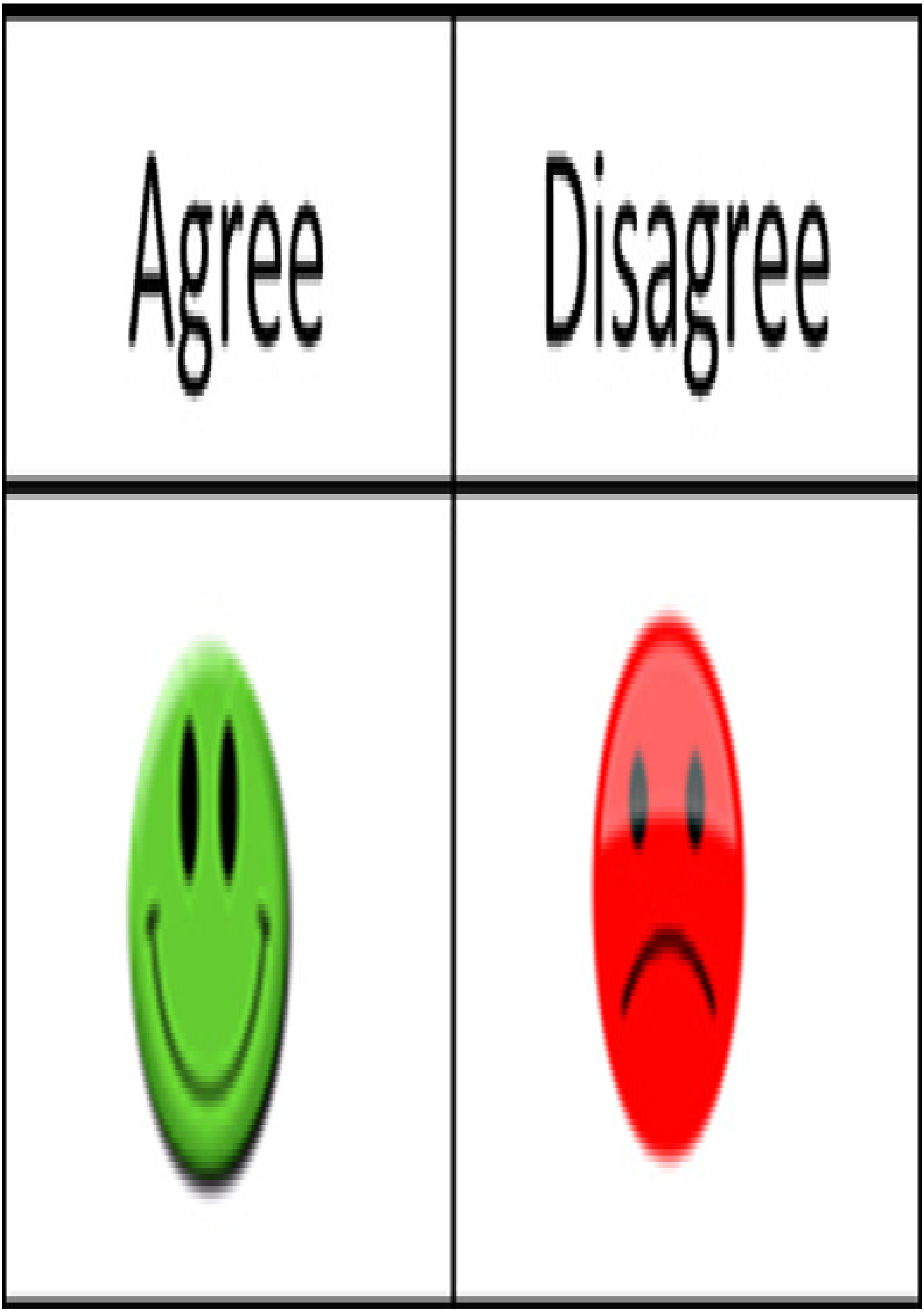
Thus, a glossary of terms difficult to understand was created with the participants to expand the information in plain language and use it in a second layer with additional information. It included the concepts of a placebo, vaccine safety, blood tests, confidentiality and the right of revocation.

In terms of format, as requested in the first session, the use of graphic components to complement the information such as icons, infographics and simple and easy-to-interpret images was added, making the written information more easily readable and understandable.[17]

The use of digital tools and/or multimedia components[18] and the possibility of offering the participants different formats to receive the information was worked on with the children. In both sessions, the four options most discussed were: text, video, comic and web. Through brainstorming, the children contributed their preferences and then worked on a prototype of a website to present the information.

It is important to consider the provision of information in written or digital format as a complement to, not a substitute for, face-to-face discussions with the research team. Evidence suggests that simple and brief consent forms,

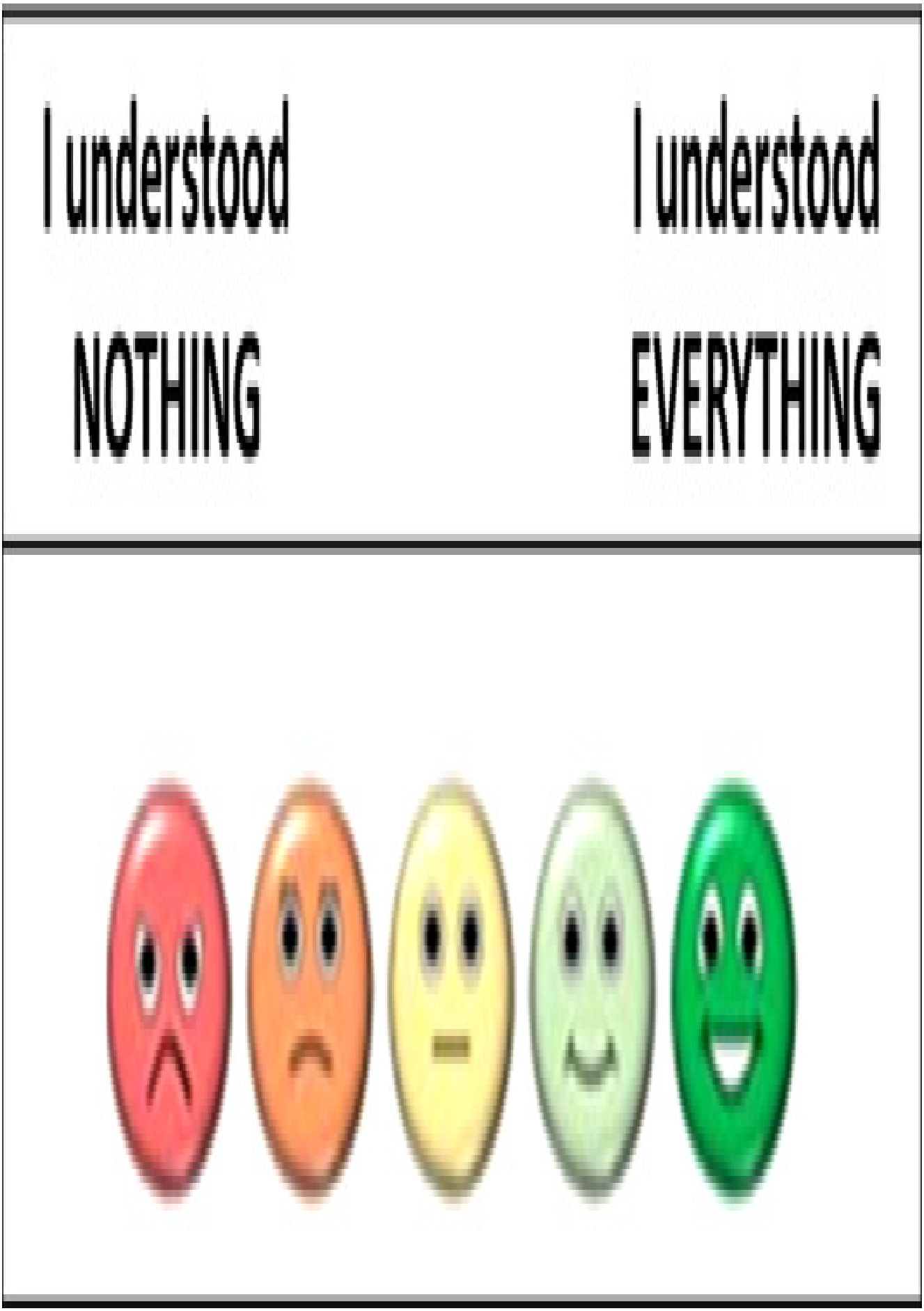
accompanied by a meaningful conversation between participants and researchers, can improve comprehension.[19]
To assess comprehension of the information, an Assent Comprehension Questionnaire for vaccine studies (abbreviated ‘C-CAsIn’ for ‘Cuestionario de Comprensión del Asentimiento Informado’) was developed in Spanish, based on the Quality of Informed Consent (QuIC).[20]
During this session, the comprehension of the items of the C-CAsIn questionnaire was analysed. Those items that raised doubts were rewritten with the children's help. The Likert-type response was adapted by changing the numbers (1–5) with small icons that graphically represented an emotion or idea (emoticons).
In the first part of the questionnaire, which assesses comprehension objectively, the response possibilities for each statement were represented by a green, smiling icon for ‘agree’ and a red, sad icon for ‘disagree’ (see Figure 1).



Enlarge this image.

In the second part of the questionnaire, which assesses comprehension subjectively, the response possibilities were

widened and broken down further, with the possibility of choosing between five degrees of comprehension between ‘I understood NOTHING’ and ‘I understood EVERYTHING’ (see Figure 2).



Enlarge this image.

The last part of the questionnaire includes a series of general questions about previous experience in a CT, satisfaction with the information received, the preferred format for receiving the information and sense of understanding of all the information.

Before closing the session, a brainstorming session was held on how to improve the information received, how they would adapt it to an interactive format and what elements they would use to support the information (links, pop-ups, embedded videos, etc.). Table 3 summarizes the work done during the second session.

Table 3 Objectives and methodology of the second design thinking session with children

Objectives	Methodology
Empathize	Narrative story and mind map explaining the previous session and placing the main issue in the centre (information in assent) and connecting the different strands or areas of improvement: information and format
Identify and define	Reread adapted information document design to identify poorly understood concepts and define glossary of terms for second layer of information
Designing prototypes	Web prototype design Brainstorming: features of narrated video
Validate/test	Test the assent comprehension assessment questionnaire
	Test the information received

Table 4 Comprehension of Assent Questionnaire (C-CAsIn) Part A

No.	Question	Agree	Disagree	Section of information
A1	I can decide to participate in this study without discussing it with my parents. Their opinion does not matter.	[IMAGE OMITTED. SEE PDF.]	[IMAGE OMITTED. SEE PDF.]	Decision-making
A2	One of the benefits of participating in this study is helping other children. What the researchers learn from me can be applied to others.	[IMAGE OMITTED. SEE PDF.]	[IMAGE OMITTED. SEE PDF.]	Indirect benefit
A3	The researchers have told me how long the study will take.	[IMAGE OMITTED. SEE PDF.]	[IMAGE OMITTED. SEE PDF.]	Procedures

A4	The study vaccine has been tested before in many girls and boys.	[IMAGE OMITTED. SEE PDF.]	[IMAGE OMITTED. SEE PDF.]	Procedures
A5	One of the objectives of this study is to see how safe the vaccine is.	[IMAGE OMITTED. SEE PDF.]	[IMAGE OMITTED. SEE PDF.]	Aim of the study
A6	One of the benefits of participating in this study could be improving my defenses against diseases.	[IMAGE OMITTED. SEE PDF.]	[IMAGE OMITTED. SEE PDF.]	Direct benefit
A7	After I decide to participate in this study, I will be randomly (like playing heads or tails) put in a group.	[IMAGE OMITTED. SEE PDF.]	[IMAGE OMITTED. SEE PDF.]	Randomization procedure
A8	I will know what group I am put in throughout the whole study.	[IMAGE OMITTED. SEE PDF.]	[IMAGE OMITTED. SEE PDF.]	Blinding Procedures
A9	If I receive the placebo, my defenses will improve.	[IMAGE OMITTED. SEE PDF.]	[IMAGE OMITTED. SEE PDF.]	Placebo Procedures
A10	Participating in this study does not involve any risk or inconvenience.	[IMAGE OMITTED. SEE PDF.]	[IMAGE OMITTED. SEE PDF.]	Risks
A11	By participating in the study, I would be helping the investigators to know more about the product they study.	[IMAGE OMITTED. SEE PDF.]	[IMAGE OMITTED. SEE PDF.]	Aim of the study
A12	The information that I have read explains who I have to talk to if I am worried or if I have any questions.	[IMAGE OMITTED. SEE PDF.]	[IMAGE OMITTED. SEE PDF.]	Further information
A13	If I do not want to participate, I can leave the study without any problem.	[IMAGE OMITTED. SEE PDF.]	[IMAGE OMITTED. SEE PDF.]	Voluntariness
A14	I have to stay in the study even if I want to quit.	[IMAGE OMITTED. SEE PDF.]	[IMAGE OMITTED. SEE PDF.]	Right to withdraw

Table 5 Comprehension of Assent Questionnaire (C-CAsIn) Part B

Num.	I understood…	I did not understand ANYTHING	I understood EVERYTHING
B1	That the study vaccine is being investigated.	[IMAGE OMITTED. SEE PDF.]	
B2	That my participation in the study will help other children.	[IMAGE OMITTED. SEE PDF.]	
B3	How long will I be in the study.	[IMAGE OMITTED. SEE PDF.]	
B4	What the researchers are trying to achieve by doing this study.	[IMAGE OMITTED. SEE PDF.]	
B5	What will be done at each visit.	[IMAGE OMITTED. SEE PDF.]	
B6	The possible risks and inconveniences of participating in this study.	[IMAGE OMITTED. SEE PDF.]	
B7	The possible benefits of participating in the study.	[IMAGE OMITTED. SEE PDF.]	
B8	Which people will know that I am participating in the study.	[IMAGE OMITTED. SEE PDF.]	
B9	Whom I will need to talk to if I have any questions or worries about the study.	[IMAGE OMITTED. SEE PDF.]	
B10	That it is not compulsory for me to participate in this study.	[IMAGE OMITTED. SEE PDF.]	

RESULTS

Ten healthy children with no previous experience in CTs and their parents participated in the design thinking sessions. All the children were 12–13 years old and lived in the Valencian Region.

The final design of the assent information materials for the hypothetical trial with minors was discussed with external design and digital communication experts.

The text was improved in terms of its linguistic readability using the Fernández-Huerta Index (IFH)[21] and the Flesch-Szigriszt Index (INFLESZ) readability scale,[22,23] using the web tool 'Legible'.[24] The full-text readability scores of the first layer were:

•(1)
IFH: 'easy' (80.46 points);

•(2)
INFLESZ index: 'fairly easy' (76.52 points);

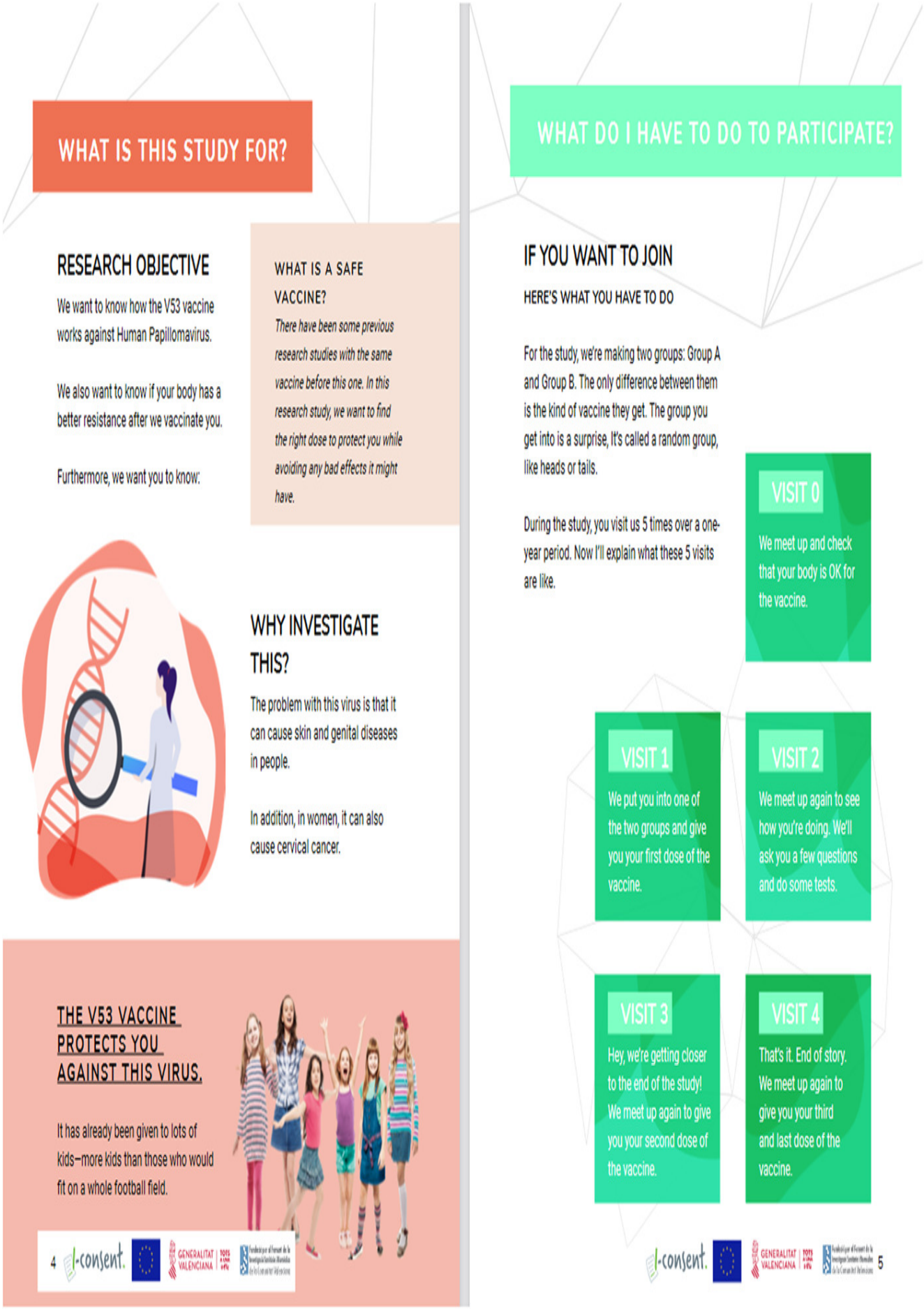
•(3)
Estimated reading time: 6 min;

•(4)
Years of schooling needed to understand Crawford's[25] formula: 4 years.

Following the suggestions of the children, visual aids were added and the text was accompanied by images, animated gifs and photographs featuring children.

The sketches made by the children on the design of the website were taken into account for the visual and navigational design of the website. The website (Figure 3), offered the possibility of obtaining the information in the website, narrated video and/or written text (document in pdf format) with icons and images (Figure 4). At the bottom of the website, at the end of the information, the comprehension evaluation questionnaire was placed.

Enlarge this image.



Enlarge this image.

The final version of the *C-CAsIn* for vaccine studies was designed in collaboration with the children in several

sections:

•(1)

Introduction: explanation of the study, objective, procedure, duration of participation, right to withdraw, voluntariness, decision making

•(2)

Part A—Objective (Table 4): 14 items written in plain language, with two response possibilities symbolized by facial expressions and colours, green for agreement and red for disagreement. The questions tested comprehension of all sections of the information provided.

•(3)

Part B—Subjective (Table 5): 10 items whose wording starts with ‘I understood…’. The response possibilities are wider, with 5 possibilities between ‘I didn't understand anything’ and ‘I understood everything’. Also symbolized by a colour code and a visual facial code.

•(4)

The last section of the C-CAsIn includes a short questionnaire with 8 items on sociodemographic data (age, sex and country of residence), previous experience of participation in a CT, the difficulty of the information received and preferred format and overall satisfaction with the information received.

The final digital assent form was created on a web page with a narrated video. All documents underwent several rounds of text adaptation, review of assent content requirements, review of the comprehension assessment tool, translation from Spanish into English and Romanian and linguistic adaptation for end users by native translators. Finally, potential participants also tested the information prototypes, providing their final improvements which were taken into account before the information was uploaded to the target website and before validation in the target population of 620 children aged 12 and 13 in Spain, England and Romania.

Before final publication, it was checked that the recommendations for the preparation of the study information in the i-CONSENT guidelines had been followed (Table 1).

It should be noted that the sessions did not seek consensus, but took into account all ideas and positions expressed in the design of the materials. Priority was given to suggestions that were common to the majority of participants.

The final materials are available on the following websites:

•(1)

Spanish version: http://iconsent.pilotvalidation.eu/estudio-adolescentes/;

•(2)

English version: http://iconsent.pilotvalidation.eu/en/teenagers-study/;

•(3)

Romanian version: http://iconsent.pilotvalidation.eu/ro/studiu-pentru-adolescenti/.

DISCUSSION

The process of designing the information materials for an ICP is perhaps the central part of any research study since it determines the potential participants' understanding of the information and, therefore, their autonomy in making free and informed decisions. This is also important to make the study population feel that they are at the centre of the research and that they participate and collaborate consciously and voluntarily.

There are various factors that influence the understanding and interpretation of the information a person receives, but it is the task of sponsors, industry and researchers to ensure that each and every participant understands it. The amount of information received by children before participating in a CT is overwhelming, as was seen in the two design thinking sessions conducted in this study. But, according to Regulation (EU)536/2014,[17] it should include the nature, objectives, benefits, implications, risks and inconvenience of the CT, rights and guarantees of their protection, right to withdraw at any time without any problem and without justification, the conditions of the study, including the duration of participation and treatment alternatives. Faced with this large amount of information, the proposal developed in this study is to use a layered approach to present it. The first layer would contain brief information on the aspects covered by the legislation, and the second and successive layers would allow for further information. In this way, the child who wishes to know more about a specific aspect can expand on this information. All this information should be clear, concise and adapted to the child's capacity to understand, but little account is taken of the information that children really want and need to know, as Roth-Cline and Nelson[26] pointed out. The systematic review carried out by Fons-Martínez et al.[27] shows that information needs are not the same for legislators, children, their parents and members of the research team. Focusing attention on the needs of children, it is observed that their interest is especially directed towards procedures, confidentiality and benefits[28]; knowing why they have been chosen to participate and if other children like them have already participated to ask them about their experience.[29] In the study conducted by Tait et al.,[28] slight differences were found with respect to gender at ages 13–17, with girls showing more interest in obtaining more detailed information about the procedure, objective, benefits, voluntariness and right to withdraw, and boys more interest in the alternatives.

But the amount of information is one thing; the difficulty of reading and understanding it is another. The urgent need to improve the readability of the information a minor receives before giving consent was already highlighted by Grootens-Wiegers et al.,[30] following a systematic review where the gap between the readability of the information and the reading level of minors was observed. Documents are often long, their readability low[31] and the language complex, negatively impacting the ICP.[32] What may seem simple to read and understand for trial sponsors and researchers can be complex for participants. In the present study, the readability of the initial information was improved by constructing shorter sentences with simpler terms, fewer syllables and more direct grammatical structures.[33,34] In this process, the contributions made by the children were of great help, as they participated in the drafting of the aspects that were more difficult for them to understand after being explained by the researchers.

To facilitate reading, the text was accompanied by simple pictures which, although not proven to significantly improve comprehension of the information, do improve satisfaction and the child's subjective belief that their understanding is improved.[35]

Attempts to improve the formats of information materials presented to children participating in research have been numerous in recent years, but none of them conclusive. Although the improved readability of written text and the comic format were shown to improve the comprehension of some aspects of the information presented to children compared to a traditional text format,[36,37] children participating in our design thinking sessions preferred other more interactive formats. The video format and the combination with multimedia tools[18] have also shown improvements in understanding and satisfaction with the information received by children in numerous previous studies,[38–41] as preferred by the children who participated in the co-creation process of the present study.

It is possible that all of these novel proposals in previous studies would have shown a greater positive impact on children's understanding and acceptance if they had also been involved in the design process.[13] In this way, the information and format would have been better adapted to their needs and preferences. It is not about offering a wide variety, but about offering what each age group prefers. Even making information more readable and attractive

to children does not ensure that they will understand it.

One of the fundamental problems is the lack of validated tools to assess the comprehension of information in minors participating in an assent process. Although it is best to assess the level of comprehension of information through a natural conversation between the potential participant and the researcher,[42] these tools make it possible to homogenize the process of verifying comprehension, provide an objective record of comprehension during the assent process and serve as a support for those researchers who are less skilled in carrying out this assessment through a natural conversation. Several studies have developed and validated tools, such as the MacArthur competence assessment tool for clinical research (MacCAT-CR)[43] to assess the competence of minors, and the QuIC,[20] which measures comprehension objectively and subjectively, in cancer patients involved in CTs. Other studies such as Chaisson et al.'s,[44] Lee et al.'s[45] and Blake et al.'s[46] have developed ad-hoc questionnaires with true/false items, to measure comprehension improvement after an intervention; none of these tools have been validated.

Based on the QuIC, as it is the most widely used questionnaire in different studies to measure comprehension, we adapted and created a new version for children, with the children's participation. Their participation at this point was crucial, as all their contributions to the items and the presentation format resulted in a new questionnaire (C-CAsIn) that was shorter, more comprehensible and simpler in its response format.

Co-creating by involving children increases the complexity of the process of designing information materials, but the benefit for them is direct, as it is adapted to their needs, increases their understanding and autonomy and therefore improves the decision-making process.

The limitations found in the present study were related to the fact that the children were not real participants in the CT for which the materials were being developed, which could generate a bias in their response. Working with a sample of children living in the Valencian Region may affect the transferability of the results.

CONCLUSION

This article describes the methodology for the design and elaboration of IC materials for CTs with children (assent) and defines the specific tools to be used.

To ensure that the informational materials are tailored to the child's maturity, preferences and needs, it is recommended that a representative group of the target population be included in the design of the materials.

The design thinking methodology has proven to be an easy and useful tool to involve children in the design of information adapted to their needs and preferences.

It is recommended to conduct two working sessions focusing on three main topics:

- 1. what information is relevant to them;
- 2. which concepts are difficult for them to understand and
- 3. in what format they prefer to receive this information.

This will improve their understanding and promote their autonomy.

In addition, as part of the assent process in a CT, it is necessary to confirm that the information provided to the child has been understood. The C-CAsIn survey has been designed, together with the children, to test understanding of information in the assent process of vaccine CTs, however, it should always be checked for its suitability to the

particular study design.

ACKNOWLEDGEMENTS

We would like to thank Dafina Petrova Dobreva, Júlia García Bayarri, Sally Jackson and María Cubillo Díaz-Valdés for their contribution to the design of the design thinking sessions. Our thanks also go to the paediatricians of the VIVA network who facilitated the recruitment of participants for the study. Special thanks to the children who participated in the design thinking sessions and to their parents. Our thanks to the European Commission for funding the i-CONSENT project and to the partners who helped us to develop the guidelines followed in this study.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DETAILS

Subject:	Gender differences; Clinical trials; Vaccines; Guidelines; Collaboration; Medical research; Communication; Clinical research; Questionnaires; Pharmaceutical industry; Children; Internet; Evaluation; Patients; Human papillomavirus; Wording; Design thinking; Comprehension; Community involvement; Participation; Documents; Informed consent; Empowerment; Computer based; Treatment preferences; Autonomy
Business indexing term:	Subject: Pharmaceutical industry
Location:	Spain
Identifier / keyword:	assent; design thinking; ethics; information materials; informed consent process; participant-centred design
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	429-439
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford

Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-23
Milestone dates:	2022-08-29 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2021-09-21 (manuscriptReceived); 2022-11-23 (publishedOnlineEarlyUnpaginated); 2022-11-10 (manuscriptAccepted)
Publication history :	
First posting date:	23 Nov 2022
DOI:	https://doi.org/10.1111/hex.13675
ProQuest document ID:	2767101959
Document URL:	https://www.proquest.com/scholarly-journals/co-creation-information-materials-within-assent/docview/2767101959/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the “License”). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-05-18
Database:	Public Health Database,Publicly Available Content Database

How do clients with multiple problems and (in)formal caretakers coproduce integrated care and support? A longitudinal study on integrated care trajectories of clients with multiple problems

Lieke Reinhoudt-den Boer [1]
; Jeroen van Wijngaarden [1] ; Huijsman, Robbert [1] [1] Department of Health Services Management &Organisation, Erasmus School of Health Policy and Management, Rotterdam, The Netherlands

ABSTRACT (ENGLISH)

Introduction
Integrated care can create several advantages, such as better quality of care and better outcomes. These advantages apply especially to clients with multiple problems (CWMPs) who have multiple, interconnected needs that span health and social issues and require different health care (e.g., mental health care or addiction care), social care (e.g., social benefits) and welfare services at the same time. Integrated care is most often studied as a phenomenon taking place at the system, organizational, professional and clinical levels. Therefore, in many studies, clients seem to be implicitly conceptualized as passive recipients of care. Less research has been conducted on how clients and (in)formal caretakers coproduce integrated care.
Methods
We performed a longitudinal study to investigate how CWPMs and (in)formal caretakers coproduce integrated care. Data were collected among CWMPs and their (in)formal caretakers in Rotterdam, the Netherlands. CWMPs' care trajectories were followed for 1–1.5 years. CWMPs were interviewed three times at an interval of 6 months (T0, T1, T2). Informal caretakers were interviewed three times (T0, T1, T2), and formal caretakers of 16 clients were interviewed twice (T1, T2). Data in the municipal record systems about participating CWMPs were also included.
Results
Our study shows that the CWMPs' multidimensional needs, which should function as the organizing principle of integrated care, are rarely completely assessed at the start (first 6 weeks) of CWMPs' care trajectories. Important drivers behind this shortcoming are the urgent problems CWMPs enter the support trajectory with, their lack of trust in ‘the government’ and the complexity of their situations. We subsequently found two distinct types of cases. The highest level of integrated care is achieved when formal caretakers initiate an iterative process in which the CWMP's multidimensional needs are constantly further mapped out and interventions are attuned to this new information.
Conclusions
Our study indicates that integrated care is the joint product of formal caretakers and CWMPs. Integrated care however does not come naturally when CWMPs are ‘put at the center’. Professionals need to play a leading role in engaging CWMPs to coproduce integrated care.
Patient Contribution
CWMPs and their (in)formal caretakers participated in this study via interviews and contributed with their experiences of the process.

FULL TEXT

INTRODUCTION

Integrated care has the potential to generate several advantages, including better quality of care (experienced by the client), better continuity of service, better outcomes and better cost efficiency.[1–4] Integrated care has been defined as ‘an approach to strengthen people-centered health systems through the promotion of the comprehensive delivery of quality services across the life-course, designed according to the multidimensional needs of the population and the individual and delivered by a coordinated multidisciplinary team of providers working across settings and levels of care’.[5] This holistic personalized perspective on clients pays attention to the origin of clients' symptoms on a psychological, mental, medical and (psycho)social level and consciously adopts their needs, preferences and perspectives.[6] The advantages of integrated care apply especially to clients with multiple problems (CWMPs), as they need different services from different social support and care providers at the same time to address all their

needs.[7,8] CWMPs are people who experience various combinations of mental illness, intellectual disability, acquired brain injury, physical disability, physical conditions, behavioural difficulties, homelessness, social isolation, family dysfunction and addiction.[8]

Integrated care has been studied extensively. Nevertheless, despite numerous studies, the evidence that integrated care leads to improved outcomes is dispersed and inconsistent.[4,6] Integrated care is most often studied as a phenomenon taking place at the system, organizational, professional and clinical levels, including functional and normative dimensions.[9] Many studies have focused on the barriers, difficulties and effects of cross-sectoral, cross-organizational and interprofessional collaboration.[4,7] With the main focus on these levels of integration, clients often seem to be implicitly conceptualized as passive recipients of care, not as active coproducers of services.[4,10] Consequently, clients' impact on the establishment and outcomes of integrated care may be overlooked.[10]

In recent years, there is an increasing call in the literature on integrated care for stimulating coproduction. Coproduction in this context is described somewhat ‘idealistic’ as ‘engaging clients, their families and communities in the design, implementation and improvement of services through partnership in collaboration with professionals and providers’.[11] Active involvement of clients, their families and the community is in this type of literature regarded as an essential condition for the success of integrated care.[12–14] Coproduction or actively engaging clients, families and communities are seen as a valuable route to harness their power, attune services to their needs and increase their ability to self-care (especially for unserved populations and marginalized groups).[11,14]

While coproduction is seen in the literature on integrated care as something to strive for, in service management literature coproduction is regarded as inevitable and intrinsic to any service experience.[15–18] Services have four distinctive characteristics: intangibility (services are intangible before delivery), inseparability (the production and consumption occur during the interaction between professional and client), variability (the service's quality and outcomes are shaped within the interaction between professional and client) and perishability (services cannot be stored).[15,18] In this body of literature, it is underpinned that services do not have any intrinsic value to their users in advance of their usage. Service organizations can only ‘promise’ a certain experience, but their actual performance is coproduced in the interaction with their users.[15,18] In that sense, the delivery of integrated care services is always a coproduction, although the level of involvement of both (in)formal caretakers and clients may vary.

To add to the literature on integrated care, we focus on how CWMPs, informal caretakers and formal caretakers coproduce integrated care. In this study, informal caretakers are people who provide unpaid care to the CWMP with whom they have a social relationship, such as a spouse, parent, child, other relatives, neighbour, friend or other nonkin. This informal care involves, for example, help with household chores or other practical errands, transport to doctors or social visits, social companionship, emotional guidance or help with arranging professional care.[19] In accordance with the service management literature, we consider integrated care as inevitably coproduced, although the level of involvement of the participants may vary. Our main question is as follows: How do CWMPs and (in)formal caretakers coproduce integrated care and support? We use data gathered among CWMPs and their (in)formal caretakers in Rotterdam, the Netherlands.

METHODS

We chose a qualitative research design for this study because the coproduction of integrated care is a complex and multidimensional phenomenon, which is hardly studied. Qualitative methods help us provide rich descriptions of this phenomenon and will help enhance our understanding of the context as well as the underlying mechanisms.[20]

Setting

Data were collected among ambulatory CWMPs. CWMPs are an interesting group of clients to study how integrated care is coproduced. It is widely acknowledged that people who have problems on psychological, mental, medical and (psycho)social levels need a continuum of care designed according to their multidimensional needs delivered by different actors, services and facilities involved on multiple levels of welfare, health care and social services to address all their needs.[5,21]

Data were collected in five districts in Rotterdam, the Netherlands: Bloemhof, Hillesluis, Lage Land, Ommoord and Lombardijen. Rotterdam is the second largest city in the Netherlands and is known for its large population of people

with socioeconomic and (psycho)social problems. In the selected districts, large concentrations of these people can be found, although Ommoord scores slightly better compared to the other four districts.[22]

Since 2015, as part of a major welfare state reform in the Netherlands, responsibility for social care and support, basic income provisions and youth care have been decentralized from the central government to municipalities. The idea behind this decentralization is that municipalities are more capable than the national government of being responsive to local needs and can provide tailored, integrated care as they are (literally) closer to clients. The reform was envisioned as a transition from a welfare state to a participation society, which places greater emphasis on citizens' individual responsibility, engaging civil society and shrinking the role of the state.[23,24] Traditional roles (citizen as client) were reshaped (citizen as coproducer).[25]

Participants

CWMPs were recruited via community-based primary care teams (CT) professionals (CPs). As part of the implementation of the welfare state reform, a community-based primary CT was established in every district in Rotterdam. Community-based primary CPs are assigned by the municipality of Rotterdam to completely assess the multidimensional needs of CWMPs and organize integrated care. Citizens can only turn to CPs when they are faced with multiple problems. CPs have different disciplinary backgrounds, for example, social psychiatric nurses, youth care workers, social workers, community workers, counsellors for elderly individuals and intercultural workers. The procedures prescribe that CPs map out the CWMP's multidimensional needs within the first 6 weeks. Based on this assessment, the CPs, together with CWMPs and their informal network (if available), are expected to organize integrated care. CPs provide support themselves and work together with professionals in their teams and with professionals across the boundaries of their teams, such as housing corporations, general practitioners, addiction therapists, mental health organizations, charity and religious organizations and CWMPs' informal networks. CPs have 6–9 months to organize care and support and refer the CWMPs to the appropriate professionals and institutions for follow-up, if necessary.

Our aim was to follow CWPMs for 1 year, from the start of their involvement with a CT, until several months after a referral from the CT. This allowed us not only to track and reconstruct the entire coproduction process but also to see the longer-term effects. CPs were asked to inform CWMPs within the first 6 weeks of their involvement with CWMPs. A period of 6 weeks was chosen in coordination with CPs. CPs indicated that 6 weeks were needed to introduce the study properly, for example, to establish an initial trust relationship. As inclusion was difficult at this study's start, an incentive (a 10-euro gift card) was introduced. Incentives increase the likelihood of participation but could negatively affect the data collection or the human subject.[26,27] We, however, think that the conditions that may lead to a negative impact were absent in our study: subjects were not in a dependent relationship with the researcher, the study is not degrading and the incentive was not that high that it would overrule participants possible aversions.[27]

All CWMPs signed a declaration of consent before participation. CPs ensured that CWMPs understood the study's content via an extensive oral explanation. Figure 1 gives an overview of the data collection process.

Figure 1. Data collection process

Due to different types of circumstances, such as imprisonment, mental breakdown, stress overload of the participating CWMPs and struggles to contact them (e.g., disconnected phones or CWMPs not answering their phone), our interview planning was not always attainable. This challenge is inherent to CWMPs' situation and characteristics.[26] Most interviews were held around the scheduled date, with a maximum deviation of 3 months. The clients' characteristics and reasons for nonparticipation in T0, T1 and T2 can be found in Appendix A.

(In)formal caretakers were recruited via participating CWMPs. All CWMPs were asked whether the researchers could interview their informal caretakers at T0. Although we aimed to also include informal caretakers in our study, the reality was that many CWMPs did not have informal caretakers (e.g., they lost their informal network as their problems increased), did not want to involve their informal network in the care trajectory or they did not have an informal network that could contribute to the care trajectory (e.g., informal network occupied with their own (multiple) problems or consisted of criminals or addicts).

At T1, via a purposeful sampling strategy, 16 CWMPs were asked whether all involved formal caretakers could be interviewed. Cases varied, such as excellent or rich examples of cases, cases representing a variety of typical situations and cases meeting predetermined criteria (e.g., CWMP).[28,29] The inclusion of involved formal caretakers at T1 was decided after experiences with their inclusion at T0. A trust relationship was necessary for CWMPs to feel confident that it would not harm their support trajectory or privacy. Forty-six formal caretakers participated in this study. The (in)formal caretakers' characteristics, including reasons for nonparticipation, can be found in Appendix B. Data in the municipal record systems about participating CWMPs were also examined. In this system, CPs and other professionals working for the municipality recorded all interactions with CWMPs, informal caretakers and other professionals, CWMPs' support plan and assessment of their self-reliance. Alongside data collected via interviews, data from the municipal record system helped to get an overview of the timing, frequency and nature of interactions among CWMPs, informal caretakers and other professionals. It also provided information on what professionals recorded after (re)assessing the CWMPs' situation with the CWMPs including (revised) plans and actions to deal with the CWMPs' situation during the care trajectory. This data was used to substantiate the data collected via interviews and (re)construct the coproduction process of integrated care during the care trajectory (including a timeline). The four sources of data (interviews with CWMPs, informal caretakers, formal caretakers and data from the municipal record system) collected over multiple time points allowed us to gain insight into the process of coproducing integrated care, including all participants' considerations, perceptions and evaluations during this process.

Interviews

Data were collected between September 2015 and March 2018 using a semistructured interview guide. The central themes in all interviews were the interviewee's understanding of the CWMPs' situation and problems, their perspective on the CWMP's support needs, their evaluation of the quality and adequacy of care provided, their (evaluation of their) role and those of others involved in the support trajectory, the level to which integrated care was provided and their perspective on CWMPs' future. Formal caretakers were also asked about the circumstances under which they can provide CWMPs the care needed, their interaction with other (in)formal caretakers and their knowledge about the care provided by other (in)formal caretakers. Each theme relates to aspects of integrated care and coproduction. Especially, the themes that focus on the interviewee's understanding of their role and those of others involved in the support trajectory, formal caretakers' interaction with other (in)formal caretakers and their knowledge about the care provided by other (in)formal caretakers used to delve deeper into the coproduction aspect of integrated care.

Interviews with CWMPs and informal caretakers lasted between 45 min and 2 h, and those with formal caretakers lasted between 30 min and 1 h. All interviews were audiotaped and transcribed verbatim.

Data analysis

Data were pooled and analysed by two authors (LR-dB and JvW) using Luborsky's[30] technique for thematic analyses. This process includes becoming acquainted with the data by reading the texts, the development of preliminary themes (open coding), axial coding and selective coding. At each step, the data and developed themes were discussed among the two authors, and an intercoder agreement was reached. The data analysis followed a deductive and inductive analysis process. Deductive in the sense that we, for example, analysed in each care trajectory how the CWMP's multidimensional needs were assessed, how care was designed and implemented according to these needs (aspects of integrated care) and how this process was the result of active involvement or engagement of CWMPs and their (in)formal caretakers (coproduction). Inductive in the sense that new themes and codes were created through the analytical process. Themes that were inductively developed related among others to ‘crisis, stress, complexity, trust, reflexivity and iterative’. Based on these themes two types of cases were identified in which professionals dealt differently with these issues and clients were involved differently. Data were analysed using Atlas.ti.

Ethics

The ethics review board confirmed that our study was outside the scope of the Netherlands' Medical Research

Involving Human Subjects Act and that the rights and privacy of study participants were sufficiently considered (MEC-2017–348). All participants signed a declaration of consent and could withdraw from the study at any moment for any reason. One CWMP withdrew from the study during an interview due to emotional instability; other reasons for withdrawing can be found in Appendix A.

RESULTS

To outline our findings, we follow the timeline of our cases. Our involvement starts when CWMPs reach out for help from the municipality (start care trajectory) and stops after 1–1.5 years. The start of the care trajectory is a relative concept in this context. Most CWMPs have been involved with many (public) services and care trajectories, often from early childhood, before we start to follow them. Therefore, some CWMPs reach out for help from the municipality while actively following another care trajectory, and not all care trajectories are completed when our involvement stops. Following the timelines of our cases, we first outline how the client's multidimensional needs are mapped out. We then outline two distinct types of cases in which various levels of integrated care are coproduced.

Assessing multidimensional needs

Our data indicate that CWMPs' multidimensional needs are rarely completely assessed at the start of care trajectories. We found several reasons for this.

The crisis first

Most CWMPs enter the support trajectory with massive problems, mostly acute needs, which require immediate action to avoid further escalation. For example, CWMPs are confronted with pending house evictions, have had their utilities turned off, have escalating debts, are homeless, have no income, have no health insurance, have no ID or are heavily addicted. CWMPs feel highly anxious and want their urgent problems to be solved and have their stress level reduced. Consequently, CWMPs' initial problem description focuses on their urgent problems in which they emphasize the need to have these issues resolved.

I had so many problems, so many problems, also debts. I had to write letters … couldn't do it myself. (…) I have a wife, a baby on the way, those financial problems made me crazy and had to be solved. (C36)

Additionally, many CPs (and other professionals) believe that the multidimensionality of CWMPs' situations can only be truly assessed when their urgent problems are addressed and their stress level has decreased.

My first focus was to calm things down. Her financial problems caused a lot of stress and increased her physical problems. (…). She [C23] had no insurance, and her utilities were going to be turned off. These are such basic needs. Those matters had priority. The other things would take more time [other underlying problems, such as her mental health]. It was not immediately made an important topic. (Community-based primary care team worker C23a)

Some formal caretakers also notice that CWMPs attract formal caretakers with a hands-on mentality who enjoy managing crises, causing them to overlook the multidimensionality of CWMPs' situations.

I think that we as caretakers overlook things [already involved caretakers or problems] because we dive into problems too quickly and get to work. We are often dealing with crises that cause us to BAM!, start acting. Then, halfway through, we find out all types of things [problems, involved people, interventions that do not work out]. That's a shame (…) We want to help. (…) I like crises. There must be pressure. (Community-based primary care team worker C23b)

Partnership is built on trust

Another complicating factor for assessing CWMPs' multidimensional needs is the lack of trust among CPs (and other professionals) and CWMPs at the start of the care trajectory. Almost all CWMPs in this study have a deep-rooted distrust of public service providers or ‘the government’, mostly due to negative experiences with the public service system in the past. Their distrust prevents them from sharing information beyond the (urgent) problems they want to be helped with.

In my situation, it's all caused by the municipality [of Rotterdam]. Because of the municipality, I ended up having rent arrears. Social services gave me too little money [income earned months before was deducted from his social benefits]; if I get too little money, I cannot pay my rent. It is called ‘social service’ and not ‘social misery services’. (C54)

Therefore, most CWMPs are reluctant to share information about, for example, things they are ashamed of, illegal activities they are involved with or more private matters. This withheld information can be potentially relevant information for assessing CWMPs' multidimensional needs.

C80 enters the support trajectory with massive debts. She says that after she ended her beauty salon, her accountant appeared to have never paid taxes. C80's community-based primary care team worker starts to help C80 with her debts. After a couple of months, C80's community-based primary care team worker finds out that C80's debts are not caused by her accountant but by C80's criminal activities and related conflicts.

Many formal caretakers are aware of the importance of a good relationship with CWMPs. At the start, for many of them safeguarding the relationship outweighs the importance of obtaining insight into CWMPs' multidimensional needs. When CWMPs are reluctant to share information, many professionals respect this.

A veil of complexity

The complexity of CWMPs' problems also hinders the understanding of CWMPs' multidimensional needs.

C23 has had problems in several areas of her life. She used to have a cocaine addiction, had bladder cancer, had several abusive relationships, went through several traumatic events, had Gilles de la Tourette, and had major financial problems (e.g., threats to shut off her utilities).

As in C23's case, CWMPs deal with problems in many areas of their lives. What makes it difficult to see through the (veil of) complexity of these problems is that they often have a great number of problems (e.g., it is difficult to map out all problems), CWMPs' problems are intertwined (e.g., making it challenging to unravel them) and it is difficult to understand how these problems affect daily life and current problems. Additionally, CWMPs' attitudes towards potential underlying problems vary. Many CWMPs do not want to explore the multidimensionality of their problems. For example, they ignore the layeredness of their problems, lack insight into their disease or are afraid of diving deeper into the origins of their problems (e.g., afraid of mental instability and traumas). Others are more open to exploring their underlying problems but, together with formal caretakers, struggle to see through this complex puzzle.

The crisis is not curbed quickly

Our data show that all care trajectories start with addressing the urgent problems first but also show that this ‘crisis phase’ is often of long duration (several months to a year). Solving urgent problems usually implies going through several interdependent (bureaucratic) procedures, such as the application for social benefits, a municipal postal address and debt rescheduling. These bureaucratic procedures use predefined steps with limited forgiveness for CWMPs' mistakes or deviant behaviour. CWMPs struggle to successfully complete these processes, and formal caretakers must invest a great deal of time to help CWMPs with this.

[C56] had no money at all, nothing. The woman would not accept our help if it cost her money [support would cost her health insurance excess]. We arranged funds to pay for this for her. We left her psychiatric situation for what it was, until the basics were rearranged [woman has schizophrenia] (…) We have arranged special administration, reconnected her utilities [utilities were turned off]. Her finances are now arranged. (…) Before you can write to all money claimants, special administration must be arranged, many steps must be taken. [We must] collect all necessary documents, bank account statements, make copies of these documents, etc. She also needed to be seen by an independent psychiatrist [for the application of special administration]. Then, it is up to the court, which takes a few weeks before the judge decides. (…) This is a process of months, not something done in a couple of weeks. (Psychiatric nurse C56)

In only two cases in this study were the most urgent problems of CWMPs relatively quickly solved, creating room to further analyse the multidimensionality of these CWMPs' situations.

In sum, our data indicate that CWMPs' multidimensional needs are rarely completely assessed at the start of CWMPs' care trajectories. Additionally, starting from the client's perspective does not automatically lead to an integrated approach.

The coproduction of integrated care

Nevertheless, our findings show that despite the absence of a full understanding of CWMPs' multidimensional needs

at the start and reluctant clients, integrated care can be achieved. We found two types of cases in which different levels of understanding of CWMPs' multidimensional needs and integrated care were finally established. Table 1 gives an overview of the key elements of the two types of cases.

Table 1 Overview of key elements case types 1 and 2

Case type 1	Case type 2
CWMPs' multidimensional needs are not completely assessed at the start of the care trajectory.	CWMPs' multidimensional needs are not completely assessed at the start of the care trajectory.
Both CWMPs' and formal caretakers' actions are aimed at addressing urgent problems first. CWMP's multidimensional needs are ignored until urgent problems are solved.	From the start, formal caretakers take initiative to not only address the CWMP's urgent problems, but also to explore the multidimensionality of CWMP's needs together with other formal caretakers.
Solving urgent problems takes more time than anticipated beforehand due to CWMP's underlying problems in combination with the complexity of bureaucratic procedures.	Experiences gained during the first period, in which both urgent problems are addressed, and the multidimensionality of CWMPs' needs is explored, are used to revise involved actors understanding of CWMPs' multidimensional needs and tailor interventions.
The care trajectory's progress and approach are reconsidered by both formal caretakers and CWMPs. At this moment in time, many CWMPs get disappointed, lose motivation and even leave the care trajectory. Formal caretakers take more initiative to redirect the course of the care trajectory. Collaboration with other formal caretakers is intensified and formal caretakers try to redirect the client to get the urgent problems solved. Focus remains on solving urgent problems first, and multidimensionality of CWMPs' needs are not explored (yet).	Urgent problems are often more quickly addressed than in type 1 cases.
Finally, formal caretakers and CWMPs manage to solve the urgent problems, yet this takes more time than anticipated. Underlying problems are usually not addressed, and CWMPs are still very vulnerable. This vulnerability makes them susceptible to new crises. Several relapse into similar problems within the 1–1.5 years we followed these CWMPs.	In successful type 2 cases, CWMPs seem to leave the care trajectory less vulnerable than in type 1 cases. CWMPs have more often gained (some) insight into the multidimensionality of their situation and have a more positive image about public services.

Abbreviation: CWMPs, clients with multiple problems.

Case type 1: Solutions to problems

C60 is addicted to heroin, has war traumas, is homeless, has no income, struggles with feelings of loss, and stays in a religious community. C60 wants a normal life. C60's community-based primary care team worker starts to help C60 regain his necessities. She concludes that he needs a postal address to be able to apply for social benefits and social housing. She also notes his war traumas and addiction.

Case type 1 cases represent most cases in our study (80% of the cases). In these cases, at the start, solving urgent problems is the sole focus of CWMPs and formal caretakers (‘solutions to problems focus’). In C60's case, this

implies getting him a postal address so he can apply for social benefits. In case type 1, the multidimensionality of CWMPs' situation is ignored until the urgent problems are solved. The care trajectory is approached as a linear process (urgent problems first, then diving deeper into the multidimensionality of CWMP's situation).
As multidimensionality is ignored, the help CWMPs receive and the interactions among CWMPs and formal caretakers have a practical focus, for example, how the CWMP can apply for social benefits, what documents need to be collected and how to best interact with formal bodies (e.g., creditors or social services). During interactions, formal caretakers and CWMPs mostly exchange practical information. The same applies to interactions among formal caretakers. Formal caretakers most often exchange information about what has been and still needs to be done to address urgent problems. It also stood out that in type 1 cases, formal caretakers more often tend to work solo.
All formal caretakers involved with C60 have contact with each other about practical matters (who does what, what has been done), except his addiction therapist and people from the religious community. His addiction therapist does not want to be involved (he thinks it is not necessary to do his work). People from the religious community are not considered relevant for the care trajectory by other formal caretakers.
However, this often changes when it becomes clear that urgent problems are more difficult to solve than expected. From the start, C60 does not keep appointments with any formal caretaker involved. He also struggles to collect the documents necessary to apply for social benefits. C60's behaviour delays the application for social benefits. C60's challenges with engaging in the care trajectory leads the involved formal caretakers to wonder why.
When progress is not being made, formal caretakers start to look beyond the most urgent problems. This triggers the need to align actions with other formal caretakers and go beyond practical matters. Contact among formal caretakers is intensified and starts to become more reflexive; what may be the underlying causes? Interactions between formal caretakers and CWMPs also start to change. However, CWMPs often become disappointed at this point and lose their motivation. Some CWMPs even decide to exit the care process. This attitude is reflected in the way they express themselves to formal caretakers. Formal caretakers start to initiate conversations with CWMPs about why progress is not being made and try to reflect on potential reasons, for example, they confront CWMPs with their (destructive) patterns and own responsibility and try to determine what is hindering CWMPs from moving forward. The initial linear process becomes more iterative and reflexive.
After 6 months, C60's social benefits are granted. His debt counsellor has been replaced. In hindsight, she believes C60 should have received more specialized support. C60's community-based primary care team worker is not sure what is truly going on with C60, possibly his heroin addiction or brain damage due to his addiction. She continues to encourage C60 to show up to appointments and collect his documents with little success.
During the summer holiday, fewer people are in the religious community, and C60 increases his drug use and lies in bed a lot. He misses more appointments, and involved formal caretakers struggle to contact him. C60's community-based primary care team worker and the debt counsellor arrange a meeting with C60 to reconfirm their agreements. C60 says it is chaotic in his head, and he feels overburdened.
However, this reflexivity continues to have a practical focus, namely, on what needs to be altered to solve the urgent problems (still a solution to problems focus). In C60's case, the focus on arranging his social benefits continues. C60 is encouraged to show up at meetings, answer his phone and put effort into collecting his documents. Formal caretakers and C60 do not reflect upon his increased drug use (this is even ignored). An in-depth or comprehensive understanding of the multidimensionality of the CWMP's situation is usually not gained.
In type 1 cases, formal caretakers and CWMPs manage to solve the crisis, yet this takes more time than anticipated beforehand. Underlying problems are usually not addressed, and CWMPs are still very vulnerable. This vulnerability makes them susceptible to new crises. Even during the time in which we followed CWMPs, we saw several of them relapse into similar problems, as occurred with C60. The crisis often leaves lasting marks: making CWMPs feel less competent to deal with challenges in life and less in control.
After 1 year and after a period of six months of having social benefits, C60's social benefits have ended. He did not comply with appointments made (he left the country and missed several appointments). In hindsight, C60's

community-based primary care team workers believe that he should have received more specialized care, and more attention should have been paid to underlying problems, such as C60's mental welfare. C60's community-based primary care team worker was not aware of C60's increased heroin use. After 14 months, C60 is referred to an organization specializing in people in recovery and ex-cons. C60 feels unfairly treated; he has no idea what was expected of him and seems unable to reflect on his own role.

Case type 2: An iterative process

C39 lives on the proceeds of a house he previously sold, is in arrears (eviction pending), has troubled relationships, and has severe health problems (e.g., has approximately 5% vision due to cataracts).

One day, C39's is evicted by the housing association. C39 is surprised. He did not know about the debts (never opened his letterbox). The eviction is averted when C39 accepts C39's community-based primary care team worker's help.

In type 2 cases (20% of the cases), CWMPs and formal caretakers also start with solving urgent problems.

C39's community-based primary care team worker starts to immediately deal with C39's urgent problems. She starts to organize his mail and debts, plans an appointment with a trustee, and reaches out to formal caretakers from the housing association. She also reaches out to people in C39's informal network (with C39's consent). Initially, C39 doubts whether this is necessary, but C39's community-based primary care team worker convinces him it is.

In this case, from the start and alongside interventions to address urgent problems, formal caretakers take the initiative to come to a shared insight into the multidimensionality of the CWMP's situation. Formal caretakers take the initiative to contact other involved formal caretakers and people from the CWMP's informal network. They have conversations about practical matters but also initiate discussions about potential underlying problems and the adequacy of interventions. For example, C39's community-based primary CT worker reaches out to C39's friends and children. She invites them to share their perspectives on C39's situations and vice versa.

C39's community-based primary care team worker makes an appointment with C39's GP for his eye problems and feelings of depression. C39's community-based primary care team worker goes with C39 to his GP and ophthalmologist. She picks him up in her car. C39 appreciates this a lot. When C39 is truly short of breath, C39's community-based primary care team worker brings him to the hospital and stays with him until the treatment is finished in the evening.

C39's community-based primary care team worker is compassionate but also direct and confrontational. For example, she confronts C39 with a potential unhealthy relationship with a woman and her belief that C39 dwells in feelings of grief. C39 appreciates his community-based primary care team worker's directness and thoroughness.

Formal caretakers also take initiative during interactions with CWMPs to come to a shared understanding of the multidimensionality of their situation. Our study shows that CWMPs mostly consider external reasons as causes for their problems. These formal caretakers also confront them by discussing the CWMPs' own involvement in their problems.

Several interventions are implemented, not all equally successful. For example, the trustee is formally assigned by the court. This is a massive relief for C39. He appreciates he no longer receives mail, and his finances are arranged. Domestic support is arranged to help C39 keep his house clean (C39 is not open to this).

In type 2 cases, solving urgent problems is not a linear process. Although many of these formal caretakers also believe the CWMP's multidimensional needs could only be truly assessed when their urgent problems are addressed, many view this period as helpful to gain more insight into the multidimensionality of CWMPs' situation. Experiences gained during this period are used to continuously revise involved actors' understanding of CWMP's multidimensional needs and tailor interventions (iterative process).

C39's ex-wife dies. He is shattered by the news. C39 gets into another conflict with his GP. His debts are solved, although with some hiccups. C39's eye problems are solved with surgery. C39's community-based primary care team worker ends her support. In hindsight, C39's community-based primary care team worker hoped to address more of C39's problems, but he was not open to this. For example, his inguinal hernia, his teeth, and potential mental problems which caused him to get in trouble. During the interviews, C39 shares that he knows he could have

more help, and C39's community-based primary care team worker thinks he should address more problems, but he does things at his own pace. When needed, he will reach out for help again.

In type 2 cases, multidimensional needs are often more completely assessed than in type 1 cases. However, formal caretakers can only encourage CWMPs to address their needs, and CWMPs ultimately decide on what needs are addressed. If CWMPs do not want to address certain needs, formal caretakers cannot force them to do so. However, in successful type 2 cases, CWMPs seem to leave the care trajectory less vulnerable than in type 1 cases. CWMPs have more often gained (some) insight into the multidimensionality of their situation and have a more positive image of public services.

DISCUSSION

In recent years, there is an increasing call in the literature on integrated care for stimulating coproduction. Coproduction in this literature is described as actively engaging clients, families and communities and is seen as a valuable route to harness their power, attune services to their needs and increase their ability to self-care (especially for unserved populations and marginalized groups).[12–14] It is also part of a fundamental paradigm shift in which people are put at the heart of services and paternalistic care is abandoned.[11,14] In this study, we show that there is always a level of coproduction required to establish integrated care, especially for CWMPs. Client involvement is indispensable to assess their complex needs, but also during service delivery. However, stimulating a more active role of CWPMs in coproduction does not seem to increase, but may even hinder the delivery of integrated care. Foremost, our study shows that in practice, the multidimensionality of CWMPs needs, which should function as the organizing principle of integrated care, are often not completely assessed at the start of CWMPs' care trajectories. Important reasons behind this are the urgency of the specific problems with which CWMPs enter the support trajectory, their lack of trust in government institutions and the complexity of their problems. Basically, CWMPs are at the start often unwilling and unable to look beyond their most urgent problem(s). We furthermore identified two types of cases. In both types, we see professionals trying to coproduce integrated care with clients. But only in one case type, do they seem to succeed. In case type 1, formal carers follow the wishes of the CWPM to only focus on the problems they consider urgent. At the start, CWPM and carers have more or less equal roles. However, when progress is not forthcoming, caregivers feel obliged to take the lead and also look at underlying problems (a more paternalistic approach). As the focus remains however on solving urgent problems, this does not result in integrated care. In case type 2, from the start, formal caretakers direct the care trajectory, and in a sense, take the lead. CWMPs' expressed needs (get urgent problems solved) are respected and actions are taken to get these solved. However, from the start, formal caretakers also direct and prepare the process to further analyse the multidimensionality of CWMPs' needs (although this is not what CWMPs ask for) together with other formal caretakers. Later, in the process, they also motivate CWPMs to work on other problems, thereby stimulating the delivery of integrated care and support. These observations raise questions about the extent to which paternalistic care is something to leave behind for this group of unserved and marginalized clients. It seems that to stimulate integrated care for these clients, formal caretakers must take the lead in exploring the multidimensionality of CWMPs' needs and in designing and implementing care according to these needs. Another important finding is that for this client group especially, the coproduction of integrated care cannot be approached as a simple linear process, which starts with a diagnosis (identifying multidimensional needs) and is then followed by the delivery of care and support. Our study indicates that the coproduction of integrated care should be viewed as an iterative process. It is something that needs to be worked towards via iterative steps in which the CWMP's multidimensional needs and interventions are continuously revised, deepened and sharpened.

These conclusions lead to several reflections on the literature on integrated care, the role of formal caretakers, current policies aimed at integrated care and bureaucratic processes.

One of the core principles in integrated care is that clients should be put at the centre and care should be organized in line with clients' multidimensional needs.[9,11,31,32] These principles are not disputed in this study. We see that when the multidimensionality of CWMPs' situation is not considered an urgent problem are approached in isolation, care trajectories often fail. Most studies on integrated care implicitly conceptualize clients as passive care recipients,

while we found that integrated care delivery is very much dependent on the willingness of clients to participate in its coproduction. At the same time, our study shows that involving clients and putting them in the centre does not automatically stimulate an integrated approach. As we have seen, CWMPs do not initiate (and may even hinder) a multidimensional assessment of their situation and are often not expecting (or even wanting) an integrated approach. Formal caretakers seem to have a key role in initiating integrated care for this client group. This approach requires formal caretakers who can build strong trust relationships with CWMPs, can organize shared reflexivity to unravel the complexity of CWMPs' situations, and can take on supportive, compassionate and confrontational roles (coaching). However, even then, there are no guarantees that this will result in integrated care delivery, as not all clients will be enticed to participate in coproducing integrated care.[17]

Furthermore, our study shows that for delivering integrated care, formal caretakers experience difficulties not only because of the fragmented delivery system, as is often discussed in the literature but also because bureaucratic procedures mostly follow a linear logic.[4,7,9,32–35] These procedures stipulate that in predefined steps, starting with a multidimensional diagnosis, CWMPs and formal caretakers (must) work towards an outcome (e.g., social benefits or debt restructuring). While these procedures safeguard equal treatment of equal cases, they do not facilitate or initiate iterative processes. Consequently, formal caretakers must invest a considerable amount of time, in bringing together the fickle processes of helping CWMPs go through these linear bureaucratic processes. The bureaucratic process also steers formal caretakers towards a linear instead of an iterative process. This could be an important insight for policymakers in the Netherlands and other European countries who implement policies aimed at integrated care.[36–38]

Limitations

In this study, we focused on a specific population, that is, CWMPs in Rotterdam, the Netherlands. Nevertheless, the specific policy context emphasizing integrated care and Rotterdam provided an interesting setting, as vast numbers of CWMPs can be found in this city, especially in the districts we focused on. We acknowledge that the specific population and setting could have affected our results. Therefore, studies on the coproduction of integrated care with other populations and in other settings could help to gain more insight into how integrated care is coproduced at a micro level. We must also acknowledge that the inclusion of people with multiple problems had its challenges. We have conducted our research in a scientifically sound manner, but we had to deal with obstacles in obtaining access to CWMPs and keeping them on board. Including clients via CTs could have created a selection bias. Knowing that CWMPs are difficult to include in research and that our study is one of a few longitudinal studies on CWMPs, we are confident that our study provides interesting insights and can stimulate more research into the care trajectories of these types of complex clients.[39,40] Another limitation of this study is that we struggled to include CWMPs' informal caretakers. Although we tried, we were only able to include a few informal caretakers. We therefore could not reflect on the role of informal caretakers in the coproduction of integrated care.

CONCLUSION

Our study shows that integrated care does not come naturally when CWMPs are put at the centre and that formal caretakers have a key role in initiating integrated care. The linearity of many bureaucratic processes does not enhance and even hinders the establishment of integrated care. Based on this study, we also conclude that clients should be considered active actors in every study on integrated care.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are not available due to privacy or ethical restrictions. The data are not available on request due to privacy and ethical restrictions.

AAPPENDIX

See Table A1.

Table A1 Participants' characteristics

	T0 all participants who signed declaration of consent	T0 participants who participated in first interview	T1	T2
Sex				
Male	44	32	23	22
Female	43	32	17	13
Total	87	64	40	35
	T0		T1	T2
Age (years)				
25–50	26		9	9
50-75	33		26	21
75–100	5 (oldest 86 years)		5	5
Total	64		40	35
Living circumstances				
Alone	28		24	21
Alone/without a partner or roommates and with children	9		2	2
With partner/roommates	6		4	4
With partner/roommates and child(ren)	13		8	7
Homeless	6		1	1
Homeless with child(ren)	2		1	0
Total	64		40	35
District				
Bloemhof	21		12	11
Hillesluis	7		3	3

Lage Land	8	7	5
Lombardijen	10	6	4
Ommoord	18	12	12
Total	64	40	35
Type of problemsa			
Finances (e.g., no income or debts)	59		
Daytime activities (e.g., no daytime activities)	30		
Housing (e.g., impending house eviction, homelessness, or contaminated house)	21		
Domestic relationships (e.g., domestic violence or parenting problems)	11		
Physical health	25		
Mental health (e.g., mental problems or mental illness)	36		
Addiction	10		
Activities of daily living	18		
Social network (e.g., absence of a social network or a destructive social network	26		
Participation in society (e.g., no job or no volunteer work)	29		
Encounters with law enforcement system (e.g., [pending] lawsuits for criminal activities)	5		
Nonparticipants: reasons for nonparticipation			
Reason for nonparticipation			

Unreachable	9	12	1
Change of mind, no longer willing to participate	6	9	2
No show	7	3	2

a

We used data gathered by the primary care teams complemented with the data from the interviews to provide an overview of the problems the participants in our study faced. We categorized the problems in line with the tool the primary care teams used to identify problems: the self-reliance matrix (in Dutch: de zelfredzaamheidsmatrix). This tool helps to identify problems in different life domains. All the participants had problems in different life domains.

BAPPENDIX

See Table B1.

Table B1 Formal and informal caretakers' characteristics

Type of professional	Organization	Number
Informal caretakers	N/A	6
Community-based primary care team professional	Municipality of Rotterdam	20
Social worker	Organization for addiction treatment Organization for people with acquired brain injury Religious social work organization Organization for sheltered living	7
Psychiatric nurse	Mental health organization Organization for addiction treatment	2
Psychiatrist	Mental health organization	1
Trustee	Trustee's office	6
Debt counselor	Organization for forensic and specialized care Voluntary organization for debt counselling Debt counselling organization	3
Spiritual caretaker	Organization for spiritual care	1

Pro bono legal counsellors	Municipality of Rotterdam	1
General Practitioner	General practice	2
General-practice-based nurse specialist specialized in mental health	General practice	1
Social support act professionals responsible for assigning care for which an indication from the municipality was necessary (in Dutch: Wmo-consulenten)	Municipality of Rotterdam	2

DETAILS

Subject:	Mental health services; Mental health; Collaboration; Needs; Patients; Health care; Welfare services; Quality of care; Clinical outcomes; Social care; Participation; Medical research; Caretakers; Addictions; Design; Integrated delivery systems; Social services; Data collection; Social issues; Professionals; Clients; Qualitative research; Integrated services; Mental health care
Business indexing term:	Subject: Professionals
Location:	Netherlands
Identifier / keyword:	care trajectory; clients with multiple problems (CWMPs); coproduction; integrated care; iterative processes
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	268-281
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety

ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-12-15
Milestone dates:	2022-10-07 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-03-31 (manuscriptReceived); 2022-12-15 (publishedOnlineEarlyUnpaginated); 2022-10-23 (manuscriptAccepted)
Publication history :	
First posting date:	15 Dec 2022
DOI:	https://doi.org/10.1111/hex.13653
ProQuest document ID:	2767101957
Document URL:	https://www.proquest.com/scholarly-journals/how-do-clients-with-multiple-problems-formal/docview/2767101957/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the “License”). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-05-10
Database:	Public Health Database,Publicly Available Content Database

Integrating Patient-reported Experience (PRE) in a multistage approach to study access to health services for women with chronic illness and migration experience

Abel, Thomas [1] ; Tadesse, Lidya [2] ; Frahsa, Annika [1] ; Sakarya, Sibel [3 1] Institute of Social and Preventive Medicine (ISPM), University of Bern, Bern, Switzerland [2] Institute of Social and Preventive Medicine (ISPM), University of Bern, Bern, Switzerland; Johns Hopkins Bloomberg School of Public Health, Baltimore, Maryland, USA [3] Department of Public Health, School of Medicine, Koç University,

Istanbul, Turkey

ABSTRACT (ENGLISH)

Background

Patient-reported Experience (PRE) is an emerging concept integrating patient perspectives and amplifying voices often marginalized in discussions surrounding health systems. However, it remains a challenge to use and integrate PREs when studying patient agency and access to quality services, particularly with data from multiple sources. In this article, using study materials from the Swiss MIWOCA project, we present and reflect upon a multistage PRE approach to study healthcare access.

Methods

The MIWOCA project, a study on healthcare access and quality among immigrant women with chronic illnesses living in Switzerland, provided data from multiple sources for the integration of PRE data. These sources included interviews with women (n=48), two focus group discussions with women (n=15), interviews with service providers (n=12) and observations from stakeholder dialogues (n=3). In addition, we utilized field notes, focus group illustration maps, patient vignettes and policy briefs to develop a multistage data linking model. PRE data served as starting themes and reference topics in each of the interlinked stages of knowledge production.

Results

Deploying PREs, we coherently linked the data from preceding stages and used them to inform subsequent stages. This, in turn, enabled us to identify, reflect and rectify factors limiting immigrant women's agency and access to quality services. Ultimately, the approach engaged patients as knowledge co-producers for system-level changes. This knowledge was transformed into a set of practice recommendations and a policy brief addressing ways to improve health systems to better serve immigrant women in Switzerland.

Conclusions

Building on PREs to systematically combine multiple data sources and engage patients continuously can improve our understanding of barriers in health systems. Beyond individual patient-doctor encounters, a multistage PRE approach can identify structural problems and provide clues for resolving them at the systems level. The PREs approach presented may serve as an example and encourage more public health experts to consider PREs in future research and practice.

Patient and Public Contribution

Women with chronic illness and immigration experience contributed to interview-guideline development, provided PREs in interviews, identified priority areas for health-service change and actively participated in the development of practice recommendations.

FULL TEXT

INTRODUCTION

Patient-reported Experiences (PREs) are an emerging concept that addresses how patients view and interpret their interactions with healthcare systems. PREs reflect how patients perceive their access to and quality of care.[1,2] PREs are particularly valuable when considering interactions of various dimensions of care that can include (but are not limited to) patient satisfaction, patient perception, patient preferences and patient engagement.[3] Although broad in their scope, most current PRE approaches focus on their potential for improving the individual healthcare experience in clinical settings, during or within a patient's encounters with services.[4,5]

PREs are still in their early stages of conceptual development and remain a topic of wide discourse amongst health researchers and professionals. Many issues are currently on the research agenda, including the appropriateness and effectiveness of current methods in creating and implementing patient-reported experience measures (PREMs).[6–8] Currently, most studies exploring PREs/PREMs use quantitative approaches. This is particularly true for leading

health systems that have implemented PREs/PREMs into service evaluations; primarily in the form of practice-specific questionnaires.[9,10] However, using questionnaires to capture PREs has its limitations, especially in that survey methods and data can be difficult to administer and interpret for healthcare staff, which acts as a significant barrier to their wider use.[11] Moreover, standardized quantitative methods are limited in addressing complex issues related to patients' utilization patterns. This includes questions on how system features affect patients' perception, knowledge and ultimately their behaviours.[12,13]

A particular challenge for PRE approaches is addressing social inequalities stemming from structural disadvantages. Many health system evaluations demonstrate that patients' access to high-quality healthcare services varies significantly by their social backgrounds (i.e., gender, race, migration status, socioeconomic status, etc.). A 2019 report on social inequalities in health systems found that across all OECD countries, Indigenous peoples and ethnic minorities were more likely to face socioeconomic disadvantages, language problems, cultural barriers and discrimination—all of which increase the likelihood of experiencing health disparities.[14] Qualitative PRE approaches may better capture these issues, particularly those of intersectionality, which is important to understand in patient groups experiencing multiple disadvantages.[15,16] Moreover, as these population groups also often face a higher burden of disease, their involvement in working towards improving services is even more crucial.[17]

Given this background, the present paper demonstrates how a PRE approach can be used to link various data sources addressing healthcare system shortcomings. In doing so, we draw on data and insights from an empirical research study (Migrant Women's Health Care Needs for Chronic Illness Services in Switzerland [MIWOCA]) on immigrant women's healthcare needs and access to chronic illness care.[18] In the current paper, we illustrate how a multistage qualitative methodology can integrate patients' experiences in consecutive steps of the knowledge-production process and engage patients as co-producers of knowledge for improving care services.

METHODS

This is a sub-study of MIWOCA, short for Migrant Women's Health Care Needs for Chronic Illness Services in Switzerland (SNF NRP74 2017-2020), a larger research project in which we researched access to and quality of healthcare service among women in Switzerland with chronic illness and migration experience.[18,19] MIWOCA included migrant and native-born women with chronic illnesses across diverse cultural and social backgrounds as well as care providers and other relevant stakeholders. Women participated in interviews and focus group discussions (FGDs). Subsequently, they were invited to participate in a series of stakeholder dialogues convened to develop policy recommendations. Detailed information about the study population, sampling strategies and data collection has been described previously.[18] In the current paper, we present a methodological approach to linking data sources based on the empirical fieldwork conducted during the MIWOCA project. We used data from multiple sources: observations and field notes from regular project meetings and three stakeholder dialogues, an analysis of project documents (such as minutes, reports, notes, patient vignettes, MIWOCA evidence and policy briefs), as well as a re-analysis of qualitative semi-structured interviews with women with chronic illnesses and health/social service providers (n = 48; n = 12) and two FGDs (n = 15). A survey among study participants provided auxiliary data on participants' perception and assessment of the multistage approach.

To develop our data linking model, we analysed field notes, documents and interview and focus group data, the latter ones had been analysed using the framework method,[20] supported by Atlas.ti software. We applied the following steps: transcription, familiarization with the data, coding and categorizing, identifying themes, developing a working analytical framework, applying an analytical framework, charting the data into the framework matrix and interpreting the data. We used both inductive and deductive approaches to help create an analytical framework. Throughout the data analysis process, we held regular peer debriefings among co-authors to discuss and refine findings and to interpret the meanings of data.

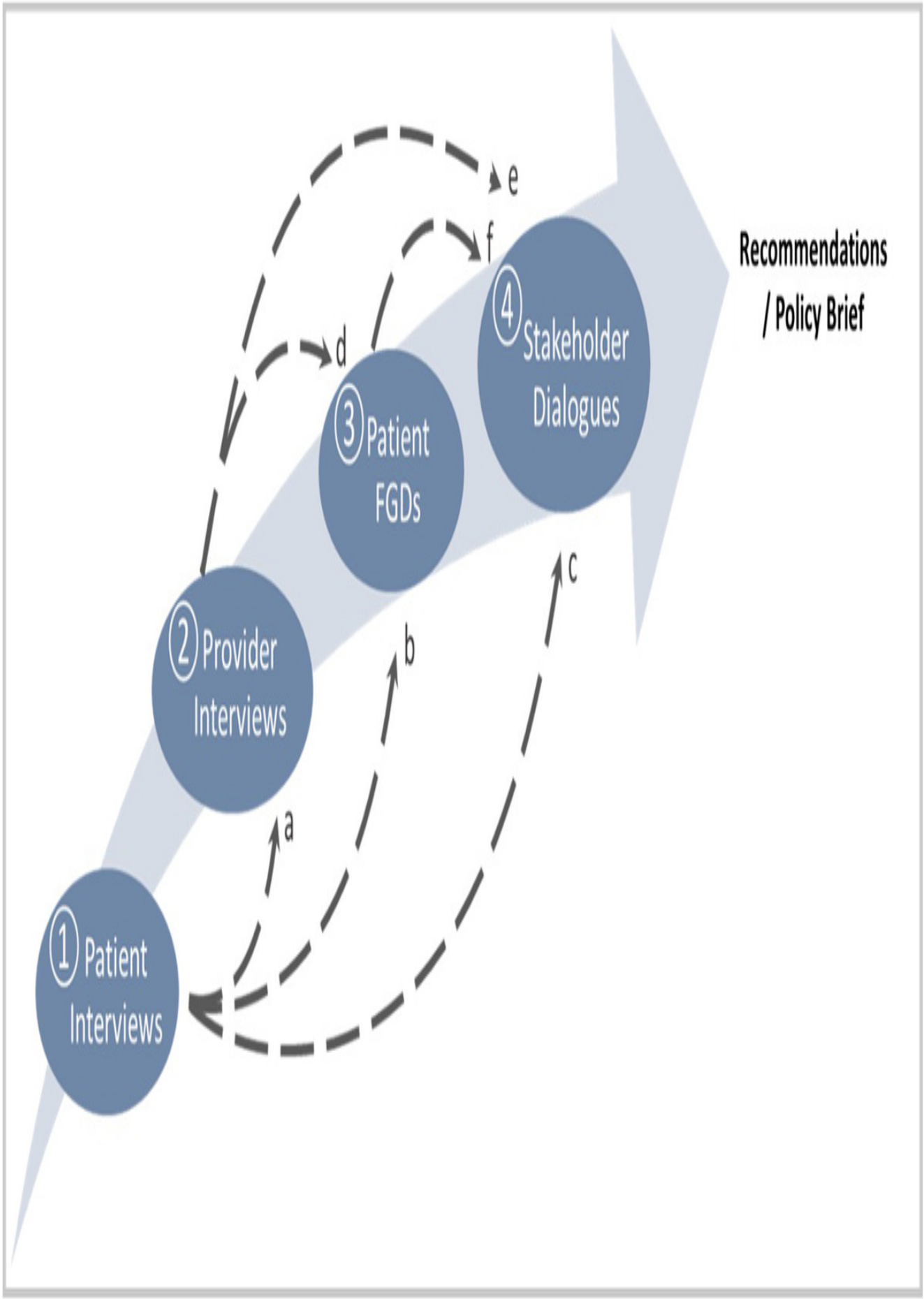
RESULTS

In the following, we present (1) the multistage approach from MIWOCA that integrates PRE data, and thus patient involvement, at each step. We also present (2) selected examples of specific PREs on access to and quality of healthcare services. We will use those concrete examples to demonstrate how PREs were continuously reflected

and considered throughout the multistage methodology to ultimately generate recommendations for improving access to and quality of healthcare services for this patient population. In addition to that, we present (3) participants' perceptions of the multistage PRE approach developed here.

The multistage approach to integrating PREs

Figure 1 depicts how PREs were synthesized, analysed and applied between these main stages: (1) interviews with patients, (2) interviews with providers, (3) FGDs with patients and (4) stakeholder dialogues. In each stage based on a different data source, researchers applied findings from previous stages to inform subsequent steps.



Enlarge this image.

In Stage 1, researchers conducted interviews with patients. Findings from these interviews informed the themes addressed in interviews with providers in Stage 2 (stream a). These findings were also used in later parts of the

study (streams b and c). Interviews in Stage 2 allowed researchers to contrast perspectives between patients and providers and identify structural factors that impact access to and continuum of care. Findings from interviews with patients and providers informed the themes addressed in FGDs with patients in Stage 3 (streams b and d). In FGDs, patients reflected upon these findings and presented their views on priority topics to be discussed in stakeholder dialogues (Stream f). In Stage 4, researchers conducted stakeholder dialogues that incorporated diverse perspectives from relevant parties, including those from previous steps (streams c, e and f). Ultimately, the key findings generated in this data flow were used to formulate and finalize a set of practice recommendations for health professionals and policymakers. The recommendations were later included in a policy brief and distributed among key actors in Swiss healthcare policy and organizations (see Supporting Information: Additional Files 1 and 2).

We used findings from data analysis in different formats: PREs from Stage 1 were used as substantive findings that were analysed and reported (e.g., identified barriers to accessing particular healthcare services [lack of system knowledge, stigma, etc.]). PREs from Stages 1 and 3 were also used to develop complementary formats, namely 'vignettes' and 'focus group illustration maps'.[21] These formats aimed to represent 'thick descriptions'[22] of patients' experiences and were presented and discussed as part of the stakeholder dialogues in Stage 4.

Each stage combined additional and different kinds of data. As a consequence, the increased richness and complexity of this data facilitated data triangulation and advanced analyses. Likewise, findings from each stage served to inform the content and focus of the next stage. For example, parts of the analysis of interviews with patients were used to help create the interview guide for interviews with providers. In principle, insights from PREs were carried forward to each mode of data production and analysis. In integrating data from all stages, we identified barriers and resources to accessing care (e.g., women's unfamiliarity with the complex healthcare system; informal social support networks). These were confirmed when merging the different sources, including other patients, care providers and third-party stakeholders (e.g., insurance experts). Through applying this model, we were able to gain insights on (1) why and how accessibility barriers are linked to specific healthcare utilization behaviours and limitations in the patient agency, (2) how such patterns are distributed among migrant women with chronic illnesses and (3) how these factors and processes can be improved.

Specific PREs and their integration throughout the multistage approachPREs identified from interviews with patients

PREs often addressed women's knowledge of the health system's available health services and how to access those services, health insurance options and issues of how to mobilize social support. Those issues were identified as key elements for a patient's ability to make choices and use health services effectively thus, facilitating their agency. Here, the patient agency is defined as the ability of patients to influence and contribute to the decision-making process behind their care.[23]

Through PREs, women interviewed provided rich context information yielding a better understanding of issues such as limited health knowledge among certain population subgroups or lack of financial resources needed to utilize the healthcare services. In regard to the former, women described how their perspectives relate to both sides of the provider–patient relationship, specifically in the context of receiving and understanding medical information. On the one hand, patients critically evaluate the quality of information providers offer. On the other hand, they also reflect on their positions as receivers of this information. Patients highlight issues such as the lack of information, lack of clarity of the information or lack of details. For example, one woman notes her disapproval:

It's the 8th day now, but how aware am I [about the medication]. How much information was given to me? They didn't provide information about anything; this is used for these diseases; this is used for these people …do you really want it 100% or not …they didn't say that.

Apart from direct interaction between medical service providers and patients and its impact on knowledge of medication, the women interviewed provided context on the reasons why they do not have complementary health insurance. One patient explains:

And it's not that I don't trust the public health care system that I want a complementary plan. For a long time, I didn't have any complementary insurance because I had trusted the hospital so much. I have always found the quality of care to be good.

Participants also expressed a need for better knowledge about navigating the health system and how this lack of knowledge ultimately limits their abilities to access health services and make informed decisions. For example, one participant noted:

There [should] be more openness. And that this support [should] be made available in several languages, not only German […] when I arrived in Switzerland, it was 'and now, what doctor am I going to?' The options are reduced. So sometimes I ended up picking a doctor just because they spoke English.

These examples highlight how PREs help to address 'how' and 'why' questions on potentially widely known aspects, such as alleged health illiteracy and lack of financial resources.

PREs applied in the providers' interviews

To facilitate the utilization of different data sources and further explore those issues around information deficits, language problems and access to care, researchers included the following questions in the interview guide for interviews with providers (*Stage 2*):

- (1)
  Do you think the current healthcare system works equally well for
- (i) women compared to men with chronic illnesses?
- (ii) For Swiss women compared to foreign-born women?
- (iii) For women with lower versus higher educational attainment?
- (2)
  Are there any consequences of those differences?

Providers echoed not only that immigrant women with chronic illnesses have less information on how the health system operates, but also that this negatively impacts the ways in which they access care. For example, one provider stated:

If you put literacy problems, language barrier issues, lack of knowledge of the health care system, fear of seeking care […] How many people are not seeking care! […] In relation to insurance too, it's sometimes complicated to understand […] It can be a little complicated sometimes […] Knowing what you're doing, what you need to do…

Likewise, another provider (based in the French-speaking part of Switzerland) noted:

Yes, I think [it's complicated]. Yes, especially for a non-French-speaking woman. It's complicated. For me, the ones I see, they were helped, e.g., by a friend who spoke French. But how else do they call, find a phone number, make an appointment, know where to go? It's complicated […]

PREs applied in the FGDs

Researchers synthesized and analysed initial findings from Stages 1 and 2 in a short report and presented this to immigrant women (streams b and d) participating in FGDs (*Stage 3*). During FGDs, these issues acted as three stimuli to the participants: (1) access to health care, (2) interactions with healthcare providers and (3) potential solutions to problems as proposed by interview participants. More specifically, researchers presented condensed findings from previous stages. We used a knowledge mapping approach[21] to visualize discussion results and to have participants prioritize issues by assigning points to them.

Participating women reported three key demands for change at the system level:

- (1)
  better access to transcultural communication with healthcare providers,

•(2)
more support and counselling on the day-to-day struggles of managing a chronic illness,

•(3)
and early access to user-friendly information about health insurance, particularly policies and contracts.

PREs applied in the stakeholder dialogues

Finally, stakeholder dialogues were conducted (*Stage 4*). Findings from the previous stages were synthesized into a MIWOCA brief and vignettes (cf. Supporting Information: Additional Files). Vignettes addressed (1) communicative competence in terms of language, culture and lifestyle, (2) being understood without prejudice, (3) transparent communication and adequate information about basic insurance policies, and (4) information about supplementary insurance policies, especially for those new to Switzerland. Vignettes would be read out aloud to make findings more amenable and palpable. They were orally presented during the stakeholder dialogues, as in the following:

She is also not satisfied with her current insurance, she complains. The tall woman, wrapped in countless layers of sweaters and cardigans, pulls out a considerable stack of documents. She immediately adds that she printed out all these files in the office ‘on a private budget, of course!’ They are documents from a wide variety of health insurance companies, all of them annotated. ‘How can anyone wade through this insurance jungle? Where is the best offer for me, both in terms of the costs to pay and the services I need with my illness?’ she laughs somewhat helplessly. (Excerpt from patient vignette, presented in the stakeholder dialogues, cf. Supporting Information: additional file 3)

Discussing the findings in the stakeholder dialogues, participants recognized that, while immigrant women had good self-perception and self-efficacy concerning individual health, practitioners and policymakers should prioritize improving immigrants' health system knowledge and the accessibility of health system information. Moreover, they acknowledged this would improve patients' overall agency.

Last, participants transformed the findings from the stakeholder dialogues into a list of final recommendations to be distributed to relevant parties. To give just one example: In addressing immigrant women's lack of access to relevant health system information specifically, these recommendations included the following:

Low-threshold information services should be promoted in communities and neighbourhoods. Communities should offer orientation aids for patients with chronic diseases at the neighbourhood level, especially for: the search of social services, health care services, and self-help groups; navigation in complex systems (e.g., adequate insurance models and services); questions concerning patient rights. (cf. more detail additional files)

The recommendations were thus developed including the initial PREs, advanced in various stages. They were afterwards transformed into a policy brief on the access to and quality of health care for immigrant women with chronic illnesses living in Switzerland (see Supporting Information: Additional File 2). This was systematically distributed via media relations of the University of Bern and the MIWOCA project webpage.

Study participants' perspectives on the multistage approach

Study participants varied in their perception of the multistage approach to integrating PREs. Participating women tended to express a great level of enthusiasm for engagement as well as gratitude ‘for being really heard’ as expressed by one woman. At the same time, some women discontinued participation or voiced disappointment about the scope of the project over time. They had hoped to receive individually tailored recommendations for their own health and context, concrete advice concerning their own situation rather than to contribute to a system-level discussion and recommendations.

Participating service providers stated that it is crucial but nevertheless not very common in their contexts to involve

people concerned from the beginning and in all phases of the research. However, some participants also expressed doubts about the usability and interpretation of PREs beyond an individual patient's experience in a broader context. I think it is important to focus on the patients' experiences for once. However, this approach leaves it somewhat open whether these are individual cases or whether these experiences can be generalized. Interpretation is also difficult because we only know the view of those affected. But basically, an exciting approach! (Response in the follow-up survey of the stakeholder dialogue)

DISCUSSION

This study presented an example of how PRE data can be linked and utilized in health services research. PREs from women with chronic illnesses formed the core of data collection, linking and analysis, and in the development of practice recommendations. PRE data from multiple sources were integrated into multiple stages to produce a more comprehensive understanding of the conditions, factors, and processes impacting patients' agency in access to care services. In a different function, PREs from interviews and FGDs served to focus and complement perspectives gathered through interviews with providers, jointly offering a more comprehensive view of systemic and institutional factors affecting the quality of services. Moreover, PREs functioned as trigger points and guided the agenda during stakeholder dialogues. In presenting PREs to health experts who participated in these dialogues, PREs finally facilitated meaningful discussions on how relevant parties can improve the health system to provide better care for immigrant women with chronic illnesses. While PREs varied in their function and formats over the course of the study, they remained the driving force behind this data flow and the knowledge base used to formulate final recommendations.

The multistage qualitative approach combining multiple data sources allowed to concretely involve patients in exploring existing problems and give them a voice in creating solutions. From a wider perspective, the use of the PREs methodology presented here aligns with the concept of co-production, which Greenhalgh et al.[24] define as, ‘the collaborative generation of knowledge by academics working alongside stakeholders from other sectors’. In the current approach, co-production of knowledge allowed researchers to identify systems-level issues while maintaining a focus on the actual lived experiences of patients.[24] The use of PREs exemplified how they can be applied to capture more of the complexity that emerges from the manifold social and economic challenges typically seen in immigrant populations, especially those facing chronic health problems. Within the context of immigrant health specifically, linking data from different sources while keeping a focus on PREs can help researchers better understand the challenges and barriers that are otherwise difficult to identify (e.g., language problems, lack of familiarity with health and social care services, effects of discrimination, etc.).

In the case of Switzerland, examples of these obstacles include challenging administrative conditions, complicated health and social insurance schemes and difficulties in communicating with care providers in the absence of translation services. It is likely that other countries face similar challenges, and that PRE research can help to describe and understand such structural conditions and how they interact.

The multistage approach of integrating PREs was developed with a focus on chronic illness care and included women living in Switzerland most of them with a migration background. This poses limitations for transferring the approach to other healthcare settings. However, the basic approach of using PRE in participatory research on structural conditions and patients' agency can still be feasible in different contexts-adjusted, though, to each country's unique conditions. As the current paper's primary focus was to introduce a new methodological approach that builds on the systematic linking of different data sources, it could not address the pending issue of how to best assess PREs in different contexts. The current use of PREs is often disease- or area-specific.[9,10] The findings presented here indicate that a more needs- and context-specific use of PREs may contribute to a more differentiated

understanding of the interplay of the structural conditions and patient agency—that is, PREs for specific populations or with specific needs in mind. Thus, the current methodological approach might aid researchers and practitioners in developing new measures, which are particularly useful for evaluating health services. Last, PRE approaches can and should consider other current developments in the field, namely those focusing on patients as social actors in and for their health. The approach presented here aligns with current research on people-centred services, which the WHO defines as care that emphasizes the '[…] perspectives of individuals, families and communities', and views people as '[…] participants as well as beneficiaries of trusted health systems'.[25] Moreover, the use of PREs in advancing people-centred care will allow institutions to identify problems with the delivery of care, implement new changes and interventions based on patient feedback and might promote the transparency and accountability of healthcare providers.[26]

As researchers in the current study integrated perspectives from various stakeholders, this did not come without limitations. Although patient engagement was present in every stage of the knowledge-building process, patients' personal participation declined while moving from identification of care deficits to analysing the data and drafting recommendations in stakeholder dialogues. In addition, women who were rather fluent in German tended to be overrepresented when it comes to continuous participation, despite translation services offered at meetings and dialogues. At the same time, the women who continuously participated in the MIWOCA study had been in close interaction with other patients through the group discussions and perceived themselves as representatives, particularly for the ones who might not have felt ready to speak up. Additionally, while stakeholder dialogues included representatives from a wide variety of sectors, future studies might consider including patients' families and caregivers as they can provide unique perspectives on relevant topics.

CONCLUSION

Integrating patients' experiences proved a useful approach in the current system-oriented research on access to health services. Applying a multiple-stages process that featured PRE data allowed us to successfully link different sources and formats of data. This integrative PRE approach nurtured a process of co-production allowing to engage chronic disease patients in each step of the research process, from the identification of problems to the development of recommendations to mitigate them. While the focus was on women with a migration background in Switzerland, the methodology presented may facilitate future PRE studies reaching beyond the themes and contexts addressed here. The method presented may thus serve as an example and encourage more public health experts to consider a PRE approach for patients' involvement in health systems research.

AUTHOR CONTRIBUTIONS

Thomas Abel defined the main research question, developed the structure of the paper, wrote major parts of the text and lead the author team. Lidya Tadesse conducted the literature search and its integration into the text, contributed to creating a focus for the argument and wrote major parts of the text. Annika Frahsa contributed to the development of the argument, analysed and interpreted data sources and wrote major parts of the methodology description. Sibel Sakarya helped in developing the focus and the structure of the paper. All authors contributed to the revision of the manuscript. All authors read and approved the final manuscript.

ACKNOWLEDGEMENTS

This paper is part of the MIWOCA study (www.miwoca.ch). We would like to thank all women who agreed to participate in the study, to be interviewed, to join group discussions and to bring in their expertise into the stakeholder dialogues. We would like to acknowledge the input of MIWOCA team members Luna De Araujo (University of Geneva), Tevfik Bayram (Maramara University, Istanbul), Romaine Farquet (University of Bern) and Sandro Cattacin (University of Geneva). We would like to thank Leonie Mugglin who participated in the

conduct and analysis of the women's interviews and also carried out the providers' interviews in Bern. Funding for this research was provided by the Swiss National Science Foundation (SNsF) as part of the National Research Program 74. Grant No. 407440_167428.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data sets generated and analysed during the study cannot be shared as participant confidentiality could be compromised if full interview transcripts were released.

ETHICS STATEMENT

We submitted this study to the Ethics Commission of the canton of Bern who determined no formal ethics approval was needed. All participants provided written informed consent, except for two women, who instead provided oral consent.

DETAILS

Subject:	Womens health; Socioeconomic factors; Public health; Health care policy; Immigration; Maps; Health care; Patients; Women; Chronic illnesses; Questionnaires; Health care access; Data analysis; Health services; Interviews; Patient satisfaction; Migration; Change agents; Focus groups; Illnesses; Health professional-Patient communication; Vignettes; Stakeholders
Business indexing term:	Subject: Stakeholders
Location:	Switzerland
Identifier / keyword:	agency; health services; immigrant women; inequity; methodology; patient experience
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	237-244
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford

Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-23
Milestone dates:	2022-09-14 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-03-21 (manuscriptReceived); 2022-11-23 (publishedOnlineEarlyUnpaginated); 2022-10-23 (manuscriptAccepted)
Publication history :	
First posting date:	23 Nov 2022
DOI:	htt ps://doi.org/10.1111/hex.13649
ProQuest document ID:	2767101956
Document URL:	https://www.proquest.com/scholarly-journals/integrating-patient-reported-experience-pre/docview/2767101956/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the “License”). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-05-25
Database:	Public Health Database,Publicly Available Content Database

A qualitative investigation of perceptions towards antibiotics by members of the public after choosing to pledge as an Antibiotic Guardian

Flintham, Lorna [1] ; Ashiru-Oredope, Diane [2] ; Charlesworth, Jordan [2] ; Harrison, Roger [1] ; Dalgarno, Elizabeth [1]

[1] Epidemiology and Public Health Group, School of Health Sciences, Faculty of Biology, Medicine and Health, The University of Manchester, Manchester, UK [2] HCAI, Fungal, AMR, AMU &Sepsis Division, UK

Health Security Agency, London, UK

ABSTRACT (ENGLISH)

Introduction

Antimicrobial resistance is one of the biggest threats facing global humanity. In 2014, Public Health England (now the UK Health Security Agency) launched the Antibiotic Guardian (AG) campaign as a national health promotion initiative to increase public and health professionals' commitment to reducing the threat of antibiotic resistance (ABR). The aim of this research study was to gain a snapshot of public AG attitudes towards antibiotic use, the AG campaign and illness postpledge.

Methodology

This research used an exploratory study design using thematic and framework analysis of semistructured, in-depth interviews. A purposive convenience sampling strategy was used to recruit 10 participants; adults in the general population who had registered with and chosen an AG pledge via the AG online platform during November 2020 were eligible for inclusion. Interviews were conducted via Zoom.

Results

Six main themes were identified: campaign awareness, motivators to pledge (uncertainty about the future of ABR, personal gratification, personal responsibility, moral obligation and COVID-19), perceptions of personal responsibility (and patient perspectives of moral obligation in clinicians), the impact of the campaign and campaign promotion. Pledging appeared to solidify existing perceptions AGs held. Behavioural motivations for responsible antibiotic behaviours stemmed from perceptions of personal responsibility, moral obligation and concerns about ABR. AGs attributed responsibility to variable patterns in overprescribing. Perceptions towards COVID-19, coinciding with the previously established study period, appeared mixed. AGs were keen to promote responsible perceptions in relation to antibiotics, resistance and the AG campaign. However, poor social acceptability of ABR concern was raised as a barrier to campaign promotion.

Discussion

The AGs' longstanding commitment to antimicrobial resistance demonstrates the importance of a pre-existing interest in the public's self-reported judicious behaviours and decision to pledge to an ABR-focused campaign. Presenting the local and global threat to human mortality and morbidity in a more relatable format in public messaging should be considered in future strategies promoting ABR awareness and shifts in public perceptions. More frequent messaging to existing AGs is further recommended to propagate positive behaviour change among a wider audience.

Patient or Public Contribution

This study was based on interviews with adult members of the public who had pledged to be AGs via the website www.AntibioticGuardian.com. Interviews were based on the public's perceptions of the AG campaign, antibiotic use and ABR.

FULL TEXT

INTRODUCTION

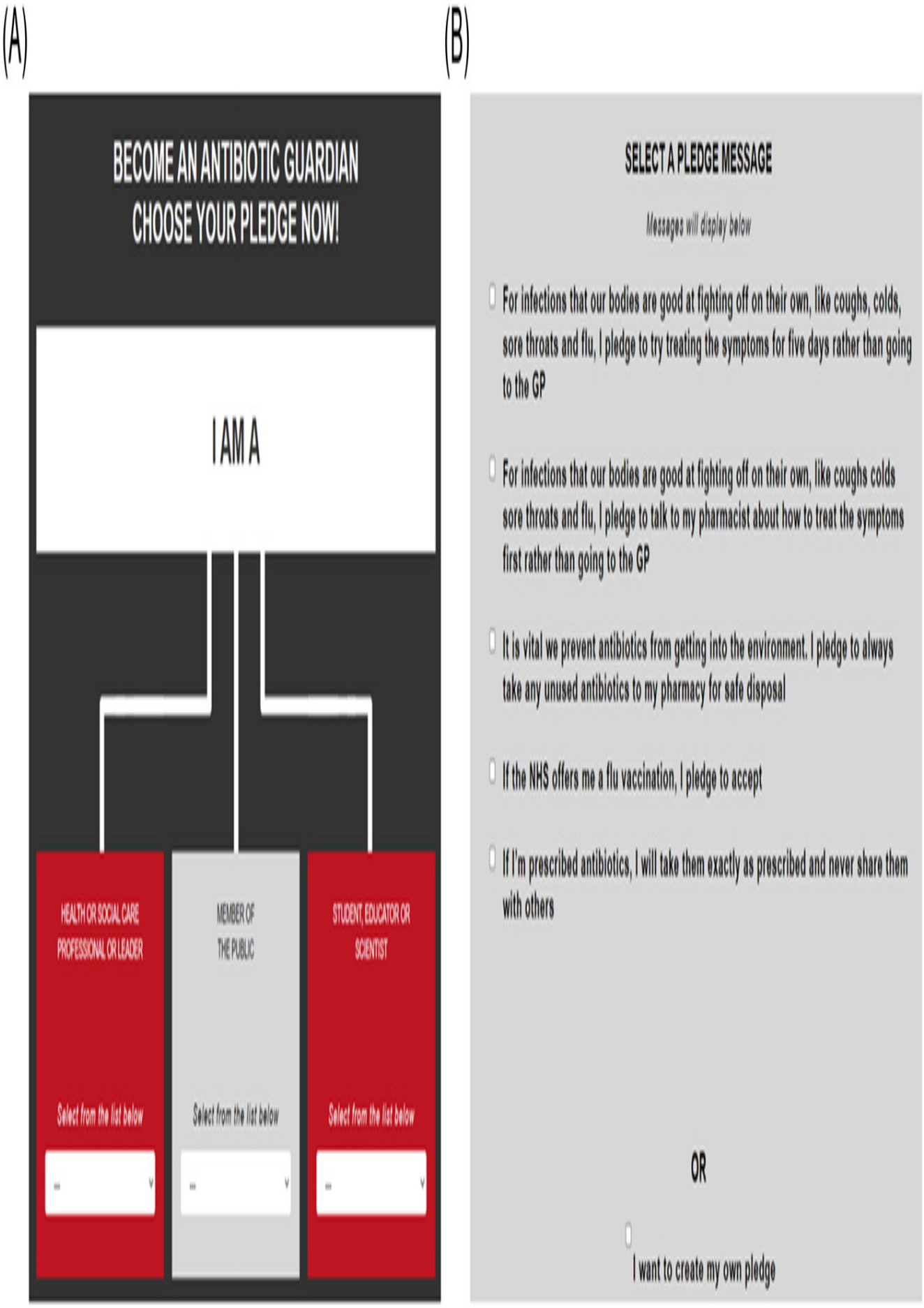
Since their wide-scale introduction in the 1940s, antimicrobials have remained the most effective and widely used drugs for the prevention and treatment of many bacterial, viral and fungal infections in people, animals and the environment. Yet, the growing presence of antimicrobial resistance, in which pathogens naturally develop resistance to the therapeutic effect of antimicrobials, is now the biggest risk to the survival of humans and nonhuman animals. The most recent, comprehensive study estimated that, in 2019, 4.95 million (3.62–6.57) deaths were *associated* with bacterial antimicrobial resistance, of which 1.27 million (95% UI 0.911–1.71) were *attributable* to bacterial antimicrobial resistance.[1] The research findings support previous estimates predicting 10 million annual deaths

worldwide by 2050.[2] The COVID-19 pandemic may have exacerbated this risk, with as many as 70% of COVID-19 patients initially receiving antibiotics, despite a lack of clinical indication.[3]

In 2021, the World Health Organization (WHO), the Food and Agriculture Organization of the United Nations (FAO), the World Organization for Animal Health (OIE) and the United Nations Environmental Programme (UNEP) established a united strategy, the Tripartite Plus, to reduce the magnitude of antimicrobial resistance in terms of associated and attributable deaths.[4] This follows Sustainable Development Goal objectives set in 2020 to reduce bloodstream infections associated with antimicrobial resistance and earlier initiatives established by the WHO Global Action Plan in 2015.[5,6] Despite such targets, around 30% of antibiotic prescriptions in outpatient settings have been deemed unnecessary on clinical basis.[7] In UK Primary care alone, approximately two million NHS antibiotic prescriptions are distributed a month,[8] of which 20% were thought inappropriate.[9] When also incorporating inappropriate antibiotic selection, duration and dosage, the total proportion of unnecessary antibiotic use has been estimated at a staggering 50%.[10]

Undoubtedly, COVID-19 has had a differential impact on the use of antibiotics around the world and placed selective pressures (or lack thereof) on antibiotic resistance (ABR). Reductions in bacterial infections were likely to follow through country-level ‘lockdown’ measures, major restrictions in travel and public-focused infection control measures.[11] Comparatively, misunderstanding of COVID-19 and poor antimicrobial stewardship resulted in considerable use of antibiotics, often without any clinical need.[12] A meta-analysis of data derived from Asia found that broad-spectrum empirical antimicrobial prescriptions were frequent and 72% of COVID-19 patients received antimicrobial therapy despite less than 10%, on average, having a fungal or bacterial coinfection.[13] To complement this known, quantitative evidence of selective pressure on ABR development, this study has qualitatively sourced public viewpoints to add to the field's limited exploration of public perceptions of antibiotic use in relation to COVID-19.

The initial WHO's Global Action Plan on Antimicrobial Resistance includes five objectives to reduce the risk from antimicrobial resistance, of which Object One made a clear mandate to ‘Improve awareness and understanding of antimicrobial resistance through effective communication, education and training’.[6] To support the UK's 5-year antimicrobial resistance strategy, Public Health England (PHE, now the UK Health Security Agency [UKHSA]) established the national AG campaign in 2014. With the ambition of reducing the unnecessary use and demand for antimicrobials, the campaign was developed, in part, to involve, educate and engage the public. Like many public health interventions, the involvement of the general public and patients in identifying, developing and implementing interventions is vital for subsequent effectiveness.[14] The campaign seeks to increase public and health professional commitment to reducing ABR and provide prospective patients with the motivation required for subsequent education.[15] This study sought to appraise public commitment by gaining a small cohort's individual perspectives. The campaign focuses on gaining commitment from those accessing it and reducing the risks of antimicrobial resistance by encouraging appropriate health behaviours, especially when seeking clinical input and subsequent use of antibiotics. Anyone with internet access can use this global campaign and pledge to become a ‘guardian’ of antibiotics. The whole campaign is underpinned by evidence of behavioural change and reflects positive approaches of similar strategies open to the public.[16,17] The online AG campaign collects basic information on the person's background (coded as a ‘member of the public’, ‘health or social care professional or leader’ or ‘student, educator or scientist’). The individual may select one of the available tailored pledges, as shown in Figure 1.



Enlarge this image.

Some similarities with the AG Campaign are shared with previous approaches to public engagement with regard to ABR in the United Kingdom. Notably, the national 'Andybiotic' and 'Keep Antibiotics Working'.[19,20] However, direct

engagement was missing and such approaches focused on more passive health education/awareness raising. More formal, systematic evidence on the value of public awareness interventions, such as 1 including 19 interventions, identified that campaigns trigger a significant effect on public knowledge and antimicrobial stewardship behaviours.[21] Only one previous study has qualitatively examined AG perceptions in campaign exposure, motivations to pledge and impact of the campaign.[15] Through semistructured interviews, the researchers found that public and health professional AGs were motivated to pledge due to personal or professional concerns for ABR, but most could not recall their specific pledge.[15] There remains a dearth of valuable evidence in terms of the distinction between public and health professional perspectives and the potential influence of COVID-19. This study intends to build on the work of Kesten et al.[15] by exploring public AG perceptions in isolation while appraising the potential influence of the COVID-19 pandemic.

Aims/objectives

The aim of this research study was to gain a snapshot of public AG attitudes towards antibiotic use following a pledge made during the month of World Antimicrobial Awareness Week (November) 2020 with specific objectives to explore (1) campaign and antimicrobial resistance awareness; (2) perceptions towards illness, antibiotic treatment and COVID-19.

MATERIALS AND METHODSEligibility criteria

Participants were eligible for inclusion if they were aged 18 years or over, consented to participate, selected 'Member of the public' at the point of pledge, had provided informed consent to be contacted by PHE in the future, were UK-based and were able to use Zoom. Only those who made their pledge in November 2020, the month including World Antimicrobial Awareness Week, were selected.[22]

Sampling strategy and data collection

A purposive convenience sampling strategy was used with a gender and age balance similar to the main AG cohort.[23] AG demographic information, such as age, gender and ethnicity, are not collected as part of the current campaign strategy; therefore, it was not possible to stratify the contacted AG sample.

Recruitment

A recruitment email was sent to all eligible AGs, as identified from the central database for the AG campaign, on the 15th June 2021 with a further follow-up 2 weeks later. The recruitment email presented the participant information sheet, a written consent form and a link to an online survey (Qualtrics.co.uk). The survey sought the participant's first name, gender, age band (18–20, 21–30, 31–40 etc.), ethnicity and email address for further contact. Interview slots were offered between 8 AM and 8 PM 7 days a week to avoid selection bias associated with only allowing individuals with certain working hours or shift patterns to participate.

Ethical considerations

A timetable of available interview times was provided; participants were asked to select slots that would be convenient for the Zoom interview. All participants were informed of how their data may be used before the interview started and all were reassured of their right to withdraw their data from the project at any point before a specified date (31 July 2021).

Participant interviews

All participant interviews took place across June and July 2021, 7–8 months after their pledge. Given the diverse geographic location of AGs and COVID-19 restrictions present at the time of data collection, interviews were conducted via the web-based Zoom platform. Participants chose whether their camera was on or off during the interview depending on their personal preference and device camera capabilities. This choice, alongside allowing participants to reside in a comfortable and familiar environment, may have led to more forthcoming responses.[24] A topic guide was developed after a review of the study published by Kesten et al.,[15] the only other published qualitative study examining the campaign, and questions were assessed by leaders of the campaign and qualitative research experts for content, purpose, design and relevance. The topic guide was used to direct the semistructured interviews towards relevant areas of questioning: pledging to the campaign, prescribing, antibiotic use and propagating campaign awareness.

Data analysis

Data surrounding AG pledges were analysed according to previously published methodology.[25] One researcher (L. F.) recorded and transcribed all interviews before analysis. The transcription function within the Zoom software was used to generate a transcript; this transcript was later checked for accuracy and recordings were deleted immediately after transcription to comply with research governance. Framework analysis was used to analyse the transcribed interview data. Framework analysis initially involved familiarization with the data, by re-examining the recorded interviews and rereading through the corrected transcripts until the researcher was familiar with them in their entirety.[26]

In the second step of framework analysis, thematic analysis was conducted by L. F. to develop a coding scheme. The thematic analysis involved analysing the data to identify common or recurrent themes in the participants' responses.[26] This was a comparative process by which different participants' responses were gathered and compared. A ‘scissors and paste’ approach was taken and coding schemes were developed to identify core ideas. Themes in the data were identified and these became labels for the codes. Next, as per framework analysis guidance,[26] codes were applied to the full data set in a process of indexing. In the fourth step, charting, the data was rearranged by thematic content. Charts were developed to contain summaries of the data, by theme, with the range of interview data systematically documented under each theme. This was useful for comparing data across codes and allowing the whole range of phenomena to be observed while examining relationships between codes.[26] While themes were discussed and refined with supervisor E. D., as this was conducted as a Master's dissertation, the coding, development of themes and analysis was conducted by L. F.

RESULTS

The number of public adults who pledged to be an AG in November 2020 and agreed to further contact was 135, of whom all were emailed the information about the study and participation. The pool consisted of 102 members of the public who pledged as an ‘Adult’, 25 ‘Family Members’, 7 ‘Pet/Horse Owners’ and 1 ‘Farmer’.

Of the 135 AGs who were contacted, 14 gave consent, of whom 2 did not respond to emails from L. F. and 2 respondents did not provide a correct email address. The remaining 10 participants were interviewed via Zoom. The sample size is representative of all public individuals who pledged during the most recent month of World Antibiotic Awareness Week (and during the height of the COVID-19 pandemic) who were willing and able to participate in the research. Likewise, smaller sample sizes promote a closer association between the researcher and respondents, increasing the validity of in-depth, fine-grained enquiry.[27] Further recruitment from pledges made outside of this time was not necessary for this qualitative, exploratory study as data saturation was reached and no new themes or data became apparent in the final interviews.[28] The mean interview duration was 48 min (with a range of 20–74 min). Table 1 provides information on the participants' characteristics, pledge and campaign exposure.

Table 1 Interview participant characteristics

	n
Members of the public	
Adults	10
Gender	
Male	6
Female	4

Ethnicity	
White	9
Black, African, Caribbean or Black British	1
Pledge recall	1
Selected pledge	
For infections that our bodies are good at fighting off on their own, like coughs, colds, sore throats and flu, I pledge to try treating the symptoms for 5 days rather than going to the GP	4
If I'm prescribed antibiotics, I will take them exactly as prescribed and never share them with others	3
If the NHS offers me a flu vaccination, I pledge to accept	1
It is vital we prevent antibiotics from getting into the environment. I pledge to always take any unused antibiotics to my pharmacy for safe disposal	1
[Created own pledge] To only seek to have antibiotics prescribed as a last resort, to reduce over use and the risk of increased antibiotic resistance.	1
Selected response to ‘How did you hear about us?’	
NHS	4
Community pharmacy	2
News Media	1
Colleague	1
Internet search	1
Family	1

The semistructured interviews were directed towards core topics of pledging to the campaign, prescribing, antibiotic use and propagating campaign awareness. This resulted in themes of campaign awareness, motivators to pledge (uncertainty about the future of ABR, personal gratification, personal responsibility, moral obligation and COVID-19), perceptions of personal responsibility and moral obligation in clinicians, the impact of the campaign and campaign promotion. For a full list of themes and corresponding subthemes, see Table 2. Participant perceptions could fall into more than one theme. For example, participants could be motivated to pledge due to both uncertainties surrounding the future of ABR and personal responsibility.

Table 2 Themes and subthemes identified through a thematic and framework analysis of the transcribed interview data

Theme	Subtheme	*n*
Campaign awareness	Incidental internet searching	3
	Pre-existing scientific interest and endeavours	3
	Professional networks	2
	Social media	2
	Cannot remember	2
Motivation to pledge	Uncertainty about the future of ABR	6
	Personal gratification	7
	Personal responsibility and moral obligation	4
Perceptions of prescribing	Widespread overprescribing	9
	Clinician-dependent prescribing values	5
	Age-related prescribing	6
Impact of campaign	Solidified existing beliefs	10
	Increased commitment to minimize ABR	6
	Increased antimicrobial resistance knowledge	3
Impact of COVID-19	COVID-19 irrelevant to pre-existing beliefs	5
	Appreciation for infection prevention	3
	ABR deprioritized	3
Campaign promotion	Promotion to friends and family	5
	Antimicrobial resistance Stigma	5
	Wider targeting	4
	Frequency of messaging	9

Note: '*n*' represents the participant frequency of theme occurrence.

Abbreviation: ABR, antibiotic resistance.

Campaign awareness

The majority of the group initially struggled to remember exactly where they became aware of the campaign but suggested a number of possible options, including through work, social media, friends and through scientific or academic groups they were already involved with. Routes of campaign exposure included World Antibiotic Awareness Week initiatives in the workplace that offered competition entry with AG sign-up, routine emails, COVID-19-related Google searching and encouragement from friends who were AGs themselves.

Uncertainty about the future of ABR as a motivator to pledge

Following initial campaign exposure, participants were probed for perceptions that triggered a subsequent pledge and commitment to ABR. Motivations that stemmed from a fear of the future consequences of ABR were common.

Participant 8

We still need that sort of comfort blanket to fall back on, to know that if we need the Doctors, then the antibiotics are going to work.

Participant 5

It could happen so quickly. You won't have a choice, you can't backpedal.

This reflected a possible relationship between pledging and fear centred on the uncertainty of a future without antibiotics and the inability to reverse this fate should it manifest. Additionally, participants would often translate their fear of the consequences of ABR to their own personal lives and the needs of their loved ones. When talking about a family member, Participant 4 noted:

Participant 4

She seems to get a lot of antibiotics for nothing and it really concerns me, and I wanted a bit more of an insight into antibiotics and what they do and if there would be an issue for her later on in life.

Participant 4 shared a fear that if antibiotics were required for that family member in the future, they would not be effective due to historic excess use. Beyond fear of ABR, one individual also noted a fear of the cost of care in ABR, on personal and national levels, as a motivating factor that drove a desire to become involved in PHE's efforts. This shows a concern that supersedes the consequences of ineffective treatment in bacterial infections and spans broader to the eventualities of needing other support from the NHS if it becomes stringently strained for resources. However, personal concern for ABR was not noted universally across the group and was not a predominant ‘general’ motivator to action. Additionally, there were indicators that the threat of ABR and statistics that may attempt to trigger an emotional response in fact deliver indifference. Participant 6 talked about hearing the figure that 10 million lives could be lost each year by 2050.

Participant 6

Is it actually too big, you know, to actually comprehend and how do you make that real? …We've become numb to the numbers …Sometimes people equate it [the death rate] to the equivalent of plane crashes and it's like ‘crikey, we wouldn't stand for that’. Yet, we accept this.

This suggests a potential disassociation from the data in the public realm and a numbness to the numbers when they are not tangible or perceived on a personal level.

Personal gratification as a motivator to pledge

A perception of personal gratification derived from a desire to champion positive behaviours was a second key motivator that drove the participants' antibiotic-related behaviours and choose to involve themselves in the AG campaign. Many of the participants reported a personal gratification associated with being part of a larger collective action that transcended their own day-to-day antibiotic-related behaviours.

Participant 1

It gave me that feeling that, you know, I was doing something good and being part of something that was big.

This demonstrates the influence of perceived gratification on perceptions. Altruistic intentions acted as an implied reason for engaging in judicious behaviour among AGs with a perceived fulfilment of these intentions achieved by pledging. By extension, this suggests that AGs' altruistic antibiotic-related behaviours and the choice to pledge go together with solidarity and the concept that people depend on one another. Personal gratification also arose from

personal interest.

Participant 3

I've been following issues on antimicrobial resistance …So, getting interested and getting to know more about how I can provide my help to prevent antimicrobial resistance.

This suggests that personal gratification may be derived from personal interest and the perception they were helping others. AGs' scientific interest in an area of public concern and a desire for driving action seemed to underpin campaign-related perceptions in the cohort.

Participant 6 suggested that observed public ignorance stems from publications with mixed messaging and frequently changing perceptions towards health threats in the media.

Participant 6

This week it's that, now next week they'll have changed their minds and it'll be something else. Last week it was bad and this week it's alright.

Thus, a lack of commitment may stem from mixed messaging.

Personal responsibility and moral obligation as a motivator to pledge

Personal responsibility and moral obligation along with a perception of frustration towards others who did not share this sense of responsibility were also common.

A moral obligation in the context of personal responsibility to others/society was raised as a key motivator when pledging and, more generally, when following responsible antibiotic-related beliefs. As a comparison to ABR responsibility, Participant 10 discussed a moral obligation to get the flu jab to avoid contracting or spreading the virus. This implies a perceived duty to prevent others from becoming ill and requiring health service support.

Participant 2

As someone who is advocating an end to antimicrobial resistance, therefore, it is upon me to act as an example.

A further participant in the 71–80 age group (Participant 7) expanded on this and suggested that their disinclination to visit the doctor unnecessarily arose from their childhood, implying that those who overuse doctor services and antibiotics are a less responsible sort of person:

Participant 7

I've always been disinclined to go to the doctors unless I really needed to. I think that stems from as a child, we had private doctors so we had to pay for them. And you didn't go for just trivial, silly things and I'm always irritated by people who do.

Others theorized that the perceived lack of knowledge and appreciation for antibiotic use among other members of the public is because it is assumed that some other party would take responsibility.

Participant 6

Somebody else can deal with that one … they'll do it for us.

Such views were observed with verbalized frustration and anger. Overall, this indicated a moral principle that bound the participant's actions to their perceptions of antibiotic use.

Perceptions of personal responsibility and moral obligation in clinicians

The perceived overprescribing of antibiotics by prescribers as a driver of ABR was an opinion reported by almost all participants. This too was linked to personal responsibility and moral obligation, but an obligation relative to the clinicians providing antibiotics. However, within this concept, viewpoints varied considerably, from perceptions that irresponsible overprescribing was common to some who perceived that overprescribing occurred, but it depended largely on the clinician, to others who felt overprescribing was rare and only occurred in cases of unavoidable patient pressures. The concept of pervasive antibiotic distribution was described as follows by Participant 1:

Participant 1

I think they're giving them out just sort of willy-nilly …I wish they'd go and take the pledge as well and just say that they're not gonna give them out, sort of, you know, like Smarties. Because a lot of them do.

The time constraints of the consultation along with the influence of the patient on the GP were also acknowledged as influential factors, with Participant 9 noting:

Participant 9

Even if the GP is reluctant to prescribe, you know, they can be put under lots of pressure by patients …GPs, you know, they have seven minutes per patient.

The more general perception that the health service comprises both responsible and irresponsible prescribers was also common, and participants reported that the likelihood of antibiotic distribution depended on the individual clinician.

Participant 9

I think that a hundred percent it boils down to whom it is in question.

Participant 10

I was prescribed amoxicillin like smarties when I had recurring tonsillitis as a teenager, to the point where amoxicillin became totally useless on me.

Participant 10 went on to describe how, following recurrent tonsillitis, they had their tonsils removed through a private health service, a surgery not permitted through NHS services. Consequently, the procedure terminated any further need for tonsillitis-related amoxicillin and emphasizes the role of other avenues of preventative medicine.

Impact of the campaign

Although there were subtle changes to the strength of AGs' motivations postpledge, in all cases, the participants reported pre-existing perceptions and suggested that involvement in the campaign reinforced these perceptions:

Participant 1

I've always been one for not having antibiotics if I can help it.

Participant 6

I'd already been convinced to know what antibiotics are for.

Many of the participants had completed previous publicly accessible online courses on antimicrobial resistance, watched online presentations and been part of patient feedback groups. These actions are potentially the root of Participant 2's and Participant 5's comments:

Participant 2

It's a subject I related to, I understood the pros and cons.

Participant 5

It [pledging] was a bit easy for me to do actually.

When questioned about their perceived level of antimicrobial resistance knowledge, responses varied, with seven participants reporting that their knowledge had remained the same following a pledge. This was largely attributed to pre-existing knowledge of antimicrobial resistance received through professional friends, extracurricular organization involvement or personal research related to their interest in the issue.

However, three of the participants reported increasing their knowledge and moral obligation to commit to tackling antimicrobial resistance following the pledge, reinforcing the idea that pledging is linked to personal gratification, responsibility and moral obligation:

Participant 1

Having actually signed the pledge and knowing that there is an organisation that's involved with that side of things, its made me want to, sort of, want to look into it more and get involved more.

Participant 9

I definitely did [increase my knowledge] at the time, when it was first mentioned, I definitely went and had a little Google.

Impact of COVID-19 on motivation to pledge

Half of the cohort reported that the pandemic had not influenced their perceptions of ABR, the AG campaign or antibiotic use. Comparatively, in three cases, COVID-19, including the associated restrictions such as lockdown, appeared to increase participants' appreciation of the ABR issue and AG campaign, as shown in the two examples below:

Participant 1

I was put on furlough when it first started, I'd got a lot more time on my hands anyway. So, I used that time to sort of go on the internet a lot more …Before I went on the internet, you know, I never knew about it at all. So, perhaps if COVID hadn't happened, perhaps I still wouldn't.

Participant 5

One of the upsides of the pandemic is that people are more focused on infection prevention.

Participant 1's comments highlight how the pandemic beneficially allowed them to actively come into contact with the AG campaign.

In direct contrast to an increased appreciation for infection prevention and ABR were opinions that ABR is a quieter message than it was before COVID-19 and less important as a result, as noted by three AGs. One participant suggested that COVID-19 had deprioritized the issue in the public's eye:

Participant 2

[ABR] is a smaller argument now, so, you know, it's like it's been replaced with ‘you need the vaccination’.

Likewise, it was perceived that COVID-19 may have exacerbated the overprescribing of antibiotics that some participants attributed to patient pressures:

Participant 2

More antibiotics being used because people [prescribers] are panicking, ‘gosh I don't know what it is but I'll give them antibiotics’. So, sort of making the resistance situation worse.

This indicates a view that prescribers' commitments to weighing risk with nonmaleficence have become sensitized to the added risk of COVID-19.

Campaign promotion

All of the cohorts reported discussing antibiotic-related behaviours and perceptions with friends, family or co-workers and five AGs had recommended the campaign to others. Participant 1 disclosed discussing the campaign with their spouse, while Participant 9 discussed guiding others along a thought process if it was raised. However, Participant 2 felt that this was equally true of misinformation:

Participant 2

People say ‘my Mums always sworn by antibiotics for everything’ and you're like, ‘hmm, ok’.

This suggests the contrasting challenge of social networks in ABR-related perceptions. Other AGs, when asked if they promoted the campaign to others, replied that they had not due to not remembering their pledge to the campaign:

Participant 7

Until I got your email, I had totally forgot about it [the campaign].

Thus, this potentially indicates the need for more regular messaging in campaign promotion.

When touching on campaign promotion, the concept of ABR pledge stigma and disapproval of those who discussed ABR became apparent. This was evidenced by the comments below as AGs deliberated the prospect of discussing ABR and the AG campaign:

Participant 1

I sort of drop it into the conversations that I'm having with people and do it subtly so I don't sort of, people don't think ‘Oh my God, here he comes again, it's the antibiotic man’. I don't want that impression of me!

Participant 5

I would, yeah, risk being unpopular

Participant 7

It depends on the situation obviously, you don't want to lose too many friends over these things.

Thus, three AGs considered their views and the notion of discussing the topic as controversial. This indicates an experienced or anticipated rebuttal of opinions if they were to discuss antibiotic-related consumption and disapproval for doing so.

When discussing the promotion of the campaign, the majority of participants communicated a desire for the AG campaign and other similar efforts to be communicated more widely and for contact to be more frequent:

Participant 4

I don't see much about it to be honest …People aren't coming to me to warn me about it.

This suggests a need and demand for broader targeting strategies. Contrastingly, Participant 7 reported that there is possibly too much messaging in the public realm:

Participant 7

You become saturated or people go ‘this is too much’ then shut off.

This raises the potentially challenging notion that there is already too much ‘noise’ in health messaging for campaigns to be heard and that the bombardment of the sheer volume of messaging in health can generate disengagement.

DISCUSSIONCampaign awareness and promotion

The public became aware of the AG campaign through various routes, emphasizing the broad, cross-community campaign exposure that has been reported in earlier research.[15] Most participants struggled to remember exactly where or how they encountered the campaign but provided various avenues of potential exposure, which reflected the actual varied routes of the exposure recorded in the participants' pledge data. Social media is undoubtedly a key part of this, with around one-third of initial contacts made this way.[23] Consequently, in the wider field of public health, this evidence highlights the importance of widespread marketing across communities to allow campaign dissemination to feed public commitment to ABR. The current study also supports the presence of self-motivated individuals joining the campaign and making varied pledges, as opposed to top-down interventions.

Likewise, most AGs could not recall their pledge, a finding similarly found in previous interview data,[15] but in contrast to existing quantitative research.[29] Kesten et al.[15] attributed the difference in findings to the discrepancies in time between the individual's pledge and the time of research, with the researchers examining AGs 10–16 months postpledge compared to 5 months in the quantitative evaluation.[15,29] However, in this research, AGs were interviewed 7–8 months postpledge and the majority could still not recall their pledge. Being unable to remember one's pledge may potentially be because becoming an AG was not usually a commitment to new responsible behaviours but a solidifying of existing approaches, thus reducing the memorability of a specific pledged commitment. The significance of this finding to the current body of literature indicates that the act of pledging itself appeared to solidify beliefs in ABR, rather than the details surrounding the pledge. This is an important factor to note in future campaign development and further research could investigate whether this phenomenon is dependent on pre-existing beliefs or reproducible in non-AG cohorts.

Pledge motivators

Pledging in the AG campaign appeared to foster the solidification of responsible perceptions that predated the individual's involvement in the campaign. However, there were also indicators that pledging acted as a trigger that encouraged participants to increase their knowledge of ABR. This reflects the results of previous AG knowledge research, which showed that ABR knowledge is greater in public AGs than in the EU Eurobarometer survey.[30] This suggests that AGs hold a place as a valuable source of knowledge within the public, which has so far been untapped as part of the current campaign.

Furthermore, fears regarding their own health, and that of others, acted as a motivating catalyst for pledging to the AG campaign. It is perhaps unsurprising that some people taking time to visit the campaign website or pledge support, had some notion of the consequences of a world with increasingly ineffective antibiotics. Our findings suggest that individual members of the public are sufficiently concerned about ABR and this is an important feature to incorporate in ongoing and future health promotion interventions. Yet, using fear to promote healthy behaviours is historically controversial and has not been associated with desired outcomes in ABR.[31] Thus, national and international campaigns could utilize this study's evidence for personal gratification as a motivator in ABR commitment and prudently utilize the Wellcome Trust's endorsement of a more open, upfront account of ABR, which makes it clear that individuals can take ‘immediate action’ to a ‘solvable’ problem.[32]

Similar to previous qualitative work, this study reinforces the significant role that perception of personal responsibility plays in motivating positive direction.[15] The notion of altruistic intentions in personal responsibility was raised and is

distinguished from egoistic intentions to use antibiotics judiciously. However, having interviewed just the public in this study, it is worth noting that a personal responsibility to address the ABR issue is not isolated to either healthcare professionals or the public but is instead a universal motivator in this pledge-based campaign. Furthermore, although the AG campaign pledge system is positive for triggering a commitment and interest in ABR, drip-feed messaging in the current AG and future campaigns are recommended to maintain engagement among the public, who, unlike health professionals, may not be exposed to the issue regularly.

Perceptions of prescribing

Some AG perceptions indicated that their clinicians' reasoning for prescribing antibiotics was not always congruent with their own, whether their perceptions were true or not. Previous research on the AG Campaign found the public was more likely to exhibit behaviours that reflected their pledge than healthcare professionals.[29] From the perspective of members of the public, these amalgamated viewpoints stress that an AG status is one immersed in frustration towards the actions of others and unshared principles. This emphasizes the need for joint decision-making and clear dialogue between prescribers and patients. However, perceptions of prescribing were highly varied; it was common for participants themselves to report variance in prescribing practices.

Impact of COVID-19

Participants verbalized two somewhat contradictory perceptions towards COVID-19, the first being a heightened appreciation for infection prevention and the second a deprioritizing of ABR in the face of a larger public health threat. The risk of COVID-19 in the United Kingdom, as measured by death and infection rates, was significantly higher at the participants' point of pledge (November 2020) compared to the time of interview (June–July 2021).[33,34] Legal limits on social interactions also differed between the national lockdown through much of November 2020 and the staged relaxation of limits through the summer of 2021. Thus, it is theorized that views may have been influenced by public freedoms as well as the perceived threat of the virus as a by-product of the case and death rate at the time, epidemiological spread and media-conveyed data.[35]

Strengths and limitations

It is thought that the integrity and interparticipant dependability of this research was encouraged through a consistent approach to semistructured questioning and written communications.[36] This study allowed the researcher to compare the perceptions and experiences of AGs who are nonhealthcare professionals, allowing for a comprehensive analysis of perceptions towards antibiotic use, the AG campaign and illness.[37] Additionally, this study has provided insight into the real-life experiences of AGs as members of the public and patients, an outlook that has not been formally captured in isolation.

Comparatively, the discrepancy between participants who activated their camera (seven participants) versus those who did not (three participants) may have led to inhomogeneity in the participant pool, given that the face-to-face element of interviews comes with the advantage of social cues that can guide the discussion.[38] A relatively small sample size was used, a reflection of the eligibility criteria employed, to gain a snapshot of perspectives and data saturation was reached. The research may have inevitably been affected by recall bias and social desirability bias; participants may have been more likely to remember instances of positive antibiotic-related perceptions and behaviours given the nature of the research.[39] Similarly, participants were recruited on a volunteer basis and therefore this research may have been affected by volunteer bias, one form of selection bias.[40] This is particularly so as AGs who had a previous interest in the subject may have been more likely to pledge as an AG and volunteer for the study.

CONCLUSION

For the first time, this research has provided direct insight into the reasons why members of the public pledge to support a continuing AG campaign. People pledging were clearly motivated to continue optimizing antibiotic use as a way of containing resistance, at times driven by their own fears of the consequences if they and others failed to take action. In many cases, pledging solidified existing values and behaviours, while in other cases, this study found personal gratification to be important to inspiring action. Resultantly, improved collaboration and more frequent messaging between leading campaign makers and the public will be vital to acknowledge the relationship between

ABR development, social barriers, antimicrobial prescribing and human health.

AUTHOR CONTRIBUTIONS

Lorna Flintham carried out the study, developed the main conceptual ideas and led the data analysis. Lorna Flintham, Diane Ashiru-Oredope and Elizabeth Dalgarno conceived and designed the study. Elizabeth Dalgarno developed the theory, supported the analysis and interpretation of results, provided guidance in all study aspects and supervised Lorna Flintham throughout the project. Diane Ashiru-Oredope and Jordan Charlesworth contributed to the methodology and Jordan Charlesworth implemented the participant eligibility criteria, and population targeting and led initial participant communications. R.H. and Diane Ashiru-Oredope devised the project and facilitated interorganization collaboration and communications. Roger Harrison had considerable input into the design and content of the paper, working with all other authors as part of an academic team. This included approving the results and their wider implications. Lorna Flintham, Elizabeth Dalgarno, Diane Ashiru-Oredope, Jordan Charlesworth and Roger Harrison contributed to the interpretation of the results. Lorna Flintham wrote the manuscript with input from all authors. All authors provided critical feedback and helped shape the research, analysis and manuscript.

ACKNOWLEDGEMENTS

We would like to acknowledge the contribution of all the Antibiotic Guardians who were involved as participants in this study, and the research opportunity as part of a Masters in Research (Public Health and Primary Care), at The University of Manchester. All views and opinions stated in this work are those of the authors and not necessarily those of Public Health England/UK Health Security Agency or The University of Manchester.

CONFLICTS OF INTEREST

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

The anonymized data sets, free from personal identifiers, generated during and/or analysed during the current study are available from the corresponding author on reasonable request.

ETHICS STATEMENT

Ethical permission for the research was gained from The University of Manchester's University Research Ethics Committee (UREC), which granted proportionate UREC approval (reference number 2021-11584-19056). Written consent was gained from all participants before the interview.

DETAILS

Subject:	Health promotion; Drug resistance; Public health; Threats; Professional attitudes; Campaigns; Gender; Websites; Gratification; Antimicrobial resistance; Antibiotics; COVID-19; Motivation; Medical research; Coronaviruses; Security; Infections; Antibiotic resistance; Morbidity; Moral responsibility; Ethnicity; Interviews; Uncertainty; Medical personnel; Pandemics; Bacterial infections; Perceptions; Antimicrobial agents; Obligations; Data collection; Overprescribing; Qualitative research; Public opinion; Behavior change
Location:	United Kingdom--UK
Company / organization:	Name: United Nations--UN; NAICS: 928120
Identifier / keyword:	Antibiotic Guardian; antibiotic resistance; behavioural change; education; engagement
Publication title:	Health Expectations; Oxford
Volume:	26

Issue:	1
Pages:	440-451
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-23
Milestone dates:	2022-11-01 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-07-27 (manuscriptReceived); 2022-11-23 (publishedOnlineEarlyUnpaginated); 2022-11-13 (manuscriptAccepted)
Publication history :	
First posting date:	23 Nov 2022
DOI:	https://doi.org/10.1111/hex.13677
ProQuest document ID:	2767101954
Document URL:	https://www.proquest.com/scholarly-journals/qualitative-investigation-perceptions-towards/docview/2767101954/se-2?accountid=211160

Last updated:	2023-08-04

Preferences of patients with chronic low back pain about nonsurgical treatments: Results of a discrete choice experiment

Morillon, Gabin F [1]
; Benkhalti, Maria [2] ; Dagenais, Pierre [3] ; Poder, Thomas G [4]
[1] Montpellier Recherche en Economie, University of Montpellier, Montpellier, France [2] CIUSSS de l'Estrie—CHUS, Sherbrooke, Quebec, Canada [3] CIUSSS de l'Estrie—CHUS, Sherbrooke, Quebec, Canada; Department of Medicine, Faculty of Medicine and Health Science, University of Sherbrooke, Sherbrooke, Quebec, Canada [4] Department of Management, Evaluation and Health Policy, School of Public Health, University of Montreal, Montreal, Quebec, Canada; Centre de recherche de l'Institut Universitaire en Santé Mentale de Montréal, CIUSSS de l'Est de l'île de Montréal, Montreal, Quebec, Canada

ABSTRACT (ENGLISH)

Introduction
This study aimed to assess patients' preferences of nonsurgical treatments for chronic low back pain (CLBP).
Method
We conducted a discrete choice experiment (DCE) in Quebec, Canada, in 2018. Seven attributes were included: treatment modality, pain reduction, the onset of treatment efficacy, duration effectiveness, difficulties with daily activities, sleep problems, and knowledge of the patient's body and pain location. Treatment modalities were corticosteroid injections, supervised body-mind physical activities, supervised sports physical activities, physical manipulations, self-management courses, and psychotherapy. Utility levels were estimated using a logit model, a latent class model and a Bayesian hierarchical model.
Results
Analyses were conducted on 424 424 individuals. According to the Bayesian hierarchical model, the conditional relative importance weights of attributes were as follows: (1) treatment modality (34.79%), (2) pain reduction (18.73%), (3) difficulties with daily activities (11.71%), (4) duration effectiveness (10.06%), (5) sleep problems (10.05%), (6) onset of treatment efficacy (8.60%) and (7) knowledge of the patient's body and pain location (6.06%). According to the latent class model that found six classes of respondents with different behaviours (using Akaike and Bayesian criteria), the treatment modality was the most important attribute for all classes, except for class 4 for which pain reduction was the most important. In addition, classes 2 and 5 refused corticosteroid injections, while psychotherapy was preferred only in class 3.
Conclusion
Given the preference heterogeneity found in the analysis, it is important that patient preferences are discussed and considered by the physicians. This will help to improve the patient care pathway in a context of a patient-centred model for a disease with growing prevalence.
Patient or Public Contribution
A small group of patients was involved in the conception, design and interpretation of data. Participants in the

DCE were all CLBP patients.

FULL TEXT

INTRODUCTION

Low back pain (LBP) refers to pain occurring in the lumbar region (vertebrae L1–L5) and affects individuals of all ages, genders and conditions.[1] LBP is classified according to its duration[2]: acute or short-term back pain (lasting less than 6 weeks), subchronic back pain (lasting between 6 and 12 weeks) and chronic back pain (lasting 12 weeks or longer). As reported by Nieminen et al.,[3] risk factors of chronic low back pain (CLBP) are physical (e.g., overweighting, hypertension, smoking, congenital problems), social (e.g., work environment) and psychological (e.g., depression, anxiety, behaviour). Besides its cost to society (e.g., USD 100 billion in the United States[4]), LPB impairs many of the various aspects of individuals' daily lives. Its lifetime prevalence is estimated between 60% and 70% in industrialized countries with a peak between the ages of 35 35 and 55, $55,$[5] and according to the literature review conducted by Meucci et al.,[6] the average prevalence of CLBP is 11.84% (from 1% in China to 25.4% in Brazil). Moreover, four-fifths of individuals will experience back pain in their lifetime, and one-fifth experiencing acute back pain will develop CLBP.[7]

Individuals with CLBP face several treatment alternatives. As recommended by the guideline of the Institute of Health Economics (Alberta, Canada),[8] treatments facing CLBP include exercise (e.g., physical, therapeutic, aquatic exercises), yoga therapy, active rehabilitation and self-management programmes, massage therapy, acupuncture, medication (i.e., nonsteroidal anti-inflammatory drugs, muscle relaxants, antidepressants and opioids), herbal medicine, behavioural therapy, injection therapy and surgery. Note that nonpharmacologic treatments should be used as first-line treatments.[9] However, the lack of certainty about recovery and adverse events is a major issue in the choice of treatment.[10]

Patients' decisions and beliefs are taking a growing place in their care pathways.[11,12] Patients' preferences are driven by a trade-off between the perceived risks and the perceived benefits, and considering their preferences, especially when these are strong and linked to social, environmental or psychological factors, and when the outcomes are uncertain, is beneficial to promote shared decisions. In a qualitative survey, Dima et al.[11] underlined four treatment beliefs expressed by patients in their research of care. The treatment must be credible (i.e., making sense, being in adequation with pain and delivering by the right practitioner), effective, safe and affordable and adequate to the individuals. Better comprehension and shared decisions between the practitioner and the patient often led to greater satisfaction and better results. From their side, Poder and Beffarat[13] conducted a mixed studies review on LBP to highlight treatments and attributes (i.e., treatments' characteristics), and out of the 13 articles included, they found that patients granted priority to effectiveness, followed by the capacity to realize daily life activities, fit to patient's life and the credibility of the treatment, among others. These studies indicate that knowing patients' preferences could have the potential to improve treatments' adherence and effectiveness.

However, patients' preferences on CLBP treatments are not well known. The present study was the result of a request from an academic healthcare institution in Quebec (i.e., the CIUSSS de l'Estrie—CHUS) to improve the organization of care and provision of CLBP treatments, especially nonsurgical treatments. Consequently, we conducted a discrete choice experiment (DCE) to assess patients' preferences for nonsurgical treatments for CLBP. DCE is a stated preference method that aims to elicit preferences throughout a set of choice tasks on the basis of a finite set of alternatives. This method relies on the random utility theory and on the ability of individuals to make choices between treatments with the same characteristics (attributes) but different modalities (levels).

To our knowledge, only 7 DCEs on patients' preferences specifically refer to back pain treatments with mean samples of about 337 individuals using between 5 and 8 attributes. Different treatments were studied such as pharmaceutical treatments[14] or both pharmaceutical and surgical treatments.[15] Moreover, acupuncture and low infrared treatments were studied by Chen et al.,[16] physical exercises by Aboagye et al.[17] and Ferreira et al.,[18] and other pain management programmes by Yi et al.[19] Finally, Walsh et al.[20] made focus on treatments' adverse events. The most recurrent attributes were cost, time to effect (i.e., the onset of treatment effectiveness), type of treatment

(i.e., modality, type of training, the content of the programme), frequency (i.e., the number of sessions), design (i.e., individually or in the group, the size of the group) and travel time (i.e., from the clinic or the gym). A total of 21 different attributes were used in the studies we found, and all the authors were assessing pain and exercise activities experienced by patients using a set of questions or specific questionnaires such as the Chronic Pain Grade,[21] the Self-Efficacy to Exercise Scale[22] or the Form-C of the Multidimensional Health Locus of Control,[23] which examines perceived health status as being dependent from internality, chance and powerful others items. The main difference in our study is that we only considered first-line treatments (i.e., nonsurgical treatments) and put emphasis on corticosteroid injections (which was the treatment focus in the evaluation request made by our institution) versus five other first-line treatments that have shown effectiveness in the scientific literature.

METHODOLOGYSurvey design

An online questionnaire was conducted in Quebec, Canada, at the end of 2018 among members of a provincial support group for patients with chronic pain, the Association Québécoise de la Douleur Chronique (AQDC). Respondents had to suffer from LBP for at least 3 months to be included in the survey (i.e., to have CLBP). The study was conducted in both French and English, using the online survey platform provided by Sawtooth Software (Sawtooth Software Inc.).

Among the members of the AQDC, it was estimated that about 1,500 suffered from CLBP and it was expected to recruit 15%–20% of them through an invitation letter sent by the president of the AQDC. A minimal sample of 200 200–300 300 respondents was thus expected, which is in line with the usual recommendation for a DCE[24–28] and with the usual empirical findings.[29–31] The invitation letter was sent through an email to all members of the AQDC, specifying that only members with CLBP were invited to participate.

The survey included a first part on health status and sociodemographic data, including specific questions about LBP such as the medical diagnosis, the duration of pain, the frequency of painkillers medication use, other treatments (apart from painkillers) and numeric pain rating scales (NPRS)[32] (i.e., current pain, worst and average pain in last 2 weeks) ranging from 0 0 (no pain) to 10 10 (worst pain). Three scales ranging from 0 0 to 10 10 measured personal health and life satisfaction and willingness to take risks. The second part was the DCE itself followed by choice certainty scales from 0 0 (not certain at all) to 10 10 (absolutely certain). The third part included follow-up questions (i.e., choice tasks difficulty, number of dimensions taken into account, self-ranking of dimensions, responses quality, irritation/boredom) and four health-related quality of life questionnaires (i.e., EQ-5D-5L[33] with its Visual Analogue Scale [VAS], SF-6Dv2,[34] Oswestry Disability Index [ODI][35] and Rolland-Morris Disability Questionnaire [RMDQ][36]). A Quality-Adjusted Life-Year (QALY) derived from the EQ-5D-5L and the SF-6Dv2 equal to 1 indicates a perfect health state while a QALY equal to 0 indicates a state of death. Cronbach's α were equal to .783, .801, .842 and .853, respectively. Finally, to compare the relative importance of attributes elicited from the choice exercise with respondents' personal perception and to assess the validity of their choices, we asked them to rank the seven attributes and then the six treatments proposed in the DCE, as well as to complete the four-item questionnaire from Dima et al.[37] adjusted for the treatments presented in the DCE. The latter is a validated psychometric questionnaire used to assess patients' beliefs regarding their LBP treatments and to examine the determinants of treatment uptake. Cronbach's α ranged from .651 to .818.

DCE design, attributes and levels

The DCE was constructed following the ISPOR recommendations.[24] We used a mixed-methods design including a systematic literature review, a patients' focus group (n = 4) and discussions with two patients and experts (i.e., two economists specialized in preference-based studies, one rheumatologist, one ethicist and one public health professional with expertise in equity) as well as with a health technology assessment (HTA) consultative committee.[38] After discussing a list of 40 attributes with the patients' group, 7 attributes with 3–6 levels were retained and then agreed by the HTA committee: treatment modality, pain reduction, the onset of treatment efficacy, duration effectiveness, difficulties with daily activities, sleep problems, knowledge of his/her body and pain location (Table 1). Based on this, the survey was developed by the research team. The survey was pretested for appropriateness and univocity by all patients from the focus group, as well as by one medical doctor, two patient representatives and one

public health expert. This led to minimal changes in the final survey.[38] The online field survey was also pretested before the launch. This was carried out with the same patients from the focus group and the two patient representatives, as well as with four additional patients who provided their insights about the field survey, including an understanding of the DCE tasks, ease of completion and conviviality of the format used. All levels in the DCE were dummy-coded. The treatment modality had six levels: corticosteroid injections, supervised body-mind physical activities, supervised sports physical activities, physical manipulations, self-management courses, and psychotherapy. Examples of such activities were provided to respondents (Table 1) and were presented as such to participants before the start of the DCE. When selected, the two treatment modalities presented in the DCE worked as a label since they were presented first. For more details, see Poder et al.[38] and Poder and Beffarat.[13]

Table 1 DCE attributes and levels

Attributes	Levels
Treatment modality	Corticosteroid injections
	Supervised body-mind physical activities (e.g., Yoga, Tai-Chi, Pilates)
	Supervised sports and physical activities (e.g., active walking, swimming, bike riding, weight training, CrossFit)
	Physical manipulations (e.g., chiropractic, physiotherapy, occupational therapy, kinesiotherapy)
	Self-management courses (e.g., medication, self-hypnosis, breathing techniques, relaxation)
	Psychotherapy (e.g., cognitive-behavioural therapy, posttraumatic shock, progressive muscular relaxation, motivational approach)
Pain reduction	None to very slight
	Slight
	Reduced by half
	No pain
Onset of treatment efficacy	1 month
	6 months
	12 months
Duration effectiveness	Effective for 2 months

	Effective for 6 months
	Effective for 12 months
Difficulties with daily activities	As many difficulties as before
	Fewer difficulties as before
	No difficulties
Sleep problems	As many problems as before
	Fewer problems than before
	No problems
Knowledge of his/her body and pain location	Same knowledge as before
	Knowledge somewhat better than before
	Knowledge much better than before

Abbreviation: DCE, discrete choice experiment.

About 5832 different scenarios were possible resulting from all combinations of attributes and levels, yielding a possibility of 34,006,392 $\mathrm{34,006,392}$ pairs. An orthogonal selection procedure allowed us to generate 600 600 scenarios combined into 300 300 pairs. These pairs were divided into 30 30 blocks. Each respondent had to answer to one block of 10 10 pairs plus one pair for the rationality test (i.e., one scenario dominating the other one and was presented first in the DCE) and one pair for the temporal consistency test (i.e., choice cards 2 and 12 were identical). The blocks' distribution was run randomly among participants. An illustration of a choice card is given in Supporting Information: Appendix 1.

Models

The Conditional Logit (CL) model[39] is easy to perform but suffers from two limitations: (1) The independence of irrelevant alternatives (IIA) hypotheses that states that the relative probability of selecting alternatives should not change if we introduce or eliminate another alternative[40] and (2) it does not consider preference heterogeneity (i.e., that individuals have different preferences). Thus, we used two other models for the analysis of the DCE: a Hierarchical Bayesian (HB) model and a Latent Class Logit (LC) model. The results of the CL model are however presented in Supporting Information: Appendix 2.

A dual-response none opt-out option (named ‘none’) was allowed in all models (i.e., respondents could choose neither the first nor the second scenario in each pair). The LC model considers interclass homogeneity and intraclass heterogeneity while the HB model considers that all individuals have different preferences. In the HB model, all parameters were specified as normally distributed. We also performed cross-tabulation between latent classes clusters (Supporting Information: Appendix 3.1) and segmentation via a hierarchical clustering method using the Ward's minimum variance method and L2 squared dissimilarity measure (Supporting Information: Appendix 3.2). It allowed us to provide a comparison with the LC model. For the latter, we used Akaike and Bayesian information criteria, the log-likelihood function and the interpretation of the results to determine the number of latent classes

(Supporting Information: Appendix 4). Subject preferences were expressed using part-worth utilities calculated using algorithms in Sawtooth Software Lighthouse Studio version 9.7.2. Utility values were zero-centred, and the value of the reference modality was calculated as the negative of the sum of the other modalities' values. A high positive level of utility thus indicates a high preference for the level concerned. For comparison purpose, attributes' conditional relative importance are presented for each model with standard deviation and 95% confidence intervals. [41]

Descriptive statistics

Two ways of considering the individuals' sociodemographic characteristics in such models can be used. First, the LC model does not assign individuals to a class, hence the term ‘latent’. However, it generates class membership probabilities for each individual. To assign individuals to each latent class, we can consider the maximum class membership probability (Supporting Information: Appendix 5). Second, the effects of variables of interest can directly be estimated by integrating them into the model specification as being continuous or dummy-coded (see Boeri et al. [42] for more details). Although we chose the primer option which consists in assigning an individual relative to his/her maximum class membership probability, we must be cautious because this individual does not really belong to this class.

For purposes of descriptive analysis, relevant tests (i.e., one-way analysis of variance, Kruskal–Wallis *H*-test, Bartlett's test for equality of variances, Fisher's exact test and χ^2 test of independence) were performed to assess differences between classes. The bivariate results are presented in terms of column frequencies. A *p*-value <.1 was considered significant.

Inclusion criteria

To be included in the analysis, participants had to have LBP for at least 3 months, to have completed some of the choice tasks, to have responded ‘yes’ to the question ‘Did you try to answer the choice exercises as well as possible?’, to have not declared ‘poor’ or ‘very poor’ quality of answers at the question ‘How do you judge the quality of your responses to the choice exercises?’, to do not always chose the first treatment or the second treatment.

RESULTSSurvey uptake

Among a total of 610 610 respondents who began the survey, 134 134 (21.97%) did not achieve any of the choice tasks, 6 6 (0.98%) declared not responding as well as possible to the choice exercises, 6 6 (0.98%) declared ‘poor’ or ‘very poor’ quality of answers, 6 6 (0.98%) had low back pain for less than 3 months, 31 31 (5.08%) never chose a treatment (opt-out), 2 2 (0.33%) always chose the first treatment and 1 1 (0.16%) always chose the second treatment. This yielded a total of 424 424 (69.51%) individuals included for analysis.

Descriptive analysis

All descriptive statistics presented here are available in Table 2 (Supporting Information: Appendix 6 for 4 classes). The sample had a mean age of 55 55 years (±12.62 ± 12.62) and was in majority composed of women (F/M ratio of 4.15 4.15). About 36.08% were married, 36.56% were retired and 43.40% had a higher education with an average household income of 57,883 $\mathrm{57,883}$ CAD (±37,619 $\pm \mathrm{37,619}$). Almost all individuals reported having LBP for more than 1 year (98.11%) and the principal diagnoses were lumbar disc herniation (37.74%), facet arthritis (34.91%) and fibromyalgia (35.14%). When answering the survey, individuals had an average level of CLBP of 5.39 5.39 (±2.03 ± 2.03) over 10 10. The worst pain and the average level of CLBP in the past 2 weeks were 7.53 7.53 (±1.85 ± 1.85) over 10 10 and 5.53 5.53 (±1.88 ± 1.88) over 10 10, respectively. Almost one-third used painkillers every day. Other treatments used were massage therapy sessions (41.98%), infiltrations of corticosteroid products (36.08%) and physiotherapy sessions (34.20%). The average amount of treatment expenditure was 1542 CAD (±3,519 $\pm \mathrm{3,519}$) per year and 10.38% were not covered by insurance (public or private one). More than two third of respondents reported having a health problem affecting their quality of life (e.g., pain, 60.38%; tiredness, 50.47%; insomnia, 40.33%; anxiety and stress, 37.50%) and half declared having a fair or poor health status. Both satisfactions with health and life, and the willingness to take risks were 4.12 4.12 (±2.42 ± 2.42), 5.53 5.53 (±2.42 ± 2.42) and 4.31 4.31 (±

2.50 ± 2.50) over 10 10, respectively. Finally, the RMDQ, the ODI, the health utilities of the SF-6Dv2 and the EQ-5D-5L and the EQ-5D VAS were, respectively, 10.02 10.02 (±5.18 ± 5.18) over 24 24 (14 14 or more indicates a poor outcome), 42.49 42.49 (±15.36 ± 15.36) over 100 100 (41 41–60 60 indicates severe disability), 0.33 0.33 (±0.23 ± 0.23) and 0.58 0.58 (±0.23 ± 0.23) over 1, and 52 52 (±22.39 ± 22.39) over 100 100.

Table 2 Sociodemographic characteristics by latent classes (c=6 $c=6$) and full sample

Sociodemographic characteristics	Class 1	Class 2	Class 3	Class 4	Class 5	Class 6	Total	*p* Value a
Observations	48	57	43	67	159	50	424	
Absolute share	11.32%	13.44%	10.14%	15.80%	37.50%	11.79%	100.00 %	
Class share	12.60%	14.10%	10.60%	15.70%	34.90%	12.00%	-	
Gender								
Male	18.75%	19.30%	25.58%	14.93%	15.09%	34.00%	19.34%	**.031**
Female	81.25%	80.70%	72.09%	85.07%	84.91%	64.00%	80.19%	
Intersex	0.00%	0.00%	2.33%	0.00%	0.00%	2.00%	0.47%	
Female/male ratio	4.33	4.18	2.82	5.70	5.63	1.88	4.15	-
Age (years)								
Mean	54.69	56.53	56.67	54.73	53.11	61.66	55.38	**.002**
Standard deviation	14.14	12.07	11.82	13.95	12	10.42	12.62	
Range	(28–85)	(26–87)	(32–78)	(25–75)	(20–85)	(32–82)	(20–87)	
Less than 35	8.33%	3.51%	2.33%	7.46%	5.03%	2.00%	4.95%	**.004**
35–39	12.50%	3.51%	2.33%	7.46%	6.92%	0.00%	5.90%	
40–44	6.25%	12.28%	11.63%	13.43%	16.98%	4.00%	12.50%	

45–49	6.25%	5.26%	13.95%	2.99%	8.81%	10.00%	7.78%	
50–54	10.42%	14.04%	13.95%	19.40%	15.09%	6.00%	13.92%	
55–59	16.67%	17.54%	18.60%	8.96%	16.35%	12.00%	15.09%	
60–64	10.42%	21.05%	4.65%	2.99%	14.47%	22.00%	12.97%	
65–69	16.67%	12.28%	13.95%	17.91%	9.43%	18.00%	13.44%	
70–74	6.25%	3.51%	11.63%	16.42%	3.14%	16.00%	8.02%	
75 or more	6.25%	7.02%	6.98%	2.99%	3.77%	10.00%	5.42%	
Body mass index (BMI)								
Mean	28	30.97	29.13	29.56	29.05	29.08	29.28	.316
Standard Deviation	5.78	6.92	7.16	6.61	6.66	6.22	6.6	
Range	(18.42–54.69)	(19.44–58.37)	(18.42–62.75)	(19.96–47.18)	(13.71–50.81)	(19.07–49.94)	(13.71–62.75)	
Marital status								
Married	47.92%	35.09%	32.56%	32.84%	34.59%	38.00%	36.08%	.808
Living with a partner	25.00%	28.07%	23.26%	22.39%	24.53%	18.00%	23.82%	
Single	8.33%	14.04%	20.93%	20.90%	19.50%	12.00%	16.98%	
Separated	6.25%	5.26%	6.98%	5.97%	6.92%	6.00%	6.37%	
Divorced	10.42%	10.53%	9.30%	16.42%	11.32%	24.00%	13.21%	
Widowed	2.08%	7.02%	6.98%	1.49%	3.14%	2.00%	3.54%	
Occupational status								
Employed	18.75%	17.54%	23.26%	19.40%	25.79%	4.00%	20.05%	**.083**
Self-employed	0.00%	1.75%	9.30%	5.97%	6.92%	8.00%	5.66%	

Retired	39.58%	45.61%	34.88%	38.81%	27.04%	52.00%	36.56%	
At home	4.17%	5.26%	4.65%	4.48%	5.03%	6.00%	4.95%	
Student	0.00%	1.75%	0.00%	4.48%	3.77%	0.00%	2.36%	
Unemployed	2.08%	7.02%	2.33%	2.99%	1.26%	4.00%	2.83%	
Sick leave	20.83%	8.77%	11.63%	8.96%	13.21%	18.00%	13.21%	
Parental leave	0.00%	0.00%	0.00%	1.49%	0.00%	0.00%	0.24%	
Other (e.g., disability)	14.58%	12.28%	13.95%	13.43%	16.98%	8.00%	14.15%	
Educational level								
Secondary or less and Diploma of professional studies	25.00%	22.81%	30.23%	26.87%	25.79%	36.00%	27.12%	**.046**
College and CEGEP	35.42%	35.09%	34.88%	29.85%	18.87%	36.00%	28.30%	
Baccalaureate, Masters and PhD	39.58%	42.11%	32.56%	43.28%	53.46%	26.00%	43.40%	
Other	0.00%	0.00%	2.33%	0.00%	1.89%	2.00%	1.18%	
Annual household income (CAD)								
Mean	72,604	63,158	52,965	52,985	56,509	52,900	57,883	**.042**
Standard deviation	36,796	36,421	38,704	34,395	39,345	34,683	37,619	
Range	(40,000–102,500)	(40,000–85,000)	(17,500–75,000)	(30,000–75,000)	(22,500–75,000)	(22,500–75,000)	(30,000–75,000)	
Living with an adult								
Yes	68.75%	71.93%	58.14%	65.67%	66.04%	64.00%	66.04%	.802
No	31.25%	28.07%	41.86%	34.33%	33.96%	36.00%	33.96%	
Underage dependent children (at least one)								

Yes	16.67%	8.77%	18.60%	13.43%	19.50%	12.00%	15.80%	.434
No	83.33%	91.23%	81.40%	86.57%	80.50%	88.00%	84.20%	
Type of residence								
Rural	20.83%	38.60%	20.93%	34.33%	26.42%	34.00%	29.01%	.185
Urban	79.17%	61.40%	79.07%	65.67%	73.58%	66.00%	70.99%	
Owning a home								
Yes	77.08%	68.42%	55.81%	59.70%	66.04%	58.00%	64.62%	.229
No	22.92%	31.58%	44.19%	40.30%	33.96%	42.00%	35.38%	
Smoking								
Yes	14.58%	10.53%	16.28%	10.45%	12.58%	18.00%	13.21%	.807
No	85.42%	89.47%	83.72%	89.55%	87.42%	82.00%	86.79%	
Diagnosis given by a medical doctor								
No diagnosis	0.00%	1.75%	4.65%	5.97%	4.40%	8.00%	4.25%	.364
Muscle and/or ligament sprain	12.50%	7.02%	4.65%	10.45%	7.55%	16.00%	9.20%	.381
Sciatica	27.08%	24.56%	18.60%	31.34%	23.90%	36.00%	26.42%	.394
Lumbar disc herniation	39.58%	42.11%	39.53%	37.31%	33.96%	42.00%	37.74%	.856
Degenerative disc disease	18.75%	24.56%	13.95%	16.42%	16.98%	14.00%	17.45%	.714
Facet arthritis	47.92%	21.05%	44.19%	34.33%	33.33%	36.00%	34.91%	**.069**

Vertebral arthritis or spondylarthrosis	18.75%	21.05%	9.30%	14.93%	8.81%	22.00%	14.15%	**.069**
Spondylolisthesis	8.33%	5.26%	6.98%	4.48%	7.55%	4.00%	6.37%	.914
Deformation (e.g., scoliosis, kyphosis)	14.58%	3.51%	9.30%	13.43%	11.32%	8.00%	10.38%	.381
Osteoporosis	2.08%	8.77%	6.98%	8.96%	7.55%	8.00%	7.31%	.758
Osteoporosis with spinal fracture	0.00%	0.00%	2.33%	1.49%	0.00%	4.00%	0.94%	**.077**
Fracture or dislocation of the spine	0.00%	0.00%	6.98%	1.49%	1.26%	6.00%	2.12%	**.046**
Autoimmune inflammatory disease	10.42%	1.75%	6.98%	2.99%	3.77%	2.00%	4.25%	.266
Fibromyalgia	35.42%	42.11%	27.91%	32.84%	35.22%	36.00%	35.14%	.795
Other diagnosis	27.08%	24.56%	32.56%	22.39%	24.53%	24.00%	25.24%	.890
How long are you suffering from low back pain?								
Between 3 months and 1 year	0.00%	1.75%	6.98%	0.00%	2.52%	0.00%	1.89%	.133
More than 1 year	100.00 %	98.25%	93.02%	100.00 %	97.48%	100.00 %	98.11%	
Today low back pain (ranging from 0 to 10)								
Mean	5.71	5.60	5.65	5.00	5.17	5.86	5.39	**.090**
Standard deviation	1.83	2.11	2.11	2.06	1.98	2.06	2.03	
Range	(2–9)	(0–10)	(2–10)	(0–9)	(0–10)	(1–10)	(0–10)	

Worst level of low back pain in the past 2 weeks (ranging from 0 to 10)								
Mean	7.83	7.56	7.49	7.34	7.45	7.78	7.53	.666
Standard deviation	1.73	2.07	1.82	2.13	1.69	1.87	1.85	
Range	(4–10)	(2–10)	(2–10)	(0–10)	(4–10)	(3–10)	(0–10)	
Average level of low back pain in the past 2 weeks (ranging from 0 to 10)								
Mean	5.75	5.46	5.72	5.4	5.35	6.02	5.53	.267
Standard deviation	1.54	1.88	1.92	2.02	1.83	2.07	1.88	
Range	(2–10)	(1–10)	(1–10)	(0–10)	(1–10)	(1–10)	(0–10)	
Frequency of use of painkillers								
Several times a day	33.33%	29.82%	51.16%	22.39%	22.64%	40.00%	29.72%	.196
Every day	33.33%	33.33%	23.26%	31.34%	31.45%	32.00%	31.13%	
Several times a week	18.75%	5.26%	9.30%	16.42%	18.24%	10.00%	14.39%	
Once a week	2.08%	3.51%	4.65%	2.99%	1.26%	2.00%	2.36%	
Several times a month	4.17%	8.77%	4.65%	5.97%	9.43%	2.00%	6.84%	
Once a month	2.08%	5.26%	4.65%	0.00%	3.77%	2.00%	3.07%	
Several times a year	2.08%	7.02%	2.33%	7.46%	6.92%	8.00%	6.13%	
Once a year	0.00%	1.75%	0.00%	1.49%	0.63%	0.00%	0.71%	
Never	4.17%	5.26%	0.00%	11.94%	5.66%	4.00%	5.66%	
Treatments for reducing pain other than painkillers								
Homoeopathic products	2.08%	8.77%	2.33%	2.99%	10.69%	10.00%	7.31%	.124

Infiltration of corticosteroid products	62.50%	12.28%	53.49%	34.33%	31.45%	40.00%	36.08%	**<.001**
Chiropractic sessions	16.67%	15.79%	4.65%	17.91%	13.84%	12.00%	13.92%	.467
Physiotherapy sessions	43.75%	29.82%	27.91%	26.87%	39.62%	28.00%	34.20%	.172
Osteopathy sessions	12.50%	26.32%	23.26%	23.88%	28.93%	24.00%	24.76%	.355
Occupational therapy sessions	10.42%	3.51%	4.65%	1.49%	5.03%	16.00%	6.13%	**.027**
Psychotherapy sessions	18.75%	5.26%	13.95%	5.97%	13.21%	10.00%	11.32%	.176
Reflexology sessions	0.00%	1.75%	2.33%	2.99%	3.77%	6.00%	3.07%	.675
Massage therapy sessions	50.00%	35.09%	44.19%	46.27%	41.51%	36.00%	41.98%	.590
Yoga sessions	12.50%	19.30%	6.98%	10.45%	21.38%	12.00%	15.80%	.103
Stretching sessions	16.67%	12.28%	23.26%	14.93%	20.75%	20.00%	18.40%	.638
Acupuncture sessions	10.42%	7.02%	13.95%	11.94%	15.72%	18.00%	13.44%	.529
Cupping sessions	0.00%	1.75%	0.00%	2.99%	5.03%	0.00%	2.59%	.291
Infrared frequency sessions	4.17%	0.00%	0.00%	1.49%	2.52%	0.00%	1.65%	.522
Bodybuilding	6.25%	14.04%	4.65%	7.46%	12.58%	16.00%	10.85%	.322
Endurance activities (aerobic)	10.42%	7.02%	6.98%	5.97%	11.32%	6.00%	8.73%	.774

Consumption of medical cannabis	12.50%	15.79%	13.95%	20.90%	16.98%	10.00%	15.80%	.663
Others	37.50%	28.07%	34.88%	29.85%	32.08%	36.00%	32.55%	.892
Treatment expenditure per year (CAD)								
Mean	2107.29	961.93	1770.93	1905.82	1433.45	1323.60	1542.26	.547
Standard deviation	4821.23	1245.30	3602.08	6679.35	1648.68	1364.97	3519.43	
Range	(0–34,000)	(0–7000)	(0–20,000)	(0–55,000)	(0–10,000)	(20–8000)	(0–55,000)	
Insurance								
RAMQ (carte soleil)	39.58%	36.84%	39.53%	44.78%	33.96%	46.00%	38.68%	**.091**
Private insurance	60.42%	57.89%	46.51%	44.78%	52.20%	42.00%	50.94%	
No insurance	0.00%	5.26%	13.95%	10.45%	13.84%	12.00%	10.38%	
Do you suffer from a disease or a physical or mental problem that reduces your quality of life (e.g., diabetes, cancer, osteoarthritis)?								
Yes	75.00%	68.42%	83.72%	65.67%	64.78%	60.00%	67.92%	.138
No	25.00%	31.58%	16.28%	34.33%	35.22%	40.00%	32.08%	
Tiredness	52.08%	49.12%	60.47%	50.75%	49.69%	44.00%	50.47%	.947
Insomnia	52.08%	38.60%	55.81%	26.87%	37.74%	44.00%	40.33%	**.049**
Pain	70.83%	57.89%	72.09%	61.19%	57.23%	52.00%	60.38%	.667
Anxiety/stress	39.58%	31.58%	51.16%	40.30%	35.22%	34.00%	37.50%	.759

Depression	27.08%	19.30%	30.23%	26.87%	24.53%	18.00%	24.29%	.833
Other mental disorder	6.25%	3.51%	6.98%	4.48%	3.14%	4.00%	4.25%	.925
Osteoarthritis	52.08%	47.37%	58.14%	38.81%	40.88%	34.00%	43.63%	.763
Arthritis	10.42%	8.77%	18.60%	11.94%	8.81%	14.00%	11.08%	.673
Unintentional injury	2.08%	3.51%	4.65%	4.48%	5.03%	2.00%	4.01%	.949
Musculoskeletal problem	25.00%	17.54%	18.60%	20.90%	21.38%	18.00%	20.52%	.838
Disease of the central nervous system	6.25%	5.26%	13.95%	4.48%	6.92%	6.00%	6.84%	.793
Thyroid problem	8.33%	14.04%	9.30%	20.90%	13.84%	4.00%	12.74%	**.057**
Other endocrine problem	6.25%	1.75%	2.33%	5.97%	3.77%	0.00%	3.54%	.505
Genital-urinary problem	6.25%	7.02%	9.30%	5.97%	5.66%	4.00%	6.13%	.991
Hypertension	22.92%	31.58%	37.21%	25.37%	17.61%	28.00%	24.53%	.136
Cardiac disease	4.17%	3.51%	13.95%	5.97%	1.26%	6.00%	4.48%	**.033**
Stroke	2.08%	0.00%	2.33%	0.00%	0.63%	0.00%	0.71%	.473
Digestive disorder	10.42%	15.79%	27.91%	26.87%	16.98%	12.00%	18.16%	**.097**
Other gastrointestinal problem	16.67%	19.30%	16.28%	14.93%	11.32%	10.00%	13.92%	.777

Diabetes	8.33%	19.30%	27.91%	19.40%	8.18%	22.00%	15.09%	**.006**
Cancer/tumour	0.00%	1.75%	0.00%	7.46%	0.63%	2.00%	1.89%	**.021**
Chronic obstructive pulmonary disease (COPD)	0.00%	0.00%	0.00%	1.49%	1.89%	2.00%	1.18%	.790
Other breathing problems (asthma, emphysema)	6.25%	5.26%	4.65%	17.91%	11.95%	14.00%	10.85%	**.031**
Other medical disorder	27.08%	31.58%	20.93%	20.90%	31.45%	34.00%	28.54%	**.044**
Health status								
Mean	3.58	3.42	3.81	3.51	3.39	3.62	3.50	**.093**
Standard deviation	0.90	1.03	0.82	0.84	0.89	0.83	0.90	
Range	(2–5)	(2–5)	(2–5)	(1–5)	(1–5)	(2–5)	(1–5)	
Excellent	0.00%	0.00%	0.00%	1.49%	1.26%	0.00%	0.71%	**.066**
Very good	6.25%	21.05%	6.98%	7.46%	13.21%	8.00%	11.32%	
Good	50.00%	35.09%	23.26%	40.30%	41.51%	36.00%	38.92%	
Fair	22.92%	24.56%	51.16%	40.30%	33.33%	42.00%	34.91%	
Poor	20.83%	19.30%	18.60%	10.45%	10.69%	14.00%	14.15%	
Satisfaction with health								
Mean	3.96	4.26	3.26	4.27	4.30	4.10	4.12	.215
Standard deviation	2.18	2.61	2.32	2.44	2.4	2.44	2.42	
Range	(0–8)	(0–9)	(0–8)	(0–9)	(0–10)	(0–9)	(0–10)	
Satisfaction with life								

Mean	5.73	5.63	5.33	5.55	5.74	4.72	5.53	.187
Standard deviation	2.01	2.44	2.84	2.26	2.38	2.62	2.42	
Range	(1–9)	(0–10)	(0–10)	(0–10)	(0–10)	(0–9)	(0–10)	
Willingness to take risks								
Mean	4.42	4.00	4.07	4.33	4.38	4.54	4.31	.863
Standard deviation	2.43	2.51	2.54	2.34	2.53	2.67	2.5	
Range	(0–9)	(0–9)	(0–8)	(0–8)	(0–9)	(0–10)	(0–10)	
Roland-Morris Disability Questionnaireb								
Mean	9.76	9.69	10.46	10.32	9.58	11.38	10.02	.475
Standard deviation	4.94	4.65	4.92	5.43	5.32	5.37	5.18	
Range	(1–22)	(1–19)	(1–20)	(1–23)	(1–23)	(1–22)	(1–23)	
Oswestry Disability Indexc								
Mean	44.45	43.57	45.74	42.65	38.89	48.39	42.49	**.005**
Standard deviation	12.21	16.20	13.68	15.38	16.27	13.14	15.36	
Range	(20–66)	(6–80)	(14–92)	(6–78)	(6–76)	(24–86)	(6–92)	
Minimal disability (0–20)	2.27%	14.89%	2.63%	8.06%	16.90%	0.00%	10.16%	**<.001**
Moderate disability (21–40)	38.64%	25.53%	31.58%	35.48%	40.85%	24.39%	35.03%	
Severe disability (41–60)	43.18%	48.94%	60.53%	43.55%	31.69%	63.41%	43.58%	
Crippled (61–80)	15.91%	10.64%	2.63%	12.90%	10.56%	9.76%	10.70%	
Bed-bound (81–100)	0.00%	0.00%	2.63%	0.00%	0.00%	2.44%	0.53%	

SF-6Dv2d								
Mean	0.337	0.343	0.246	0.332	0.356	0.306	0.332	.200
Standard deviation	0.218	0.220	0.271	0.234	0.223	0.234	0.231	
Range	(−0.245–0.703)	(−0.191–0.840)	(−0.373–0.759)	(−0.317–0.843)	(−0.320–0.882)	(−0.320–0.799)	(−0.373–0.882)	
EQ-5D-5Le								
Mean	0.563	0.565	0.519	0.613	0.597	0.515	0.575	.137
Standard deviation	0.200	0.230	0.257	0.226	0.227	0.230	0.229	
Range	(0.088–0.885)	(0.129–0.885)	(−0.072–0.860)	(0.045–0.904)	(−0.044–0.904)	(−0.064–0.828)	(−0.072–0.904)	
EQ-5D-5L-VAS								
Mean	46.42	53.79	51.57	55.21	54.99	41.27	52.00	**.006**
Standard deviation	18.81	21.63	23.70	24.33	21.77	21.51	22.39	
First quartile	33	36	31	36	40	24	34	
Median	46	56	51	59	54	40	53	
Third quartile	59	68	68	72	71	58	69	
Range	(11–89)	(0–88)	(0–100)	(5–100)	(0–100)	(2–83)	(0–100)	

Note: Bold values are statistiscally significant at p <.1 level.

Abbreviations: CEGEP, Collège d'enseignement général et professionnel; QALY, Quality-Adjusted Life-Year; RAMQ, Régie de l'assurance maladie du Québec.

a

The p-values refer to tests between classes using the one-way analysis of variance, Kruskal–Wallis H-test, Bartlett's test for equality of variances, Fisher's exact test and χ^2 test of independence.

b

The Roland-Morris Disability Questionnaire is a 24-item questionnaire measuring self-assessed back pain with a yes/no format and ranging from 0 (no back pain) to 24 (worst back pain).

c

The Oswestry Disability Index is a 10-item questionnaire with a 6-point Likert scaling and rescaled from 0 to 100.

d

The Short Form 6-Dimension version 2 (SF-6Dv2) is a 6-dimension generic health-related quality of life questionnaire.

e

The EuroQol 5-Dimension 5-Level (EQ-5D-5L) is a 5-dimension generic health-related quality of life questionnaire.

Preference analysis

The ranking of the attributes' conditional relative importance was the same in two models (HB and LC). It was as follows: (1) treatment modality, (2) pain reduction, (3) difficulties with daily activities, (4) duration effectiveness, (5) sleep problems, (6) onset of treatment effectiveness and (7) knowledge of his/her body and pain location (Table 3).

Table 3 Hierarchical Bayesian and Latent Class Logit models with the conditional relative importance of attributes

Part-worth utilities								
Attribute	Bayesian Hierarchical model				Latent Class model			
	Standardized utilitiesa	Standard deviation	Lower 95% CI	Upper 95% CI	Standardized utilitiesa	Standard deviation	Lower 95% CI	Upper 95% CI
Noneb	−45.63	159.19	−60.78	−30.48	−129.97	254.71	−154.22	−105.73
Treatment modality								
Corticosteroid injections	−52.18	121.81	−63.78	−40.59	−63.54	147.25	−77.56	−49.53
Supervised body-mind physical activities	23.30	61.09	17.48	29.11	24.94	57.25	19.49	30.39
Supervised sports physical activities	6.15	89.92	−2.41	14.71	20.84	68.64	14.30	27.37
Physical manipulations	52.57	49.66	47.85	57.30	59.39	28.24	56.70	62.08
Self-management courses	21.47	59.66	15.80	27.15	20.62	47.74	16.07	25.16
Psychotherapy	−51.31	76.19	−58.56	−44.06	−62.24	60.73	−68.02	−56.46
Pain reduction								
None to very slight	−61.18	40.95	−65.07	−57.28	−69.92	50.02	−74.69	−65.16
Slight	−16.69	25.55	−19.12	−14.25	−18.81	21.77	−20.89	−16.74

Reduced by half	33.67	23.75	31.41	35.93	40.25	18.55	38.49	42.02
No pain	44.19	44.36	39.97	48.41	48.48	47.76	43.94	53.03
Onset of treatment efficacy								
1 month	12.97	30.37	10.08	15.87	16.29	18.57	14.53	18.06
6 months	7.18	16.08	5.65	8.71	3.76	10.77	2.73	4.78
12 months	−20.15	28.58	−22.87	−17.43	−20.05	20.07	−21.96	−18.14
Effectiveness duration								
Effective for 2 months	−26.61	28.10	−29.28	−23.93	−24.75	28.68	−27.48	−22.02
Effective for 6 months	1.83	25.50	−0.59	4.26	−2.43	11.80	−3.55	−1.30
Effective for 12 months	24.78	23.89	22.50	27.05	27.17	20.31	25.24	29.11
Difficulties with daily activities								
As many difficulties as before	−42.64	29.23	−45.42	−39.85	−45.61	29.69	−48.44	−42.78
Fewer difficulties as before	17.30	19.41	15.45	19.15	18.97	19.98	17.07	20.87
No difficulties	25.33	23.58	23.09	27.58	26.64	16.49	25.07	28.21
Sleep problems								
As many problems as before	−25.25	32.14	−28.31	−22.19	−24.71	30.48	−27.61	−21.81
Fewer problems than before	14.45	18.99	12.65	16.26	18.70	10.62	17.69	19.71
No problems	10.79	30.61	7.88	13.71	6.00	36.97	2.48	9.52
Knowledge of his/her body and pain location								

Same knowledge as before	−8.74	19.77	−10.62	−6.86	−11.36	10.94	−12.40	−10.32
Knowledge somewhat better than before	−1.86	17.41	−3.51	−0.20	1.86	15.63	0.37	3.35
Knowledge much better than before	10.59	19.34	8.75	12.44	9.50	20.82	7.52	11.48
Observations	424				424			
McFadden R^2	30.01				26.04			
χ^2	2830.41				2730.48			
Log-likelihood	−3701.45				−3876.09			
Log-likelihood (null)	−5233.01				−5243.68			
AIC	7901.67				7994.88			
BIC	8567.87				8764.89			
Relative weight of attributes								
Attribute	Bayesian Hierarchical model				Latent Class model			
	Weight (%)	Standard deviation	Lower 95% CI	Upper 95% CI	Weight (%)	Standard deviation	Lower 95% CI	Upper 95% CI
Treatment modality	34.79	11.94	33.65	35.93	36.06	12.74	34.85	37.28
Pain reduction	18.73	7.13	18.05	19.41	20.43	8.32	19.63	21.22
Onset of treatment efficacy	8.60	5.12	8.11	9.09	6.65	3.59	6.31	6.99
Effectiveness duration	10.06	4.76	9.61	10.52	9.96	2.02	9.77	10.15
Difficulties with daily activities	11.71	5.41	11.20	12.23	11.40	5.16	10.91	11.89
Sleep problems	10.05	4.60	9.61	10.48	10.07	4.34	9.66	10.48

Knowledge of his/her body and pain location	6.06	3.17	5.76	6.36	5.43	3.06	5.14	5.72

Abbreviations: AIC, Akaike Information Criterion; BIC, Bayesian Information Criterion.

a

A value close to zero indicates an absence of preference: the further the value is from zero, the greater the preference.

b

Coefficient of the opt-out option.

According to the HB model, the conditional relative importance of these attributes ranged from 34.79% (±11.94 ± 11.94) to 6.06% (±3.17 ± 3.17) whereas it was from 36.06% to 5.43% in the LC model, showing little difference (Table 3). For treatments in the HB model, the standardized utility values (i.e., the personal satisfaction provided by chosen treatments) were as follows: −52.18 -52.18 (±121.81 ± 121.81) for corticosteroid injections, 23.30 23.30 (±61.09 ± 61.09) for supervised body-mind physical activities, 6.15 6.15 (±89.92 ± 89.92) for supervised sports physical activities, 52.57 52.57 (±49.66 ± 49.66) for physical manipulations, 21.47 21.47 (±59.66 ± 59.66) self-management courses and −51.31 -51.31 (±76.19 ± 76.19) for psychotherapy, thus showing on average a high disutility for corticosteroid injections and psychotherapy. However, in the LC model, classes 1, 3 and 6 (i.e., one-third of the full sample) expressed a high utility for corticosteroid injections, and only class 3 (i.e., one-tenth of the full sample) expressed a utility for psychotherapy (Table 4) (Supporting Information: Appendix 7 for 4 classes). Some discrepancies were identified in the utility levels, particularly in class 6 (e.g., they expressed a high disutility as the level of treatment effectiveness increased or when the level of sleep problems decreased), which was considered as a limit.

Table 4 Zero-centred utility values produced by the latent class analysis (c=6 ${\boldsymbol{c}}={\bf{6}}$) and conditional relative importance of attributes

Latent class logit						
Attributes (standardized utilities)a	Class 1	Class 2	Class 3	Class 4	Class 5	Class 6
Noneb	−266.03	30.16	−88.43	101.02	−400.98	333.90
Treatment modality						
Corticosteroid injections	98.32	−322.34	148.46	−3.90	−159.76	99.49
Supervised body-mind physical activities	−35.28	141.11	−43.22	−12.99	54.71	−31.82
Supervised sports physical activities	75.37	46.56	−175.12	10.47	39.84	83.49
Physical manipulations	42.70	82.24	−5.58	93.21	60.74	21.29
Self-management courses	−52.13	124.36	29.80	−39.60	29.61	7.70
Psychotherapy	−129.00	−71.93	45.66	−47.20	−25.16	−180.16

Pain reduction						
None to very slight	−0.78	−20.59	−104.51	−93.70	−109.22	33.74
Slight	14.81	−41.14	4.84	−36.75	−29.80	19.97
Reduced by half	15.39	26.07	44.25	64.08	46.12	−1.47
No pain	−29.43	35.67	55.42	66.38	92.91	−52.25
Onset of treatment efficacy						
1 month	43.08	14.38	−24.28	40.57	10.89	1.35
6 months	12.57	−7.24	−1.08	−12.09	15.59	−1.43
12 months	−55.65	−7.14	25.36	−28.48	−26.47	0.08
Duration effectiveness						
Effective for 2 months	−24.95	−31.63	−14.28	−44.98	−36.30	55.20
Effective for 6 months	−15.04	11.67	−3.18	10.90	−1.21	−26.97
Effective for 12 months	39.99	19.96	17.46	34.08	37.50	−28.24
Difficulties with daily activities						
As many difficulties as before	−78.14	−8.70	−18.41	−77.50	−52.76	16.43
Fewer difficulties as before	30.38	8.42	−22.07	42.03	28.83	−13.53
No difficulties	47.76	0.28	40.48	35.47	23.92	−2.90
Sleep problems						
As many problems as before	−8.47	−24.80	−27.24	−62.79	−31.52	55.11
Fewer problems than before	32.87	9.40	16.12	22.80	6.96	32.76
No problems	−24.39	15.40	11.12	39.99	24.56	−87.87
Knowledge of his/her body and pain location						
Same knowledge as before	−32.84	−12.15	14.41	−4.46	−14.32	−7.73

Knowledge somewhat better than before	−15.36	−4.90	0.39	−12.32	4.80	41.46
Knowledge much better than before	48.20	17.05	−14.80	16.78	9.52	−33.72
Number of observations	48	57	43	67	159	50
Absolute share (%)	11.32	13.44	10.14	15.80	37.50	11.79
Class share (%)	12.60	14.10	10.60	15.70	34.90	12.00
Observations	424					
McFadden R^2	26.036					
χ^2	2730.48					
Log-likelihood	−3876.09					
Log-likelihood (null)	−5243.68					
AIC	7994.88					
BIC	8764.89					
Conditional relative importance of attributes (%)						
Treatment modality	32.47	66.21	46.23	20.06	31.50	39.95
Pain reduction	6.40	10.97	22.85	22.87	28.87	12.28
Onset of treatment efficacy	14.10	3.09	7.09	9.87	6.01	0.40
Duration effectiveness	9.28	7.37	4.53	11.29	10.54	11.92
Difficulties with daily activities	17.99	2.45	8.94	17.08	11.66	4.28
Sleep problems	8.18	5.74	6.19	14.68	8.01	20.43
Knowledge of his/her body and pain location	11.58	4.17	4.17	4.16	3.41	10.74

Abbreviations: AIC, Akaike Information Criterion; BIC, Bayesian Information Criterion.

a

A value close to zero indicates an absence of preference: the further the value is from zero, the greater the preference.

b

Coefficient of the opt-out option.

With regard to other attributes, the HB model (i.e., preferred model) indicated that respondents were able to wait up to 6 6–12 12 months until their preferred treatment started to be effective and wanted an effectiveness duration of at least 6 6 months. An improvement from ‘fewer difficulties as before’ to ‘no difficulties’ in their daily life activities, as well as an improvement to ‘fewer problems than before’ in their sleep problems were appreciated whereas the attribute ‘knowledge of his/her body and pain location’ had a minor effect, that is, its improvement had to be high to influence the preference.

From these results, we can observe that the treatment modality had a great importance beside the other attributes. All attributes were always ranked at the same position. Among the treatment modality, corticosteroid injections and psychotherapy were generally rejected whereas physical manipulations were preferred followed by the supervised body-mind physical activities and the self-management courses. Supervised sports physical activities had a positive but slight effect, and its weight was smaller in the HB model than in the logit and CL models. Graphical representations of part worth utilities are given in Supporting Information: Appendix 8 and Kernel densities using Epanechnikov kernel function of the HB model' part worth utilities are provided in Supporting Information: Appendix 9 with one-sample multivariate tests of means (H0:μ=0 ${H}_{0}:\mu =0$).

Explaining preference heterogeneity

The LC model estimated preferences that are heterogeneous between classes and homogeneous within classes. It allowed us to examine significant statistical differences (at p <.10) in the sociodemographic characteristics of the six classes (Table 2). Compared to the other classes, class 1 had a higher household income (72,604 $\mathrm{72,604}$ CAD, ±36,796 $\pm \mathrm{36,796}$, p = .042), benefited more from injections of corticosteroid products (62.5%, p <.001), spent more money to treat CLBP (2107 CAD, ±4821, p = .547) and had more other problems affecting quality of life (75%, p = .138). Class 2 benefited less from corticosteroid injections (12.28%, p <.001) and spent less money to treat their CLBP (961 961 CAD, ±1245, p = .547). Class 3 was made up of more men (25.58%, p = .031), although the ratio of men over women remained very unbalanced (1 man for 2.8 women against 1 man for 4.1 women in the full sample), more individuals with pain lasting from 3 months to 1 year at the time of the survey (7%, p = .133), more often benefited from corticosteroid injections (53.49%, p <.001), did less yoga (6.98%, p = .103), presented more diseases or physical or mental problems affecting their quality of life (83.72%, p = .138), and reported having a better self-reported health state (3.81 3.81 over 5 5, ±0.82 ± 0.82, p = .093). Class 4 was more feminine (85.1%, p = .031), less in pain when responding to the survey (5 5 over 10 10, ±2.06 ± 2.06, p = .090) and spent more money to treat CLBP (1906 CAD, ±6679, p = .547). It was also the class with the most cancer (7.46%, p = .021) and respiratory problems reported (17.91%, p = .021). Class 5 was slightly younger (53 53, p = .002) and more educated (p = .046). Finally, class 6 was the oldest (62 62, p = .002), the one with the most of men (34%, p = .031), the least educated (p = .046), more diagnosed with vertebral arthritis or spondylarthrosis (22%, p = .069) and the one being the most in pain when answering the survey (5.86 5.86 over 10 10, ±2.06 ± 2.06, p = .090), but also having the fewest other problems affecting their quality of life (60%, p = .138). Respondent's personal ranking of attributes and treatment modalities are presented in Supporting Information: Appendix 10.

No significant difference was found among classes on the choice certainty score, the difficulty and the quality of responses (Supporting Information: Appendix 11). The feeling of annoyance was almost significant (p = .102), individuals in class 1 being more annoyed compared to other classes (39.13%). Only the number of dimensions/attributes considered by individuals for the choice exercises was significant (p = .004), with classes 1, 3 and 6 who considered less dimensions than the average. Class 6 rejected more than other classes the treatments offered in the choice-based exercise (7.3 7.3 opt-out selected over 11 11 possibilities [note that respondents who refused 12 times were deleted], ±2.91 ± 2.91, p <.001), whereas classes 1 and 5 strongly accepted them (0.73 0.73, ±1.03 ± 1.03 and 0.17 0.17, ±0.41 ± 0.41, respectively). About 13.68% and 34.67% failed to the rationality test and to the temporal consistency test, respectively, and differences between latent classes were significant (p <.001 and p = .003, respectively). Bivariate analyses of the hierarchical clustering derived from the HB model are provided in Supporting Information: Appendix 12.

The bivariate analyses were enhanced by a multinomial logistic regression on the classes (Supporting Information: File 1) with respondent characteristics as arguments. The results indicated that only some variables explained the class memberships. This is an important limitation that is probably due to a very limited number of observations in each class to allow a consistent multivariate analysis.

DISCUSSION

Using a DCE, we assessed patients' preferences for CLBP nonsurgical treatments. Our segmentation results showed a diversity of preferences. Indeed, classes 1, 3 and 6 preferred the corticosteroid injections (class 3 also preferred psychotherapy), class 2 preferred supervised body-mind physical activities and self-management courses and classes 4 and 5 preferred physical manipulations. More generally, patients (as a whole) expressed a disutility for corticosteroid injections and psychotherapy. They all differed in their sociodemographic status, health conditions and in their experiences with CLBP, although in a multivariate analysis this was not a predictor for class membership. These results were somewhat corroborated with a simple question of attributes self-ranking and with the four-item questionnaire of Dima et al.[37] (Table 5). According to these findings, Quebec health authorities should generalize access to physical manipulations, develop body-mind and self-management programmes, and in some specific cases, propose corticosteroid injections and psychotherapies.

Table 5 Four-item Low Back Pain Treatment Beliefs Questionnaire (LBP-TBQ) scores

Four-item LBP-TBQ	Proposed treatment					
	Corticosteroid injections	Supervised body-mind physical activities	Supervised sports physical activities	Physical manipulations	Self-management courses	Psychotherapy
Taking/having this treatment for back pain makes a lot of sense (1–5)						
Mean	3.01	3.89	3.64	4.16	4.18	3.76
Standard deviation	1.32	0.99	1.11	0.96	0.76	0.96
Range	(1–5)	(1–5)	(1–5)	(1–5)	(1–5)	(1–5)
Strongly disagree	19.17%	3.06%	6.11%	2.78%	1.11%	2.22%
Disagree	15.00%	4.72%	8.61%	3.06%	0.56%	6.67%
Neither agree nor disagree	25.28%	21.94%	23.33%	13.06%	13.06%	27.50%
Agree	26.94%	40.28%	38.61%	38.06%	49.44%	40.28%
Strongly agree	13.61%	30.00%	23.33%	43.06%	35.83%	23.33%

I think this treatment is pretty useless for people with back pain (1–5)a						
Mean	2.66	2.44	2.46	2.29	2.33	2.54
Standard deviation	1.29	1.23	1.21	1.30	1.26	1.22
Range	(1–5)	(1–5)	(1–5)	(1–5)	(1–5)	(1–5)
Strongly disagree	23.61%	26.67%	26.39%	37.78%	32.78%	24.44%
Disagree	24.44%	31.39%	29.44%	23.89%	29.72%	26.11%
Neither agree nor disagree	26.11%	19.44%	22.22%	17.78%	16.94%	28.61%
Agree	14.44%	15.83%	15.56%	12.78%	13.33%	12.78%
Strongly agree	11.39%	6.67%	6.39%	7.78%	7.22%	8.06%
I have concerns about taking/having this treatment for my back pain (1–5)a						
Mean	3.33	2.22	2.63	2.19	1.99	2.27
Standard deviation	3.33	2.22	2.63	2.19	1.99	2.27
Range	1.41	1.10	1.29	1.14	0.99	1.12
Strongly disagree	15.56%	30.56%	23.61%	33.61%	36.94%	30.56%
Disagree	15.28%	33.33%	28.33%	32.22%	36.94%	29.72%
Neither agree nor disagree	15.83%	23.33%	18.89%	19.44%	17.78%	26.11%
Agree	27.50%	8.89%	20.00%	10.56%	6.39%	9.17%
Strongly agree	25.83%	3.89%	9.17%	4.17%	1.94%	4.44%

I am confident this treatment would be a suitable treatment for my back pain (1–5)						
Mean	2.93	3.82	3.54	4.03	4.05	3.67
Standard deviation	2.93	3.82	3.54	4.03	4.05	3.67
Range	1.39	1.03	1.20	0.99	0.85	1.07
Strongly disagree	24.72%	4.72%	8.61%	2.78%	1.67%	5.00 %
Disagree	11.39%	5.00%	11.67%	6.11%	2.78%	6.94 %
Neither agree nor disagree	25.83%	20.00%	19.17%	12.78%	15.28%	28.0 6%
Agree	22.78%	44.44%	38.33%	42.50%	49.44%	36.3 9%
Strongly agree	15.28%	25.83%	22.22%	35.83%	30.83%	23.6 1%

a

The lower the score is, the more appreciated the treatment modality. For each treatment, Cronbach's α were equal to .818, .737, .766, .738, .652 and .712, respectively. See Supporting Information: File 2 for details.

We found seven DCEs on LBP treatments conducted in the United States,[14] the United Kingdom,[19,20] Denmark,[15] Sweden,[17] Australia[18] and China.[16] Contrary to these DCEs mostly focussing on a few specific treatments, our study mobilized six different first-line treatments for the management of CLBP. For example, Kløjgaard et al.[15] opposed generic surgical to nonsurgical treatments and Turk et al.[14] only included oral medication and injection as treatment modalities. About three types of exercises were considered by Aboagye et al.[17] and a mix of medication, education and physical therapy were considered by Yi et al.[19] Comparing six different treatments including psychological and various physical therapies was useful for considering the patients' preferences about CLBP treatments, particularly in an academic hospital to help organize the care pathways (e.g., to identify imbalance in resource requirements, potential breakdown of services). In addition, beyond preferences, differences in accessibility and/or affordability for some treatments may remain, particularly surgical ones, and such a study may help healthcare professionals and managers to better consider the balance between the needs of patients and what can be offered to them. While patients granted the highest conditional relative importance to treatments and manifested a high preference for physical manipulations and body-mind physical activities in our study, these types of treatment were not significantly retained by the participants in the study of Yi et al.[19] However, they were the second most important attributes in the study of Aboagye et al.[17] Chen et al.[16] and Kløjgaard et al.[15] were the only studies to examine the effectiveness duration and treatment effect on daily activities and found that these attributes were significantly retained by participants, as in our study. Both also found that the effectiveness of pain relief was a major attribute.

Differences between the six classes from the LC model could explain some preferences for the treatment choices, in particular why classes 1, 3 and 6 expressed a strong preference for corticosteroid injections. Including heterogeneity in the analysis, the LC model allowed us to consider heterogeneity between classes. We described six classes and

found relevant discriminant preferences and sociodemographic characteristics. Ferreira et al.[18] and Walsh et al.[20] also derived different classes (n=4 $n=4$) from an LC model with their own preferences and sociodemographic characteristics. For instance, the primer found that the elderly and men with lower socioeconomic status preferred exercise less whereas all other classes preferred this programme. Individuals already doing exercise also had a preference for a programme with higher frequency and lower cost. Walsh et al. found that improvement in symptom control and risk of physical dependency were of utmost importance in patients suffering from osteoarthritis and CLPB and defined four distinct groups as 'efficacy-focused', 'cost-averse', 'physical-dependence-averse' and 'needle-averse'. Yi et al.,[19] Kløjgaard et al.[15] and Turk et al.[14] also performed subgroups' analyses. They found the following, respectively: (1) significant differences between pain grades and preferences for the content (i.e., education, physical therapy, medicines and coping with pain) and for the type of provider of the programme (i.e., nurse, physiotherapist, general practitioner and psychologist), (2) some differences between sociodemographic characteristics and subgroups (i.e., age, gender, surgery experiences) and (3) few significant differences between the respondent condition (i.e., osteoarthritis, CLBP or both), the opioids use, the painful experiences or the locus of control questions on the MHLC Scale-Form C. However, all studies had different aims, with their own attributes and levels. Thus, the comparisons with common attributes may not be so relevant.

The biggest strength of our study was the number of treatment modalities, which allowed us to consider a variety of preferences. Indeed, this is the only study that considered six nonsurgical CLBP treatments. Moreover, preferences were estimated via three models and attributes were mostly coherent in their levels, when there was gradation. Also, almost 70% of the total number of respondents was included in the analysis showing the attractivity and quality of the survey. Individuals with longer CLBP were more willing to answer with 98.11% of respondents suffering for more than 1 year. Another strength of the study is that we followed a mixed method with a qualitative phase for the design of the DCE.[13,38] In addition, the survey was administered online and made it possible to relieve the social desirability bias.[43] Nevertheless, an online survey may create a self-selection bias, targeting only a certain population who have an interest and capacities and access to information technologies to answer the survey. However, this bias may be limited by the fact that 93%of Quebec households have access to the internet.[44] Another limitation referred to the DCE design. Using seven attributes may lead to a cognitive burden and cause confusion in individuals while making their choices. Indeed, it has been shown that increasing the number of attributes increases the likelihood of self-simplifying the exercise by heuristics.[45] However, the number of attributes in this study is in line with other studies.[14–19] In addition, since a very simple description of the attributes, was provided (Table 1), it could induce a lack of comprehension from the participants if not carefully considered. Yet, participants were part of an association dealing with chronic pain. Almost all were suffering from CLBP for more than 1 year, used painkillers, and had substantial experiences with treatments (Table 2). We are thus confident that participants were knowledgeable about the treatments. Also to note that as Ferreira et al.[18] discussed in the case of exercise programmes, it must be emphasized that considering the patient's preferences, he/she could prefer a less effective treatment because of his/her own socioeconomic characteristics or because of the treatment characteristics (e.g., price, risk). This underlines the importance of having a dialogue between practitioner and patient, and the need to discuss barriers to treatments (e.g., price, availability). The discrepancies identified in the LC model, particularly in class 6, could be explained by many factors and we highlighted two of them: (1) they chose the most to refuse one or the other treatment (7.30 7.30 over 11 11, ±2.91 ± 2.91), which could lead to estimation issues, and (2) they reported higher CLBP according to the NPRS, higher frequency of use of painkillers, as well as a low satisfaction with life, which could induce a poor understanding about the choice-based exercise although it was not reflected by the follow-up questions (Appendix 11). Even so, we must be careful about such interpretations. Another limitation of this study is related to the choice of the software for analysis, which allows less flexibility in the methods used as compared to other software.

Finally, DCEs are in line with patient-centred health care because they allow for consideration of patients' preferences. Socioeconomic background, illness experiences, the lifestyle of the patient and his/her preferences are now part of the care pathway. Knowing these preferences can be useful for decision-makers to tailor care delivery to

the needs of patients. Shared-decision making improves patient adherence, involvement and satisfaction, and so, treatments' outcomes.[12,46] DCEs and, more generally, stated preference methods are a way to understand and measure patients' preferences and to ensure that they are well embedded in healthcare decision-making.[47] The study allowed us to determine the preferences of patients for CLBP nonsurgical treatments whereas only the surgery and corticosteroid injections were available in our healthcare institution at the time of the survey. These results may be used by managers and clinicians as a lever to better address patients' needs by offering them a wider variety of treatments through a reallocation of funds. They can also serve physicians and practitioners by improving information in monitoring patients for their needs and choices. In our local context, this led to nonsystematically offering corticosteroid injections and discussing other options with patients. It also led to a discussion for prioritization of resources to offer the preferred nonsurgical options assessed in this study and finally to a reorganization of the patient's care pathway.

CONCLUSION

We assessed the preferences of patients suffering from CLBP for six different nonsurgical treatments. We drew different patients' profiles and highlighted several differences between preferences and sociodemographic characteristics. As such, there is no one size fits all approach and we should consider the singularity of each patient, even if some common patterns can be found. For example, physical manipulations were highly preferred by most while corticosteroid injections were strongly rejected by a large minority, thus impacting the general results found in the HB model. Because of the heterogeneity and the complexity of behaviours towards choices for treatments, patients' preferences about treatments need a clearer understanding, especially for those facing CLBP which is a growing condition in developed countries with an ageing population. An array of different CLBP treatments exists but most of them are still understudied, unavailable or underused. We hope that it can help decision-makers for organizing care access and delivery and at an individual level, to informing practitioners' sharing decisions with their patients for choosing what treatment best suits them for CLBP.

ACKNOWLEDGEMENTS

We thank the staff of the Association Québécoise de la Douleur Chronique (AQDC) for their support and all respondents who participated in the survey and preparatory steps. We acknowledge the participation of Marion Beffarat and Ginette Ladouceur in the focus groups that were held in the first step of this study. Thomas G. Poder is a former employee of the CIUSSS de l'Estrie—CHUS and is actually a member of the FRQS-funded Centre de recherche de l'IUSMM. This work was funded by internal funds from the HTA unit of the CIUSSS de l'Estrie—CHUS.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data will be made available upon reasonable request to the corresponding author.

ETHICS STATEMENT

This study was approved by our university hospital ethics committee (CIUSSS de l'Estrie—CHUS) and all participants gave their informed consent before starting the survey. All responses were anonymous.

DETAILS

Subject:	Patient-centered care; Chronic low back pain; Patients; Sports; Psychotherapy; Selfmanagement; Questionnaires; Low back pain; Chronic back pain; Personal health; Efficacy; Pain; Health services; Sleep; Heterogeneity; Acupuncture; Bayesian analysis; Back pain; Sleep disorders; Focus groups; Chronic pain; Effectiveness; Activities of daily living; Mind and body; Public health; Preferences; Logit models; Attributes; Literature reviews; Sleep problems; Treatment preferences; Discrete choice

Location:	Canada; Quebec Canada
Identifier / keyword:	chronic low back pain; discrete choice experiment; preferences
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	510-530
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-12-08
Milestone dates:	2022-11-23 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-05-28 (manuscriptReceived); 2022-12-08 (publishedOnlineEarlyUnpaginated); 2022-11-25 (manuscriptAccepted)
Publication history :	
First posting date:	08 Dec 2022
DOI:	https://doi.org/10.1111/hex.13685
ProQuest document ID:	2767101931

Incorporation of the emotional indicators of the patient journey into healthcare organization management

Rodríguez-Fuertes, Alfredo [1] ; Reinares-Lara, Pedro [1] ; Garcia-Henche, Blanca [2] [1] Department of Business Economics, Universidad Rey Juan Carlos, Madrid, Spain [2] Department of Economy and Business Management, Universidad de Alcalá, Madrid, Spain

ABSTRACT (ENGLISH)

Background

In recent years, attempts have been made to incorporate patients' experiences into healthcare processes, to complement clinical indicators, with what are known as patient-reported outcome measures (PROMs) and patient-reported experience measures (PREMs). While the research into PROMs is more developed, the application of PREMs faces some difficulties. The incorporation of emotional indicators into assessments of the experience is an area that remains to be explored.

Objectives

This study proposes a new technique to analyse the emotions experienced by patients during the care process, examines how these emotions influence their satisfaction and propose that if healthcare services focus more on patients' emotions, they can improve the effectiveness of the sector.

Methods

The first, qualitative stage, gathered data from patients to design a patient journey (PJ). The PJ was then reproduced as a video. In a subsequent, quantitative stage, the video was shown to experimental participants, and their emotions were measured through facial expression analysis and a questionnaire.

Results

A new technique to gather emotional data showed that the emotions patients experience do not affect their satisfaction with their clinical care or the physical aspects of the process. However, their emotions did affect their satisfaction with people and organizations.

Conclusions

The importance of the emotional component of patients' experiences was underlined. Therefore, healthcare

organizations should take account of this dimension, as well as the cognitive, to increase patient satisfaction and improve their care processes. Understanding the impact of the emotions identified at the subconscious level can help improve the patient experience. A new methodology was applied that may help health professionals to collect emotional data about patients' experiences and to develop PREMs.

Patient/Public Contribution

Patients were involved in all stages of this research. In the exploratory phase, some helped define the touchpoints of the PJ. The data from the subsequent experimental phase were collected from another group, and the emotions they experienced were identified through the analysis of their facial expressions. Based on the results of this study, a working group including patients has been established to work on improvements in the PJ.

FULL TEXT

INTRODUCTION

In recent years, there has been an evolution in the management of healthcare organizations, which are now less focused on their professionals, and are more focused on their users and their expectations. This evolution has seen the development of the concept of patient-centred care. This entails considering both the physical and emotional needs of patients.[1] In this move from a focus on internal organizational aspects towards a market orientation, and faced with growing demand for healthcare services, an increased understanding of patients' experiences and emotions can improve how services are delivered and increase customer satisfaction. Various authors have emphasized the importance of listening, understanding and learning from the patient experience.[2,3]

In consequence, attempts have been made to develop indicators (other than clinical results) that can allow healthcare managers to incorporate the patient's perspective; thus, patient-reported outcome measures (PROMs) and patient-reported experience measures (PREMs) have been introduced. While PROMs are more advanced and are supported by scientifically validated questionnaires, the development of PREMs lags behind and faces challenges such as the difficulty of collecting data on individuals' subjective experiences.[4]

Healthcare providers seek to achieve high levels of patient satisfaction,[5] one of the most frequently applied healthcare quality indicators. However, although satisfaction is a widely employed concept, there is no consensus on its nature, or on its evaluation.[6] One of the most accepted theories on the conceptualization of satisfaction proposes that it is a psychological state that can be represented in a double cognitive-affective dimension,[7,8] as shown in Figure 1.

Figure 1. Dimensions of satisfaction

The first dimension is cognitive satisfaction, that is, where patients assess the positive and negative aspects of the different components of a service, either by evaluating the perceived outcomes in isolation or by measuring them against a standard or preformed expectations.[6,8] Cognitive satisfaction with experiences has traditionally been measured through satisfaction questionnaires. The second dimension is the patient's affective evaluation of the experience, which takes into account subjective elements, and captures the emotions generated in the patient by the patient–organization relationship.[9] With this premise, various studies have demonstrated that, for an objective analysis of satisfaction, account must be taken of both cognitive and affective reactions, since they are different and independently influence the formation and explanation of satisfaction (e.g., Liljander and Strandvik[7]).

Maria Ugolini et al.[10] highlighted the importance of identifying the emotions that patients experience during the care process. Patients can be in intense emotional states when visiting their doctors. The recognition of, and response to, patients' emotions is related to important healthcare outcomes, including patient satisfaction with the process and adherence to treatment.[5] Thus, an important aspect of patient-centred care is the effective recognition of the emotions they go through during medical processes.[11]

Although healthcare organizations have been seen to be increasingly active in improving the emotional experiences of their patients, they still lack a specific approach to addressing these emotions to increase satisfaction levels. Traditionally, healthcare organizations have focused on evaluating the cognitive aspect of satisfaction, and ignored the emotions generated during the care process.[12] Altringer[13] highlighted the need for further research into the

patient's emotional experience during the care process. Arguably, there is no other service setting in which emotions are more important than in health care. Understanding and managing emotions during the service experience is an important area of research because emotions influence customer perceptions, future intentions and behaviours.[14] Experience-based design is a method used to capture the emotional content of patient healthcare experiences and can serve as the foundation for patient-centred healthcare. As previously noted, an important aspect of patient-centred care is the effective recognition of the emotions evoked in patients during the healthcare process. Helena Vinagre and Neves[15] showed just how important positive emotions are for patient satisfaction. Emotions have been shown to be highly predictive in consumer satisfaction models; it has been demonstrated that satisfaction can be influenced by positive and negative emotions. Positive patient experiences have a positive effect on clinical outcomes and cost-effectiveness. Surveys are commonly used to evaluate satisfaction with care experiences.[9,16] Nonetheless, while surveys are valid tools for measuring the cognitive component of satisfaction, they have some important limitations. First, surveys have difficulty in assessing the affective component of satisfaction due to the complexity of the respondents' emotions, and in predicting future behaviours[6,17] - there is evidence that emotions better predict behavioural intentions than do cognitive measures. Second, their inability to evaluate the complete experience, since they generally assess user satisfaction only at one specific moment, although emotions are experienced before, during and after service delivery.[16] Qualitative research techniques have sometimes been used in combination with quantitative techniques to identify the key determinants of the quality of health services.[7,18,19] Based on this background, the following study objectives are proposed:

- To validate a methodology that evaluates the affective dimension of patient satisfaction during the care process/patient journey (PJ) and collects data for the development of PREMs.

- To confirm if a relationship exists between the emotions recorded during the PJ and satisfaction measured through a questionnaire.

- To develop proposals for actions to improve care processes in healthcare establishments.

METHODS

The present study, undertaken sequentially, used various methods validated in previous studies, to ensure the scientificity of the results.[20] First, an exploratory phase was carried out using qualitative techniques. Second, an experiment collected neurophysiological data, which was complemented by data collected through a questionnaire (Figure 2). The main methodological novelty was combining traditional data-gathering techniques with the observation of motor behaviour through facial expression analysis (FEA).[21,22]

Figure 2. Experimental schema

In the exploratory phase, the general objective was to identify the emotions generated in patients during a healthcare experience, that is, the PJ, in one of the surgeries most frequently carried out in the Spanish health system, that of hospital-based inguinal hernia repair. The same process was subsequently examined in the main experiment. The term ‘patient journey’ here refers to ‘the processual and experiential aspects of service processes as seen from the customer viewpoint’.[23] The PJ journey is here represented graphically as a sequence of patient movements through the care process, showing the interactions between the patient and the various other agents. In addition, this qualitative phase sought to identify patients’ perspectives about the quality of, and their satisfaction with, the experience. In this qualitative stage of the research, the participation of the patients and their companions was essential. First, they described, from their perspective, how the PJ developed, and they helped identify the key

points/moments in this particular healthcare experience, and their significance for patients; and, second, they helped the researchers establish a standardized PJ.

Focus group and in-depth interview techniques were employed in two stages; these had different objectives:

•1.
To achieve a good understanding of the most important elements of the PJ by defining the most important touchpoints in the care process. Specifically, the profile of the people who participated in this stage was as follows:

•a.
Three focus groups, with a total of 21 participants:

•1.
Groups 1 and 2: The participants had to have been hospitalized (or had accompanied a hospitalized person) in the previous 3 months.

•2.
Group 3: the participants had to have attended a specialized care consultation in the previous 3 months.

•b.
Four in-depth interviews with patients/companions:

•1.
Three patients who had been hospitalized in the previous 3 months.

•2.
A companion of a person who had been hospitalized in the previous 3 months.

•c.
Eight in-depth interviews were conducted with healthcare professionals and experts in healthcare quality (as carried out in other types of research[24]) to understand the care protocols.

•2.
Individual in-depth interviews were conducted with two patients who had undergone hernia surgery in the previous 2 years to review the defined PJ and reach a consensus on the PJ that would be used in the experimental phase.

The guides used in the qualitative techniques were developed based on the objectives established for the exploratory phase of the research. The general objective was to identify the emotions generated in the patients during the healthcare experience. Other objectives were to identify the touchpoints between the patient and the health service provider and to follow the PJ of someone undergoing inguinal hernia surgery.

The objectives of the experimental phase were to obtain quantitative data to identify the emotions experienced by patients during the PJ and to establish their influence on satisfaction. It should be noted that the experiment did not focus only on major phases of the patient experience, rather it examined the complete process, that is, the entire PJ through the different levels of care.

The PJ was recorded on video and tries to show the whole process that the patient goes through, that is, from the appearance of the relevant symptoms until his/her discharge from the hospital. The methodology of collecting facial expression data from participants while they watch videos has previously been used in the analysis of care service processes.[25,26]

The videos were recorded from the patient's perspective. First-person sequences help immerse the viewer in the scene. So, in the videos, the professionals addressed themselves directly to the camera, as if the patient were in front of them. This means that no patient is featured in the videos.

The recording was divided into the three phases of the patient's experience at each healthcare level (primary care, specialized care and hospitalization) based on the touchpoints previously identified. The division of the process into three independent phases is valid as different types of emotions are identified in each phase (as was verified in the exploratory phase).

The three phases were as follows:

•1.

Phase 1: Primary care (PC): Prediagnosis.

•When the patient develops his/her first symptoms and requests an appointment and the first consultation. This took place in the patient's home and in the primary care centre (consultation and appointment area). The participants were the patient and the PC physician.

DETAILS

Subject:	Patients; Patient-centered care; Organizations; Health care; Questionnaires; Indicators; Emotions; Health services; Objectives; Health care industry; Facial expressions; Patient satisfaction; Hospitalization; Qualitative analysis; Clinical indicators; Medical personnel; Experiments; Hernias; Cognitive ability; Qualitative research
Identifier / keyword:	emotions; facial expressions analysis; patient experience; patient journey; PREMs
Publication title:	Health Ex pectations; Oxford
Volume:	26
Issue:	1
Pages:	297-306
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety

ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-06
Milestone dates:	2022-10-19 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-04-16 (manuscriptReceived); 2022-11-06 (publishedOnlineEarlyUnpaginated); 2022-10-23 (manuscriptAccepted)
Publication history :	
First posting date:	06 Nov 2022
DOI:	https://doi.org/10.1111/hex.13656
ProQuest document ID:	2767101895
Document URL:	https://www.proquest.com/scholarly-journals/incorporation-emotional-indicators-patient/docview/2767101895/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the “License”). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-09-21
Database:	Public Health Database,Publicly Available Content Database

Patient views on asthma diagnosis and how a clinical decision support system could help: A qualitative study

Canny, Anne [1] ; Donaghy, Eddie [1] ; Murray, Victoria [1] ; Campbell, Leo [1] ; Stonham, Carol [2] ; Bush, Andrew [3] ; McKinstry, Brian [4] ; Milne, Heather [5] ; Pinnock, Hilary [1] ; Daines, Luke [1]

[1] Asthma UK Centre for Applied Research, Usher Institute, University of Edinburgh, Edinburgh, UK [2] NHS Gloucestershire Clinical Commissioning Group, Gloucester, UK; Primary Care Respiratory Society (PCRS), Knowle, UK [3] Imperial Centre for Paediatrics and Child Health and National Heart and Lung Institute, Imperial College, London, UK; Department of Paediatric Respiratory Medicine, Royal Brompton Hospital, London, UK [4] Centre for Population and Health Sciences, Usher Institute, University of Edinburgh, Edinburgh, UK [5] South East GP Unit, NHS Education for Scotland, Edinburgh, UK

ABSTRACT (ENGLISH)

Introduction

Making a diagnosis of asthma can be challenging for clinicians and patients. A clinical decision support system (CDSS) for use in primary care including a patient-facing mode, could change how information is shared between patients and healthcare professionals and improve the diagnostic process.

Methods

Participants diagnosed with asthma within the last 5 years were recruited from general practices across four UK regions. In-depth interviews were used to explore patient experiences relating to their asthma diagnosis and to understand how a CDSS could be used to improve the diagnostic process for patients. Interviews were audio recorded, transcribed verbatim and analysed using a thematic approach.

Results

Seventeen participants (12 female) undertook interviews, including 14 individuals and 3 parents of children with asthma. Being diagnosed with asthma was generally considered an uncertain process. Participants felt a lack of consultation time and poor communication affected their understanding of asthma and what to expect. Had the nature of asthma and the steps required to make a diagnosis been explained more clearly, patients felt their understanding and engagement in asthma self-management could have been improved. Participants considered that a CDSS could provide resources to support the diagnostic process, prompt dialogue, aid understanding and support shared decision-making.

Conclusion

Undergoing an asthma diagnosis was uncertain for patients if their ideas and concerns were not addressed by clinicians and were influenced by a lack of consultation time and limitations in communication. An asthma diagnosis CDSS could provide structure and an interface to prompt dialogue, provide visuals about asthma to aid understanding and encourage patient involvement.

Patient and Public Contribution

Prespecified semistructured interview topic guides (young person and adult versions) were developed by the research team and piloted with members of the Asthma UK Centre for Applied Research Patient and Public Involvement (PPI) group. Findings were regularly discussed within the research group and with PPI colleagues to aid the interpretation of data.

FULL TEXT

INTRODUCTION

Asthma is a chronic respiratory disease accounting for at least 6.4 million primary care consultations each year in the United Kingdom.[1] Although a common condition, making a diagnosis of asthma is not always straightforward for clinicians and estimates from primary care suggest that asthma is often misdiagnosed.[2–4] Overdiagnosis can lead to costly, potentially harmful treatment and may affect job and lifestyle decisions; whilst underdiagnosis risks inadequate treatment and avoidable morbidity and mortality.[5,6]

Asthma is a variable condition with different phenotypes meaning that individuals with asthma can experience and present with a range of symptoms of varying severity.[7] The aim is to demonstrate objective evidence of variability

over time, and investigations such as spirometry and fractional exhaled nitric oxide (FeNO) can increase or decrease the likelihood of a diagnosis of asthma. However, in primary care, timely access to tests is not guaranteed and false positive and false negative results are common.[5] Consequently, it may take months before a clinician feels able to confirm (or refute) a diagnosis of asthma.[8] The potentially long timescale can lead to frustration and uncertainty amongst patients[9] and needs to be handled confidently and accurately by clinicians.[10]

In addition to increasing the availability and use of objective tests,[11] clinical decision support systems (CDSS) could provide a solution for improving the accuracy of an asthma diagnosis and may help improve the patient experience. CDSS are usually designed to aid the decision-making of clinicians[12] but may be utilized to facilitate shared decision-making with patients. For instance, CDSS can be used to collect, calculate and present information about the likelihood of a diagnosis or treatment benefit. For example, AsthmaTuner, a self-management system which collects lung function and symptom data via Bluetooth spirometer and patient app, respectively, provides automated treatment recommendations for patients and an interface for health professionals.[13] For diagnosis, a CDSS could be used to calculate the likelihood of a particular condition, and present the probability and options for confirming a diagnosis. In a Norwegian study, a web-based CDSS designed to aid the diagnosis and classification of chronic obstructive pulmonary disease (COPD) in primary care was found to reduce misdiagnosis and increase the number of patients receiving smoking cessation advice but did not improve the prescription of pharmacological treatment.[14]

Having derived and validated a clinical prediction model for asthma diagnosis,[15,16] we plan to implement the model in primary care as a CDSS. Being aware that a previous systematic review found that CDSS for asthma were infrequently utilized,[17] we wanted to understand patient views on asthma diagnosis and how a CDSS could help to maximize the potential value of a future CDSS for patients. Therefore, to inform the development of the CDSS, this study aimed to explore patient experiences regarding asthma diagnosis and to understand how a CDSS could also be used to improve the diagnostic process for patients.

METHODS

To inform the development of an intervention (the CDSS), the study design was guided by the Medical Research Council (MRC) framework for developing and evaluating complex interventions.[18] To understand which design features would be most important for an asthma diagnosis CDSS, the experiences and views of patients (and parents of children) who had undergone an asthma diagnosis were sought using qualitative methods. We undertook interviews with young people, adults and the parents of children, who had a recent diagnosis of asthma (ideally within the past 5 years). Interviews took place between 1 October 2020 and 31 January 2021. All participants provided informed consent before interviews were conducted.

Recruitment and sampling

Participants were recruited from general practices across four regions within the United Kingdom (Greater Glasgow and Clyde, Lothian, West Midlands, Yorkshire and Humber). Participating practices identified adults and young people (≥12 years) and parents of children (≥5 years) who had ‘active asthma’ and a diagnosis of asthma coded in the electronic health records within the last 5 years. ‘Active asthma’ was defined as a coded diagnosis of asthma and having had a prescription for any asthma treatment within the previous year.[19] Children below 5 years of age were excluded as viral-associated wheeze is common in this age group and can complicate asthma diagnosis. Based on the age of legal consent, we offered young people aged ≥12 years the chance to take part in an interview themselves. A clinician from each practice screened the list of selected patients for eligibility and excluded individuals who had COPD, were unable to give informed consent (e.g., due to cognitive impairment) or for social/clinical reasons (e.g., significant co-morbidity, recently bereaved or on a palliative care register).

Potential participants were mailed an information sheet and an expression of interest form which included questions about age, gender, age at diagnosis, how asthma was confirmed (i.e., symptoms, examination, tests) and the confidence they had in their asthma diagnosis (agree, not sure, do not agree with diagnosis). Responses to these questions were used to purposively sample individuals to represent a range of participants in terms of age, gender, length of time since asthma diagnosis, confidence in the diagnosis and who made the diagnosis.

Data collection

Semistructured in-depth interviews were conducted by telephone (to comply with social distancing during the COVID-19 pandemic). Interviews were conducted by a male (E. D.) or female (V. M.) postdoctoral researchers, both of whom have experience in health services research. Interviews lasted between 30 and 45 min and were audio recorded, transcribed verbatim and redacted of identifiable information. No repeat interviews were necessary. Transcripts were not returned to participants for comment.

Topic guides

Prespecified semistructured interview topic guides (young person and adult versions) were developed by the research team (see Supporting Information). To consider the validity and reliability of the topic guides, we conducted pilot interviews with members of the Asthma UK Centre for Applied Research Patient and Public Involvement (PPI) group. Topic guides mapped to the study objectives and were designed to allow a focused yet flexible approach[20] that facilitated exploration of experiences of asthma diagnosis; perceptions and expectations of patient involvement in the diagnostic process and how a computer system/CDSS could have helped or hindered their experiences.

Data analysis

We used a thematic approach to data analysis.[21] Using Nvivo 10 (QSR International), transcripts were read and manually coded using overarching themes. In an attempt to maximize reliability, after the initial transcripts had been coded, three researchers (V. M., L. D., H. P.) conducted a thematic analysis with selected transcripts during this iterative process. Emerging themes were discussed before deciding on an initial coding framework. Transcripts were coded on an ongoing basis concurrently with interviews and revisited as the study progressed so that new themes could be included, and the coding framework refined. The final coding framework was thus a combination of themes proposed in advance together with other themes generated during the analysis,[21] which represents both a deductive and inductive approach to qualitative analysis.[22] The consolidated criteria for reporting qualitative research was used to guide reporting (see Supporting Information).[23]

Interpretation

We took a critical-realist perspective when interpreting the data,[24] which helped when considering the experiences, motivations and meanings of participants' lived realities.[21] To aid interpretation, findings were regularly discussed within the research group and with PPI colleagues. The concept of medical dominance emerged as relevant, and we used this to guide interpretation.[25,26] Medical dominance is based on the view that in relation to health and illness, medical professionals hold power and control which shape and influence health professional/patient interactions and experiences.[27–29]

RESULTS

We received 53 expressions of interest within the study period and using purposive sampling, 27 individuals were invited to take part. 17 participants contributed to interviews, including 14 individuals with asthma and 3 parents of children with asthma (Table 1). All participants had been diagnosed with asthma before the COVID-19 pandemic. Of the 26 individuals not invited, 23 had been diagnosed several years before and 3 children had been diagnosed before 5 years of age.

Table 1 Patient demographics

Characteristics	Participant (*n*)
Sex	
Female	12
Male	5
Age (years)	

<16	3
16–30	2
31–40	3
41–50	5
51–60	2
61–70	2
Years since diagnosis (years)	
<2	3
2–4	7
4–6	1
6–8	1
8–10	3
>10	2
Diagnosis made by	
Asthma nurse	3
General practitioner	10
Hospital staff	4
Confidence about their asthma diagnosis?	
Yes	12
Not sure	5
Parents of children interviewed	3
Location (type, practice size)	
Site 1 (urban, 10,361)	5

Site 2 (urban, 6576)	4
Site 3 (urban, 5465)	1
Site 4 (urban, 10,123)	4
Site 5 (semiurban, 12,578)	2
Site 6 (urban, 3851)	1

Overview of themes

Analysis of data sought to answer two key research questions: patient experiences during an asthma diagnosis and patient views and experiences of a CDSS. Four subthemes were identified regarding patient experiences during the diagnostic process; knowledge and understanding of asthma, communication, receiving and retaining information and self-management. An additional four themes emerged in relation to patient experiences and views of a CDSS; patient experiences of screen sharing, online health information use, patient views on an asthma CDSS and barriers and facilitators to a CDSS being used. Topics are reported in this order.

Key theme 1: Diagnosis: The patient experienceKnowledge and understanding of asthma

Several participants recalled being uncertain about what asthma was or how it might present. For instance, participant adult/1 (female, age 41–50, site 1), recalled that she ‘didn't know that coughing was a sign of asthma’, despite having a sister and a best friend who had been diagnosed with the condition as teenagers. Other participants believed asthma always started in childhood:

It was a bit weird 'cause I'd never had it before and [obviously it] was …like, I thought it was quite late. I thought it was one of those things you just had as a kid and then, like, you had it from the beginning and that was that. (P/young person/1, female, 16–30, site 5)

Participants often had their own ideas about the cause of symptoms, and without prior knowledge or experience of asthma, some individuals worried about what they viewed as the worst-case scenario such as cancer:

Somehow you associate it (asthma) with really sick people. I don't know. I didn't sort of think of it as a kind of a manageable issue. Sort of, these people who had maybe asbestos poisoning to their lungs or something like that. A very dramatic thing. (P/adult/6, male, 41–50, site 1)

In a similar vein, some participants, held a lack of familiarity with asthma symptoms leading them to assume their symptoms were a consequence of lifestyle choices or personal stresses so achieving a diagnosis was a relief:

We bought a house which we then discovered had a lot of hidden mould issues and I think that's been a contributor to all of this [….] The asthma diagnosis really helped. (P/adult/6, male, 41–50, site 1)

For some participants, an asthma diagnosis came as a surprise and was made co-incidentally during an appointment for another problem:

I was actually diagnosed accidentally, but I was glad I was diagnosed at the time. I was actually meant to go about my toe because I was arguing with my son when he was about five years old and I got my toe jammed under the door and it was bruised. So anyway, I went to the doctor about it and he noticed that I was a bit wheezy so he decided that he would do a test. And he turned round and said, yeah, you're asthmatic. (P/adult/4, female, 41–50, site 2)

Communication

The importance of communication during the assessment for an asthma diagnosis was a common theme arising from interviews. Some participants were surprised that being diagnosed with asthma had taken a long time, and another participant remained unsure if they had asthma:

Cause all along they're like, oh there's no official test so this might not be, so you'll just need to try this and try that and see if it works or not. So, it's quite a …like, unsure and quite a long process sometimes. (P/young person/1,

female, 16–30, site 5)

There was never any concrete diagnosis, so I don't know whether I have a pre-existing condition now or not. (P/adult/9, male, 41–50, site 4)

However, many patients were satisfied with the step-by-step processes they experienced and the principles of parsimony by problem-solving through the simplest means available to enable an accurate diagnosis:

‘It took a while. It was sort of an ongoing thing over …well, I'd had sort of recurring colds and kind of persistent things like that, with kind of wheezing and coughing and so on. And […] my GP, I think was quite methodical about this so there were various tests and eliminations and so on. So, it had come over a couple of years’. Yeah, but I think it's the right way to do it. I was very happy with it. (P/adult/6, male, 41–50, site 1)

A lack of communication about the variable nature of asthma during the early stages of the diagnostic process left some respondents unsure if they had asthma or not. P/young person/2, reported feeling ‘a wee bit shocked’ when diagnosed with asthma because she considered herself to be ‘the fittest I'd ever been’. Participant adult/8 (male, 61–70, site 3) was told he had asthma but said ‘it wasn't explained how my lungs work or anything’. Participant young person/2, felt disappointed with the support she had received, explaining she had been ‘just told and then left with that information’:

You sometimes feel that people are just giving you a decision but not explaining it in enough detail. [….] Even like when my mum's been there with me, it's just been, kind of […] like none of us have fully understood how I have asthma. (P/young person/2, female, 16–30, site 2)

A perceived lack of time and the use of complex language were reasons participants gave for communication being poor. Participants generally understood why time may be an issue. For instance, P/adult/6 (male, 41–50, site 1), who had been diagnosed for 1 year, felt that ‘the system is congested’. Similarly, P/adult/2 (adult, female, 21–30, site 1), talked about General Practitioners (GP) as being ‘obviously very busy’ and her GP did not have time to explain the diagnostic process or use language that the patient understood. Consequently, she preferred to see the asthma nurse who provided ‘more of an understanding about what's going to happen’:

I think the thing about the nurses are …especially the asthma nurses, they, kind of, already […] I mean, they only see you once a year but they know you a bit better so they can, kind of, explain things in a bit more user-friendly way, I guess. (P/adult/2, female, 21–30, site 1)

Parents of children undergoing the diagnostic process felt communication was particularly problematic, and reported feeling frustrated and helpless during a lengthy and often inconclusive testing process:

You know, I was really annoyed because, you know, like every second week she was ill and [had a] high temperature. And like even, you know, we went to emergency hospital appointment, and nobody could say nothing. And I thought because she was …she has a twin sister, and they were born two weeks before due. Yeah, and […], another problem, like, because she is the second twin, she's the youngest one. And when she had cough and the doctor said, because (twin 1) has asthma, (twin 2) probably has asthma as well. And for me it's like, she never had a test. (P/parent of child/2, female, age 9, site 2)

Receiving and retaining information

Whilst one participant (P/adult/4, female, 41–50, site 2) reported her diagnosis was explained to her satisfaction and she remembered everything she was told, most respondents reported leaving their appointment(s) with little information or not being able to recall the information later. For some respondents, much of the asthma information received was new to them and felt overwhelming. One individual felt relief at finally having a label for their condition. Others suggested they had appeased their GP by pretending to follow the conversation:

It takes me sometimes a while to cotton on to things. And I may say, ‘yes, I understand’ at first. I suppose I'm fairly typical of most people in that way. I say ‘yes, I understand’ but I don't think I've got a full grasp of it, you know? (P/adult/3, male, 61–70, site 1)

The provision of paper handouts to support asthma information during consultations was useful for some, met with indifference by others, and received poorly by one or two participants. Handouts were unlikely to be kept and the information on them was not well retained:

I probably got a leaflet or something like that, that had two or three pages in it, then, and then, well, a leaflet, you put it down and then it disappears. (P/adult/7, female, 51–60, site 1)

I'm a digital person. I hate bits of paper, 'cause I lose bits of paper. (P/adult/10, male, 61–70, site 4)

Self-management

In keeping with the perceived lack of information provided at the time of diagnosis, some individuals felt underconfident in managing their asthma after they had been diagnosed, for example, taking their inhaler correctly:

The only problem, if it could be said to be a problem, was I didn't know how to use the inhalers correctly. I don't believe, I don't remember being told how to use an inhaler. (P/adult/3, male, 61–70, site 1)

Although some participants talked about their personal asthma action plan (a key component of asthma self-management),[5] several respondents said they had not been provided with one, and others did not know what they were:

I know everybody talks about their asthma plan, but mine is not like …I've not got any asthma plan written down, but I mean […] I know myself and I've got an oximeter in the house as well now that I will test on these various things. (P/adult/13, female, 41–50 site 4)

Key theme 2: CDSS: Patient experience and views

Participants spoke about a range of topics relating to how a computer, the internet or a CDSS could be used to enhance a consultation for a possible asthma diagnosis. Four subthemes were identified: patient experiences of screen sharing, online information use, patient views of an asthma CDSS and barriers and facilitators to CDSS use.

Patient experiences of screen sharing

Respondents were asked about their experiences of using the screen alongside their clinician during appointments.[30] One or two participants talked about screen sharing with their asthma nurse, but most could not recall being invited to look at the computer screen during a GP consultation. Few participants realized that they could be invited to look at the screen, or even understood why they might want to see it:

GPs certainly not, I don't think they ever share screens. The asthma nurse …I think they have like, they've shown us, but they are just graphs, not really to do with asthma necessarily. They are to do with like height and weight and where you should be and then your peak flow, that stuff. (P/parent of child/1, male, age 14, site 4)

Well, I don't really think like that is a nice thing to do …Aye, I'm just thinking that (screen) was a bit private, you know, would that not be a bit private to them? (P/adult/12, male, age 51–60, site 6)

That said, some participants had experience screen sharing during clinical consultations:

Certainly, in the hospital in most sessions. I'm quite curious as an individual anyway, and dangerous because I have a little bit of knowledge, so I've been looking at the numbers they were copying down. I think in the consultant conversation he was definitely pivoting the monitor so we could look at it. I can't remember what was on it, but I do remember that seating arrangement to both look at it. (P/adult/9, male, 41–50, site 4)

Some respondents felt that a CDSS which allowed them to see how the clinician worked through their diagnosis, might have helped them to understand more about the variability of asthma and other aspects to help understand the condition.

So, yes anything that provides better, broader information from a multitude of directions, so not just ‘Here is a piece of writing for you’. Like you are seeing with visuals, you know, I think is only going to make it better. (P/parent of child/1, male, age 14, site 4)

Online health information use

When asked about accessing health information online pre-diagnosis, a lack of trust in the quality of information online alongside patient perceptions of GP dislike of the practice, meaning that most respondents avoided using the internet to try and self-diagnose.

I think I'm of the generation that what the doctor tells me I believe him. I tend not to look up illnesses myself. (P/adult/11, female, 61–70, site 4)

I don't go online so much […] because I work for a health organisation. And I know that doctors get annoyed with, sort of, patients looking up symptoms online before actually going to see them; and then thinking they've got

something when they've not actually got it. So, that's maybe one of the reasons I don't tend to sort of go online to look out for health problems and things like that. (P/adult/1, female, 41–50, site 1)

However, participants noted that they accessed information online post-diagnosis to expand their knowledge or define their condition better:

I looked it up, which I never normally do, asthma symptoms. And it's because I was still coughing and I'm …the thing I says to my brother and sister, I'm not convinced I've got asthma. I think it's a chest infection […] So …what I read on the Internet, sadly […] confirmed what I was feeling [was asthma]. (P/adult/8, male, 61–70, site 3)

There was a perception that negative clinical attitudes existed towards patients exploring online information before a GP diagnostic appointment (P/adult/1, female, 41–50, site 1). Conversely, some held the view that 'Dr Google' was useful particularly in terms of searching for groups to exchange views and experiences of asthma.

Everything seems to be online, and everybody seems to have an opinion and so easily accessible […] information that you need, and you know, you've got your asthma, you know, groups online. (P/adult/13, female, 51–60, site 4)

Patient views on an asthma CDSS

The most popular output for the CDSS was the ability to provide the probability of an asthma (ideally visually) during diagnostic consultations. Moreover, respondents agreed that being able to see the factors which could lead towards an asthma diagnosis would be useful alongside further information to improve treatment management:

And he could say, I don't know, let's say there's various fields on your screen, if five out of these ten fields are ticked, the chances are, that you've got asthma or whatever disease and as you can see you've got seven of them ticked; you know, something like that. A visual representation. (P/adult/10, male, 61–70, site 4)

In keeping with the lack of confidence that individuals had about their understanding of asthma, participants suggested that incorporating an educational section within the CDSS which could be used during the consultation to show a visual representation of how asthma affects the lungs would be of interest and could assist communication and their understanding:

I actually think that kind of thing would be really helpful for children and young people…. because it's very abstract, and especially if it's just something that you think is just how your body is, you never question it, you never really think about it in terms of the actual physiological processes that are happening, you are just like, 'Oh I've got asthma, right'. (P/parent of child/1, male, age 14, site 4)

If there's a simulation or something like that, 'Here's how it looks when it's really bad' and 'Here's how, what', 'Here's how an inhaler, what it does to your lungs', 'Here's what specific medication does' and stuff like, yeah, I think that would be very interesting […] just looking at it on a piece of paper, is not the best. I think seeing some kind of simulation would be much more helpful. (P/adult/7, female, 51–60, site 1)

Respondents also noted that it would be beneficial to understand where they fit into an overall picture of asthma severity, with P/adult/9 (male, 41–50, site 4), two years since diagnosis asking, 'What's normal and where am I versus normal?' Some participants felt that understanding the significance of their diagnosis could have helped them take the diagnosis more seriously from the beginning. Participants also suggested that a website associated with the CDSS which could be used after the consultation would be more beneficial to them in the long run than the traditional handouts:

I think it's a good idea. I think it would help quite a lot 'cause the big problem I had was that I wasn't using my inhaler correctly and then I wasn't seeing an improvement on …kind of, on my, like, lung capacity essentially. So, I think if I'd, kind of, had that understanding earlier on then I would have been more dedicated to using my inhaler the way that I'm meant to. (P/adult/2, female, 21–30, site 1)

There were others who were sceptical, believing their diagnosis would not have been speedier or different with the aid of a CDSS.

I mean, I have to say that on these indicators alone, my family history was 'no none'. At that point I didn't really have any allergies, they have come on since. Also, my coughing had, ironically, stopped by the time …after the first episode my coughing had stopped because of the operation. Also, I didn't have a wheeze. (P/adult/13, female, 51–60, site 4)

Barriers and facilitators to a CDSS being used

Respondents expressed interest in the potential role of the CDSS and could see areas where the CDSS might improve the diagnostic experience. However, respondents also highlighted that whether the potential was realized depended on how the CDSS was used:

So, I think this system would be good but if it's just the system and then a very overworked GP that doesn't make eye contact, it's not really going to work. It would be, kind of …you know, you'd have to have the right person who was interacting in …on it with you. (P/adult/2, female, 21–30, site 1)

To this end, most respondents viewed the CDSS as an avenue through which communication between patients and clinicians could be facilitated:

Between yourself and the health professionals, this might be a little bit of a focal point for the conversation. So, I think that's likely to work well. (P/adult/6, male, 41–50, site 1)

The ability to aid understanding between clinicians and patients was viewed as the most important aspect of the CDSS, especially for those respondents who found the initial diagnosis 'daunting' (P/adult/7, female, 51–60, site 1). Moreover, screen-sharing was viewed as an opportunity to be 'treated like an intelligent adult' (P/adult/6, adult male, 41–50, site 1). Using the CDSS could provide a framework for clinicians and patients to use together to provide a better-shared understanding of potential routes to diagnosis.

DISCUSSION

Being diagnosed with asthma could feel like an uncertain process for participants in this study, who felt that limited consultation time or poor communication made it difficult to understand how and why the diagnosis had been made. Some participants felt they retained information about asthma diagnosis poorly and considered online or digital resources more useful than paper handouts. Participants felt possible advantages of a CDSS for asthma diagnosis may be prompting dialogue, improving understanding and encouraging a shared diagnostic process between patients and clinicians.

Interpretation

The hallmark of asthma is variability. Symptoms vary over time and in severity and making a diagnosis of asthma can require time or repeated investigations to build up the information required.[5,8,9] For patients, preconceived concepts about what asthma is, and who is at risk of developing the condition influence the credibility of an asthma diagnosis. For clinicians, weighing up the probability of an asthma diagnosis, differentiating between asthma and other conditions and excluding red flags can all influence how a consultation is conducted.[8,10] Thus, the perceptions of both clinician and patient can shape a consultation and a mismatch in these perceptions may lead to dissatisfaction.[30] Involvement of patients in consultations to allow shared decision-making is widely accepted in medical practice and may lead to better asthma control, quality of life, adherence to medication and patient satisfaction.[31] Yet for shared decision-making to occur, clinicians need to have time and resources to provide information on the pros and cons of a particular course and patients need to feel able to understand and question the medical explanations while contributing what is important for them.[32–34]

Amongst the diagnostic experiences recalled in this study, there were instances where participants felt their diagnosis was a *fait accompli* or did not feel empowered to ask questions or engage in meaningful interactions, believing the professional opinion was final. Additionally, and often because of perceived time constraints during GP appointments, patients were reluctant to ask for clarification or explore their diagnosis further, even though they were often dissatisfied with the information provided. One patient deliberately refrained from seeking further asthma information online, believing that doing so would be annoying for their GP. These examples may indicate a mismatch in the perceptions of health between patients and clinicians during the consultation. One influencing factor may be patient/clinician power imbalance, whereby the health professional was perceived to hold power within the consultation.[26–28] A perceived superiority of clinicians in the eyes of patients can impact their willingness to share their opinions,[34] and engage in consultation because they trusted that the clinician knew best.[35] On the other hand, some patients may prefer a more direct consulting style and not be actively involved in decision-making.[36] Consequently, strategies/interventions to support a more egalitarian partnership between patient and professional

may encourage more supportive patient care, an increased understanding of individual illness and facilitate patient empowerment.[28,29] In this study, there were a few examples of reduced medical dominance; participants described screen sharing and the layout of seating in the consultation room. In keeping with a prior study[30]; screen sharing did contribute to patients feeling involved in the consultation, yet few participants had direct experience of it occurring. Whilst the theory of medical dominance extends beyond doctors to allied health professionals and nurses,[28,29] some participants in this study preferred to see their asthma nurse (compared to a GP) because they found the consultation more understandable. Other factors such as the length of the appointment may also have influenced this view.

To promote shared decision-making, Agoritsas et al.[33] suggested clinicians need 'skills and tools' while patients require 'information and support'. CDSS has traditionally been seen as technology to support clinicians, but can also promote patient-focussed practice[37] through the involvement of patients in decision-making about their health and well-being.[38,39] In this study, participants liked the idea of visualizing the probability of asthma, seeing simulations of lung physiology and being able to see how the clinician worked through their diagnosis. However, some participants had reservations, explaining there would be no point in a CDSS if the clinician did not have time to engage. In keeping with this view, some have argued that the barriers to using CDSS set them up for failure.[40–42] For instance, with a lack of guidance on how decision support systems could be used, clinicians were more likely to rely on their training and experience than on new technologies.[43] Additionally, a lack of appointment time could result in reduced patient involvement, as the clinician focuses on the CDSS rather than inviting the patient to become part of the decision-making.[40]

Implications for research and practice

The importance of diagnostic tests was noted by participants in this sample, and the lack of access to tests is a source of frustration and uncertainty for patients and health professionals alike.[10] In Germany and Sweden, spirometry can be achieved at the time of presentation, or within 2 weeks, respectively.[10] Yet in other health systems, including the United Kingdom, the time between the first presentation and achieving spirometry or FeNO can be months. Therefore, in the United Kingdom, one implication of this work is to improve capacity and timely access to diagnostic tests for asthma.[44,45] Digital solutions, such as the AsthmaTuner self-management system, could transform the diagnosis and management of asthma.[13,46] The use of connected technologies such as wearable sensors, Bluetooth spirometers and digital peak expiratory flow devices could increase access to diagnostic information and allow measurements to be performed when a patient is symptomatic.[13,46] CDSS which collates data from such devices and supports interpretation could lead to improvements in the diagnostic accuracy of asthma though further high-quality studies are needed.

In situations where testing remains difficult to achieve, where the outcome of tests makes the diagnostic process protracted (i.e., false negatives), or variable symptoms occur over an extended timescale, considering how best to achieve shared *diagnosis-making* through explanation of the current situation and deciding on the most appropriate next steps may help patients remain involved. The role that an asthma diagnosis CDSS may have in engaging patients through the diagnostic process is planned to be evaluated during a feasibility pilot study.

Strengths and limitations

The study was designed and piloted using input from a multidisciplinary advisory group and PPI members to develop topic guides and trial interviews which ensured that the topics covered were important to those with asthma and the study team. We sought views from a wide range of individuals who had been recently diagnosed with asthma from different areas across England and Scotland. Despite invitations being sent to those who had a diagnosis of asthma coded in the electronic health records within the last 5 years, we received many expressions of interest from individuals who had been diagnosed over 5 years before, and from individuals over 50 years of age.

Recruitment was severely hampered by the COVID-19 pandemic for the following reasons: non-COVID studies (such as ours) were paused to prioritize urgent research which meant the planned study period was restricted; GP practices faced high workload and reduced staffing which made it more difficult to recruit practices; it took longer for clinical research network staff to gain access to GP practices and send out invitations to patients. Despite these

challenges, we managed to recruit parents of children with asthma, young people and older adults, with a range of diagnostic experiences. Having interviewed 14 participants, we considered that with respect to adults over 30 years, no new information was being collected and no new codes were developed.[47] In line with our purposive sampling approach, we chose to complete further interviews to enhance the diversity of the sample, specifically parents of children and young people. Before the study period closed, we were able to include more parents of children and one participant in the 16–30 age group.[47] Overall, we felt that data saturation had been achieved because the themes had been fully described with no new information being obtained in the later interviews. However, we acknowledge that had we been able to recruit more males, participants aged 16–30 years and individuals from rural GP practices we may have heard about different experiences.

This study sought views on a proposed CDSS being developed by the research team. Regarding reflexivity, we acknowledge the desire to create a successful CDSS may have influenced data collection and interpretation by being eager to pick up on positive aspects during interviews and identifying favourable opinions when analysing the data. We attempted to minimize the influence of any one individual by having two researchers conduct interviews and several team members (including a steering group) contribute to the interpretation of results.

CONCLUSIONS

The process of diagnosing asthma was uncertain for patients if their ideas and concerns were not addressed by clinicians and were often related to a perceived lack of consultation time and limitations in communication. A CDSS designed with patients' needs in mind could encourage a more shared diagnostic process between patients and clinicians, and improved communication relating to the nature of the condition and its management, including the patient's role in self-management.

AUTHOR CONTRIBUTIONS

Luke Daines and Hilary Pinnock conceived the idea and achieved funding. Victoria Murray and Eddie Donaghy conducted patient interviews. Anne Canny, Eddie Donaghy, Luke Daines, Hilary Pinnock and Victoria Murray wrote the first draft and Leo Campbell, Carol Stonham, Andrew Bush, Brian McKinstry and Heather Milne were involved in the interpretation of data, revising it critically. All authors gave final approval for the article to be published.

ACKNOWLEDGEMENTS

We are grateful to Optimum Patient Care and Tactuum, our collaborators on the Asthma + Lung UK/Innovate UK-funded research grant to develop an asthma diagnosis clinical decision support system. We thank the Asthma UK Centre for Applied Research Patient and Public Involvement team for their help during the study. This study was funded by Asthma + Lung UK/Innovate UK grant number: AUK-PG-2018-404. L. D. was supported by a clinical academic fellowship from the Chief Scientist Office (CAF/17/01).

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

The study received ethical approval from London Stanmore Research Ethics Committee (ref: 19/LO/1722).

DETAILS

Subject:	Parents &parenting; Asthma; Patients; Medical diagnosis; Selfmanagement; Applied research; Young adults; Diagnosis; Data analysis; Primary care; Teams; Chronic obstructive pulmonary disease; Interviews; COVID-19; Consent; Citizen participation; Public involvement; Decision support systems; Health care; Age; Clinical decision making; Pandemics; Medical research; Diagnostic systems; Patient participation; Communication; Medical personnel; Group decision making; Professionals; Qualitative research; Understanding; Coronaviruses; Decision making
Business indexing term:	Subject: Professionals
Location:	United Kingdom--UK
Identifier / keyword:	asthma diagnosis; clinical decision sup port system (cdss); GPs and asthma nurses; patient perspectives; primary care
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	307-317
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-12

Milestone dates:	2022-09-22 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-07-06 (manuscriptReceived); 2022-11-12 (publishedOnlineEarlyUnpaginated); 2022-10-23 (manuscriptAccepted)
Publication history :	
First posting date:	12 Nov 2022
DOI:	https://doi.org/10.1111/hex.13657
ProQuest document ID:	2767101886
Document URL:	https://www.proquest.com/scholarly-journals/patient-views-on-asthma-diagnosis-how-clinical/docview/2767101886/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the "License"). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-11-08
Database:	Public Health Database,Publicly Available Content Database

Document 31 of 46

What are the reasons for unfinished nursing care as perceived by hospitalized patients? Findings from a qualitative study

Chiappinotto, Stefania [1]
; Coppe, Alberto [2] ; Palese, Alvisa [3]
[1] Medical and Surgical Department, University of Tor Vergata, Roma, Italy [2] Health Care Professionals Service, AULSS 2 Marca Trevigiana, Treviso, Italy [3] Department of Medical Sciences, University of Udine, Udine, Italy

ABSTRACT (ENGLISH)

Introduction
Unfinished nursing care (UNC), as the care required by patients that delayed or not delivered, has been investigated mainly from the perspective of nurses, while little is still known from the side of patients. Some studies have involved patients to measure which elements of care are mostly unfinished (e.g., mouth care), whereas a few studies have investigated the reasons for UNC as perceived by them. Their involvement in understanding the reasons for UNC is crucial to advance the knowledge and co-develop possible strategies to prevent or minimize UNC.
Methods
This is a descriptive qualitative study performed according to COnsolidated criteria for REporting Qualitative

research guidelines in 2022. A purposeful sample of Italian hospitalized patients in two medical and two surgical units was involved. A face-to-face semistructured interview was used to merge reasons for UNC. Qualitative content analysis was conducted to merge subthemes and themes as factors leading to UNC according to the experience of patients.

Results

A total of 23 patients (12 surgical and 11 medical) were involved (12/23 male) with an age average of 66.2 years, educated mainly at secondary school, and with previous hospitalizations (20/23), and dependent on nursing care in daily activities (14/23). Reasons for UNC have been identified at four levels: (1) 'New health-care system priorities' and 'Pre-existing frailty of health-care facilities' were reasons identified at the health-care system level; (2) 'Lack of resources attributed to wards', 'Ineffective ward organization' and 'Leadership' were identified at the unit level; (3) 'Nurses' attitudes and behaviour' were reported at the nurses' level and (4) 'Increased nursing care expectations' were pinpointed at the patient level.

Conclusion

Patients can be involved in identifying UNC, but also in recognizing the underlying reasons. Engaging them in such investigations might broaden our understanding of the phenomenon and the possibility of identifying strategies to minimize and prevent UNC.

Patient or Public Contribution

Patients from four hospital units (two medical and two surgical) were involved in face-to-face interviews to merge the reasons perceived by them as triggering UNC. All factors (as themes and subthemes) have derived from their words, thus enhancing the evidence available from the side of the patients.

FULL TEXT

INTRODUCTION

Unfinished nursing care (UNC)[1] has been widely investigated as a concept,[2] in terms of its antecedents[3] and consequences (e.g., Wieczorek-Wojcik et al.[4]). This phenomenon has been documented in the literature under different terms, such as 'tasks left undone',[5] 'implicit rationing of nursing care'[6] and 'missed nursing care'.[7] Over time, these different frameworks have highlighted various aspects of the phenomenon,[8] but emerging evidence agrees on the fact that UNC is experienced by nurses and patients as an important issue for the quality of care,[9–11] as it refers to nursing care that is required but completely omitted or delayed. However, this phenomenon has been investigated mainly from the nurses' angle, whereas little is still known from the patients' side.

The first study involving patients was performed by Kalisch et al.[12] using a qualitative method to explore the extent and type of missed nursing care as experienced by them. The authors developed a list of questions centred on activities that patients should have received, asking them if nurses had been able to complete them. Findings have highlighted that some activities were fully reportable as missed by patients (e.g., mouth care and bathing), others were partially reportable (e.g., hand washing and assessing vital signs) while others were not (e.g., nursing care process). Therefore, patients were not able to report all UNC elements, but their perceptions were considered important to gain the global picture of the phenomenon as perceived by them; however, the reasons for missed nursing care were not investigated.

Two years later, another study[13] assessed the missed nursing care phenomenon using the MISSCARE Survey-patient, by developing it from that already validated among nurses.[14] This quantitative study was aimed at investigating the amount and type of missed nursing care as perceived by patients and the patient-reported outcomes. Patients referred to basic care, communication and time to respond to needs as the most omitted or postponed care. However, the 'Reason for Missed Nursing Care' section of the MISSCARE Survey for nurses, which assessed the perceived causes, was not administered to patients because during the pilot testing the most frequent answer to various items was 'I do not know'.

Three further studies quantify patients' perceptions of missed nursing care under the Kalisch framework. First, Dabney and Kalisch[15] performed a study aimed at investigating the relationship between nursing staffing and the patients' report on missed nursing care, using the MISSCARE Survey-patient. They found a correlation between the

total nursing staff hours of care per patient-day, registered nurse hours per patient-day, registered nurse skills mix and the occurrence of missed care; however, no exploration was conducted on the reasons for missed care.
By using the MISSCARE Survey-patient to investigate patients' perceptions, Cho et al.[16] explored the mediating effects of missed nursing care as reported by patients on the relationship between nursing staffing and patients' experiences. An association between better staffing adequacy, less missed care and better patient experiences emerged, indicating that patient perception of missed care mediated the relationship between staffing adequacy and their own experiences.
However, only Moreno-Monsiváis et al.[17] investigated nurses' and patients' perceptions using the MISSCARE Nursing Survey, providing some modifications to the tool to collect patients' points of view regarding causes. In the attempt to expand the knowledge available, the section concerning the reasons perceived was retained and adapted by asking to patients 'Why do you think nurses do not "always" provide some aspects of care?'. Patients reported the lack of staff, the insufficient experience of the staff, the lack of organization and teamwork, the lack of staff communication from one shift to another and the attitude of staff members as reasons for missed nursing care.
In addition to studies based on the Kalish framework, Orique et al.[18] administered the Hospital Consumer Assessment of Healthcare Providers and Systems (HCAHPS) tool,[19] a national standardized instrument developed by the Centers for Medicare &Medicaid Services and the Agency for Healthcare Research and Quality for assessing patients' perception of hospital care. Researchers used some items that were useful for investigating elements of missed nursing care in an acute care setting, but again in this case no questions were raised about the reasons for missed care.
Therefore, according to our best knowledge, the only qualitative study to have investigated UNC with a missed nursing care framework as perceived by patients was the one by Kalisch et al.[12] Some other quantitative studies have collected patients' perceptions about this phenomenon, but underlying reasons as perceived by them were little investigated. Second, available data have been collected mainly under the missed nursing care conceptualization, while in recent years a more comprehensive framework capable of including all different concepts in this field by establishing the UNC umbrella concept.[20] Moreover, involving patients in investigating the reasons for UNC might contribute to expanding the knowledge available by including a wider perspective. The concept of patient engagement has also assumed a fundamental role in detecting issues and in promoting the quality of care.[21,22] In a world where citizens require a health system to be transparent, open and responsive, patients' engagement has become imperative and an effective strategy for understanding their experiences and for promoting alliances with them with a view to achieving better care.[23] Evidence has documented that engaging patient increases their safety,[24,25] their satisfaction with care[26] and last but not least, their healthcare outcomes.[27] Moreover, deepening an understanding of patients' experiences has been seen as the first step towards patients' engagement.[23] Following this perspective of involvement and engagement, the purpose of this study was to explore the reasons for UNC as perceived by patients, thus going beyond the point of view of nurses as mainly perspective included to date.

METHODSStudy design

This is a descriptive qualitative study,[28] performed in 2022 and reported here according to the COnsolidated criteria for REporting Qualitative research guidelines[29] (Supporting Information: Table 1). Moreover, the study was designed under the UNC framework[1] to ensure (a) inclusiveness of all different conceptual traditions in the field and (b) an updated approach is given that tools and investigations in recent years have been conducted under the UNC framework.[30]

Setting

A large healthcare trust of the Veneto Region Public Health Care Service, comprised of seven hospitals and four accredited facilities, equipped with a total of 2390 beds serving 880,000 citizens in 2021, of which 23% were >65 years old, was approached. Among those available, one large hospital was identified (35,000 admissions/year[31]), and within these, two medicals (66 beds each) and two surgical units (52 beds/each) were considered for the study.

Participants

A purposeful sample of patients[32] with rich knowledge about, or experience with, the phenomenon of interest was

chosen. Specifically, patients were included if they (a) were adults (>18 years); (b) had been hospitalized for more than 48 h; (c) was on discharge or with a planned discharge, and thus were not unstable or in their acute phase; (d) were able to participate in an interview and (e) were willing to participate in the study. Therefore, those patients not meeting the inclusion criteria were excluded.

The recruitment process was conducted daily from the start of the study: a researcher (S. C., see authors), who was an advanced educated nurse (PhD student and research fellow), and was not involved in the care of patients, consulted the nurse responsible for the nursing care or the nurse manager to decide on the patient to approach. The recruitment ended when data saturation was achieved,[33] as judged independently by two researchers (S. C. and A. P.; see authors), when dominant themes were perceived as completed and no others emerged from the interviews. None of the identified patients refused to participate.

Data collection

According to the only study available that collected patients' perceptions about UNC with a qualitative approach,[12] and considering the most recent studies investigating reasons for UNC as perceived by nurses,[34–36] a semistructured interview was designed. The interview was composed of the following open-ended questions:

- (1)
  Demographic data and the perceived degree of dependence in the activity of daily living (e.g., I am independent; I need help in some daily activities [eating, hygiene]);
- (2)
  A recall of a particular UNC episode; and
- (3)
  A full description of the underlying reasons according to the perceptions/experience of the patient (Table 1). The interview guide was pilot tested on the first four participants. No changes were necessary.

Table 1 Interview guide

Interview guide
Introductory section
Researcher self-presentation
Presentation of the study aim and of data collection procedures
Acquisition of written consent for the interview and the audio-recording
First section
Demographic data, perceived degree of dependence in activities of daily living and previous hospitalization
Age
Gender

Education
Working profile
Functional dependence (yes/no)
Previous hospitalizations (yes/no)
Second section
Unfinished nursing care and reasons
Recall of a particular episode of UNC
Narration of the perceived reasons triggering the episode narrated according to personal experience
Additional elements considered relevant in the context of UNC experienced

Abbreviation: UNC, unfinished nursing care.

The interviews were scheduled for between April and June 2022 and were conducted face-to-face on a day and at a time preferred by each patient. No relationship with participants was established before the commencement of the study. They were informed only about the working position of the researcher and the aims of the study, which were illustrated in a detailed fashion by the nurse responsible for the patient and then repeated by the researcher at the time of the interview. The interviews, which lasted for between 3 and 22 min, were carried out in a quiet setting, where only the researcher and the participant were present.

Data analysis

A qualitative content analysis[37] was used to merge subthemes and themes describing reasons for UNC as perceived by patients. Specifically, two researchers (S. C. and A. P.; see authors) performed the analysis by (a) transcribing the interviews; (b) reading and rereading the transcriptions, and also by contextually selecting the units of meaning (i.e., a word or sentence that holds a specific meaning in the context of perceived UNC reasons); (c) identifying subthemes: each researcher identified subthemes (i.e., an abstraction of the units of meaning labelled with a code) independently, as derived from the data. Then, a consensus was reached between researchers regarding the subthemes that emerged; after having reached the consensus, researchers proceeded by (d) categorizing the subthemes. As in the previous step, each researcher identified the themes by grouping subthemes independently; the agreement was reached by consensus through multiple meetings. An example of the coding tree is reported in Supporting Information: Table S2.

The data analysis was performed manually, without using any software. The coding process was initiated immediately, after three interviews, and then continued to assess the saturation when reached[33] as judged when no new subthemes emerged. The concurrent analysis of the data as immediately performed after the interview, allows to limit the number of participants, given that being involved in understanding the reasons for the poor quality of care may burden patients, especially when they are still hospitalized.

Rigour and truthfulness

Several strategies have been enacted to ensure rigour and truthfulness.[38] First, the understandability and feasibility

of the questions included in the interview were ensured throughout the pilot test. Second, the credibility of the findings and the data dependability were ensured by extracting quotations to provide concrete examples of reasons from the words of participants, and by reporting the number casual assigned to each participant (e.g., P6, Participant number 6) to ensure anonymity. Third, the end of the interviews was decided according to the data saturation as assessed by two researchers, who evaluated in an independent fashion and then compared subthemes that emerged. Fourth, to prevent the influence of preconceptions, the coding process was conducted by two researchers independently by using anonymized data and then agreeing on findings; moreover, the quality of the process was ensured by involving researchers who were experts in qualitative methods and in interviews. Furthermore, transferability was promoted by describing the settings involved and the participants' main profiles.

Ethical issues

The Ethical Committee approved the study protocol (16th December 2021, prot. n. 234258/2021; Amendment 31st March 2022). Participants were informed of the study aims, and they were free to withdraw from the study at any time without any consequence. They were also ensured that the interview would not be shared with nurses responsible for their care and all data would be anonymized. At the end of the explanation, they were asked to sign the consent form where they also agreed to be audio-recorded.

The researchers anonymized the narratives before the data analysis, assigning a casual number to each participant interviewed; moreover, the wards were anonymized, and thus their official names were changed to prevent them from being recognized. Quotations were also identified with the number of participants.

RESULTSParticipants

As reported in Table 2, a total of 23 patients were involved, most of them male (12/23) with a mean age of 66.2 years (standard deviation 14; range 40–92). Most of the patients reported a secondary school education (14/23), followed by an elementary school (6/23) and some were educated at the university level (3/23). Most participants were retired (14/23). Moreover, 20 out of 23 reported previous hospital experiences in different wards, 12 had been cared for in surgical units and 11 in medical units. The majority (14/23) perceived the need for help due to functional dependence on activities of daily living.

Table 2 Participants' characteristics

ID	Gender	Age	Education	Ward	Work position	Functional dependency	Previous hospitalization
1	F	74	Elementary school	Medical α	Retired	No	Yes
2	F	89	Elementary school	Medical α	Retired	Yes	Yes
3	M	60	Secondary school	Medical α	Designer and production manager	No	Yes
4	F	56	Secondary school	Medical α	Embroiderer	No	Yes

5	F	65	Elementary school	Medical β	Retired	Yes	Yes
6	M	69	Master Degree	Medical β	Retired	No	Yes
7	M	60	Secondary school	Medical β	Retired	No	Yes
8	M	79	Secondary school	Medical β	Retired	Yes	Yes
9	F	57	Bachelor	Surgical α	Freelancer	No	No
10	M	51	Secondary school	Surgical α	Taxi company manager	Yes	Yes
11	F	92	Elementary school	Surgical α	Retired	Yes	Yes
12	M	57	Secondary school	Surgical α	Retired	No	Yes
13	F	77	Master	Surgical β	Retired	Yes	Yes
14	M	40	Secondary school	Surgical β	Owner of a company	Yes	Yes
15	F	75	Elementary school	Surgical β	Retired	Yes	Yes
16	M	44	Secondary school	Surgical β	Truck driver	Yes	No
17	M	71	Secondary school	Medical α	Retired	Yes	Yes
18	F	59	Secondary school	Medical α	Housewife	Yes	Yes
19	F	87	Elementary school	Medical α	Retired	Yes	Yes

20	M	47	Secondary school	Surgical α	Digital video entrepreneur	No	Yes
21	F	67	Secondary school	Surgical α	Farmer	Yes	No
22	M	76	Secondary school	Surgical α	Retired	No	Yes
23	M	71	Secondary school	Surgical α	Retired	Yes	Yes

Abbreviations: F, female; M, male.

UNC reasons

As summarized in Table 3, the reasons for UNC have been identified at four levels, namely at the healthcare system, at the unit, at the nurses and at the patient levels, including seven subthemes. The ‘New healthcare system priorities’ and the ‘Pre-existing frailty of healthcare facilities’ were reasons identified at the healthcare system level, and the ‘Lack of resources attributed to wards’, the ‘Ineffective ward organization’ and ‘Leadership’ were identified at the unit levels; the ‘Nurses' attitudes’ and ‘Behavior’ were reported at the nurses' level while the ‘Increased nursing care expectations’ at the patient level. Moreover, as reported in Table 3, some reasons were reported only by patients hospitalized in medical or surgical units.

Table 3 Levels, themes and subthemes

Level	Themes	Subthemes	Medical ward	Surgical ward
Healthcare System	New healthcare system priorities	Cost restraints	a	a
		Dramatic changes due to the COVID-19 pandemic		a
	Pre-existing frailty of healthcare facilities	Unsuitable environment layout	a	a
		Old technologies	a	a
		Discrepancies in resource allocation across wards		a
Unit	Lack of resources attributed to wards	Staff shortages	a	a

		High patient-to-nurse ratio	a	a
	Ineffective ward organization	General vocation of the ward		a
		Poor nursing care delivery design	a	a
		Poor shift design		
		•Lack of staff during the day, nights and weekends •Excessive length of shifts •Lack of care continuity between shifts	•a •a	•a
		Overlapping activities	a	a
		High frequency of interruptions		a
		Limited capacity to react to unpredictable events		
		•Admissions •Emergencies		a
			a	
	Ineffective ward leaders	Inadequate nurse manager leadership		a
Nurses	Nurses' competences and attitudes	Lack of delegation skills	a	
		Lack of empathic competences	a	
		Lack of responsibility	a	a
		Low motivation	a	a

		Living in a hurry	a	a
		Expressed fatigue	a	a
Patients	Increased nursing care needs and care expectations	Worse clinical conditions	a	a
		Increased ADL dependence	a	a
		Demanding patients		a

Abbreviation: ADL, activities of daily living.

a

Reported by patients hospitalized in this ward.

UNC reasons at the healthcare system level

Two main themes have emerged at this level. Patients reported that UNC is due to the ‘New health-care system priorities’, where the quality of care has started not to be identified among the top priorities in recent years. In other words, patients reported UNC as an inevitable consequence of the ‘cost restraints’ applied in the last few decades to the entire system, reducing progressively the funding, and affecting the number of staff employed in hospitals:

You cannot always cut on the number of personnel …Health care is based on the quality and the quantity of the personnel. (P6)

Participants have also underlined the effects of the ‘dramatic changes due to the COVID-19 pandemic’, where new priorities were established marking a turning point in nursing care delivery, further reducing resources in some units, especially in medical and surgical ones, to devote them to COVID-19 wards, thereby increasing the risk of care omissions.

Also, now for the COVID situation, I have seen …I've been going inside out of hospitals for 10 years and I've seen a great negative change. (P10)

Patients reported that the emerging priorities greatly affect the ‘Pre-existing frailty of health-care facilities’; among these, the ‘unsuitable environment layout’, due to old-fashioned hospital buildings, has been reported as affecting nurses' timely responses to the needs of patients, due to the time required to reach each patient's room or the nurse station, thereby increasing the risk of delays in care.

Yes, because sometimes they are closer, sometimes they are further away. (P21)

Patients have also highlighted the role of ‘old technologies’ as a factor influencing the occurrence of UNC, where nurses are still using papers and pencils and dedicating a lot of time to filling in them, thus staying away from patients:

…the lack of the more advanced technologies. (P9)

Moreover, participants also perceived ‘discrepancies in resource allocation across wards’ as a reason for UNC, where human and material resource allocation across settings is unbalanced, leading to an excess of some resources and a paucity in others:

Therefore, I saw discrepancy in resources within the same department. (P9)

UNC reasons at the unit level

Three main themes emerged at the unit level, namely a ‘Lack of resources attributed to wards’, the ‘Ineffective ward

organization' and the 'Ineffective ward leaders', all of which led to UNC according to the patients' perceptions.
Among the first of these, participants stressed 'staff shortages', as the number of all staff, ranging from nursing aides to nurses, was below the minimum standard required to manage all care:
Few [staff], few, very few… (P11)
In the specific context of nurses, participants also reported a 'high patient-to-nurse ratio', as identified by the nurses themselves:
So, nurses themselves say: 'We are undersized, it would take more professionals'. (P3)
Alongside the resources allocated at the unit level, patients also reported the role of an 'ineffective ward organization'. The 'general vocation of the ward' was considered a reason for UNC, given that according to previous patients' experience, specialized wards were able to ensure greater attention to individual needs, delivering more complete care:
…[nurses] provide a better care in a specialized ward than a general medicine ward. (P9)
Moreover, the 'poor nursing care delivery design' was found to be a reason for UNC, due to the chaotic environment and the nonoptimal care processes, where participants relieved nurses from being responsible for their omissions:
…the service is badly organized; it is not the fault of the nurses. The organization of the service is terrible. (P6)
The poor organization has also been reported as being complicated by the 'poor shift design': the lack of nurses during the day resulting in high workloads and the need to postpone some activities was perceived as an issue preventing the completion of care, especially in the mornings.
During the day they take a little longer; in the evening they are faster. (P20)
Also, during the night and at the weekend, patients reported being cared for by a lower number of staff than expected for managing all needs. The same duration of shifts was reported as being a reason for UNC because it affected the performance of nurses:
…with shifts too long. They [nurses] could do broken shifts…. (P6)
On the other hand, patients reported a lack of continuity of care between shifts as increasing omissions, as nurses have been considered unable to share the main data about patients, leaving out needs perceived by them as important:
…those who were there have left and those who have arrived have just arrived. (P6)
In the attempt to cope with the high workloads, patients often witnessed nurses 'overlapping activities' to accelerate the process of care in the desire to ensure all the nursing care required. However, performing several activities at the same time has been reported as a source of delays or omissions:
If she sees a call, she is doing a job and she must finish for other patients, by walking she answers the first patient who has called and then she comes later. (P14)
In the same vein the 'high frequency of interruptions' because of patients' calls (P21) thus disrupting the planned activities, has been reported as increasing the number of possible omissions and the capacity to be on time in satisfying multiple needs.
The frailty of the units is further increased by the number of newly admitted patients and emergencies, limiting the nurses' capacity to respond to the needs of patients already present in the unit and in a stable condition, resulting in 'limited capacity to react to unpredictable events':
Well, she [nurse] was a little bit late, because maybe a lot of people are admitted here. (P11)
They [nurses] say there are other emergencies and I need to wait for them. (P18)
Above all is the 'ineffective ward leader' of the unit, as expressed by patients in his/her capacity to negotiate resources, allocate them properly in the shifts, implement appropriate models of care delivery and support the staff:

…It depends on the ward manager nurse, the head of the ward. That is, these kinds of responsibilities never depend on the last person, you must go up in the hierarchy. (P13)

UNC reasons at the nurses' level

Patients reported some factors also at the nurses' level, specifically highlighting the role of their ‘Competences and attitudes’ as possible reasons for UNC. First, participants referred to a ‘lack of delegation skills’ in some tasks, and thus a higher risk of omitting some relevant nursing activities when workloads increased in intensity:

The nursing aide …the nursing aide can't touch the medicines. Why can't they? (P6)

The ‘lack of empathic competencies’ has also been underlined as triggering omissions in communication, in the understanding of needs, and in responding to them in a timely manner according to the patients' priorities. In addition, patients also reported the perceived ‘lack of responsibility’ and ‘low motivation’ as leading to UNC:

Because it is so convenient for them [nurses] not to do all things. (P19)

…in recent years they [nurses] are all listless. (P17)

On the other hand, patients reported that nurses are always ‘living in a hurry’, thus preventing any contact or interruption by patients to express their needs; sometimes being in a hurry has been reported as the consequence of the excessive workloads, at other times as a question of habit/attitude.

This nurse went away immediately, not even time to finish speaking. Here, when you are still talking and the nurse is already at the door, that is…. (P6)

Moreover, nurses' ‘fatigue’, as explicitly expressed, or as interpreted by patients according to some manifested behaviour as a reaction to the high workloads and the chaotic environment, has been identified as leading to UNC:

Yes, because they are exhausted. (P2)

UNC reasons at the patients' level

Participants have recognized the role of the ‘Increased nursing care needs and care expectations’ in receiving the care required; therefore, while that was sufficient or adequate in the past, today it is never enough because of the ‘worse clinical conditions’ of patients and their ‘increased dependence in daily activities’, determined by co-morbidities, older age, complex treatments (e.g., medications) and frailty:

And well, of course, when they [nurses] see that you are more stable, they put you a little further back, because there is someone who needs them more. (P12)

I can only say that for the first five days that I could not move, they [nurses] ran here. (P12)

The explosion of nursing care needs presents nurses with a daily challenge in deciding the priorities with the same resources provided to the units years ago. In addition, they must face highly ‘demanding patients’ due to their increased expectations regarding nursing care, rising nurses' workloads and the risk of UNC:

Then I don't know if maybe some periods are different for patients too, maybe at a certain time they are more demanding. (P20)

DISCUSSION

To the best of our knowledge, this is the first qualitative study investigating the reasons for UNC, as perceived by patients. Adults and older individuals were involved, without applying strict inclusion criteria, resulting in participants educated at different levels, and with different working positions, from active to retired, nearly all with previous hospital experiences and in need of help with basic care. According to the main profile of participants, while gender bias[39] has been prevented by balancing the genders, the previous hospitalization of patients and their need for basic nursing care suggest that they based their perceptions regarding UNC on their direct experience: patients with poorer health status—as those involved in this study—have been documented as experiencing more UNC.[40]

Methodological discussion

Previous studies in the field have documented that patients are able to recognize and report aspects of UNC mainly regarding basic care, communication and timeliness (e.g., timely help in going to the bathroom).[40] However, studies investigating their perceptions by using available tools have deleted the questionnaire section regarding the perceived reasons, mainly because ‘Do not know’ was the dominant patients' answer to the items in the pilot surveys.[17] We undertook the challenge to investigate the reasons for UNC because of the following considerations:

- (1)
  Patients' perceptions reflect a valuable point of view in fully understanding healthcare issues as measured by healthcare professionals.[23]

- (2)
  In the field of patient complaints, the contributory factors leading to problems in care have been neglected, thus focusing their involvement instead on the underlying reasons or causing factors.[41]

- (3)
  Having evidence on perceived reasons for UNC among patients might help to inform them regarding the actual causes thus preventing violence and aggressiveness towards nurses when they are not able to ensure the care required.[42]

However, our study suggests that patients have some difficulties indicating and detecting the reasons for UNC: the interviews were very short in duration, thus indicating that participants were having difficulty in identifying the reasons for the phenomenon. Moreover, some of their perceptions seem to be experienced directly (e.g., overlapping activities), whereas others seem to be experienced indirectly (e.g., a large number of admissions), as reported by (a) the same nurses (e.g., nurse shortages, lack of nurses at the weekend, emergencies) while they try to excuse themselves for the UNC; (b) other patients (e.g., ‘there is a patient with bad clinical conditions’) or (c) by external sources (e.g., newspapers, television), where the information reported may acquire a meaning while hospitalized (e.g., cost restraints). Also in the field of UNC, the perceptions have been differentiated into visible (or fully reportable, or areas of nursing care patients were able to report on), partially and not reportable by patients, which refers to areas of nursing care that patients were unable to report on.[12] Future studies are recommended to investigate the sources of patients' perceptions, to understand how they develop their understanding regarding the reasons for UNC. Moreover, with the increasing evidence in the field, tools measuring UNC among patients might be completed with the list of possible reasons that emerged in our study.

Some differences have emerged in the UNC factors between medical and surgical wards with some perceived only by patients cared for in medical units (e.g., ‘lack of delegation skills’) and others by those admitted to surgical wards (e.g., ‘inadequate nurse manager leadership’). Studies investigating nurses' perceptions have also reported evidence of some differences.[3] However, more research is recommended to accumulate evidence in this field to inform different interventions to minimize UNC according to the underlying reasons.

Findings discussion

At the overall level, reasons for UNC have emerged at the healthcare system, unit, nurse and at patient level; previous studies investigating the perceptions of nurses[34–36] have identified the reasons at the system, unit, nurse manager, clinical nurse and patient levels, thus suggesting that patients are able to identify the reasons for UNC at all levels, mirroring the perceptions of nurses.

UNC is affected by several factors, where the upper system, namely the health-care service priorities, resources, emergencies and values, has been underlined as affecting the care delivered at the bedside.[43] Patients perceived

the relevance of the upper system for the care received daily, suggesting that the long-term disinvestment in the public health sector, further threatened by the COVID-19 storm, has reduced the capacity to provide the care required by medical and surgical patients. Apart from the threats to the basic principles of the public healthcare system that are underlined by our participants as compromised (e.g., discrepancies in resource allocation affecting equity), findings suggest that public involvement in setting the priorities, in allocating the resources and in giving feedback on the care ultimately delivered should be core values of policymakers.[44]

At the unit level, patients reported most of the reasons for UNC: hospitalized patients seem to gain an overall picture of factors, underling the importance of resources, models of care delivery and the relevance of the nurse manager leadership. Several reasons reported have already been documented from the side of nurses both in conceptual and empirical evidence, thus confirming the multiple unit factors involved in leading to UNC[3,36] (e.g., Kalisch &Williams).[14] However, some have emerged as new from the side of patients, namely: the general vocation of the ward and the lack of care continuity between shifts. All factors that emerged as subthemes seem to be influenced by each other in a sort of domino effect, where the implementation of single interventions to prevent UNC may affect only in part the occurrence of the phenomenon, thus requiring more complex interventions capable of targeting different structural and process elements at the unit level. Moreover, while some factors seem to be modifiable (e.g., poor design of shifts), others are directly connected with the decisions undertaken at the upper level (e.g., the number of resources devoted to nursing care). Furthermore, patients highlighted two main factors worthy of consideration for their ethical implications: the generalist vocation of the units has been reported as a source of UNC, and this should be further investigated and discussed given that most patients are admitted to general wards and they perceive themselves as being at increased risk of UNC compared to those admitted to specialized units; on the other hand, those patients that remain stable during their in-hospital stay are more at risk of their needs being neglected given that emergencies and newly admitted patients are considered priorities. Equity as well as strategies to prevent any form of discrimination are an imperative principle among nurses, suggesting that these findings should be considered carefully to address appropriate strategies.[45]

Patients also reported factors at the nurses' level by referring to their competencies and attitudes: these findings suggest that some factors rely on individuals, and these may vary across shifts and across nurses, modulating the amount of UNC according to the nurses' individual traits. Previous studies have highlighted the role of individual accountability[46] as well as that of the nurses' habits as a group.[7] However, our findings suggest some additional factors: (a) that regarding the competencies in delegating activities and in having an effective relationship with patients, both modifiable through undergraduate and postgraduate education; and (b) that concerning the attitudes of being in a hurry and expressing fatigue. Nurses have the right to demonstrate their difficulties in coping with high workloads and challenging environments, but when these attitudes prevent patients' expression of needs, their ethical implications should be discussed.[47] Moreover, nurses' attitudes may shape the behaviour of newly graduated nurses and students by encouraging them to conform to a particular approach.[48] Furthermore, nurses should discuss whether these attitudes are effective when directed to patients in promoting awareness of UNC; instead, identifying strategies to report their emotions, fatigue and difficulties to the healthcare trust headquarters and to the general citizenship rather than to those in need at any given moment might be more effective.

Participants reported that some factors appertain to changed patient profiles, as increased needs and complex clinical conditions trigger increased expectations. Also, in a recent systematic literature review,[3] patient profiles have been recognized as a factor triggering UNC as perceived by nurses. The fact that the same patients recognized that their needs and expectations have increased means that they are ready to accept all the investments in nursing care that policymakers will provide[49]; nurses should undertake this challenge by educating the future generation to deal

with these issues by exercising effective priority setting and by addressing the increased expectations of patients.

Limitations

This study is affected by several limitations. First, no repeated interviews[32] were performed to explore in greater depth the perceptions of patients during their hospitalization and after discharge—when they might progressively understand the situation and reflect on the entire experience. This decision was undertaken in order not to burden patients. Second, participants member checking[50] was also not conducted to assess the agreement with the themes and subthemes that emerged as categorized by researchers, given the ample range of reasons reported across patients. Third, only patients capable of participating in an interview were included—missing, therefore, those patients who were not able to answer as well as their close relatives not involved in the process. Relatives might report different perceptions or act as gatekeepers,[51] whereas patients not able to participate have already been highlighted as being more exposed to UNC,[52] but the quality of their reporting might be affected by their capacity to understand and interpret the complex situation. Moreover, a few demographic data have been collected and some (e.g., ethnicity, socioeconomic status) were not required to prevent any source of burden on patients. However, future studies should consider extending the data collection to describe in a more detailed fashion the profile of the patients involved.

CONCLUSIONS

To the best of our knowledge, this is the first study involving patients in identifying the reasons for UNC. Patients reported the causes of UNC at different levels: those close to them (at the unit, at the nurses' and at the patients' level) and those more distant (at the system level). Some UNC reasons reflect those already documented by nurses in the available literature, whereas others appear to be new (e.g., cost restraints, the general vocation of the ward). Moreover, some reasons appear to be perceived directly by patients, while others appear to be mediated by others (other patients, newspapers) and also by nurses when they try to excuse themselves for the omitted or delayed care. However, at the overall level, the rich findings that emerged suggest that patients can be actively involved in identifying the reasons triggering UNC in addition to the elements of nursing care omitted or delayed.

Involving patients in identifying the UNC reasons broadens the understanding of the phenomenon and the possibility of identifying strategies to minimize or prevent it. Furthermore, asking citizens about their perceptions and informing them about the reasons documented, may help them to understand the efforts of nursing staff to ensure the required care, as well as to modulate their expectations in times of resource scarcity, and to act in support of nurses in their attempts to influence policymakers on how to promote the best care.

AUTHOR CONTRIBUTIONS

This research has been designed and developed by all authors. Stefania Chiappinotto and Alvisa Palese collected the data and carried out the analysis. Results have been interpreted and discussed by Stefania Chiappinotto and Alvisa Palese. Stefania Chiappinotto and Alvisa Palese wrote the first version of the paper. Then, all authors read, commented on, made edits and approved the final manuscript.

ACKNOWLEDGEMENTS

The authors would like to thank the nurses who helped to select patients for interviews. This research received no specific grant from any funding agency in the public, commercial or not-for-profit sectors.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on reasonable request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

DETAILS

Subject:	Qualitative analysis; Qualitative research; Health care facilities; Delayed; Perceptions; Hospitalized; Leadership; Patients; Interviews; Content analysis; Nursing care; Secondary schools; Activities of daily living; Nurses; Nursing; Oral hygiene; Health services
Identifier / keyword:	causes; patient engagement; qualitative research; reasons; unfinished nursing care
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	256-267
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Sch olarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-22
Milestone dates:	2022-10-02 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-07-27 (manuscriptReceived); 2022-11-22 (publishedOnlineEarlyUnpaginated); 2022-10-23 (manuscriptAccepted)
Publication history :	

Acceptability of integrating smoking cessation treatment into routine care for people with mental illness: A qualitative study

Sawyer, Katherine [1] ; Kim Fredman Stein [1] ; Jacobsen, Pamela [2] ; Freeman, Tom P [1] ; Blackwell, Anna K M [1] ; Metcalfe, Chris [3] ; Kessler, David [4] ; Munafò, Marcus R [5] ; Aveyard, Paul [6] ; Taylor, Gemma M J [1]

[1] Department of Psychology, Addiction and Mental Health Group, University of Bath, Bath, UK [2] Department of Psychology, Addiction and Mental Health Group, Bath Centre for Mindfulness and Compassion, University of Bath, Bath, UK [3] Bristol Randomised Trials Collaboration, Population Health Sciences, Bristol Medical School, Bristol, UK [4] Department of Population Health Sciences, Bristol Medical School, Centre for Academic Primary Care, University of Bristol, Bristol, UK [5] MRC Integrative Epidemiology Unit, School of Psychological Science, University of Bristol, Bristol, UK [6] Nuffield Department of Primary Care Health Sciences, University of Oxford, Oxford, UK

ABSTRACT (ENGLISH)

Introduction

Improving Access to Psychological Therapies (IAPTs) Services could offer smoking cessation treatment to improve physical and psychological outcomes for service users, but it currently does not. This study aimed to understand participants' views and experiences of receiving a novel smoking cessation intervention as part of the ESCAPE trial (intEgrating Smoking Cessation treatment As part of usual Psychological care for dEpression and anxiety). We used the Capability, Opportunity and Motivation Model of Behaviour (COM-B) to understand the (i) acceptability of the integrated smoking cessation treatment, (ii) views of psychological well-being practitioners' (PWPs) ability to deliver

the smoking cessation treatment and (iii) positive and negative impacts of smoking cessation treatment.
Methods
This was a qualitative study embedded within a feasibility randomized-controlled trial (ESCAPE) in primary care services in the United Kingdom (IAPT). Thirty-six participants (53% female) from both usual care and intervention arms of the ESCAPE trial, including both quitters and nonquitters, were interviewed using semi-structured interviews. Data were analysed using a framework approach to thematic analysis, using the COM-B as a theoretical frame.
Results
Psychological Capability: Integrated smoking cessation treatment was acceptable and encouraged participants to reflect on their mental health. Some participants found it difficult to understand nicotine withdrawal symptoms. Motivation: Participants were open to change during the event of presenting to IAPT. Some described being motivated to take part in the intervention by curiosity, to see whether quitting smoking would help their mental health. Physical Opportunity: IAPT has a natural infrastructure for supporting integrated treatment, but there were some barriers such as session duration and interventions feeling segmented. Social Opportunity: Participants viewed PWPs as having good interpersonal skills to deliver a smoking cessation intervention.
Conclusion
People with common mental illness generally accepted integrated smoking cessation and mental health treatment. Smoking cessation treatment fits well within IAPT's structure; however, there are barriers to implementation.
Patient or Public Contribution
Before data collection, we consulted with people with lived experience of smoking and/or mental illness and lay public members regarding the aims, design and interview schedules. After analysis, two people with lived experience of smoking and mental illness individually gave feedback on the final themes and quotes.

FULL TEXT

INTRODUCTION

Smoking is the world's leading cause of cancer and death worldwide.[1–3] People with common mental illness, such as depression and anxiety, are twice as likely to smoke than those without common mental illness. In the United Kingdom, smoking prevalence in people with depression or anxiety is 32% compared to 14.1% in the general population.[4,5] People with mental illnesses have a 19% reduction in the odds of achieving abstinence when trying to quit,[6] but are as motivated to quit as those without mental illness.[7] These differences increase mortality in people with mental illness when compared to the general population resulting from cancer (mortality rate ratio: 1.92; 95% confidence interval [CI]: 1.91–1.94)[8] and cardiovascular disease (mortality hazard ratio: 1.85; 95% CI: 1.53–2.24).[9] Integrating cessation treatment into mental health settings could prevent 78,000 deaths in the next 80 years.[10] People with mental illness may use smoking to try to alleviate symptoms, for example, using smoking to relax when they feel anxious;[11] recent evidence suggests that this is counter-productive, as smoking can in fact exacerbate and maintain mental health symptoms, and stopping smoking can improve mental health.[12–15] Qualitative studies suggest that although people with mental illness do report perceived benefits of smoking, they also accept evidence that smoking tobacco may harm mental health, and quitting might benefit mental health, and suggest that framing cessation as a treatment for mental health could motivate them to quit.[11]

A cochrane review of smoking cessation treatments for people with current and historical depression found that adding psychosocial mood management to usual smoking cessation treatment (e.g., nicotine replacement therapy) increased cessation rates when compared to usual smoking treatment alone (risk ratio: 1.47; 95% CI: 1.13–1.92). In the United Kingdom, people with depression/anxiety can access psychological therapy services, known as ‘Improving Access to Psychological Therapies’ (IAPTs), in which service users receive evidence-based therapies to improve mood and well-being. IAPT receives over 1.5 million referrals a year,[16] and could offer smoking cessation treatment, but it currently does not. Integrating smoking cessation support within IAPT treatment for mental illness could improve physical and psychological outcomes for its service users. The World Health Organization recommends that countries integrate smoking cessation interventions into primary care services, such

as IAPT.[17]

Qualitative studies embedded within randomized-controlled trials (RCTs) provide the potential to gain new understandings of participant experiences of an intervention and inform the development of future interventions.[18] We have recently codesigned a smoking cessation intervention with IAPT staff and service users, and are testing the intervention in a large, acceptability and feasibility pilot RCT.[11,19] We conducted interviews with trial participants to understand their experiences and views of the integrated smoking cessation intervention. We used the Capability, Opportunity and Motivation Model of Behaviour (COM-B)[20] to understand the:

•1.

Acceptability of the integrated smoking cessation treatment.

•2.

Views of psychological well-being practitioners' (PWPs') ability to deliver the smoking cessation treatment.

•3.

Positive and negative impacts of smoking cessation treatment.

METHODS

This study was embedded within a feasibility RCT, prospectively registered on the ISRCTN registry (ISRCTN99531779).[19] The data are available to bona fide researchers via successful application to the University of Bath.

We have followed COREQ reporting guidelines.[21] Ethical approval for this study was received from the NHS Research Ethics Committee and the Health Research Authority on 19 March 2018.

Setting and participants

We conducted semi-structured interviews with IAPT service users taking part in the ESCAPE Trial, involving four NHS trusts in the United Kingdom. Further details are described in the study protocol.[19] In the ESCAPE Trial, intervention and control groups received usual care as part of IAPT (psychological therapy, such as Cognitive Behavioural Therapy (CBT), motivational interviewing etc.), lasting around 30–60 min. In the intervention group only, participants received smoking cessation support integrated within their IAPT sessions. The intervention included behavioural, psychological and pharmacological support adapted from the National Centre for Smoking Cessation and Training's (NCSCT) standard treatment programme.[22]

Recruitment procedure

We used a convenience sampling method. During trial follow-up, we asked participants if they would like to take part in an interview about their experience in the study. Participants who had withdrawn from the study or did not complete follow-ups were not approached for an interview. Informed consent was obtained verbally and recorded at the start of the interview; there was no written consent. The information sheet for the qualitative interviews was combined with the main trial information sheet.

To ensure confidentiality, with informed consent from participants, interviews were recorded using an encrypted digital voice recorder, transcribed and anonymized. Any identifying information in the transcripts was removed, but considering the risk of reidentification, researchers involved in the study were bound to confidentiality regulations set by the University of Bath and NHS. To further protect confidentiality, access to anonymized transcript data is restricted to only approved bona fide researchers after application to the University of Bath's Research Data Archive.

Sample size and selection criteria

For entry into the trial, participants fulfilled the eligibility criteria for IAPT and were daily tobacco smokers (see the trial protocol for details[19]). All trial participants were eligible for inclusion in the qualitative interviews, regardless of

whether they had quit smoking or not during their participation in the trial; the final sample included both quitters and nonquitters.

We aimed to achieve strong information power.[23] Information power was used based on the aim of the study being broad, sample specificity being moderate, use of applied theoretical frameworks (COM-B), with moderate quality of dialogue, and a case and cross-case analysis strategy.[23] We agreed as a team that we reached strong information power at 36 participants.

Interviews

Interviews were conducted between October 2018 and February 2021 over the telephone and lasted approximately 30–60 min. We used flexible interview schedules and open-ended questioning (Supporting Information: Appendix S1). Interview schedules were modified as necessary throughout the course of the interviews to explore newly occurring concepts and experiences. Interviewers (K. S. and K. F. S.) kept notes to capture any relevant codes or concepts for analysis. Participants were not paid for the interview.

AnalysisAnalytic approach

Two researchers (K. S. and G. T.) conducted the analysis and held a critical realist perspective. Data were analysed using a framework approach to thematic analysis, following Braun and Clarke's[24] method, with both deductive and inductive coding. This method was chosen as we aimed to compare the commonalities and differences in experiences of integrated treatment and relationships between experiences, both across cases and within individual cases.[25] Deductive codes were informed by the COM-B where appropriate; if constructs of the COM-B were not identified in the data, they were not included in the final theme structure.[20] Inductive codes were data-driven and remained close to participants' language where possible. An example of data coded inductively and deductively can be found in Supporting Information: Appendix S2. The software used for data analysis were Microsoft Word and Excel.

Coding process and how themes were identified

One researcher (K. S.) read each transcript and listened to the audio recordings, followed by inductive line-by-line coding. After coding three transcripts, K. S. iteratively developed a data-driven coding frame and sought feedback from the second researcher (G. T.). K. S. then grouped codes into categories, providing a working analytical framework that reflected the aims of the study, which were reviewed with G. T., and some inductive codes were added. K. S. then deductively coded the data based on the concepts from the COM-B model, with some data being coded both inductively and deductively. K. S. then actively identified themes relating to study objectives, developed around the COM-B model, which were reviewed and agreed with the wider team.

Reflexivity

Being aware of our own bias as researchers running and working on the ESCAPE trial, K. S. and G. T. kept notes and regularly checked in to discuss bias and the codes/themes being identified. Being aware of our own biases towards believing that the trial/therapy might succeed, we aimed to ensure that both positive and negative experiences and any deviant experiences are reflected in the results, such that themes are not necessarily all based on number. K. S. and G. T. both identify as females, K. S. has never smoked, G. T. is an ex-smoker and K. S. and G. T. have not received mental health therapy in IAPT before.

Patient and public involvement

Before data collection, we consulted with people with lived experience of smoking and/or mental health problems and lay public members regarding the design of aims and interview schedules. After analysis, two people with lived experience of smoking and mental illness individually gave feedback on the final themes and quotes.

RESULTSParticipant characteristics

We invited 49 trial participants to take part in qualitative interviews. Thirteen declined to participate or did not answer recruitment calls, with the remaining 36 completing an interview at either the 3-month or the 6-month follow-up, one person requested to be interviewed at both the 3- and 6-month follow-up time points. The mean age of the participants was 36.89 years (range: 20–65), 19/36 (53%) were female and the majority were White (92%) (Table 1). Most participants came from the Oxford Health NHS Foundation Trust (72%). Further demographic details are reported in Table 1, and the participant characteristics of in-text quotations are available in Supporting Information: Appendix S3. Additional quotes relating to the study themes can be found in Supporting Information: Appendix S4.

Table 1 Clinical and demographic characteristics of the participants

	n (%)
Gender	
Female	19 (53)
Age (*M*/SD)	36.9 (11.5)
Ethnicity	
Other	31 (8)
White	33 (92)
Highest education	
A-level equivalent	2 (5.6)
Apprenticeship	3 (8.3)
Degree	13 (36.1)
GCSE equivalent	5 (13.9)
Higher degree	6 (16.7)
Other vocational	7 (19.4)
Smoking status at Interview	
Quit (100% bioverified)	10 (27.8)
Smoking	26 (72.2)
Follow-up interviewed at	

3 months	21 (58.3)
6 months	14 (38.9)
3 and 6 months	1 (2.8)
Pretreatment mental health (*M* [SD])	
Patient Health Questionaire-9[26]	14.2 (6.2)
General Anxiety Disorder-7[27]	12.4 (4.7)
Mental health at follow-up (*M* [SD])	
Patient Health Questionaire-9[26]	9.4 (5.7)
General Anxiety Disorder-7[27]	8.5 (5.6)
Comorbid health conditions[a]	
Anxiety	13 (36.1)
Panic attacks	4 (11.1)
Obsessive-compulsive disorder	2 (5.6)
Depression	2 (5.6)
Insomnia	1 (2.8)
None diagnosed	17 (47.2)

Abbreviation: GCSE, general certificate of secondary education (public exams in UK taken around age 16).

a

Participants could have had more than one comorbid health condition.

We identified four themes and nine subthemes (Table 2).

Table 2 Themes and subthemes

Theme (mapped onto the COM-B framework)	Subtheme	Meets study aim

Theme 1: Psychological Capability Participants' psychological capability to accept and engage with integrated smoking cessation and IAPT treatment in the context of knowledge and stamina.	Subtheme 1.1 Integration supports mental health treatment and understanding	1, 3
	Subtheme 1.2: Knowledge and understanding of tobacco withdrawal	1, 3
Theme 2: Motivation Participants' reflective motivation and decisions to engage in integrated smoking cessation and IAPT treatment.	Subtheme 2.1: Openness to change when presenting to IAPT	1
	Subtheme 2.2: Curiosity and evaluation of previous quit attempts	1
Theme 3: Physical Opportunity Participants' physical opportunity to accept and engage in integrated smoking cessation and IAPT treatment. Physical opportunities are provided by environmental resources and time.	Subtheme 3:1 IAPT structure facilitates smoking cessation support	1
	Subtheme 3.2: Service-level barriers to integrated treatment	1, 2
	Subtheme 3.3: Introducing an opportunity to quit	1
Theme 4: Social opportunity Participants' social opportunity to engage in integrated smoking cessation and IAPT treatment provided by interpersonal and social influences.	Subtheme 4.1: The value of the therapist–client alliance	2
	Subtheme 4.2: Holistic reflections on mental health and smoking experiences	2

Abbreviations: COM-B, Capability, Opportunity and Motivation Model of Behaviour; IAPT, Improving Access to Psychological Therapies.

Theme 1: Psychological capabilitySubtheme 1.1: Integration supports mental health treatment and understanding

Participants described how they thought IAPT treatment models, such as CBT, ‘worked well’ with quitting smoking and how the integrated treatment benefitted mental health recovery.

… I think it was a really good thing to have because now I think about it, quitting and also having the support with CBT is something that are probably something that go quite well together, hand in hand. Had I not quit smoking, I don't know whether the CBT would have had as much of an impact as it did or vice versa, so I think they worked really well together. (Record 64)

Participants perceived that once the link between smoking and mental health was explained by the therapist, it ‘made sense’ that smoking and anxiety were related. Participants described how smoking acted as a ‘component’ of their anxiety, as a ‘temporary relief’, which then made them ‘feel worse’. Some participants described the physical

effects of smoking, such as increased heart rate, relating this to anxiety in a ‘vicious cycle’.

Subtheme 1.2: Knowledge and understanding of tobacco withdrawal

Some participants found that the integrated treatment affected their knowledge and capability to understand nicotine withdrawal symptoms, craving and anxiety. Participants described that they found it difficult to differentiate between withdrawal symptoms and mental health symptoms.

The downside about quitting in conjunction I'd say for me anyway was that there is a big kind of flashing question mark, am I feeling rubbish because I'm quitting smoking, or is it because I'm depressed? Not being able to answer that it's a bit problematic. I've just come to the conclusion that there's no way of knowing, I have to think of them as two totally separate things. (Record 50)

Some participants described craving and mental illness as two separate things: craving as a ‘niggling feeling’ and a want or need to go and smoke, whereas anxiety was described as overthinking, feeling panicky and nervous.

The craving to me is like a niggling little feeling at the back of the mind, ‘I really want a cigarette, I really want this’. It's a monkey on my back that's basically telling me I need to go and have one. When it comes to mental illness, for me it's been over-thinking things. Completely over-thinking things. It's the anxiety in the way I sort of panic over different bits and pieces…because when I'm feeling anxious I relate smoking to relaxing. That's how it's been previously. I don't know if that makes sense? (Record 89)

Theme 2: MotivationSubtheme 2.1: Openness to change when presenting to IAPT

When asked about their reasons for signing up to the integrated smoking cessation and IAPT treatment, participants reflected on their motivations and appeared to be open to change when first presenting to IAPT.

Yeah, I thought it would be quite interesting to see whether or not it was possible to do the two at the same time, because obviously people tend to use smoking when they're stressed or worried about something, and if you're receiving treatment for anxiety and low moods it's kind of can you do both at the same time, or is it you have to focus on one? (Record 51)

Participants described that they were pursuing a ‘change everything approach’, while they were presenting to the IAPT service. They were trying to change their mental state, their life, trying to ‘better themselves’. One participant described that at the time everything was changing so it was a ‘good a time as any’ to engage in both smoking cessation and mental health treatment. This suggests that participants accept integrated smoking cessation and IAPT treatment, and view presenting to mental health services as a good time to do both, although some participants did not find it helpful to address smoking and mental health together.

…because you're focusing on trying to get your mental health better. At the start it seems you're less worried about quitting smoking. (Record 65)

Subtheme 2.2: Curiosity and evaluation of previous quit attempts

Many participants reflected on previous quit attempts, how some had been unsuccessful and described this as a motivation to try integrated treatment to see if it would help them successfully quit smoking. Participants described being curious to see whether quitting smoking would help their mental health and whether it would be possible to do both smoking cessation and mental health therapy at the same time. Many participants described how they had ‘tried a couple of avenues before which haven't worked’ and so thought ‘Why not try another to see if it helps …?’.

I thought it was also an opportunity. I was kind of curious to see as well if the premise of quit smoking, less anxiety helps your mental health, I wanted to kind of see for myself. And I would work at it as well if it was something tangible that I could see as well. So, curiosity. (Record 50)

Theme 3: Physical opportunitySubtheme 3:1 IAPT structure facilitates smoking cessation support

Participants described how having smoking cessation treatment integrated within IAPT treatment facilitated the

opportunity to talk about any challenges when trying to quit. Participants said that having regular IAPT appointments offered a structure and opportunity to discuss smoking cessation regularly, which was ‘helpful’ and any issues did not have to ‘fester for too long’.

There's nothing I didn't like about it. What I did like about it is the fact that we, we had the ability to talk about it every month, so during the sessions that I was talking to [Name] anyway, I knew that there was going to be a certain period of time where we would sit and go through any issues that there were and anything along those lines and I knew that there was support there if I needed it. (Record 43)

The smoking cessation intervention fitted will within IAPT's delivery method of treatment programmes. Participants described how they found having integrated treatment over the phone ‘helpful’ and easier to fit into their daily lives.

Subtheme 3.2: Service-level barriers to integrated treatment

Some participants described how the length of time between appointments was too long and appointments were needed more frequently. Some participants described how there was a lot of content to fit into the appointments, which were often quite short. One participant described how they could sense their session was coming to an end as they felt their therapist was getting ‘stressed trying to fit everything into their appointment’.

I've done two sessions with [organisation] and both times if I'm looking at a clock …I can tell when the speed is going to go up and their speaking, their rate of speech really rises the closer you get to the end. The amount of information that they want from you really drops as the time gets closer …I guess the only thing I would suggest is if they had a bit more time for calls…. (Record 50)

One participant described how it felt ‘strange’ going from mental health to smoking cessation treatment. Another described how they felt that the smoking cessation treatment was an ‘add on’ and suggested scheduling smoking cessation treatment separately would allow them to give it ‘more emphasis, more thought and importance’.

I didn't feel like the two… you know I said they felt different. I think it's useful to have them at the same time, but they were noticeably different in that delivering the CBT for the health anxiety is what this person does normally, and the intervention for smoking cessation isn't, and that felt kind of noticeable. But if that could be tackled, or maybe if they were merged a bit more, then perhaps it would be useful. I suppose I've got a really specific type of anxiety which if they were merged a bit better it could be really helpful. But not everybody has that. (Record 41)

One participant also described their integrated treatment as ‘scripted’ and ‘unnatural’. Some participants described how setting clear objectives and an agenda with their therapist at the start of their session helped with the integration of the smoking cessation and IAPT treatment.

Subtheme 3.3: Introducing an opportunity to quit

Many participants described how having the opportunity to access smoking cessation treatment prompted them to take part. One participant described how the smoking cessation support being available and offered whilst already seeking help for something else was important. They described how they accepted the offer in a ‘change of life scenario’ and decided to give the integrated treatment a go.

I think it was the fact that I was seeking help for something else and this was an added benefit, so it was like I needed help for something and the offer was there to help me stop. (Record 43)

Theme 4: Social opportunitySubtheme 4.1: The value of the therapist–client alliance

One participant described how the relationship with the PWP helped remove self-blame around smoking as they could discuss their smoking as a coping mechanism in the context of their anxiety. Having the integrated treatment was described as a more positive and helpful experience than smoking cessation treatment alone.

Participants described how their PWPs encouraged them to make decisions and choices in their quit attempt, being guided in an encouraging and positive manner. Participants also emphasized how having to check in with their

therapist regularly prompted them to remain abstinent from smoking as they did not want to let them down. Participants described how they were encouraged when they had had a 'slip up'.

She was very gentle, and I think she was very encouraging and very positive, but it was very much, I feel it was subtly getting me to make the decisions and getting me to make the choices, while acknowledging that these are all going to be good, she never actually said, 'You must stop smoking', it was always, 'What benefits can you see from it? Can you think about why you don't stop, why you want to stop?', it was very much guiding rather than leading. At the beginning there were hiccups, there was no judgement or condemnation, it was just, 'These things happen, don't worry about it, it doesn't mean that you can't have another go', and it was that, it was validating in a way that it was okay to slip up, but that doesn't negate having another go. (Record 30)

Subtheme 4.2: Holistic reflections on mental health and smoking experiences

Participants described how having the integrated treatment allowed them to share a whole picture of their mental health, and how their smoking was affected by their mental health and other stressors. Participants described how their therapist was 'supportive but not like lecture-y' and their therapist tried to understand their smoking as part of their anxiety, 'which no one's ever done before, so yeah, that was really helpful'. One participant said that the therapist understood what they were going through and were patient with them.

Because she knew the difficulties I was going through as well, so rather than it being somebody talking to me from [service] and then somebody talking to me about my smoking, having two separate people, because it was the one person, she understood fully the struggles that life was bringing me, as well as trying to help me stop smoking, rather than feeling that…. (Record 20)

Participants also felt like their smoking cessation support was tailored to them, compared to people who accessed an NHS stop smoking service, who felt like they were treated as a 'generic smoker', for example, being provided with information about the products available rather than identifying what would work for them as an individual.

DISCUSSIONSummary

We aimed to understand the experience of an integrated smoking cessation and mental health treatment among people with common mental illness. We found that generally, people with mental illness accepted integrated smoking cessation and mental health treatment, and had the psychological capability, motivation, physical and social opportunity to accept and engage with the integrated treatment. However, participants also faced several barriers in understanding tobacco withdrawal and at the service level. Participants described how PWPs had the interpersonal skills for delivering the smoking cessation intervention, but it sometimes seemed scripted or unnatural.

Strength and limitations

A strength of this study is that the findings are likely transferable to other primary care services, or similar services and populations, as the services involved in this study used nationally standardized treatments and service models, such as CBT, motivational interviewing and the NCSCT's standard treatment programme.[22,28] Most participants were White British, which was representative of the communities served by the NHS Trusts that we sampled,[16] but the findings may not be generalizable to more ethnically diverse areas of the United Kingdom. However, our sample was broadly representative of general IAPT users in England, being mostly white, female and a younger age. Although the sample in this study is slightly older (average age 36.9 years) than the general IAPT service user population, those aged 18–24 years are most likely to access IAPT.[29] Our sampling method could have introduced bias into the data, as we only sampled from those who completed follow-ups, and it is possible that they could have had a more positive experience of the treatment than those who did not. Similarly, most participants completed interviews when they had completed treatment, so could have shown a recall bias where they reported mostly positive experiences of the intervention due to feeling more positive at the end of treatment, forgetting negative experiences.[30]

Use of a critical realist perspective allowed us to focus on understanding, instead of describing, social reality. Mental health treatment happens within a social reality as people and their actions influence the treatment pathways, and each is made up of, and influenced by, people's actions.[31–33] A critical realist perspective assumes that human perceptions are accounts of reality, as what we observe is a social and subjective account of reality.[31] Critical realism allows us to understand how and why interventions work within complex environments such as primary care mental health services.[31] The benefits of a critical realist perspective are that we can understand the relationship between context (the setting of an intervention), mechanisms (things that cause change) and the outcome or experience of an intervention.[31] Therefore, by using this perspective, we were able to understand how the experience of the smoking cessation intervention integrated within an IAPT service was influenced by the context in which it was experienced, and why or why not it was accepted in this environment.

Comparison with the existing literature

Integrated smoking cessation and mental health treatment was generally accepted, and participants had the capability to understand their smoking behaviour in the context of the tobacco withdrawal cycle and engage in treatment. These findings further those from our qualitative study of IAPT patients' views of integrated treatment, which found that IAPT patients accepted evidence that smoking may worsen their mental health and that quitting could improve their mental health.[11] However, similar to other research, some participants described how they used smoking as a coping mechanism and prioritized their mental health treatment over quitting smoking.[34,35]

Although most participants described their smoking using a CBT model and identified withdrawal as a component of their mental illness, some participants described withdrawal and mental illness as different experiences. This contrasts with the literature on the tobacco withdrawal cycle, which suggests that irritability and low mood from nicotine withdrawal are the same experience as mental illness.[36,37]

This study found that an important factor for smoking cessation intervention uptake is having smoking cessation treatment available and offered in primary care services. This finding is in line with a systematic review that found that offering all smokers help to quit increased quit attempts, compared to telling them to quit.[38] This study supports findings from our recent study, which suggests that IAPT services could be a suitable infrastructure for smoking cessation treatment, but there may be some service-level barriers.[11] Similar to a qualitative study of smoking cessation therapy for people with severe mental illness, we found that service users viewed PWPs as having good interpersonal skills to deliver smoking cessation interventions.[39]

Participants were motivated to accept and engage with the integrated smoking cessation and IAPT therapy, consistent with a systematic review indicating that people with mental illness are motivated to quit smoking.[7] Participants stated that receiving smoking cessation treatment at the same time as mental health treatment was ‘a good a time as any’. These findings challenge health care professionals' views that quitting smoking at the same time is too much for people with mental illness.[35,40]

Previous literature has suggested that altruism is an important motivator for participation in mental health trials.[41,42] The key motivators for engagement identified in the present study were participants' openness to change and curiosity regarding the potential impact of the combined treatment approach for smoking and their mental health. These motivations reflect a primary interest in the personal benefits of taking part rather than a desire to help others. These findings are consistent with the idea of ‘conditional altruism’,[43,44] which suggests that an interest in helping others may facilitate initial engagement; however, an expectation of some personal benefit is an important driver for enrolment and subsequent participation in trials.

Implications for research and practice

Integrating the NCSCT's standard treatment programme for smoking cessation[22] into IAPT services is possible and

accepted by people with common mental illness. Although there are financial considerations for IAPT to provide smoking cessation support on a larger scale, with funding required for training therapists, buying equipment, and so forth, it is important to remember that in the United Kimgdom, smoking costs our economy >£11bn per year.[45] Given that participants reported that sometimes the intervention felt rushed or scripted, it could be that further intervention refinement is required, or that IAPT services should reduce PWP caseload to lengthen the session duration. In this trial, smoking was treated as a separate intervention programme; for truly integrated treatment, smoking should be addressed synonymously with other lifestyle behaviours. Future research should investigate how to achieve this.

CONCLUSIONS

People with common mental illness generally accepted integrated smoking cessation and mental health treatment. Smoking cessation treatment fits well within IAPT's structure; however, there are barriers to implementation related to resources. Reducing caseloads to allow for longer sessions with smokers would support implementation. Participants were open to change when first presenting to IAPT and motivated by curiosity to see whether quitting smoking would help their mental health. Participants viewed PWPs as having good interpersonal skills to deliver smoking cessation intervention.

AUTHOR CONTRIBUTIONS

Ms Katherine Sawyer led on data curation, investigation, project administration, data analysis, writing and editing the manuscript. Dr Kim Fredman Stein contributed to data curation, investigation, project administration and data analysis. Dr Pamela Jacobsen supervised the analysis and investigation, and contributed to writing and editing the manuscript. Dr Tom P. Freeman supervised the analysis and investigation, and contributed to writing and editing the manuscript. Dr Anna K. M. Blackwell made a substantial contribution to interpretation of data, and writing and editing the manuscript. Prof Chris Metcalfe contributed to study conceptualization, methodology, analysis, investigation, writing and editing of the manuscript and funding acquisition. Prof David Kessler contributed to study conceptualization, methodology, analysis, investigation, writing and editing of the manuscript and funding acquisition. Prof Marcus Munafò supervised study conceptualization, investigation and funding acquisition, and contributed to the study methodology, analysis, writing and editing of the manuscript and project administration. Prof Paul Aveyard supervised study conceptualization, analysis, investigation, writing and editing of the manuscript and funding acquisition, and contributed to methodology and project administration. Dr Gemma Taylor led on study conceptualization, methodology and funding acquisition, contributed to analysis, investigation, administration, data curation, editing the manuscript and supervision of analysis.

ACKNOWLEDGEMENTS

The authors would like to thank everyone who took part in the ESCAPE Trial. This study was funded by Dr Gemma Taylor's Cancer Research UK Postdoctoral Fellowship (C56067/A21330).

CONFLICTS OF INTEREST

Marcus Munafò and Gemma Taylor previously received funding from Pfizer, who manufacture smoking cessation products, for research unrelated to this study. No other authors have any potential conflict of interest to declare.

DATA AVAILABILITY STATEMENT

Anonymized transcript data are available via application to the University of Bath Research Data Archive.

ETHICS STATEMENT

Ethics approval for this study was received from the NHS Research Ethics Committee and the Health Research Authority on 19 March 2018, IRAS ID 239339.

DETAILS

Subject:	Intervention; Mental health; Mental depression; Smoking; Mortality; Psychological factors; Mental disorders; Health status; Nicotine; Feasibility studies; Primary care; Capabilities; Motivation; Mental health services; Curiosity; Infrastructure; Illnesses; Well being; Feasibility; Withdrawal symptoms; Data collection; Psychological theories; Smoking cessation; Anxiety; Escape; Drug addiction; Withdrawal; Interviews; Bias; Consent; Health care; Psychological distress; Confidentiality; Cigarette smoking; Soft skills; Psychological well being; Qualitative research; Acceptability
Location:	United Kingdom--UK
Company / organization:	Name: University of Bath; NAICS: 611310
Identifier / keyword:	anxiety; depression; IAPT; primary health care; qualitative; smoking cessation
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	108-118
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-10-12

Milestone dates:	2022-07-28 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-04-13 (manuscriptReceived); 2022-10-12 (publishedOnlineEarlyUnpaginated); 2022-07-29 (manuscriptAccepted)
Publication history :	
First posting date:	12 Oct 2022
DOI:	https://doi.org/10.1111/hex.13580
ProQuest document ID:	2767101878
Document URL:	https://www.proquest.com/scholarly-journals/acceptability-integrating-smoking-cessation/docview/2767101878/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the "License"). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-11-08
Database:	Public Health Database,Publicly Available Content Database

Document 33 of 46

Understanding the cultural environment of the outpatient care setting for patients with dementia receiving cancer treatment: A qualitative study

Farrington, Naomi [1]
; Dantanus, Katherine [2] ; Richardson, Alison [1]
; Bridges, Jackie [3]
[1] Cancer Care, Division B, University Hospital Southampton NHSFT and Health Sciences, University of Southampton, Southampton, UK [2] Cancer Care, Division B, University Hospital Southampton NHSFT, Southampton, UK [3] Health Sciences, University of Southampton, Southampton, UK

ABSTRACT (ENGLISH)

Introduction

People with dementia have poorer cancer outcomes than those without dementia, and experience inequalities in access to, and quality of, care. Outpatient environments, where radiotherapy, chemotherapy and immunotherapy cancer treatments typically take place, have largely been excluded from research. This study was conducted to understand provision of treatment and support and experiences of care for people with dementia undergoing cancer treatment in the outpatient setting.

Materials and Methods

Using observation, interviews and document analysis, data were collected to scrutinize the cultural environment of ambulatory care, comprising the physical fabric of the care setting; interactions, behaviours and perceptions of those in the care setting; and the organizational, clinical and interactional processes involved in care delivery. The study was conducted in the outpatient oncology departments of two large teaching hospitals in England between January 2019 and July 2021.

Results

Data were gathered from a wide range of sources, including 15 h of observation, and interviews with patients (n=2), caregivers (n=7) and staff (n=20). Evidence from this study suggests that the cultural environment of the outpatient care setting reflects and supports the standardized processing of people for cancer treatment. Dementia introduces a wider set of care requirements not catered for by this standardized treatment model and associated processes. Data showed that the needs of patients with dementia could be addressed most effectively when individualized care, as opposed to standardized care, was offered.

Conclusion

There is work to be done in outpatient cancer services to ensure responsiveness to individual patient need. This could be achieved by having an established way (or ways) of eliciting needs, preferences and expectations, a belief that a person's needs and expectations are legitimate and that effort should be made to address them, with the ability to accommodate these needs and expectations.

Patient or Public Contribution

Patients and caregivers were involved in the study design and development of study materials including the interview topic guide. They also assisted with discussion and clarification of study findings.

FULL TEXT

INTRODUCTION

People with dementia have poorer cancer outcomes than those without dementia.[1,2] A scoping review[3] identified limited interventions to support older people with complex needs having cancer treatment. Good-quality evidence is lacking regarding the implications of comorbid cancer–dementia for people receiving cancer treatment, and little information exists about how the needs of this population are managed by healthcare teams.[4]

People with dementia experience inequalities in access to, and quality of, care.[5,6] There is a focus on the disease instead of the whole person, and a focus on a single disease alone, which can be disadvantageous for people with multimorbidity. People with dementia frequently feel denied, ignored or experience discrimination in healthcare[7] either as a direct result of the stigma associated with the diagnosis or through indirect mechanisms such as failure to provide inclusive services. It is increasingly being recognized that a tailored approach to multimorbidity is required,[8] enabling individual preferences and circumstances to be addressed.

Efforts have been made to ensure that people with dementia have access to services that meet their needs by establishing care standards.[9] A body of work exists on improving care environments for people with dementia in hospital as inpatients or in residential care.[10,11] Outpatient environments, where radiotherapy, chemotherapy and immunotherapy cancer treatments typically take place, have largely been excluded from these efforts, although evidence is emerging.[12] Care and treatment in an outpatient environment are different from that of an inpatient stay. Outpatient care involves a series of discrete interactions; patients attend the service for specific appointments, such as consultations and treatment, and in between return home. In addition, the patient (or caregiver) is expected to accept responsibility for coordinating appointments and treatment, as well as monitoring their own health and well-being for treatment-related toxicities. Outpatient treatment, while reducing time at the hospital, brings other challenges.

This study investigated the provision of treatment and support and the experiences of care for people with dementia undergoing cancer treatment in the cultural environment of the outpatient setting. It aimed to establish an empirically based conceptual foundation to inform development of innovations in service delivery, and improve the way in which treatment and support are offered to this group.

MATERIALS AND METHODS

Data were collected to scrutinize the cultural environment of outpatient cancer care characterized through the interactions, behaviours and perceptions of those in the care setting; organizational, clinical and interactional processes involved in care delivery; and the physical fabric of the care setting. Data were used to identify principles and characteristics that constitute 'good care', understand barriers and facilitators and identify aspects amenable to modification to meet the needs of this complex population. Study design was influenced by focused ethnography, which enables focus on a distinct issue or shared experience in a specific setting.[13–15] It has been used successfully in nursing research,[16,17] particularly when the researcher is known and trusted, and holds a 'privileged observer' position.[18] Two of the researchers (N. F. and K. D.) were nurses in departments involved in the study. Insights afforded to them by their clinical roles were invaluable in understanding care delivery.

The study was conducted in the outpatient oncology departments of two teaching hospitals in England. Data were collected from January 2019 to July 2021. Observation, semi-structured interviews and examination of patient case notes were used in a focused manner by two researchers (N. F., K. D.). Participants included patients, informal caregivers and healthcare staff (see Table 1). The process is described below; however, the study included more than the recruited participants and interviews. Ethnographic data cover a broad ontological range from 'hard, objective' documents, to 'soft, subjective' memories and experiences.[19] The field researchers (N. F. and K. D.) internalized the research aims for the period of data collection, and interpretation and understanding continued outside the assigned research time. The findings reflect a wider data field than the formal data collection opportunities described, as the researchers constantly participated in the cultural life of the cancer care outpatient services as clinicians as well as researchers.

Table 1 Inclusion and exclusion criteria for interviews.

	Inclusion	Exclusion
Patient participant	•Adult, aged over 18 years. •Diagnosis of any cancer. •Undergoing cancer treatment (radiotherapy or chemotherapy or other SACT delivered via any route) OR have finished treatment within the last 6 months. •Cancer treatment administered in the outpatient care setting. •Diagnosis of dementia of any type. •Mental capacity to decide to take part in the study.	•Acute or critical illness. •Inability to communicate choices and preferences either verbally or nonverbally. •No confirmed diagnosis of dementia. •Cognitive impairment as a result of aetiology not related to dementia.
Other participants	•Adult, aged over 18 years. •Informal carer of patient participant OR healthcare professional/NHS staff involved in the care and management of the patient participant or other patients with dementia having cancer treatment.	

Abbreviations: NHS, National Health Service; SACT, systemic anticancer therapy.

Patient participants

Patient participants were purposively sampled to participate in interviews and observation. The study invited

participation from people with a diagnosis of dementia who were receiving radiotherapy, or systemic anticancer therapy (SACT), or who had completed treatment within 6 months. Patient participants were identified through their clinical teams, and agreement obtained for researchers to approach them (face to face where possible, via telephone during the COVID-19 pandemic). Participation in each of the three data collection methods was not mutually exclusive, nor was it mandatory; patients could be interviewed, observed and consent to document analysis, and alternatively, they could only be observed, or only interviewed. Document analysis was undertaken if a patient participant consented to this at interview.

Caregiver participants

Caregiver participants were family or friends involved in supporting a patient through radiotherapy or SACT. They could participate alongside the patient they were supporting, or alone. As with patient participants, they were identified by the clinical teams, and provided agreement for the researchers to contact them.

Healthcare staff participants

Healthcare staff participants included oncologists, nurses, allied health professionals, support workers and management and administrative staff. Purposive sampling was used to recruit staff involved in the delivery of treatment and support to patients undergoing radiotherapy or SACT. They did not have to be directly providing care to a patient participant.

Data sources

Interviews took place in a private area of the department or at a participant's home (an option offered to patients and caregivers). A topic guide, developed with public and patient involvement volunteers, provided discussion prompts. Participants were invited to describe their experiences. Subsequent interview questions covered the treatment environment, factors that people found challenging and what they found helpful. Interviews were digitally recorded and transcribed verbatim. Where patient participants provided permission, data were extracted from case notes about their journey within and beyond cancer care, and the organizational, clinical and interactional processes involved in care delivery.

During general observations, attention was paid to the environment, behaviour and staff–patient interactions, with a focus on delivery and experiences of care. Focused observations allowed a detailed study of discrete experiences, such as pretreatment consultations. Attention was paid to the environment in which care was experienced and delivered, the behaviours of the people involved and the organizational processes enacted. Descriptive and reflective field notes were captured on an observation record form.

Ethics

Ethical approval was obtained from the South Central–Berkshire Research Ethics Committee (18/SC/0590). Informed consent was obtained for interviews, case note access and focused observations. A verbal explanation of the researchers' presence was provided when requested during general observations. The UK Mental Capacity Act[20] was used to guide researcher assessment of people's capacity to decide to take part in the research. The COREQ reporting criteria checklist for interviews and focus groups[21] informed the writing of this paper.

Analysis was conducted concurrently with data collection[22,23] by two researchers (N. F., K. D.) and discussed and iterated with the wider team once the full data set was available (A. R., J. B.). Based on the constant comparative methodology of grounded theory,[24,25] the process was as follows:

- 1. Initial coding (categorizing data).
- 2. Focused coding (concentrating on significant/frequent codes).
- 3. Theoretical coding (developing relationships between codes).

•4.

Memo-writing (analysing ideas about codes).

•5.

Theoretical saturation.

•6.

Sorting and integrating memos.

Analysis followed the same pattern regardless of the data source type and involved looking for patterns and relationships, as well as inconsistencies and contradictions. Several themes were identified, refined and reviewed to include those that captured the story being told by the data.[26] Data collection ceased when the researchers had achieved adequate depth of understanding to build theory.[27]

RESULTS

Data were gathered from observation (15 h), interviews with patients (*n* = 2), caregivers (*n* = 7) and staff (*n* = 20; see Table 2), document analysis and informal discussions. Interviews lasted between 10 and 42 min. Four patients approached declined participation.

Table 2 Participant details.

Participant	*N*	Subtype
Patient	2	1 Female, 1 male
Caregiver	7	Spouse (3) (2 female, 1 male)
		Child (2) (2 female)
		Sibling-in-law (2) (1 female, 1 male)
Staff	20	Healthcare assistant (1)
		Nurse (9)
		Doctor (3)
		Administrative staff (2)
		Management staff (2)
		Outpatient support staff (1)
		Pharmacy staff (1)
		Radiotherapy staff (1)

Total	*29*	

To set the scene for patient/caregiver experiences and the cultural setting that shapes them, we begin by outlining how relevant services were organized in the departments providing treatment. We then elaborate on the key steps that occur during a patient journey: ‘attending the hospital’; ‘the consultation’; ‘treatment’; and ‘at home’, illustrating each stage. Pseudonyms are used to protect anonymity.

The SACT department included outpatient clinics where patients attend for consultations, clinical areas where patients attend for blood tests and treatment areas. Before the COVID-19 pandemic, patients tended to see a clinician face to face for a consultation (doctor, nurse practitioner or pharmacist), and have a blood test, before either collecting oral treatment from the hospital pharmacy, or returning on a subsequent day for intravenous treatment. This system changed during the pandemic to reduce hospital attendance; most consultations were conducted remotely via telephone or video, blood tests were done at home or at GP surgeries and some treatments were administered at home. However, the overall process remained unaltered during and beyond 2020, regardless of the setting: patients generally need a consultation, a blood test and a treatment appointment. Treatment in the radiotherapy department was not altered by the pandemic, as radiotherapy can only be provided on a hospital site. Patients attended for a pretreatment planning appointment, with a computed tomography scan to determine the specificities of treatment. Patients were then given a treatment schedule, either for a one-off treatment or for a course of treatments. Radiotherapy and SACT treatments were sometimes administered concomitantly. In both areas, patients were discharged home after treatment, with contact details for a 24-h acute oncology service. Interactions between patients and the oncology service were organized to enable diverse patients to progress along these standardized pathways.

Attending the hospital

Participants cited difficulties with attending for hospital appointments. Services were organized and delivered in a standardized way, which could create problems for patients and their accompanying family members. The sensory experience of being in the clinic was shaped by the built environment and the busyness of services, and patients found being there unpleasant and disorienting. These negative experiences were exacerbated by frequently experienced long waits. Alternatives to visiting the hospital were welcomed by some when they were made available, but did not suit everyone's needs.

Participants found it difficult to arrive on time for set clinic appointment times, especially in the morning:

Getting up, breakfasted and all ready and then getting up here and finding a parking space, it's a bit of a panic if you've got to be here for 9.20 am. (Annabel, wife of person with cancer and dementia)

Long waiting times also created problems if people could not be seen in a timely way once they had arrived or had to wait between different parts of the appointment:

He gets confused by the waiting. Well, he has, I'm not quite sure whether he's just not used, he's not used to NHS and being ill and everything like that because he's not an ill person previously, so he can't understand why he has an appointment for say, I don't know, 9 o'clock, and he's still sitting there, you know, sometimes, he could be sitting there an hour later…. (Emily, wife of person with cancer and dementia)

The noisy and busy nature of the clinic environment aggravated the experience of long waits, and features of the built environment heightened patients' disorientation:

It's very busy, it's very noisy, it's very complicated, the doors look the same. Patients can get lost in sitting in a room with doors looking exactly the same everywhere. (Tracy, nurse specialist)

Noisy: staff calling out patient names for height and weight, appointments, treatment, numbers for bloods, phones

ringing, bin lids slamming. Very difficult to tell from which direction the noises are coming from, so sometimes patients are unaware where to go when they are called. (General observation of SACT clinic area)

Staff told us that sometimes the long waiting times were an inevitable part of attending the hospital for cancer treatment:

… when patients come to the hospital there are a number of waits and that could be waiting for blood results, waiting for going in for treatment, seeing the doctor, whatever those waits are, but there are waits built into the service…. But actually, I don't think until you actually come to the hospital and you are actually involved in those waits you can really actually realise how long some of those waits can be. (Maisie, SACT management team)

Efficient accommodation of large volumes of patients was managed by 'overbooking' clinic slots, that is, allocating more than one patient to be seen at a time. This meant that the capacity of clinic staff was exceeded and patients were inevitably delayed beyond the time of their booked appointment. Delays also occurred if patients earlier in the schedule took longer than the scheduled time for their appointment, a frequent occurrence as actual patient need was hard to predict and accommodate in the booking system where all clinic slots were the same length.

Patients and caregivers told us that they appreciated having fewer hospital visits, and we found examples where a reduction in visits had been made possible by appointments held over the telephone:

So we come here on Thursday, and the treatment is the following Tuesday. But now, the doctor's just said [my husband will visit the hospital] every 6 weeks, because he'll get a phone call on the third [week instead of a hospital visit], so that makes it easier. (Emily, wife of person with cancer and dementia)

However, what presented as a welcome solution for some did not fit with what others needed, highlighting how service provision needed to be individualized. Imogen, for instance, found that her father, who had advanced dementia and no longer spoke, was repeatedly sent letters inviting him to a telephone appointment, requiring her to contact the department numerous times to explain that her father was unable to use the telephone.

The frustrations and disempowerment reported by patients and caregivers were the result of a mismatch between the standardized way that services were organized and the needs and preferences of patients and caregivers. Dementia added another layer of complexity: it was harder to get to appointments on time, long waits were more onerous and the physical environment of the clinic was more disorientating. Dementia also complicated uptake of alternatives such as telephone appointments. Our findings also shed light on the impetus behind the way that services are currently organized and the primacy of organizational efficiency, at the expense of patient experience.

The consultation

Experiences of consultations during clinical visits tended to be shaped by the degree of continuity between one visit and the next, and by the extent to which the consultation aligned with patient concerns that may extend beyond cancer. Specialist roles in nursing and in older people's care were identified as having the potential to improve the quality of consultation in relation to actual patient need and to improve continuity between appointments.

Interviewees reported that continuity was disrupted when the staff in the consultation changed between visits:

Yeah, I think just, yeah, we saw different doctors every time, and I know that was quite hard. But it was a case of who's doing what? OK, who's chasing the scan? Is that going to get done? It was just that really, and tying it all together. (Melanie, daughter of person with cancer and dementia)

Case note analysis confirmed high numbers of staff to interact with as a potential challenge. For instance, Katherine had 47 discrete interactions with 24 different clinicians in oncology over 13 months. As the above quote illustrates, caregivers were not always assured that there was one professional with the overview of the case who was making sure that all the elements of care and treatment were appropriate, integrated and consistent. The impact of dementia on communication quality and on memory, in addition to high turnover of clinicians, placed additional burdens on

family caregivers who felt responsible for ensuring that information was transmitted and received during consultations and retained afterwards. Melanie found the lack of consistency with a central person difficult, as she found herself stuck in the middle during consultations with healthcare professionals who had not met her father before:

I think the initial meeting…It didn't seem, it wasn't rushed, but I had to audio record [the meeting], because things got lost, and Mum would hear what she wanted to hear, Dad wasn't listening at all, I was trying to interpret for the doctor and understand Dad, because he was so bad at the beginning, and then we had the nurse in there as well, saying things, that was a bit too much, that was overwhelming.

Emily, supporting her husband with cancer and dementia, found it difficult to remember the names of the different doctors they had seen, referring to one as 'Dr, whose name begins with [x]'. Her husband found the lack of continuity problematic:

He'll say afterwards, or later on, 'I keep seeing different people', and he finds that a bit confusing.

The high numbers of patients needing to be seen meant that individual consultants could not see all their patients every time they came to clinic. Patients who were viewed as continuing well on treatment and having no additional need would instead be seen by others, such as advanced nurse practitioners. The pressure on clinics and their clear remit for cancer also constrained the topic of discussions to the cancer, with more pressing concerns of patients and caregivers not being aired. During a conversation with Paul, who had cancer and dementia, and his wife Annabel, it became apparent that the couple's primary concerns were not about cancer:

Wife becomes very tearful and says that it has been a difficult month. Says that patient's sister has died at the beginning of March from vascular dementia, and that the patient has been diagnosed with vascular dementia yesterday. In addition, he had a driving assessment and is no longer safe to drive. On being reminded of all this the patient starts to become visibly tearful. Wife goes on to describe that the diagnosis of vascular dementia arrived by letter yesterday with no notification or suggestion of follow up. She appears very upset by this. (Field note)

Paul's diagnosis of vascular dementia, and the fact that he could no longer drive, were having a greater impact on Paul's life than his well-controlled cancer. The consulting clinician was sympathetic, but the primary concern was assessing treatment toxicities and prescribing further treatment, as this was where the clinician's expertize and responsibilities lay. Staff participants acknowledged that cancer would not be the only health issue for many patients, and suggested that involvement from specialists in medicine for older people could be beneficial, and allow conditions such as dementia to be addressed alongside cancer treatment:

So you also have a geriatrician involved in their care so you are looking more holistically at everything that is going on with them because they are a group of older people so they don't just have, 90% of the time they don't just have a cancer they have other medical problems going on. (Tracy, specialist nurse)

Caregivers consistently cited the specialist nurse role as key in promoting continuity of cancer treatment:

Yes we're all sorted now, I know that I can ring [specialist nurse] to find out anything about my dad's blood or blood test or anything like that at all I know any questions or anything I can ring and if [specialist nurse] doesn't know she'll find out and then she'll ring me back. So since I found that I have got someone that I can speak to it's a lot better now. (Imogen, daughter of patient with cancer and dementia)

Specialist nurses were able to operate outside of the standardized service processes to address the complexities that dementia added. For instance, Sheila (sister-in-law of a person with cancer and dementia) appreciated the fact that a nurse rang her after consultations to summarize the content. Melanie welcomed the hospital arranging for appointment letters to be sent out both to her and her father's care home, so she was kept up to date with her father's treatment.

These findings highlight that high volumes of patients in relation to consultant capacity were managed by substituting consultants with other staff members. This strategy disrupted continuity between visits, creating confusion and stress for patients and caregivers, and impacting negatively on relationship quality and information transmission. Nurse specialists had the autonomy to operate outside of the standardized service to promote continuity. In addition, the high pressure on clinics and the service focus on cancer alone constrained opportunities for patients and caregivers to raise non-cancer issues, however pressing they were for health and well-being. The broader focus of medicine for older people specialists was cited as having the potential to address such issues, but was not available to patients in the study setting.

Treatment

Patient experiences of receiving treatment were shaped by the readiness of the department to accommodate their dementia at the same time as cancer treatment. We found variation in whether or not departments were notified in advance that someone had dementia but also in the extent to which effort had been put into making the service 'dementia-friendly'.

At one study site, the booking team highlighted people with dementia to radiotherapy staff so that the appointment could be planned accordingly. Staff in other departments told us that they were not always aware that patients had dementia before they arrived:

…because the problem is quite often we're just chasing our tails. We don't know any issues until we've already booked the patient and they walk in the door and suddenly we find that it's not suitable anymore. (Louise, administrative team)

In the radiotherapy department, radiographers with specialist dementia training oversaw the care of patients with dementia. Before the appointment, they contacted family or friends to see how best to support the patient. They then met the patient at the planning appointment, giving them a chance to identify what adaptations might be needed to the treatment plan. In departments without dementia champions or where links were not in place with the hospital's dementia nurse specialists, staff were required to work in a more reactive way, but struggled to provide the quality of care that they felt was needed. If the patient needed more time with staff because of their dementia, staffing levels did not allow for this:

…when you are the nurse who is trying to concentrate and with chemo you've got to really concentrate on checking the bags, the dosage, and sometimes when they're called away for other things there's a risk of error and this is what the nurses say, and you'd hear them say it and I know awful, oh God they said Dorothy is in today. Because she was so labour intensive and it's not the physical side it's the emotional side because she will be constantly saying, why am I here, why am I here. Well again if you are working and you are trying to concentrate and she's constantly, they don't allow for that side of it for nursing staff. (Natalie, healthcare support worker)

Some staff members were not confident that they had the skills for supporting people with dementia:

One of the little nurses in there even turned around and said I've never spoken or dealt with a person with dementia before so I don't really know what I'm supposed to do. (Imogen, daughter of person with cancer and dementia)

I probably wouldn't be confident in knowing where to go and how to find out specifically related to the dementia side rather than the oncology side. (Tracy, specialist nurse)

Accommodating someone's dementia while delivering their cancer treatment depended on the motivation, knowledge and skills of staff in individual departments. Where departments were ready for patients with dementia, an individualized approach could be planned. Where departments were not ready, the resulting standardized approach to staffing and care meant that patient experiences and potentially outcomes were negatively affected.

At home

Our findings highlight the key role that family caregivers play in supporting people with dementia through their cancer treatment, especially in the home setting, and in turn, the ways in which family caregivers can be supported in their role by cancer services. Doctors told us that the presence or absence of supportive family at home affected their decision to provide certain treatments:

I think it's a safety thing from our point of view if we know that we've got a family that's going to remind that patient what the diagnosis is, what the plan is, what the treatment is, then we would feel much safer perhaps prescribing a more intensive treatment that suits them physically than for a patient that has nobody at home that's not going to remind them to take their tablets…. to come in when they're unwell. (Teresa, doctor)

Caregivers found home care challenging during cancer treatment, especially managing medicines, a role made necessary by the memory problems that accompanied dementia:

I have to do all his pills because otherwise they might get forgotten. He has five pills in the morning, two pills at 11 am, another pill after 12 pm and then his evening pills he's got three more during the evening, one with his meal and two before he goes to bed. So it's pill, pill, pill all the time. (Annabel, wife of person with cancer and dementia)

Staff recognized their role in supporting caregivers, including giving them more advance information about the whole treatment plan than would usually be provided:

Yes, I guess that as nurses we've got to care for the patient and also their support network and their loved ones so we've got to really make sure that we're looking after the whole package because if a carer is struggling they're not going to be able to be there for the patient. (Leila, nurse)

A lot of people don't appreciate actually 12 cycles could mean you are into February next year and it doesn't quite compute until you see it on paper and go oh my God. So if you can give them that right from the beginning then they can work to a plan. (Louise, administrative team)

This advanced notice enabled families to plan ahead, but whether or not this information was provided depended on the discretion of the individual staff member.

Our data showed that the needs of patients with dementia (and those of their caregivers) could be addressed most effectively when nonstandardized (individualized) care was offered. Evidence from this study suggests that the cultural environment of the outpatient care setting reflects and supports the standardized processing of people for cancer treatment. Dementia introduced a wider set of requirements that were not catered for by this standardized treatment model. The discussion below considers how healthcare systems could address this gap in provision.

DISCUSSION

This study investigated the provision of treatment and support, and experiences of care for people with dementia receiving cancer treatment in an outpatient setting. Attending the hospital to arrive at a scheduled time and waiting to be seen were stressful for patients and caregivers. Not seeing the same staff member at every visit disrupted informational and relational continuity[28] and was burdensome for caregivers, whose role become one of mitigating for lack of continuity. The focus of consultations on cancer limited opportunities to raise other concerns. If treatment departments were not readied to treat people with dementia, staff stress and poor patient experiences resulted. The findings illustrate how routine, standardized approaches to the organization and delivery of cancer care and treatment were in tension with the needs of people with dementia. A personalized approach was possible when staff had the skills, discretion, flexibility and resources to plan ahead, to elicit what an individual and their caregiver needed, to accommodate health needs beyond cancer and modify plans and treatment to fit individual needs. Participants indicated that nurse specialists, dementia champions and specialists in medicine for older people were helpful ‘anchors’ in this regard, as they had broader skills and knowledge and took into account the totality of a patient's needs. However, there were clear variations in the extent to which such roles were routinely accessible to

people with dementia and their caregivers.

Balancing the needs of a person with dementia and the requirements of a cancer service is a recognized challenge.[29] High-quality dementia care is dependent not just on the efforts of individual workers at the point of care but also on the extent to which the wider infrastructure enables high-quality care to be delivered.[30,31] Systems that enable this can be described as *responsive*.[32] This concept conveys how health systems can dynamically respond to changing needs and encounter the patient as a genuine partner, identifying and meeting each person's needs and expectations in the context of their personal goals and preferences.[33] Although responsiveness can exist alongside standardization,[34] the findings from this study suggest that this is a challenge.

Standardized packages of cancer care specify a predictable patient journey through the system from diagnosis to completion of treatment.[35] A standardized approach allows for the staff and technologies that deliver treatment to be assembled in place at the right time so that care is delivered as planned and the patient moves on, while the assemblage of staff and technologies moves efficiently to the next patient.[36] Movement is timetabled on the assumption that every patient needs an appointment of equal length, but our findings show that this rational plan is disrupted as patient needs are not uniform. Although patients further down the list are ready for their appointment, they must wait until the appointment is ready for them, suggesting that the distribution of waiting time coincides with the distribution of power.[37] We observed the practice of organizing clinic visits in a ‘hyperrational’ way, allocating more than one patient to be seen at a time, exceeding the capacity of available staff and resources, thus increasing the likelihood of waiting and inconvenience to the patient and caregiver. Any resulting poor experiences for the patient and caregiver are not accounted for when a rational model of work dominates. Primacy is given to the need to keep the clinic running efficiently: patients wait so that staff do not have to.

Standardization may contribute towards efficiency, but is not automatically equated with quality.[38] In spite of rational approaches being used to organize care delivery, there can be high variation and unpredictability at an individual patient level,[36] and our findings illustrate how dementia can be a source of unpredictability. Dementia may also constrain a patient's ability to self-manage outside the hospital environment, which is key to outpatient cancer treatment. The dementia is inconvenient in a bureaucratic system because of its potential to disrupt patient flow through the system and interfere with efficient use of resources.

Healthcare work such as cancer treatment is increasingly specialized, with different parts of the care trajectory being handled by different teams of people distributed across time and space.[35] This fragmentation means that patients can only present a particular element of their health in the context of each individual encounter with a service. This allows the clinic to keep to time, but the outcome for the patient is that only their cancer is a legitimate topic of interest. The structural lack of coordination described here is known to exacerbate treatment burden.[39] Our findings reveal the impact of this fragmented experience for patients and caregivers. In the absence of professional roles that provide the anchor point, they must provide the continuity of information and care management that otherwise feels absent.

The fragmented experience and focus on cancer alone marginalizes the dementia and renders it invisible in standardized approaches to cancer care. As such, decision-making in the oncology clinic can only relate to cancer, even if patients have more pressing health concerns beyond this. The dementia therefore seems less relevant, and is less likely to be part of the conversation, which prevents highly specialized practitioners from having a full understanding of the patient's body and personal experience.[35] In turn, this inhibits how responsive they can be to patient need.

Our findings identified key system issues that impeded the capacity of oncology staff to deliver responsive care. First, assuming that every patient will need the same amount of appointment time risks poor experiences for those

left waiting, which is particularly difficult for people with dementia. A rushed approach to appointments means that patients may not be able to take full part in decision-making, as people with dementia may need more time to consider information. Second, the single-disease focus is problematic for patients with multimorbidity. Our findings show that it is difficult for people to raise issues that are not about cancer, and for staff to respond effectively. Poor outcomes may result because services do not address all the relevant needs and how they interact with each other. There are potential solutions to reduce tension between the desire for efficiency and the requirement for responsiveness. Our findings suggest that strategies could include identifying in advance people with dementia so that arrangements can be made for them; longer clinic appointments; and geriatric oncology clinics where specialties work together. Practitioners need resources, discretion and autonomy to be able to act outside of the standardized model, but are usually constrained. However, as shown by this study, some departments (such as the radiotherapy department who made effective use of dementia champions) were able to act differently, suggesting that change is possible at the team level, given the right conditions.

Limitations and strengths

The study was suspended during the first wave of the COVID-19 pandemic. After restarting, it was challenging to recruit patient and caregiver participants; fewer patients were attending hospital, and the primary researcher (X) was unable to spend time with patients to build rapport. It was not possible to conduct observation, as no nonessential persons could accompany patients during treatment. However, the study protocol was altered to allow for telephone contact and interviews. The relatively low number of formal patient interviews was to some extent mitigated by the inclusion of alternative data collection methods.

CONCLUSION

This study contributes to the evidence base around support for people with dementia having cancer treatment. It has offered suggestions for practical and cultural modifications to increase the responsiveness of services, aiming to improve the quality of health services, and decrease health inequalities. Further work is needed to:

- 1. Identify context-specific tools for eliciting the needs of patients and caregivers;
- 2. Embed principles of personalized care in cancer–dementia services; and
- 3. Establish and test interventions to target appropriate resources, and enhance autonomy and independence of frontline practitioners.

ACKNOWLEDGEMENTS

The authors would like to thank the patients, caregivers and staff who generously gave their time to participate in this study. Naomi Farrington is funded by a NIHR Clinical Lectureship (ICA-CL-2015–01-003). The views expressed are those of the authors and not necessarily those of the NHS, the NIHR or the Department of Health and Social Care.

CONFLICTS OF INTEREST

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

DETAILS

Subject:	Research; Comorbidity; Ethnography; Oncology; Responsiveness; Immunotherapy; Patients; Cancer; Cancer therapies; Outpatient treatment; Radiation therapy; Teaching hospitals; In care; Researchers; Caregivers; Inequality; Health services; Radiotherapy; Cultural factors; Dementia; Interviews; Chemotherapy; Dementia disorders; Participation; Ambulatory health care; Individualized; Data collection; Qualitative research
Identifier / keyword:	cancer; culture; dementia; outpatient; qualitative; responsiveness; treatment
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	98-107
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-15
Milestone dates:	2022-04-28 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-03-08 (manuscriptReceived); 2022-11-15 (publishedOnlineEarlyUnpaginated); 2022-04-28 (manuscriptAccepted)

Publication history :

First posting date:	15 Nov 2022
DOI:	https://doi.org/10.1111/hex.13523
ProQuest document ID:	2767101875
Document URL:	https://www.proquest.com/scholarly-journals/understanding-cultural-environment-outpatient/docview/2767101875/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the "License"). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-11-06
Database:	Public Health Database,Publicly Available Content Database

Document 34 of 46

Implementation of a patient-reported experience measure in a Dutch disability care organization: A process evaluation of cocreated tailored strategies

Marjolein van Rooijen [1]
; Anneke van Dijk-de Vries [1]
; Lenzen, Stephanie [2]
; Dalemans, Ruth [2] ; Moser, Albine [2] ; Anna J. H. M. Beurskens [1] [1] Department of Family Medicine, CAPHRI School for Public Health and Primary Care, Maastricht University, Maastricht, The Netherlands [2] Research Centre for Autonomy and Participation of Persons with a Chronic Illness, Zuyd University of Applied Sciences, Heerlen, The Netherlands

ABSTRACT (ENGLISH)

Introduction

In 24/7 disability care facilities, patient-reported experience measures (PREMs) are important to help healthcare professionals understand what matters to care users and to improve the quality of care. However, the successful implementation of a PREM is complex. In a Dutch disability care organization, stakeholders cocreated tailored implementation strategies aimed at improving the use and integration of a qualitative PREM. This study gives insights into the uptake and experiences with these cocreated implementation strategies and the perceived impact of the set of strategies.

Methods

We performed a prospective process evaluation between February 2020 and February 2021. We collected data in three disability care facilities from 35 care users, 11 professionals, 3 facility managers and 4 organization

representatives. Data collection included observations during kick-offs and learning goal meetings and several attendance checklists. We collected 133 questionnaires (Time 0 and Time 1). We conducted 35 individual semistructured interviews and an online focus group interview. Quantitative data were analysed using descriptive statistics and qualitative data using directed content analysis.

Results

The exposure to and adoption of strategies was between 76% and 100%. Participants were positive about tailoring the strategies to each facility. Implementation was hindered by challenges in care users' communication and COVID-19. The perceived impact referred to an improved understanding of the goal and added value of the PREM and better preparation and execution of the PREM. The impact of the set of strategies was mainly experienced on the micro level.

Conclusion

The uptake of the cocreated implementation strategies was acceptable. The participants valued the tailored approach, which enabled them to focus on facility-specific learning goals. Stakeholder engagement and co-created strategies may have strengthened the adoption of and experiences with the implementation.

Patient or Public Contribution

In this article, we present the process evaluation of implementation strategies for the integrated use of a PREM in disability care. A development group consisting of communication vulnerable care users, trainers and professionals developed the implementation strategies. The disability care organization was responsible for the planning and organization of the implementation process. During the process evaluation the end users, trainers, professionals and managers tailored the implementation strategies to their own settings and needs. Researchers observed this implementation process and interviewed the stakeholders about their experiences and the perceived impact.

FULL TEXT

INTRODUCTION

Over the past 20 years, the importance of quality of care and its transparency has increased in the disability sector.[1,2] Next to measures of physical quality, such as malnutrition and medicine effectiveness, the emphasis is increasingly on care users' perceptions of their health status and quality of life, as measured by patient-reported outcome measures, and on care users' experiences of the care, as measured by patient-reported experience measures (PREMs).[3,4] Insights into care users' experiences can lead to more involvement of them in decisions about care preferences and to more effective relationships between them and healthcare professionals.[3,5–8] PREMs are essential to understand what kind of support care users wish to make their life meaningful, especially in the disability sector, in which care users rely on 24-h care.[9] Most PREMs are structured questionnaires[10]; however, there are also more qualitative instruments that support a structured conversation between care users and professionals. The advantage of qualitative PREMs is their potential to uncover, in more depth, individual care users' experiences with the received care.[11–13]

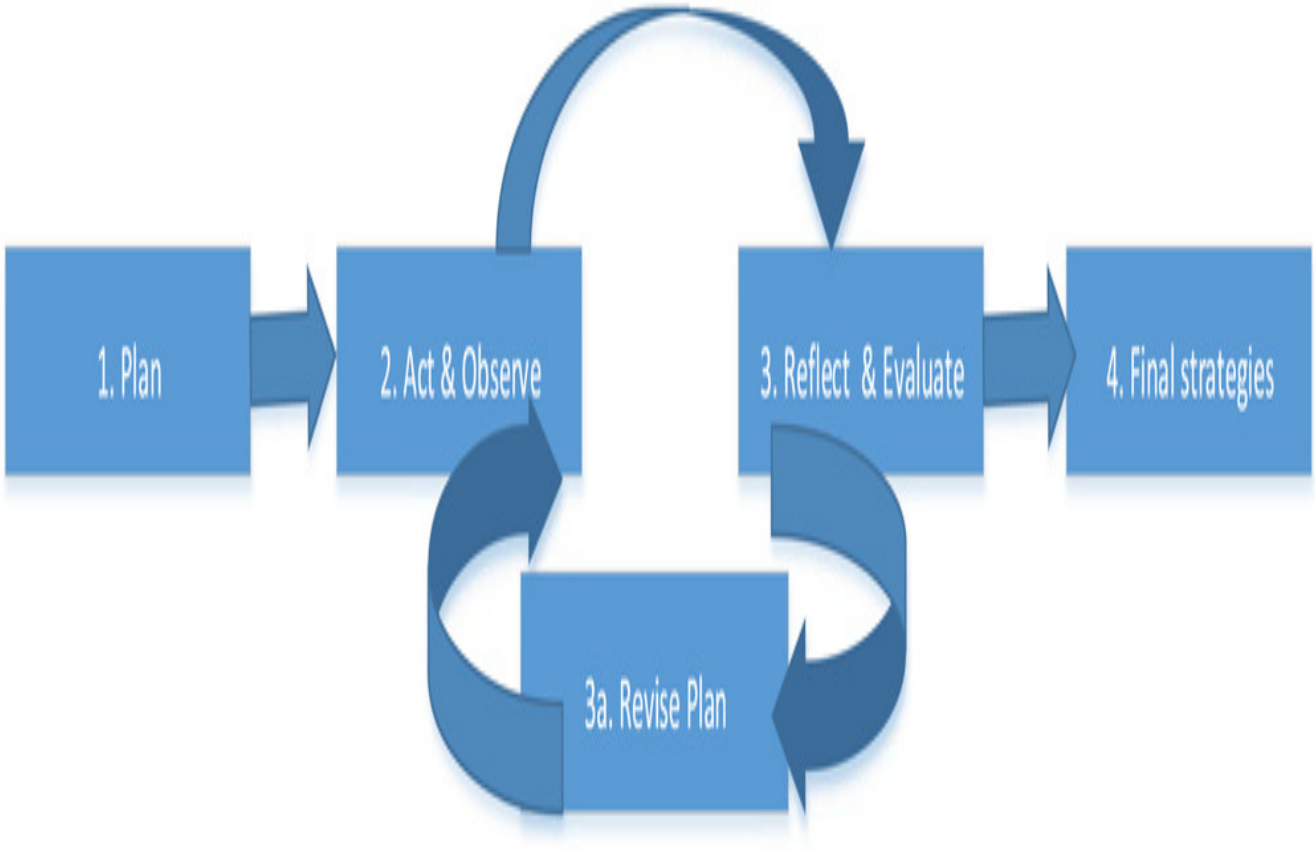
In 2017, a new quality framework for the Dutch disability sector was introduced.[9] This framework emphasises the integrated use of PREMs to improve the quality of care.[9,11] This implies that information about care users' experiences needs to include quality information on three levels: (1) the micro level (care user–professional level), to enhance the delivery of appropriate care and the development of individual care plans; (2) the meso level (organizational level), to monitor the quality of care and enhance team reflection and (3) the macro level, to facilitate organization-wide improvements and external reporting about quality of care.

The successful uptake of a qualitative PREM in routine practice demands both collecting meaningful experiences of care users and integrated use of the outcomes at the micro, meso and macro levels. However, there are various challenges to successfully integrating a PREM into routine care. These challenges include proper preparation of implementation, an efficient work process and a doable way of entering outcomes in the electronic patient records and using the outcomes for quality reports.[11,14–16]

Stichting Gehandicaptenzorg Limburg (SGL) is a Dutch disability care organization for people with acquired brain injuries. Many SGL care users experience communication vulnerability, which encompasses elements of speech,

language, hearing disorders, gestures or semantics, resulting in experienced functional communication difficulties and difficulties in expressing themselves and in understanding professionals.[17] SGL uses a qualitative PREM called 'Dit vind ik ervan!' ('This is how I feel about it!'; see Supporting Information: Appendix I). This PREM facilitates a structured dialogue that encompasses 10 themes.[18]

This study is part of a larger research project in which we systematically developed and evaluated an implementation strategy process together with all relevant stakeholders by means of a participatory action research design. This bigger study is composed of four smaller studies. The steps that have been taken are depicted in Figure 1.



Enlarge this image.

In Study 1, we identified several implementation barriers at SGL.[19] On the basis of the identified barriers, four goals for improvement were formulated: (1) goal clarity and added value of the PREM; (2) being prepared for the PREM

dialogue; (3) successful execution of the PREM and (4) integrated use of outcomes at the micro, meso and macro levels. The process of drafting strategies based on the problem analysis was described in Study 2 (see Figure 1: act and observe).[20] In this step, all stakeholders were engaged in developing strategies that are specific and tailored to the facilities' available resources and implementation context.[21–23] These stakeholders included communication-vulnerable care users, professionals, managers and our research team. Stakeholders then tested strategies, reflected on the application and evaluated the strategies. This provided information to revise the strategies. This cycle was repeated until a consensus among the included stakeholders was reached. Study 3 provided insight into the impact of each stakeholder on the final strategies.[24]

The process evaluation we describe in this article is the fourth and last step of our participatory action research design. We evaluated the uptake, experiences and perceived impact of the tailored implementation strategies. These strategies are aimed at improving the formulated goals, that is, goal clarity, preparation, execution and the integrated use of outcomes of the PREM in a disability care organization. We formulated the following questions:

- Question 1a: To what extent are the implementation strategies applied as intended in terms of fidelity, dose, adaptations and reach?
- Question 1b: What are the experiences of care users, professionals and facility managers with the tailored implementation strategies, and which factors contribute or hinder implementation uptake?
- Question 2: How do stakeholders perceive the impact of the set of implementation strategies on the use of the PREM ‘Dit vind ik ervan!’ as an integrated measure at the micro, meso and macro levels?

METHODDesign

We conducted a prospective process evaluation using a mixed-methods approach based on the Medical Research Council Process Evaluation Framework,[25] which guides the conduct and report of process evaluations of complex interventions. We used this framework because of the complex nature of both the qualitative PREM and the set of 11 implementation strategies to improve PREM uptake and integrated use of the outcomes. In this framework, the focus of the process evaluation of the implementation is on the delivery of the strategies in terms of fidelity (the extent to which the strategies were provided as intended), dose (exposure), adaptations and reach (number of participants). The study had a concurrent mixed-methods design in which both components (quantitative and qualitative) are performed simultaneously.[26]

Setting and participants

The study took place at SGL, a Dutch disability care organization offering daily activities, treatment, supported living and living arrangements to people with severe (acquired) intellectual and developmental disabilities, mostly people with acquired brain injuries. SGL has 18 facilities spread out over the Dutch province of Limburg.

A policy officer at SGL purposively selected 3 of the 18 facilities. These facilities were spread out over the province to include multiple context variables, for example, facility management and culture and challenges faced by care users. Facilities had not contributed in earlier phases of the research project, to prevent knowledge bias.[19,20,24] To safeguard the inclusion of a variety of care users, facilities had at least 12 care users. All care users living at the selected facilities were invited to participate in the study. Professionals and managers had to have worked at SGL for at least 1 year to be familiar with the PREM. Professionals also needed to be case managers of at least one care user, thus responsible for conducting the PREM.

To evaluate strategy uptake at the macro level, we selected representatives of the organization. We involved a manager of the SGL organization, a regional leader, a PREM trainer and a care user representative. All were actively involved in the planning and implementation phase.

Implementation strategies and process

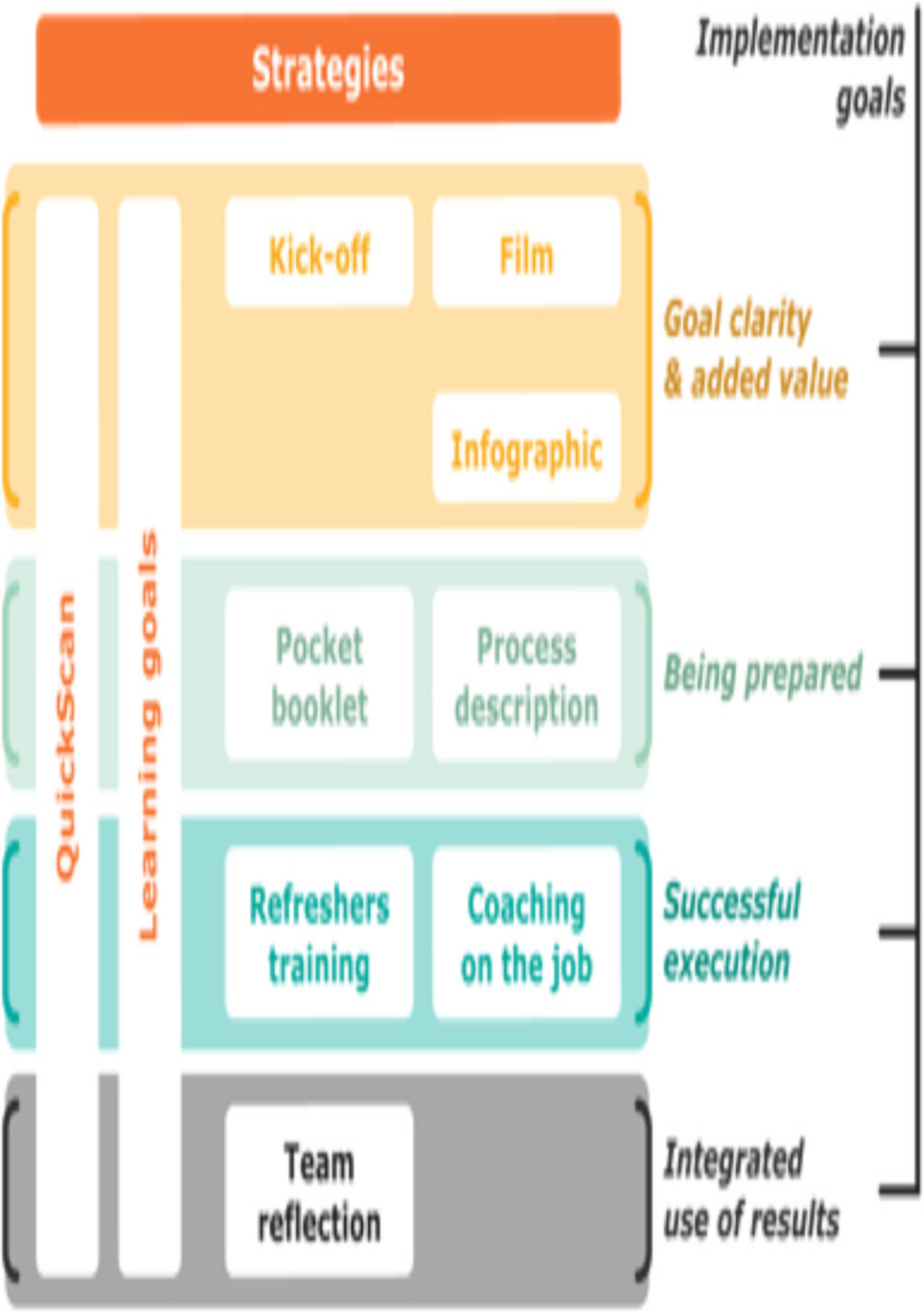
Ten implementation strategies (see Table 1) were executed by SGL to reach the four implementation goals: (1) goal clarity and added value of the PREM; (2) being prepared for the PREM dialogue; (3) successful execution of the PREM and (4) integrated use of outcomes at the micro, meso and macro levels. The implementation strategy process is illustrated in Figure 2.

Table 1 Description of the implementation strategies and the users of these strategies

Strategies	Participants	Content	Adaptive
Quickscan	Care users, professionals, facility managers	Questionnaire exploring the state of working with the PREM, tailored to all participants' scope of interest. Quickscans are filled out at the start and the end of the process.	No
Learning goal meeting	Care users, professionals, facility managers	Meeting to formulate facility-specific learning goals based on quickscan results, using summaries of quickscan results and learning goals guide. Facility-specific learning goals are added to the second quickscan at the end of the process.	Yes
Kick-off	Care users, professionals, facility managers, care user representatives	Session to introduce the facility's specific learning goals and implementation strategies, the infographics and film.	Yes
Film	Care users, professionals, facility managers	Short figurative story showing PREMs' added value (https://www.youtube.com/watch?v=hCsRuv3Bz1g&t=8s).	No
Infographic	Professionals, facility managers	Illustration explaining PREMs' goal for care users and explaining PREMs' goal and relation to other used measurements for professionals and facility managers.	No
Pocket booklet	Care users, professionals	A6 booklet to help care users prepare, execute and reflect on PREM dialogue.	No
Process description	Professionals, facility managers	Illustration explaining PREM integration into the annual cycle of care.	No

Refreshers' training	Professionals, facility managers (optional), PREM trainer	2-h training session addressing facility-specific learning goals and discussing PREMs' added value, process (using process description) and use (using pocket booklet and addressing communication supportive tools, e.g., talking mats, pictos and pen and paper).	Yes
Coaching on the job	One care user per professional, professionals, PREM trainer	Observation of professionals' PREM execution and provision of feedback to improve PREM execution by the PREM trainer.	Yes
Team reflection	Professionals, complete care team, facility managers	Team reflection on PREM execution and/or outcomes and formulation of potential actions for facilities organized using manual facilitating reflection.	Yes

Abbreviation: PREM, patient-reported experience measure.



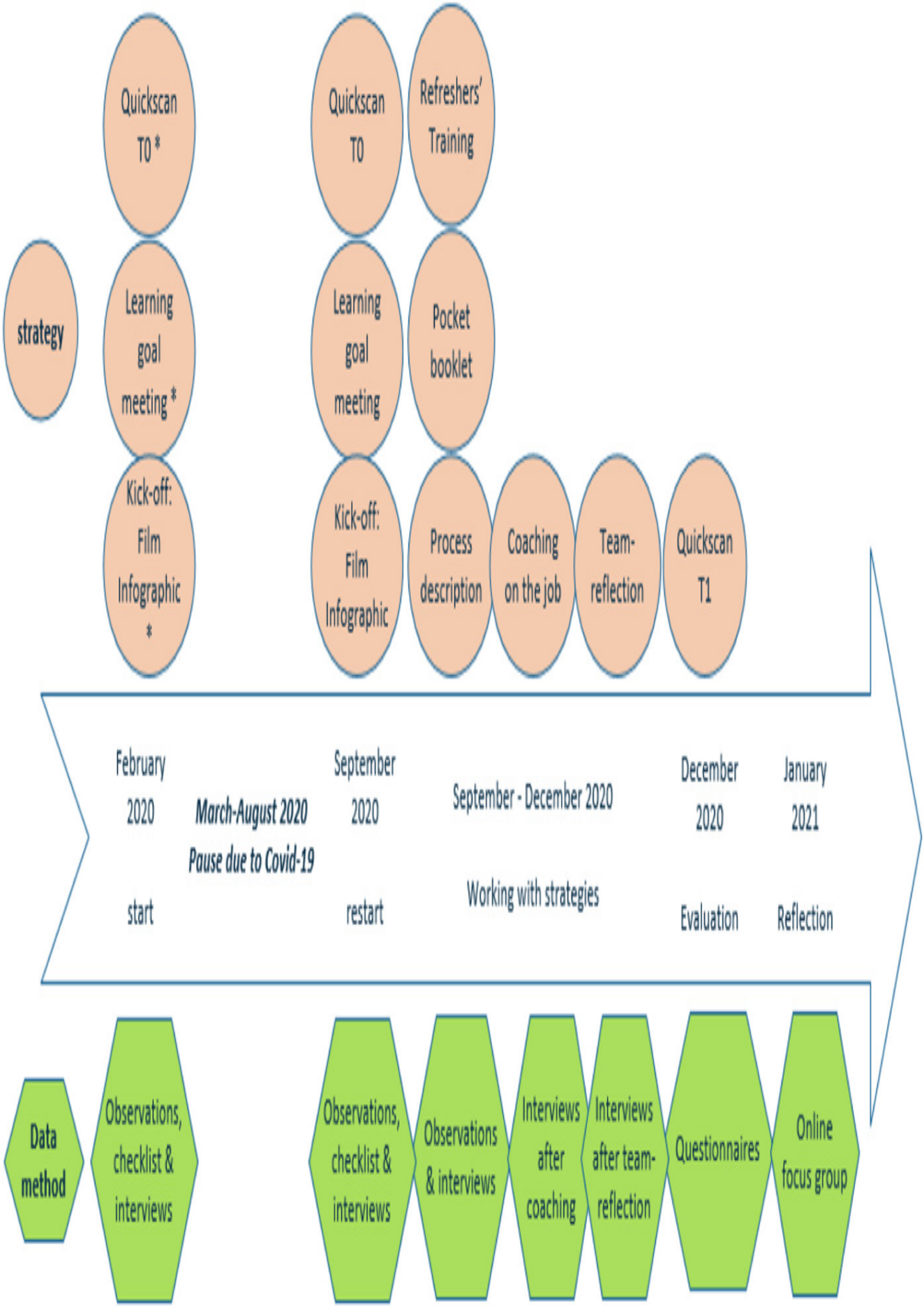
Enlarge this image.

First, the quickscan (Time 0 [T0]) was filled out to guide the formulation of facility-specific learning goals during a

learning goal meeting. A kick-off meeting was organized to show an introduction film of the PREM and to provide an infographic. The professionals attended a refreshers' training, which was tailored to facility-specific learning goals. The PREM trainer asked in-depth questions to clarify the ‘why’ behind a learning goal. Furthermore, the trainer used different techniques, such as role modelling, to improve the skills of the professionals. Moreover, they received a process description to further explain the PREM. Furthermore, the pocket booklet was handed out to professionals. The professionals could introduce this booklet to care users as a way to prepare themselves for the dialogue. During this dialogue between professionals and care users, coaching on the job took place in which the coaching was adapted to the professionals' learning points. Professionals also organized a team meeting to reflect on the execution of the PREM and improvements made to the learning goals. Finally, care users, professionals and team managers filled out the quickscan again after 4 months (Time 1 [T1]).

Data collection

Data were collected between February 2020 and February 2021 by means of observations, checklists, semistructured interviews and questionnaires, the quickscans (T0 and T1) and an online focus group interview. See Figure 3 for the timeframe, strategies and data collection methods. In February, one facility started with the implementation. Because of the COVID-19 pandemic, we had to pause the implementation process between March and September 2020. In September 2020, all participants of the three facilities had completed the quickscan at the start of the implementation (T0). Trained students from nursing, occupational and social sciences assisted care users with filling out the quickscans. They determined the care users' communication vulnerability using the following website: https://www.communicatiekeuzehulp.nl.[27] (This website was developed by Zuyd University of Applied Sciences, Research Centre of Autonomy and Participation of People with a Chronic Illness. The list is based on the ‘Communication Success Screening’ of Dynavox Mayer-Johnson; the screening list ‘starten met ondersteunde communicatie?’ by Modem and the developmental model of ‘Taal Centraal’ (2009) of Prof. van Balkom.) It provided information about the current implementation status of the PREM, which was input for the facility-specific learning goals. These learning goals were added to the quickscan at T1 to evaluate whether goals were reached. With observations and checklists, we observed how the learning goal meeting and kick-off went. After the kick-off meeting, learning goal meeting, refreshers' training, coaching on the job and team reflection, we conducted interviews to explore the participants' experiences.



Enlarge this image.

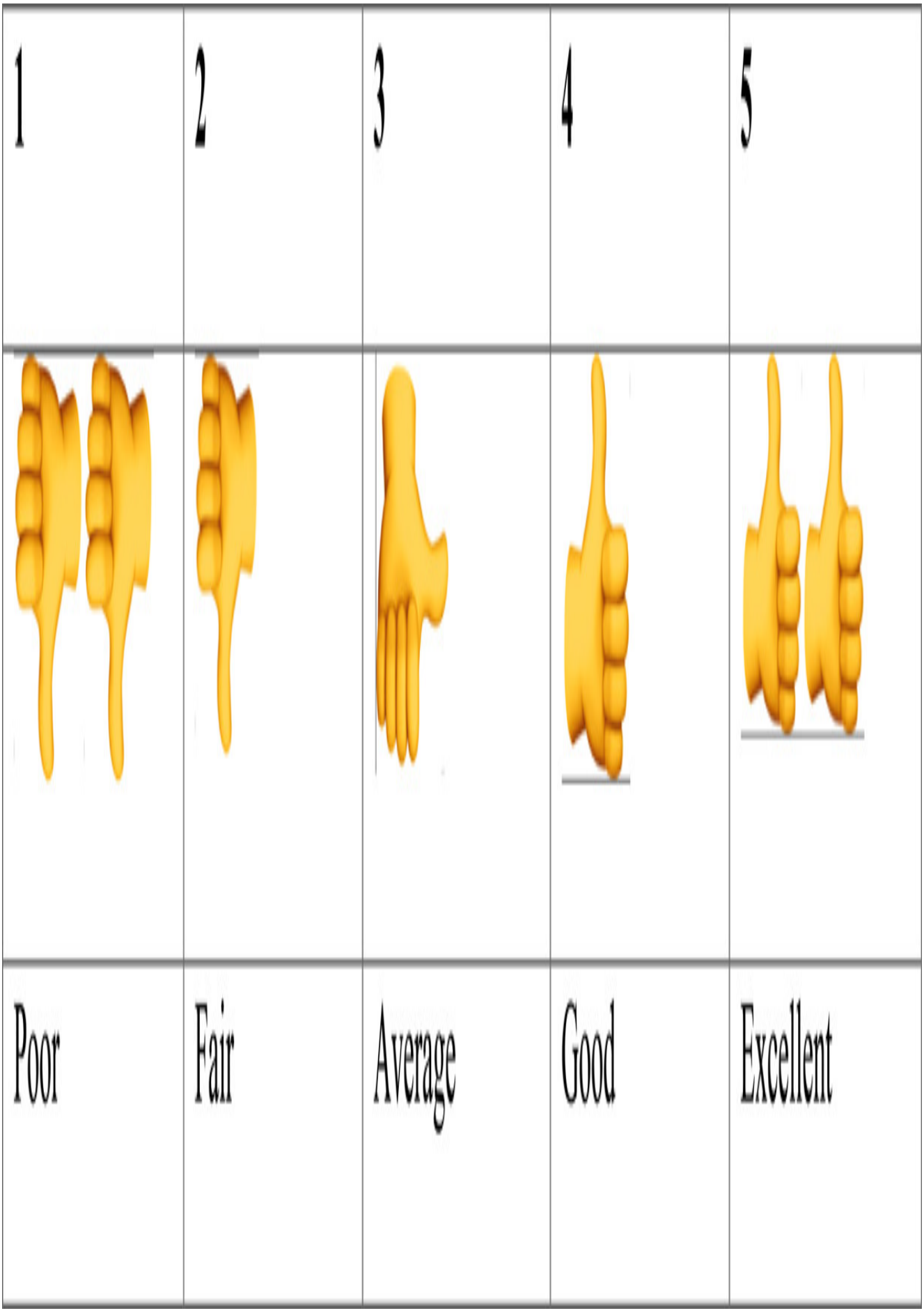
- Question 1a: *Strategy applied as intended*. We observed kick-offs and learning goals meetings to examine participant attendance (reach) and to understand how participants were exposed to the strategy (fidelity, dose, adaptations). Regarding exposure to the refresher training, coaching on the job and team reflections, a researcher (M. v. R.) contacted the trainer or a professional to verify attendance (reach) and to ask how the training went (dose, adaptations).

- Question 1b: *Experiences and factors hindering or contributing to strategy uptake*. To explore experiences with strategies and to identify which factors hindered or contributed to the application of the implementation strategies, we conducted semistructured interviews with care users (*n* = 35), professionals (*n* = 11) and facility managers (*n* = 3) at three time points.

First, 1 week after the learning goal meeting and the kick-off, a researcher (M. v. R.) interviewed care users (*n* = 6) about their experiences. The interviewer used communication-supportive tools (e.g., icons and pictures). She followed strict COVID-19 regulations, such as frequent hand-washing, keeping a 1.5-m distance and wearing a face mask and gloves. Per the facility, she also interviewed a professional and a facility manager about their experience with the learning goal meeting and the kick-offs. The professionals and facility managers were interviewed by phone to limit the risks of COVID-19 spread.

Second, the researcher (M. v. R.) interviewed all professionals and facility managers who took part in the refresher training, coaching on the job and team reflection (*n* = 13) about their experiences, within 2 weeks after the strategies took place. These semistructured interviews by phone started with items that respondents could rate on a scale that ranged from 1 (*poor*) to 5 (*excellent*), such as ‘How would you rate your knowledge about the PREM before the refreshers training?’ This was followed by open-ended questions to elaborate on each item.

Third, after all implementation strategies were applied, all care users (*n* = 35) were interviewed in person about their experiences with the quickscans, the film, the pocket booklet and coaching on the job. The interviews were conducted by trained students from nursing, occupational and social sciences. The interviews started with respondents rating items on a scale that ranged from 1 (*poor*) to 5 (*excellent*), such as ‘How did you experience the kick-off meeting?’ This was followed by open-ended questions about each strategy. Care users could use thumbs-up or thumbs-down gestures to rank their experiences, as shown in Figure 4.



Enlarge this image.

The interviewers also used reminders—for example, the infographic—to facilitate communication with the care

users. All interviews took between 5 and 26 min. COVID-19 regulations were followed.

Professionals and facility managers (*n* = 14) completed a questionnaire with (scoring/open) questions about their experiences with the quickscan, infographics, film and process description. For example, 'How would you rate the information load of the infographics?' 'How could this be improved?' In addition, we interviewed the facilitator of the learning goal meetings (*n* = 1) and the trainer who conducted the refresher training and on-the-job coaching (*n* = 1). Table 2 provides an overview of the data collection of Questions 1 and 2.

Table 2 Data collection evaluation strategy exposure and experiences

Strategy	Qual/quant	Observation	Questionnaire	Interview
Quickscans	Quant	X	X	
Learning goal meeting	Qual/quant	X	X	X
Kick-off	Quant	X	X	X
Film	Quant	X		
	Qual		X	
Infographic	Quant	X		
	Qual		X	
Pocket booklet	Qual		X	
Process description	Qual/quant		x	X
Refresher training	Qual/quant		X	X
Coaching on the job	Qual/quant		X	X
Team reflection	Qual/quant			X

Abbreviations: Qual, qualitative; Quant, quantitative.

- Question 2: *Perceived impact of the set of strategies*. We evaluated the perceived impact of the set of implementation strategies on the four implementation goals in an online focus group. Participants (*n* = 9) were two professionals, three facility managers, a deputy of the board of the SGL organization, a regional leader, a PREM trainer and a care user representative. Beforehand, participants watched a short video presentation as a refresher of all steps of the participatory action research project. A senior researcher (A. J. H. M. B.) moderated the focus group. To facilitate individual input and group discussion, we used an interactive online tool.[28] For each implementation goal, participants individually determined whether they noticed a negative change, no change or a positive change. This was followed by a discussion of each implementation goal. The focus group took 110 min. It was audio-recorded and transcribed verbatim.

Data analysis

The quantitative and qualitative analyses were performed by two researchers (M. v. R. and A. v. D.). We analysed independently (1) each data set and (2) each type of participant. Next, we integrated the results of these different analyses as a joint display. This process was guided by the research questions. In the joint display, we integrated results across data sources and different participants. No subanalysis of different participants was carried out.

We used descriptive statistics in Excel 2016 to analyse all scoring questions in the quickscans and interviews and data from checklists. For the analysis of the qualitative data, we used NVivo 12 software.[29] Open questions in the quickscans and interviews were analysed using directed content analysis, based on whether factors in strategies were experienced as contributing to or limiting PREM application.[30] First, open-ended questions and interview transcripts were read to become familiar with the data; then, contributing and hindering factors were coded and categorized. For the analysis of the perceived impact of the strategies, we conducted a deductive analysis by using the four implementation goals as the main categories in the matrix analysis. Relevant text fragments were selected and assigned to the main categories in the analysis framework.

Trustworthiness

To safeguard principles of trustworthiness, we used a variety of methods regarding credibility and transferability. Credibility was enhanced by using prolonged engagement. During the data collection period of 1 year, we became familiar with the setting and context, could test for misinformation and were able to build trust and gain deep insights into the data.[31] Data triangulation was carried out by using multiple data sources (observations, checklists, questionnaires, interviews and a focus group). In addition, we did a member-check by presenting study outcomes to participants and asking them for their reflections on the outcomes. Method triangulation took place by using both quantitative and qualitative methods. Investigator triangulation took place given that three researchers were involved in data collection. To enhance transferability, we provided rich data about the setting, sample, data collection and data analysis procedures.[31]

Ethics

All participants in the process evaluation received written, visual and verbal information about the study. We applied communication-supportive strategies in the information letters, such as the use of short sentences (a maximum of 10 high-frequency words); one message per sentence and visualizations of the keywords using drawings, photos, icons and bright colours. Participants could ask questions before giving informed consent. Confidential and anonymous handling of data was guaranteed to all participants before data collection. Anonymity was guaranteed through the use of codes. During data collection, participants were given enough time to ask questions. Participants were free to indicate if they wanted to stop or would need a break. The study was reviewed and approved by a research ethics committee.

RESULTSParticipants

Of all care users of the participating facilities, 47% did not want to participate. Some were hesitant because they found the informed consent process too long or too difficult to understand. Others felt that taking part would be too intense. Nevertheless, they could still join the kick-off and receive a pocket booklet or infographic. Thirty-five care users, 11 professionals and 3 facility managers participated in the process evaluation; their characteristics are shown in Table 3. Care users' communication vulnerability is shown in Table 4. Out of the 35 care users, 2 did not experience any difficulties in their communication skills.

Table 3 Characteristics of participants engaged in data collection

Characteristic	Care users (*n* = 35)	Professionals (*n* = 11)	Facility managers (*n* = 3)
Participation rate (% of all care users or professionals living or working with 'Dit vind ik ervan!' at SGL)	53%	100%	100%
Female, *n* (%)	20 (57)	11 (100)	2 (67)
Age in years, *M* (SD)	51 (13)	39 (15)	54 (6)
Time in years at SGL, *M* (SD)	13 (12)	9 (4)	16 (9)
Education level			
Primary school, *n* (%)	14 (40)	8 (73)	3 (100)
Secondary education, *n* (%)	11 (31)	3 (27)	
Secondary vocational training, *n* (%)	8 (23)		
Higher professional education, *n* (%)	1 (3)		
University education, *n* (%)	2 (6)		

Abbreviation: SGL, Stichting Gehandicaptenzorg Limburg.

Table 4 Communication vulnerability level of care users

Communication challenges	Care-users (*n* = 35)
	n (%)
Speaking clearly	16 (46)
Understanding	15 (43)
Remembering	15 (43)
Using pencil and pen	12 (34)
Typing	12 (34)
Speaking loudly	10 (29)
Attention	10 (29)

Seeing	10 (29)
Talking	9 (26)
Signs and facial expression	8 (23)
Hearing	3 (9)
No communication challenges	2 (6)

Findings

- Question 1a: *Strategy applied as intended*. The data in Table 5 indicate the extent to which the implementation strategies were applied as intended in terms of reach.

Table 5 Reach

Reach	Care users (*n* = 35)	Professionals (*n* = 11)	Facility managers (*n* = 3)
	n (%) or %	*n* (%) or %	*n* (%) or %
Quickscan T0	34 (97%)	7 (63%)	3 (100%)
Quickscan T1	31 (89%)	11 (100%)	3 (100%)
Learning goal meeting	100%	100%	100%
Kick-off with film	28 (76%)	6 (54%)	3 (100%)
Infographics	35 (100%)	11 (100%)	3 (100%)
Pocket booklet	35 (100%)	11 (100%)	3 (100%)
Process description	NA	11 (100%)	3 (100%)
Refresher training	NA	11 (100%)	2 (67%) nonintended
Coaching on the job	NA	11 (100%)	1 (33%) nonintended
Team reflection	NA	9 (82%)	2 (67%)

Abbreviations: NA, not applicable; T0, Time 0; T1, Time 1.

At the start of the implementation (T0), the quickscan was filled out by almost all care users and facility managers and two thirds of the professionals. In the end, three care users did not want to complete the quickscan (T1) because of COVID-19-related stress. The outcomes of quickscans (T0) provided input when the facility-specific learning goals were formulated. These learning goal meetings were attended by at least one care user, one

professional and the manager in each facility. Facility managers could invite more care users or professionals. One facility manager invited 14 extra care users. At another facility, two extra professionals attended the learning goal meeting. The kick-off was tailored to the setting. Two facility managers organized the kick-off during a regular monthly meeting with care users. In the other facility, there was a special kick-off night. The majority of care users attended the kick-offs, watched the film and received an infographic. All professionals participated in the refresher training and on-the-job coaching. Even though the refresher training and on-the-job coaching were developed for professionals, two facility managers joined as well, and one of them received on-the-job coaching. During the refresher training, all attendees received process descriptions and a sufficient number of pocket booklets. They were instructed to hand out the pocket booklets to their care users in preparation for the PREM. One team reflection was cancelled because of COVID-19 restrictions.

- Question 1b: *Experiences and factors hindering or contributing to strategy uptake*. All participants rated their experiences with each implementation strategy on a scale that ranged from 1 (*poor*) to 5 (*excellent*). The results are presented in Table 6. Because of possible memory problems on the part of care users, we asked them what they remembered of the strategy before they shared their experiences. As Table 6 shows, not all care users could remember the film, infographic and pocket booklet, despite their exposure to this strategy. If care users did not remember the strategy, no further questions were asked.

Table 6 Results of questionnaires exploring experiences with strategies (between 1 [*poor*] and 5 [*excellent*])

Strategies	Statements	Care users (*n* = 31)	Professionals (*n* = 11)	Facility managers (*n* = 3)
Quickscan	How did you experience the quickscan statements? Mdn (range) How did you experience the quickscan visuals? Mdn (range)	4.5 (4–5)	4 (4–5)	4 (4)
		4 (1–5)	4 (4–5)	4 (4–5)
Learning goal meeting	How did you experience the learning goal meeting? Mdn (range) How did you experience the facilitator during the learning goal meeting? Mdn (range)	5 (5)	4 (4)	4 (4)
		5 (5)	4.5 (4–5)	4 (4–5)
Kick-off with film	Do you remember the film? *N* (% yes) How did you experience the film? Mdn (range)	21 (59%)	NA	NA
		4 (3–5)	4.5 (4–5)	4 (4)

Infographic	Do you remember the infographic? *N* (% yes) How did you experience the infographic? Mdn (range)	22 (65%)	NA	NA
		4 (1–5)	4(4–5)	4 (4)
Pocket booklet	Do you remember the pocket booklet? *N* (% yes) How did you experience the use of the pocket booklet? Mdn (range)	25 (72%)	NA	NA
		4 (2–5)	5 (4–5)	4 (4–5)
Process description	Do you remember the process description? *N* (% yes) How did you experience the process description? Mdn (range)	NA	NA	NA
			4 (3–5)	4 (4–5)
Refreshers training	How did you experience the refresher training? Mdn (range)	NA	4 (3–5)	4 (4–4.5)
Coaching on the job	How did you experience the coaching on the job? Mdn (range) Would you recommend the coaching on the job? *N* (% yes)	NA	4.5 (4–5)	4 (4)
			11 (100%)	1 (100%)
Team reflection	How did you experience the team reflection? Mdn (range)	NA	4.5 (4–5)	NA

Abbreviations: Mdn, median; NA, not applicable or not asked.

Participants mentioned several contributing factors. First, the active involvement of different stakeholders during the kick-off and the learning goal meeting was appreciated. Participants valued the combination of input from the quickscan and the contribution of each participant during the learning goal meeting. A care user was surprised to be able to give valuable input during the learning goal meeting: ‘I expected that this would be above my capabilities, but I was able to contribute using my experiences’. A region manager facilitated the learning goal meetings. A facility manager: ‘Especially for care users this must have felt more special because of the region manager facilitating the learning goal meeting. That adds body to the session’.

Second, participants experienced the film, the process description, the infographic and the pocket booklet as practical, providing both an overview of the PREM topics and being an easy way to refresh or transfer knowledge about the PREM within the care team. They mentioned an increased understanding of the goal and value of the

PREM due to these tools. The pocket booklet in particular was helpful for care users to prepare the PREM dialogue. A care user: ‘The pocket booklet offered a comprehensive overview of all topics that could be discussed’.
Furthermore, the participants felt that the learning goal meeting, refresher training and on-the-job coaching were sufficiently tailored to their context and needs (guided by the location-specific learning goals). Professionals and facility managers appreciated the focus on practical skills and experienced an increase in their knowledge regarding the why and the how of the PREM. They became aware of their own behaviour and attitude during the PREM dialogue. During on-the-job coaching, the coach wrote down sentences as spoken by the professional. Professionals experienced this as confrontational but helpful for reflection. A professional:
I became more aware of how to ask questions without already filling in the care-user's answer. Before, I quickly started filling in solution-oriented answers but now I have learnt to let care-users fill in their own answers. It was nice to have someone say something about that.
Moreover, professionals liked the team reflection because it enabled them to share training and coaching experiences and to transfer knowledge about the PREM dialogue with facility employees who did not join the refresher training and on-the-job coaching. A professional: ‘All team members were involved, listened to each other and added personal experiences to the discussion about using the PREM’.
Respondents also experienced several hindering factors for strategy uptake. First, a major hindering factor was implementing the strategies during the COVID-19 pandemic. We originally planned a 9-month process evaluation, but we had to pause the implementation process. The evaluation period was reduced to 5 months. Participation became challenging because of restrictions and care users and professionals who tested positive for COVID-19. This complicated the organization of the refresher training, on-the-job coaching and team reflection.
A second experienced barrier was related to the care users' communication challenges. Some care users found it difficult to understand the whole discussion during the learning goal meetings or the kick-off. Some struggled to express themselves because of the group size. Even though the quickscan, infographic and pocket booklet had been developed together with care users who were communication vulnerable, some care users found it still difficult to read and suggested some improvements. They preferred bigger characters in the infographic and pocket booklet. Furthermore, they asked for an iPad version of the pocket booklet to help care users who could not skim pages because of a physical disability. A care user: ‘It would be helpful to have the pocket booklet on my iPad. Then I would have been able to increase the font size’.

- Question 2: Perceived impact of the set of strategies. Guided by the outcomes of the quickscan (T0), all facilities developed three learning goal topics. These facility-specific learning goals were added to the quickscan at T1 to evaluate whether they had been met. The stakeholders responded as follows.

In the first facility, the learning goals were (1) understanding differences between the measurements used at SGL, (2) knowing and sharing the care-users' specific needs to perform the PREM and (3) discussing PREM outcomes and experiences with PREM execution in team meetings. These learning goals were met: range = 83%–100%, median = 100%.
In the second facility, the learning goals were (1) knowing care users' individual needs to perform the PREM, (2) care users and professionals do no longer experience the PREM as a pointless task and (3) professionals ask follow-up questions to better understand the scores of the care users on the PREM. These learning goals were met: range = 87%–100%, median = 91%.
Topics at the third facility were (1) care users know a week in advance that the PREM will take place and have the opportunity to prepare themselves using a pocket booklet, (2) a successfully performed PREM does not necessarily

need to be translated in an action items list and (3) PREM reports are discussed with the care users if they want to. In the third facility, the facility-specific learning goals were met: range = 50%–100%, median = 100%.

In the online focus group, we evaluated the experienced impact of the set of strategies on the four implementation goals. The first goal, ‘Purpose, clarity, and added value’, was unanimous positively evaluated. The attitude of the team toward the PREM had changed, according to the participants. Professional: ‘It is no longer a pointless task’. The representative of the care users shared enthusiastic stories that she had heard from other care users: ‘I noticed a positive change from care users I've spoken with, which made me happy…. I think because now all people know better what “Dit vind ik ervan!” is, we're all facing the same direction’. Facility managers and professionals experienced an increase in sharing the ‘Purpose clarity and added value’ due to the refresher training. The pocket booklet was experienced as very helpful for preparing the PREM dialogue for both care users and professionals. These results had a positive impact on the second implementation goal: ‘Being prepared’ in a way that both care users and professionals felt ready to engage in a dialogue about the care. The perspectives on the impact of the strategies on the third goal, ‘Successful execution’, varied. On the one hand, professionals desired to learn more about conversation techniques to improve PREM dialogue execution. Professional: ‘I have learnt more about using conversation techniques’. On the other hand, professionals still found it challenging to plan and execute the PREM in a limited amount of time.

Professional: It is still difficult to plan the execution of the PREM. During a dialogue, you write down things care users say, but then you have to put it in the electronic care users' files. This file needs to be evaluated. All these steps together are a lot for some care users.

According to the professionals, 5 months of the process evaluation were too short to conduct a PREM with all care users.

In the focus group, the perspectives on the fourth implementation goal, ‘Integrated use of outcomes’, varied. Professionals experienced an improvement in sharing PREM outcomes with care users. They discussed action items with the care users and registered this in the daily care plans. On the team level, professionals perceived an opportunity to discuss outcomes in a team meeting.

Professional: Previously, outdated reports were read like an eight o'clock newsreader, and for the team there was no value in it. I think we should discuss the outcomes among ourselves more frequently in the teams, because this is part of what we do!

SGL management missed feedback from facility managers and could not yet see results at the organizational level. Overall, focus group participants felt that the strategies created a beginning for a sustainable uptake of the PREM in their organization; however, using the PREM on a macro level has been considered a ‘work in progress’. Their expectant attitude was expressed by one of the managers: ‘If facilities with positive PREM experiences share those experiences in different ways with other facilities, they will be stimulated. This way the speed of sharing positive experiences can be increased’.

DISCUSSION

This study aimed to evaluate the extent to which the strategies were applied as intended, the experiences regarding tailored implementation strategies and the perceived impact of this set of strategies on the integrated use of a qualitative PREM in disability care. The process evaluation took place at three facilities of a Dutch disability care organization. The reach of the strategies to the care users, professionals and managers was acceptable—between 76% and 100%. The participants valued the tailored approach, which enabled them to focus on facility-specific learning goals. Hindering factors were complications in the planning of the strategy rollout because of COVID-19 and the communication vulnerability of care users. The perceived impact of the set of implementation strategies was

noted mainly at the micro level, improving goal clarity and added value and preparation of the PREM.

This process evaluation was the last step of a participatory action research project in which we developed implementation strategies in continuous cocreation with all relevant stakeholders, including communication-vulnerable care users.[19,20,32] This research approach enabled us to generate both research knowledge and knowledge for practice, which could immediately change practice in a positive way. Stakeholders were involved at the micro level (care users, professionals and PREM trainers), the meso level (professionals, PREM trainers and managers) and the macro level (managers and quality advisors). This stakeholder engagement and the use of continuous iterations may have strengthened the uptake and experiences with the implementation. The participants found that the strategies aligned with their practice and provided an answer to problems they face. In particular, the combination of fixed and adaptive strategies was appreciated because the adaptive strategies could be tailored to the facility-specific learning goals. This seems to be a promising and feasible approach that promotes implementation. This approach can be replicated by other organizations, but only after context-specific problem analyses and the selection of strategies. Other organizations working with the ‘Dit vind ik ervan!’ PREM can use the quickscan to determine their learning goals and improve implementation in their organization with the available strategies.

Because the region manager was involved as a facilitator during the learning goal meetings, one felt the support and importance of improving the PREM within SGL. This highlights that implementation uptake highly depends on our systematic approach and involvement of stakeholders from all levels in the organization, and needs to be embedded and embraced.[22,23,33]

Even though our implementation strategies were cocreated,[19,20,32] some care users with communication challenges still expressed difficulties working with some of the strategies. They gave concrete suggestions for improvement, for example, bigger characters in the pocket booklet or an iPad version. This shows that cocreation and tailoring implementation strategies are a continuous process. Koshy et al.[34] assumed that interventions are never finished and can be adapted over time in accordance with changes in practice or context. Thus, cocreation is significant for making plans and developing strategies, and valuable suggestions for the adaption of the implementation strategies can be obtained after evaluation.[14,35] In this regard, Bentzen[35] showed that ownership of strategies and strategy outcomes are strengthened if cocreation continues in later stages of implementation. This underscores the importance of implementers remaining open to feedback provided by the strategies' users.

In our study, the tailoring of strategies (refresher training, on-the-job coaching and team reflection) was guided by facility-specific learning goals. These learning goals were based on outcomes that were derived from the quickscan. Baker et al.[37] and Lewis et al.[36] have shown the power of tailoring strategies to contextual factors to improve intervention uptake.

The stakeholders questioned the impact of the tailored implementation strategies on the integrated use of the PREM outcomes. Although professionals experienced that PREM outcomes were more often discussed, and actions were taken with both the care users (micro) and the care teams (meso), the outcomes could not be translated into actions for quality improvements at the organizational (macro) level. This is not surprising because the challenges that were identified during the problem analysis mainly addressed the micro level.[19] In this regard, Foster et al.[14] recommended starting with planning the organizational aspects to administer the PREM and preparing the staff for PREM use. Professionals need to be convinced about the value of the PREM at the micro level. The next step is to use the results of the PREM at the meso and macro levels.

Strengths and limitations

A strength of this study is the use of diverse qualitative methods (observations, checklists, questionnaires,

interviews and focus groups) and quantitative data to gain insight into the adoption and experience of the strategies. A second strong point is the systematic and stakeholder approach. During the problem analysis and strategy development phase, the research team was actively engaged in the process together with the other stakeholders at SGL. The role of the research team changed from partners to one that involved collecting data and evaluating the process as academic researchers. SGL had full ownership of the implementation. Moreover, the research team had to stay at a physical distance because of COVID-19. We were still able to collect the information we aimed for by means of phone interviews and web-based questionnaires. Fortunately, care users could still be supported by filling out questionnaires and the quickscans. This was delegated to independent, trained healthcare students. We only missed nonverbal clues in the collection of data from professionals and managers.

The set of implementation strategies was rolled out in three SGL facilities that were not engaged in the development of the strategies. Nevertheless, because of the 4-year-long engagement of the SGL organizations, management perceived a coownership of the strategies. This resulted in no dropouts and the continuous engagement of these three facilities even though the context of COVID-19 was challenging. This may be a result of all the time and energy that had been invested by the researchers to understand the organization and to involve all the stakeholders in the development process.

This study is also subject to weaknesses. This process evaluation was originally planned for a period of 9 months. Because of COVID-19, we had to shorten the study to a period of 5 months. Only 53% of all care users living at the three facilities participated in the data collection part of the process evaluation. One reason for this low participation rate is the informed-consent procedure. To participate in the process evaluation, participants needed to be informed about all the pros and cons of participation. Many care users have communication vulnerabilities, which hindered them in reading through all the legally required 20 pages of informed consent, although this was written and visualized using communication-supportive methods.[38] For some care users, this comprehensive document was a barrier to participation. The other complication of including care users was the COVID-19 situation. Because of the mental impact (e.g., anxiety, stress) of the pandemic, some care users felt discouraged and did not want to take part in a research study.

The findings of our process evaluation are promising regarding better PREM uptake at the micro level. Given the idea that implementation processes depend on a continuous learning process that changes both interventions and organizations,[35] it would have been valuable to determine the impact on integrated use of PREM outcomes and sustainability of strategies for a longer period of time.

CONCLUSION

This study of the implementation of a PREM, in which the strategies were cocreated and tailored to the three disability care facilities, shows good uptakes in daily practice. The impact was mainly experienced on the level of the care users and professionals, regarding goal clarity and the added value of the PREM and preparation for the PREM dialogue. More time and effort are needed for the integrated use of the PREM at the meso level to monitor the quality of care and enhance team reflection and at the macro level to facilitate organization-wide improvements and external reporting about quality of care. The stakeholder engagement in the whole process, from problem analysis to cocreated implementation strategies, may have strengthened the adoption of and experiences with the implementation.

AUTHOR CONTRIBUTIONS

Study conception and design: Marjolein van Rooijen, Anneke van Dijk-de Vries, Stephanie Lenzen, Albine Moser, Ruth Dalemans and Anna J. H. M. Beurskens. *Data collection*: Marjolein van Rooijen, Anneke van Dijk-de Vries and Anna J. H. M. Beurskens. *Analysis and interpretation of results*: Marjolein van Rooijen, Anneke van Dijk-de Vries,

Albine Moser and Anna J. H. M. Beurskens. *Draft manuscript preparation*: Marjolein van Rooijen, Anneke van Dijk-de Vries, Albine Moser and Anna J. H. M. Beurskens. All authors reviewed the results and approved the final version of the manuscript.

ACKNOWLEDGEMENTS

We wish to thank the three participating locations, their care users, professionals and facility managers. We also thank the organizations' representatives who participated to answer the third research question and facilitated the researchers in conducting their research at the disability care organization.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

The study was reviewed by a Research Ethics Committee of Zuyderland and Zuyd University (METCZ20200001).

DETAILS

Subject:	Participatory research; Managers; Communication; Patients; Content analysis; Evaluation; In care; End users; COVID-19; Statistical analysis; Quality of care; Disability; Data collection; Trainers; Qualitative analysis; Data analysis; Stakeholders; Action research; Learning; Interviews; Check lists; Research design; Implementation; Health professional-Patient communication; Uptake; Medical personnel; Professionals; Focus groups; Traumatic brain injury; Data
Business indexing term:	Subject: Professionals
Identifier / keyword:	care user; cocreation; communication-vulnerable; disability care; implementation
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	132-145
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford

Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-08
Milestone dates:	2022-09-29 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-04-28 (manuscriptReceived); 2022-11-08 (publishedOnlineEarlyUnpaginated); 2022-10-02 (manuscriptAccepted)
Publication history :	
First posting date:	08 Nov 2022
DOI:	https://doi.org/10.1111/hex.13628
ProQuest document ID:	2767101874
Document URL:	https://www.proquest.com/scholarly-journals/implementation-patient-reported-experience/docview/2767101874/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the "License"). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-09-28
Database:	Public Health Database,Publicly Available Content Database

For autistic persons by autistic persons: Acceptability of a structured peer support service according to key stakeholders

Valderrama, Alena [1]
; Martinez, Alejandra [2] ; Charlebois, Kathleen [3]
; Guerrero, Lucila [3]
; Baudouin Forgeot d'Arc [4]

[1] Department of Social and Preventive Medicine, School of Public Health, Université de Montréal, Montréal, Quebec, Canada; Research Center of the Sainte-Justine University Hospital, Montréal, Quebec, Canada [2] Research Center of the Sainte-Justine University Hospital, Montréal, Quebec, Canada; Faculty of Medicine and Health Sciences, McGill University, Montréal, Quebec, Canada [3] Research Center of the Sainte-Justine University Hospital, Montréal, Quebec, Canada [4] Research Center of the Sainte-Justine University Hospital, Montréal, Quebec, Canada; Centre Intégré Universitaire de Santé et Services Sociaux de Nord-de-l'Île-de-Montréal, Montréal, Quebec, Canada; Département de Psychiatrie, Université de Montréal, Montréal, Quebec, Canada

ABSTRACT (ENGLISH)

Introduction

Social support is a protective factor in the mental health of autistic people. Furthermore, prejudice regarding autistic people is a constraint for the development of social support programmes by autistic peers.

Methods

The objective of this study is to describe the anticipated acceptability of structured peer support programmes for and by autistic persons. Fifteen key stakeholders (six autistic adults, four caregivers and five service providers) participated in in-depth semistructured interviews. A qualitative thematic analysis of the content of the verbatim was carried out.

Findings

We found that while a structured peer social support programme is acceptable to autistic people and caregivers, there was no consensus among service providers. The latter expressed doubts about the ability of autistic people to offer support. The framing of discussions between peers, the training of peer helpers, the support for autistic leadership and an organization that considers the communicational and sensory characteristics of autistic persons, could influence adherence to such a programme. Moreover, a space without service providers is an important condition for the acceptability of a peer support programme.

Conclusion

A structured peer support service for and by autistic persons could be an innovative way to answer the unmet support needs of autistic people. It seems essential to anticipate potential barriers and facilitators and to communicate among health professionals to promote this approach and reduce possible prejudice about the ability of autistic people to offer support to their peers. More studies are necessary.

Patient or Public Contribution

Fifteen key stakeholders who are involved in autistic people's trajectory of service and support participated in this research. We are a research team composed of healthcare professionals and researchers, in addition to one member of our team being an autistic advocate and a mental health peer-support mentor. Two members of our team are also parents of autistic children. The comprehensibility of the questions for the interview was consulted and discussed with one autistic advocate-collaborator.

FULL TEXT

INTRODUCTION

Approximately 1 in 66 children and youth are diagnosed as autistic in Canada.[1] Autistic people are at high risk of having a low quality of life[2] and of undergoing negative social experiences, health problems and mental health issues during their life course.[3,4] Whereas social participation at work and during leisure time along with the quality of social support networks are important protective factors,[2,5–8] their needs in that regard remain unmet.[9] Despite all this, to the best of our knowledge, there are no services with the primary objective of providing social support to autistic people.

What is social support?

Widely recognized as an important determinant of health,[10,11] social support is a process of social interaction that increases coping strategies, self-esteem, sense of belonging and competence through the real or predictable exchange of practical or psychosocial resources.[12–14] Indeed, social support is a mediator between stressful events and health. Supported persons would have more positive perceptions of their environment such as the belief that others can and will provide the necessary resources to help them and the perception of their ability to cope with various consequences of stressful events.[15] Social support can be broken down into four main dimensions: emotional, appraisal, instrumental and informational. Specifically, emotional support involves receiving feelings of reassurance, protection or comfort. Appraisal support is about reassuring a person about their skills and values. Instrumental support is tangible aid, either material goods or services that directly assist people in need. Finally, informational support refers to the sharing of information and advice for solving problems.[12,15]

Social support can be deployed by peers

In fact, social support can be deployed through formal (e.g., health professionals), semiformal (e.g., community organizations) and informal networks (e.g., families and friends).[15] In particular, social support provided by peers with a common lived experience is known as peer support. The latter strategy has been proven in several chronic conditions to improve health outcomes and adherence to treatment and to reduce healthcare costs.[16,17] Efforts to integrate peer helpers into the healthcare system are underway in several areas.[18,19] In autism, peer support interventions have been proposed in the school setting as a way to promote the development of social skills and academic engagement, rather than for the benefit of social support as such.[20–23] These programmes, mainly consist of neurotypical students offering support to autistic persons, rather than autistic persons supporting autistic persons.

Social support by and for autistic people

Many autistic people appreciate and demand opportunities for socialization among autistic people and exchange in an autistic space of acceptance.[24,25] In research, there are promising experiences concerning autistic peer support.[26–30] They suggest that a safe, structured space in a positive and welcoming environment would allow autistic people to feel accepted and understood (emotional support)[26]; to socialize around shared interests, and to recognize themselves in others (socialization and belonging support),[29] while allowing for a better understanding of what autism is and developing coping strategies (informational and instrumental support) and a more positive perception of autism.[26]

These promising experiences are adolescent peer support groups of Weidle et al.,[30] mentoring dyads of Martin et al.,[29] and support groups for newly diagnosed adults of Crane et al.[26] The purpose of the autistic adolescent support groups was to provide a space for the development of autistic peer relationships in a hospital setting. Mentoring dyads aimed to improve the well-being of mentees in a school setting. The adult support groups were designed to evaluate the benefits of an autism training tool offered following diagnosis in a community setting. In each of these programmes, participants carried out a qualitative evaluation of satisfaction and an evaluation of the benefits received, and their feedback was positive overall. Even more, Hotez et al. co-developed a programme with autistic people to support students to transition into postsecondary education. In this programme, two autistic people successfully acted as mentors.[28] Finally, Gillespie-Lynch et al.[27] co-developed another mentorship programme for autistic students in postsecondary education where autistic students acted also as mentors. In fact, autistic people, possibly due to their particularities in terms of communication and social cognition, often experience difficulties in establishing informal relationships of social support.[31] Even more, myths and prejudices about autistic people such as lack of empathy and interest in social relations[32] might hinder the establishment of social support services for and by autistic peers among services providers or services users like families. That is why, from a perspective to evaluate and develop complex interventions,[33] we wanted to study the acceptability of a structured support programme for and by autistic peers in the Quebec context.

Acceptability assessments for the development of a structured peer support service

Acceptability can be and should be assessed before engaging in an intervention. We define a structured programme as an organized set of activities and services carried out simultaneously or successively with dedicated resources to

achieve specific objectives in relation to specific health problems, in a defined population.[34] Therefore, the objective of this research is to describe the anticipated perception of the acceptability of such a programme, from the perspective of key stakeholders like autistic persons, caregivers or social workers who are involved in autistic people's trajectory of service and support.

Sekhon et al.[35] define acceptability as the extent to which people delivering or receiving a healthcare intervention consider it to be appropriate, based on their anticipated or experienced cognitive and emotional responses to the intervention.[35] For the purposes of this study, acceptability is defined in terms of what is required for a structured social support programme provided by autistic peers for it to be considered a source of support for autistic people. Several factors can influence participants' perceptions of acceptability before participating in the intervention. Therefore, we used the theoretical framework of acceptability (TFA) which consists of seven component constructs: *Affective attitude* towards the intervention. *Ethicality* or the extent to which the intervention has a good fit with an individual's value system. *Intervention coherence* or the extent to which the participant understands the intervention, and how the intervention works. *Perceived effectiveness* or the extent to which the intervention is perceived as likely to achieve its purpose. *Self-efficacy* or the participant's 'confidence that they can perform the behaviour(s) required to participate in the intervention'. *Opportunity costs* or the extent to which benefits, profits or values must be renounced to engage in an intervention which influences adherence and participation. *Burden* or the perceived amount of effort that is required to participate in the intervention and reasons for discontinuation.[35]

METHODSStudy design

A qualitative descriptive design[36–38] underpinned this study with the aim of ascertaining perceptions as to the acceptability of structured peer social support for and by autistic persons by key stakeholders. We are a research team composed of healthcare professionals in public health, service organization, mental health and qualitative research, in addition to one member of our team being an autistic person and a mental health peer-support mentor. Two members of our team are also parents of autistic children.

Since our focus was on life experiences and subjective perceptions, the use of qualitative methodology was appropriate.[39,40] In-depth semistructured interviews with key stakeholders were carried out. Participants did not receive incentives for participation. Table 1 shows only some characteristics of the participants (key stakeholders) to preserve their anonymity.

Table 1 Characteristics of the participants

Key stakeholders	Main characteristics
Seven autistic adults (AA)	Six females and one male, who communicate orally, four having received a diagnosis in the last 2 years.
Four caregivers (CV)	All females, two of the children were nonverbal, three children lived with their caregivers, three of the children were adults and one was an adolescent.
Four service providers (SP), it means health professionals or social workers	All females, two persons of community organizations that offer support to autistic people and three psychoeducators from the health system.
Total of 15 key stakeholders	

Sampling and recruitment of participants

Participants were recruited using a purposive criterion sampling strategy.[41] Potential participants included (1) people that self-identified as autistic people (AA), (2) caregivers of autistic persons (CV) and (3) service providers (SP) such

as health professionals or social workers. More specifically, service providers were required to have at least 5 years of experience working with autistic people (adolescents or adults) in the health and social services network in Quebec to be eligible to participate. Moreover, the participants should be living in Quebec and speaking fluently in French. In addition, email invitations were sent to two autistic people, three health professionals and two community organizations known for their involvement in supporting autistic people. All of them accepted to participate in an interview except one autistic person who did not respond to the invitation. Thus, eight people were recruited by invitation and seven more through CHU Sainte-Justine mother and child Hospital centre social media platforms (six AA and two CV). These people were considered key stakeholders because they have valuable information, as health professionals, as people directly concerned or as caregivers involved in their community. The cases chosen should provide the greatest possible wealth of information for an in-depth study of the research question.[42] A. M. carried out the recruitment in June and July 2020. We conducted data analysis in parallel with ongoing data collection. We stopped at 15 interviews. The sample size was not predetermined and was based on the richness of the data. This is because we were able to answer the research questions.

Data collection

Fifteen in-depth semistructured interviews with these key stakeholders were carried out. After receiving training in qualitative research, A. M. conducted the interviews in July and August 2020. While most interviews were conducted via Zoom, one was by phone. One autistic person was asked to answer the questions in writing, and another person also added answers in writing after the interview. Interviews were conducted in French and lasted approximately 1 h. Finally, the interviews (by Zoom and by telephone) were recorded and transcribed by A. M.

We developed an interview guide based on the TFA and adapted it to each group. Autistic persons were asked about their needs surrounding the four types of social support: emotional, appraisal, instrumental and informational, [12] to understand their attempts to respond to those needs. An example of a question was, *when you are going through a difficult situation or you are going through negative emotions, what do you do to get better? Has this happened to you lately? How did that happen?* We also wanted to examine the role of peer support in the course of their efforts to respond to their needs in terms of support.

The second topic centred on how a peer support service would be received. An example of a question was, *could the life experiences of another autistic person be a reference, a model, an inspiration or a source of support that could help you better face problems if you have them? If yes, why? If not, why?* Based on research experiences of modalities of peer support, we also explored the participants' perception of three possible forms of social support for peers: support groups, mentors and social support online.[26–30] An example of a question was, *what would be the challenge to overcome in a possible service?* The comprehensibility of the questions was consulted and discussed with one autistic person collaborator. Changes were made afterwards. While different interview guides were used for caregivers and service providers/healthcare professionals, respectively, both interview guides comprised the following topics: (1) what they consider to be autistic persons' social support needs and (2) the acceptability of a structured programme. The interview guides were developed in French (available alongside data in Dataverse).

Data analysis

A qualitative thematic analysis[43] of the content of the verbatim was carried out by three researchers (A. M., A. V. and K. C.) using the NVivo 12 software according to the TFA.[35] The purpose of the thematic analysis was to identify the semantic units that constitute the discursive universe of the statement to reformulate the content in a condensed form.[43] The analysis was carried out in four stages: (1) the identification of significant ideas, (2) their categorization, (3) within each group of targeted actors (intragroup analysis) and (4) between the groups of targeted actors (analysis intergroups).

To enhance the trustworthiness of the data analysis we went through the next stages.[44] A number has been provided to each participant to maintain anonymity. Then, the first author reads the data several times to repair emerging issues, trends and identify patterns among coded categories. Therefore, we developed an initial codebook based on these initial readings of the interview data, along with aspects of the theoretical framework. The first coding was carried out by A. M. A. V. performed 60% of the encodings. Double-coded interviews were compared, and

disagreements were discussed until consensus was achieved. So, interview data were thus initially coded, based on: (1) the needs of autistic people in terms of social support (i.e., emotional support, informational support, appraisal and instrumental support),[12] (2) the past experiences of social peer support: positive experiences and drawbacks, (3) the acceptability of a structured service of social peer support for and by autistic persons (more precisely, around the seven constructs of the TFA: affective attitude, burden, perceived effectiveness, ethicality, intervention coherence, opportunity costs and self-efficacy)[35] and (4) the possible barriers and facilitators of a peer support service.

Afterwards, for the purposes of theme development and refinement, a template or framework was then developed following adjustments stemming from discussion among all the researchers involved in the study, until consensus was reached.[45,46] Theme development and refinement were characterized by an iterative process in which interview data and theoretical constructs are confronted and contrasted.[47] Ambiguous verbatim were also discussed as a team and reclassified. Then, K. C. performed a third data analysis. Finally, results were presented to the rest of the team and further discussed until a consensus was reached.

FINDINGS

Theme development was guided by concepts stemming from the Acceptability framework (TFA) along with concepts pertaining to various types of social support.[12] Table 2 shows how the framework was adapted for the data analyses (Table 2). Findings point to four major themes: (1) Social support needs and what peer support might mean; (2) The underpinnings of a structured social support programme provided by peers: ethicality and perceived effectiveness; (3) Practical considerations for implementing a structured peer social support programme: self-efficacy and intervention coherence and (4) The costs and potential barriers around the development of such a service: burden and opportunity costs.

Table 2 Acceptability framework (Sekhon et al.) adapted to issues around social support provided by autistic persons

Concept	Definition	Issues pertaining to social support provided by autistic people
Affective attitude	Feelings about an intervention (social support programme)	Interest in or resistance towards social support given by autistic peers Understandings of social support and social support needs Perceived social support needs (emotional, informational, appraisal and instrumental support) Recognition around the need for social support Gaps in social support
Ethicality	Fit between intervention and individual's value system	Sharing and understanding of similar experiences with other autistic peers

Intervention coherence	Understanding of the intervention by the individual and how it works; fit between components of intervention and intended aim	Understandings of the intervention; ways of defining what the intervention does Potential and limits of social support by autistic peers Comparisons with professional services Views (preconceived notions) around ability of autistic people to understand intervention among clinicians and healthcare professionals Partnerships and gaining credibility in the community
Perceived effectiveness	Perception of likelihood of intervention to achieve its purpose	Possibility to engage in mutual understanding Flexibility in meeting different social support needs Role of peer supporters Role of and support from healthcare professionals around social support by autistic peers
Self-efficacy	Participant's confidence in taking part in the intervention	Frequency of intervention Routine of intervention Format of meetings (e.g., videoconferencing) Type of discussions
Opportunity costs	Benefits, profits and values given to participate in the intervention; what has to be or was given up to engage in the intervention	Potential barriers to participating in and implementing the intervention Need for training
Burden	Perceived amount of effort required to participate in the intervention	Pressure to share experiences Compatibility among autistic persons taking part in social support intervention Extent to which the intervention is adapted to sensory characteristics of autistic peers Extent to which the intervention is adapted to communication characteristics of autistic peers

Social support needs and what peer support might mean

Underlying perspectives surrounding the acceptability of structured social support programmes by and for autistic people were participants' experiences seeking social support, the lack of social support in their daily lives as well as the gaps in terms of social support needs for autistic people.

Seeking social support and sometimes finding it

First of all, the need for social support was recognized by participants in this study, as reflected in this comment by an autistic person:

Of course, one thing that is common to everyone is that we all have a need to talk about what we are going through personally, regardless of the level of difficulty …Even if things are going well, it is certainly relevant to express our daily concerns. (AA5)

Autistic participants mentioned encountering issues with instrumental support (material and financial resources). More specifically, they experienced challenges accessing services access, for example, to transportation, something that was echoed by caregivers and healthcare professionals. Also, the need for informational support was raised by autistic persons as well as parents/caregivers who took part in this study. Some autistic participants mentioned the internet as a source of information as well as documentation found through the healthcare system. Among the information that was sought, one autistic person mentioned the need for information on how to manage one's emotions, food sensitivity and sensory perceptions. Information needs pertaining to finances, work, education, love, sexuality as well as identity were raised by caregivers interviewed in this study.

Two autistic persons mentioned being partially successful in meeting their support needs, saying that they were counting on someone in the family. Their mothers primarily were the source of emotional and instrumental support. However, for two service providers, the need for social support could be met rather through social integration, especially at school or at work, but also through rehabilitation services. Therefore, the perspective of the social support needs of autistic persons was not the same, whether it was the people concerned or the other key informants.

Service providers and caregivers interviewed in this study tended to focus on the need for socialization and noted the loneliness experienced by the autistic people they encountered. Thus, socialization groups with other autistic persons, facilitated by professionals and social media groups such as Facebook were the sources of socialization outlined by some autistic persons. Above all, unmet needs for social support were mentioned among autistic people interviewed, as reflected by one of the participants:

So I don't feel support, I don't feel anything useful. I don't feel an understanding or anything. So I can say very clearly that I don't feel any support. (AA2)

How an autistic peer might respond to social support needs

One caregiver considered providing informational support as central to the role of an autistic peer support worker. Another mother considered them (peer support worker) to be best equipped to help autistic persons compose with a neurotypical society in a pragmatic way.

With regard to emotional support, autistic participants expressed the need to find the ‘ideal’ autistic peer support worker (as put by one autistic person interviewed for this study), that is someone who can understand their feelings from the perspective of an autistic person, as reflected by one mother interviewed in this study:

If it could be someone who can explain well according to his perception of the environment […] I think that would be a win for him [hers son]. (CV4)

Healthcare professionals also mentioned the role autistic peer support workers could play in providing emotional support, with caregivers acknowledging the need for emotional support to prevent psychological distress. Alongside emotional support was the need to develop autistic persons' self-esteem. Autistic peer support worker was deemed suitable to provide such support, particularly according to the healthcare professionals interviewed for this study.

Finally, stigmatization remained an issue according to the autistic people and caregivers who took part in this study, hindering social support as well as the self-esteem of autistic persons. Autistic participants mentioned feeling judged on account of their autistic particularities. Further aggravating the situation was the categorization of autism along with false beliefs around the condition.

Autistic adults and caregivers interviewed indicated that such a structured social support service would be accepted, as this verbatim shows:

A peer support service, I would certainly make good use of it. I could even tell you that I am in contact with adults in

my condition, and we help each other a lot…. (AA5)

Peer support was deemed relevant for autistic people as well as for parents. Peer support was underlined by one parent as a form of social support for autistic persons:

I think there is nothing better than one person seeking to understand another person with the same condition. (CV4)

All autistic people and caregivers had a positive attitude towards structured peer social support services. Such a service was deemed compatible with their values of mutual aid. Participants understood that peer support might be possible due to the life experience of peers and their ability to share it. Table 3 shows verbatim according to the perceptions of the participants and the different types of support plus their affective perception towards this service (Table 3).

Table 3 Social support needs and what peer support might mean

Social support needs/verbatims	
Seeking social support and sometimes finding it.	
Emotional support	‘I cry a lot, I call in different places but I have a feeling that people don't really understand what autism is. Just to make yourself understood creates conflict’ (AA1).
	‘The only person who understands me is my son because he is also atypical’ (AA2).
	‘I read on forums …I am on Facebook groups where I chat. Recently, for example, I had a big concern to share …so uh, I wanted to share that in order to evacuate’ (AA5).
Instrumental support	‘I call on people, generally strangers, for a service’ (AA4).
	‘I have no one to help me in an emergency’ (AA6).
Appraisal support	‘I have a feeling that for a part of society, no matter how good or successful I am at something, I am the “incapable autistic”’ (AA6).
Informational support	‘My social worker. Otherwise, I found it in books or on websites. I also have an autistic friend who has read everything and who gives me good summaries with all the details’ (AA4).
Social support needs/verbatims	
The perspective of the social support needs of autistic persons was not the same, whether it was the people concerned or the other key informants.	

Social support	‘It is against their nature to be in a social environment. The best support for a young adult with autism is individual support from a professional, whether an educator or a social worker or a psychologist’ (SP4).
Affective attitude to a peer support service/verbatims	
An autistic peer might respond to social support need of other autistic persons.	
	Autistic person: ‘Even I think about it. Maybe later becoming a peer helper, because I have a certain lived experience and I can give to others. I think it's more through experience that we can share, so that things get better with people. Instigate light in them’ (AA1).
	Family member: ‘I think there is nothing better than one person seeking to understand another person with the same condition’ (CV4).
	Healthcare professional: ‘I think it's a great way to provide quick and effective support for people with peers who experience much the same reality’ (SP01).

The underpinnings of a structured social support programme provided by autistic peers: Ethicality and perceived effectiveness

Past experiences with social support initiatives underpinned participants' concerns around the ethicality and effectiveness of a structured social support programme. More specifically, experiences among some of the autistic persons interviewed with unstructured spaces (whether through the internet or community organizations) led them to emphasize the possibility to share their experiences in the absence of healthcare professionals. They also underlined the need for guidance and training among those moderating a support group. Table 4 outlines autistic persons' perspectives based on the requirements of social support initiatives stemming from their past experiences, which centred on the possibility to share one's concerns and experiences freely. This not only spoke to the ethicality of social support initiatives but also their effectiveness.

Table 4 What should underpin peer social support: Ethicality, perceive effectiveness and past experiences with unstructured spaces

	Verbatim
Ethicality	‘I participate in a social group; we exhibit what we are experiencing and then everyone can give a comment. We meet between us and then if someone has exposed a situation, we can validate saying yes. I experienced a situation like that or not, then we come to understand each other’ (AA4).

	‘I like my socialization group because we can talk about whatever topics we want, there are no service providers’ (AA5).
	‘People who have gone through adversity who are able to share their life experience …Without even giving advice that could indeed give me ideas’ (AA2).
Perceived effectiveness and past experiences with unstructured spaces	‘It does me a lot of good to have friends who are autistic like me. It's better than therapy. At least I can have a social network’ (AA2).
	‘We understand each other, we don't need to look each other in the eyes to talk to each other, we won't use all of our efforts like when talking to a non-autistic person’ (AA4).
	‘The difficulties in terms of theory of mind, in terms of reading social issues, or in terms of cognitive flexibility precisely make them more vulnerable in terms of mentoring attitude so it's not just anyone can be a mentor and the person must be trained and at the limit supervised or coached’ (SP1).

Furthermore, the need for a structured social support programme to be adaptable and flexible to individual needs were the focus among some autistic participants. Among the autistic persons interviewed, the effectiveness of such a service resided in the possibility for autistic people to engage in mutual understanding of their experiences. Autistic participants shared a similar view of the role of peer helpers as primarily a source of comfort, informational and instrumental support, in addition to encompassing a socialization role. For service providers, peer helpers represented rather a functional resource to health services, as underlined by one service provider:

The peer helpers could certainly play a role of reception, validation and comfort. At best, it might play a role of a bit of guidance, to the right services, the right resources. (SP5)

With the exception of some service providers, effectiveness and ethics went hand in hand when it came to peer-to-peer social support programmes (Table 4).

Practical considerations for implementing a structured peer social support programme: Self-efficacy and intervention coherence

Practical considerations over the implementation of a structured peer social support programme entailed both the possibility for autistic people to participate as well as the need to disseminate such a programme within the wider community. Of particular concern was the location. In general, autistic persons reported that the location of the service was irrelevant, provided it is easy to access and sensory perceptions are considered. An autistic participant and a service provider mentioned videoconferencing as an option, particularly within the context of the COVID-19 pandemic and even after. Some parents made a similar suggestion, with one mother mentioning that it made little difference to her whether the support programme occurred in a hospital, a clinic or a community organization:

[The autistic person] must be able to identify to the group […], but for the rest, I honestly think that it would not matter if [the service] was in a hospital, a clinic or a community organization. I think that is a detail. (CV4)

Compared to the autistic participants, healthcare professionals interviewed in this study expressed more concern over the location of the support programme, preferring it to take place within a local government health agency (centre local de services communautaire [CLSC]). The latter was thought to ensure the accessibility and continuity of such a programme as a service. Nonetheless, a community organization was also deemed acceptable. However,

healthcare professionals interviewed for this study remained divided with regard to a support programme being situated within a hospital, for fear that the programme would be perceived as a medical service by autistic persons:
It's all in the art of presenting the service not as …a health service, but as a support service. (SP01)
Some autistic people interviewed prioritized a space without health professionals, supervised discussions and support for autistic leadership as factors that would influence their adherence to a potential social support programme (Table 5). However, there was a lack of consensus among the service provider participants. Some expressed doubts about the ability of autistic people to offer support to others. In addition, regarding the component *intervention coherence* or the extent to which the participant understands the intervention, and how the intervention works, some suggested that such a programme would be a low-quality health service.

Table 5 What a structured peer social support programme requires: Practical considerations

Intervention coherence	
Dissemination and promotion of the support programme	'The dissemination and promotion on the internet through our own services, to integrate it to our service and inform schools that it is a service we are offering' (SP01).
	'Community organizations, promotion through social media […] promote it to parents by presenting the project. I also think it could be in the school system and in the workplace where there are autistic people' (SP02).
	'Promote it in hospitals where psychiatrists receive adolescents or adults' (PS02).
	'Maybe it could be done through the meeting with the doctor, so […] he would have a presentation I think during that meeting, well …The person would already be less afraid I think of seeing this person again then it would give credibility to the person if they were presented by the doctor as being a really useful service and that they could therefore benefit from using it' (CV4).
Comparison with professional service	'The service offered by the peer helper will not be able to go as far as a professional service. It will not be able to give the same depth' (SP1).
Self-efficacy	
A framework for discussion	'A lot of people talk a lot about their difficulties, it's good to hear it once, there is compassion …we offer compassion because the idea is also to empty your bag, but then I would like us to move on to something else' (AA2).

Support to autistic leadership	‘She tried to intervene the time I said in the group of friends that I was afraid of catching Covid-19 by picking up my recycling bin and she told me that she finds my overreaction. Afterwards, Madam leads a support group without having experience in intervention. I felt judged’ (AA4).
Predictable	‘Once a week is good [.] because I make an appointment. So, I'm going to put it in my routine and then I have no problem [.]’ (AA2).
	‘A pleasant, quiet room, no matter where’ (AA6).
A space without service providers	‘For example, we meet with friends, then we just talk to each other, without there being a predefined topic. Probably a trained person, who knows autism, who does not judge, who does not act by intervening, that is to say who does not give orders or tools, for example’ (AA4).

Other than the location was the frequency of the social support meetings, with emphasis on a fixed routine and a regular schedule, particularly among autistic people interviewed for this study as well as parents. Structured meetings with clear objectives and themes were also considered important in the design of a social support programme for autistic people, both by healthcare professionals and autistic participants. In addition, for one autistic person, a mentor available in times of crisis was noted:

Once a week is good […] because it is a fixed appointment. I then have a routine and I don't have any problems […], but it would also be [good to have a mentor if I go through a crisis for example. (PA02)

Concerns over how best to promote or disseminate a structured peer support programme in the community were raised among healthcare professionals interviewed for this study. They expressed some reluctance regarding the benefit of a peer support programme considering it often as improvised. Underlying such concerns was the need to recruit potential autistic peer support workers.

The importance of forging partnerships with other social and health organizations as well as the school system was emphasized by several healthcare professionals interviewed, namely as a way for such a service to gain credibility. This was thought of as carrying the potential to offset any reluctance around its implementation and ensure buy-in around such a programme. The need to educate healthcare professionals and clinicians about peer support was considered another strategy to obtain buy-in around such a programme.

Finally, a parent highlighted the benefit of the programme being presented by a doctor as a way to augment its credibility. One autistic person emphasized the need for support immediately following a diagnosis corroborating the relevance of having doctors suggest the programme to patients.

The costs and potential barriers around the development of such a service: Burden and opportunity costs

Regarding the burden and opportunity cost components of the TFA model, autistic persons reported untrained or skilled peer helpers and/or an organization that does not consider the communicational and sensory peculiarities of autistic people as reasons for dropping out of a social support programme. Among healthcare professionals interviewed for this study, training for potential autistic peer support workers was nonnegotiable. As to what this training would comprise, autistic participants emphasized the need for a peer support worker to know when to intervene and how, for example, when an autistic person experienced sensory overload. Parents also pointed out the need for autistic peer support workers to accompany the autistic person as well as to be knowledgeable of existing services and resources for autistic people. Healthcare professionals raised the latter point, along with the need to train autistic peer support workers about communication techniques, stigmatization issues as well as about autism in general.

Even with trained autistic peer support workers and a programme adapted to the communication and sensory peculiarities of autistic people, some autistic participants remained sceptical given issues around compatibility among autistic people taking part in such a programme. Adding to this scepticism was the pressure to share one's experiences with other members of the group. Finally, apart from possible issues accessing the service, autistic persons interviewed did not view any further difficulty using such a service. Table 6 shows the costs and potential barriers for a structured social support programme for and by autistic peers according to the participants.

Table 6 Possible barriers for a structured social support programme by autistic peers

Opportunity costs	
Training peer helpers	‘That depends on the people. I know a girl, when she tells her stories, she picks up bad comments, judgmental reports. That's why I said that intervention training for these peer helpers would be good’ (AA4).
	‘It has to be autistic qualified to listen and to put things into perspective. He must be able to transform what we are saying into a concrete tool for improvement. If we manage to progress, to develop tips, it has to be beneficial anyway to follow me on the long term. That interests me. So, it must be a qualified autistic, who has a qualification’ (AA2).
	‘The negative point I could say is maybe someone who doesn't have a lot of training to help you or it's someone who has a bigger problem’ (AA4).
Burden	
Adapting to communication particularities	‘A conference via an internet link will tire me out. My tolerance level is 30 minutes, but when I chat in writing I can chat for hours’ (AA2).
Adapting to sensory particularities	‘Because of the sensory perceptions, the light level, the temperature and all that stuff, I know these things are things that I couldn't stand, so it is not even worth thinking about going to a particular environment’ (AA1).
	‘A place that is easy to access and not too noisy and with nonaggressive lighting’ (AA4).
No obligation to share	‘It is as if in these groups it is obligatory to speak about our sufferings and not to speak about what we were proud of and what we accomplished’ (AA2).
Considering compatibility issues	‘With compatible people. Not all autistic people are compatible’ (AA5).
	‘The big disadvantage is falling into a relationship of convenience with the peer helper. For example, “I had a lot of difficulty finding a job.—Ah yes me too, uh I understand you but me it's worse”’ (SP1).

DISCUSSIONA structured peer support service for and by autistic persons is acceptable for autistic persons and caregivers and for some service providers

To our best knowledge, this research is the first to study the acceptability of a structured programme of social support for and by autistic peers from the perspective of different key stakeholders. The interviews of autistic people and caregivers carried out in this research showed their acceptance of a structured autism peer social support programmes.

They self-identified with the values of solidarity of peer support and underlined the importance of the experiential knowledge of autistic peers. However, at the same time, some service providers expressed their doubts about the ability of autistic people to offer this support. Furthermore, the success conditions could be the training of peer helpers, the framing of discussions, the support for autistic leadership of the service and an organization that considers the communicational and sensory characteristics of autistic persons. Finally, a space without service providers was an important condition for the acceptability of a peer support programme.

There are many reasons why peer support for and by autistic persons could be effective. The Double Empathy Problem suggests a higher rapport during interactions between pairs of the same neurotype,[48] indicating a smoother relationship between autistic people. Autistic people possess a distinct mode of social interaction style, rather than demonstrating social skills deficits.[49,50] In addition there is a greater affinity between them and a sense of belonging as well as some preference on their part for interaction with other autistic people.[51,52] Even more, information sharing between autistic people could be more effective than the information transfer between autistic and nonautistic people.[49] Although the autistic participants in our study did not request a space without nonautistic people, they requested a space without health professionals (service providers), who are almost always nonautistic.

Possible barriers and facilitators for the organization of a structured peer support service for and by autistic persons

In our study, all autistic participants had experiences with spaces of spontaneous exchanges between autistic people. Our study shows that autistic people make use of unstructured spaces of peer support through social media, which is in agreement with Zhao et al.[53] They express positive experiences, but also negative ones, for example, the lack of framing of discussions.[54] Therefore, as reported by Martin et al.,[29] training peer helpers and support to autistic leadership, could be a facilitator to improve the disadvantages of unstructured spaces through social media or others. Even more, as described by Cherba et al.[55] the direct implication of health professionals in peer support services could be sensed as a manifestation of authority or of an unequal power relationship.

Our study also shows that a peer support service might encounter scepticism from service providers, related to the capacity of autistic persons to offer support to others. This might be related to the experience of participants with verbal communication skills of autistic persons, which is greatly heterogeneous. It could also be related to the implicit stigma of autistic people in health care,[56,57] and could underlie this lower acceptability. Further inhibiting the establishment of a peer support service is a misunderstanding of the social support needs of autistic people as well as insufficient knowledge of peer helpers' role on the part of service providers.[18] Efforts should also be made to combat the stigma of autism in healthcare settings and to improve the acceptability of peer helper services in health care. Therefore, improving acceptability by health professionals is important to integrate a service like this across the continuum of healthcare services or into the community.

Next steps

There are further challenges in planning and organizing a structured programme of social peer support for and by autistic persons. There is a need of developing adequate training for peer helpers, of offering them the necessary mentoring and support for safe services, like resources for risk management, abilities for community management or access to communities of practice of autistic peer helpers.

Peer support workers in mental health are aimed at alleviating symptoms and risks associated with illness, disease control and well-being with illness.[19,58] The autistic participants in this study did not perceive themselves as sick people. The kind of peer support asked for focuses mainly on concepts of health promotion in a vulnerable population. This is why we need to understand how a recovery approach can be complementary to strategies of health promotion for promoting positive mental health[59] with social support to autistic persons by autistic peers.

Study limitations

The study highlights a gap between service providers and autistic participants regarding the perceived capacity of

autistic persons to offer peer support. Some service providers might have been influenced by general stereotypes about autism (e.g., 'It is against their nature to be in a social environment'), while autistic participants might have been referring to their own abilities. Or maybe autistic participants and service providers referred to different situations when talking about autism, due to the heterogeneity of autistic people receiving social or health services. We did not explore the particularities of autistic people to whom these workers offered services.

We have to admit that the meaning of social support for autistic people was not enough explored in our study. We used one of the most accepted definitions of social support.[12] A more contextualized approach to social support would be necessary to improve research for useful interventions and practices in a particular context,[14] like in autism.

In addition, while this study did not clarify how saturation was established, it was nonetheless possible to capture a certain level of wealth and depth of information based on the interviews that had been conducted, rendering the sample size appropriate for the purposes of this study.[60,61] We interviewed different groups of key stakeholders to account for diverse perspectives. We are aware of the fact that the sample is not representative of any of these groups. This is possible that the characteristics of this sample of autistic persons like being an adult, who was recruited by social media influenced their interest in using social support. We only interviewed autistic adults living in Quebec and speaking fluently in French who expressed themselves orally. Even if these results cannot be generalized to all autistic persons and stakeholders, this research gives the first insight into this important topic. Additional co-construction work is needed to confirm our findings and assess our proposals.

CONCLUSION

A structured peer support service for and by autistic persons is acceptable for autistic persons and caregivers and for some service providers. To organize a structured autism peer social support programme, we assessed the acceptability of providers and recipients. The training of peer helpers to facilitate discussions among peers, an organization that considers communicational and sensory characteristics of autistic persons and support for autistic leadership of the programme appear as factors that would influence adherence to such a programme. In addition, a provider-free space is an important condition for the acceptability of such a service. It seems essential to communicate among health professionals to promote this approach and reduce possible prejudice about the ability of autistic people to offer support to their peers. Finally, a structured peer support service for and by autistic persons could be an innovative way to answer the unmet support needs of autistic people. More studies are necessary.

AUTHOR CONTRIBUTIONS

Alena Valderrama designed the study. Alejandra Martinez oversaw recruitment and data collection. Alena Valderrama, Kathleen Charlebois and Alejandra Martinez carried out the data analyses. All authors discussed the results and contributed to the article draft. All authors have reviewed and approved the article before submission. This article has been submitted solely to this journal and is not published, in press or submitted elsewhere.

ACKNOWLEDGEMENTS

We thank Mathieu Giroux for reviewing and providing suggestions to the interview grid. We thank Dr. Lila Amirali for her support to this research. This is nonfinanced research.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Alena Valderrama—Dataverse (Université de Montréal). Valderrama, Alena, 2022, 'For Autistic Persons by Autistic Persons: Acceptability of a Structured Peer Support Service According to Key Stakeholders', https://doi.org/10.5683/SP3/3L2FXU, Borealis.

ETHICS STATEMENT

The study obtained approval from the Research Ethics Board of the Sainte-Justine University Health Center (CHU Sainte-Justine) project 2020-2858. Online informed consent was obtained from all the participants. All authors have reviewed and approved the article before submission. This article has been submitted solely to this journal and is not published, in press or submitted elsewhere.

DETAILS

Subject:	Students; Mental health; Mentors; Leadership; Intervention; Families &family life; Peer tutoring; Protective factors; Prejudice; Advocacy; Autism; Qualitative analysis; Stakeholders; Social support; Participation; Social workers; Health education; Peers; Caregivers; Health information; Higher education; Teams; Socialization; Autistic children; Health services; Facilitators; Social development; Medical personnel; Acceptability; Interest groups; Unmet needs; Support groups; Perceptions; Autistic adults; Interviews; Sensory properties; Health professional-Patient communication; Social interactions; Qualitative research
Business indexing term:	Subject: Mentors
Location:	Canada; Quebec Canada
Identifier / keyword:	acceptability assessment; mental health; mental health promotion; peer support; social support
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	463-475
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article

Publication history :	
Online publication date:	2022-11-29
Milestone dates:	2022-10-29 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-08-08 (manuscriptReceived); 2022-11-29 (publishedOnlineEarlyUnpaginated); 2022-11-08 (manuscriptAccepted)
Publication history :	
First posting date:	29 Nov 2022
DOI:	https://doi.org/10.1111/hex.13680
ProQuest document ID:	2767101873
Document URL:	https://www.proquest.com/scholarly-journals/autistic-persons-acceptability-structured-peer/docview/2767101873/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the "License"). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-11-01
Database:	Public Health Database,Publicly Available Content Database

Public contributors' preferences for the organization of remote public involvement meetings in health and social care: A discrete choice experiment study

Loria-Rebolledo, Luis E [1] ; Watson, Verity [1] ; Hassan, Shaima [2] ; Gabbay, Mark [2] ; Tahir, Naheed [3] ; Hossain, Muhammad [4] ; Goodall, Mark [2] ; Frith, Lucy [5]

[1] Health Economics Research Unit, University of Aberdeen, Aberdeen, UK [2] Department of Primary Care &Mental Health, University of Liverpool, Liverpool, UK; National Institute for Health and Care Research ARC North West Coast, Liverpool, UK [3] National Institute for Health and Care Research ARC North West Coast, Liverpool, UK [4] Health and Social Care, University of Wales Trinity Saint David, Carmarthen, UK [5] Department of Law, Centre for Social Ethics &Policy, University of Manchester, Manchester, UK

ABSTRACT (ENGLISH)

Introduction

Covid-19 expanded the use of remote working to engage with public contributors in health and social care research. These changes have the potential to limit the ability to participate in patient and public involvement and engagement (PPIE) for some public contributors. It is therefore important to understand public contributors' preferences, so that remote working can be organized in an optimal way to encourage rather than discourage participation.

Methods

We use an economic preference elicitation tool, a discrete choice experiment (DCE), via an online survey, to estimate public contributors' preferences for and trade-offs between different features of remote meetings. The features were informed by previous research to include aspects of remote meetings that were relevant to public contributors and amenable to change by PPIE organizers.

Results

We found that public contributors are more likely to participate in a PPIE project involving remote meetings if they are given feedback about participation; allowed to switch their camera off during meetings and step away if/when needed; were under 2.5h long; organized during working hours, and are chaired by a moderator who can ensure that everyone contributes. Different combinations of these features can cause estimated project participation to range from 23% to 94%. When planning PPIE and engaging public contributors, we suggest that resources are focused on training moderators and ensuring public contributors receive meeting feedback.

Discussion and Conclusion

Project resources should be allocated to maximize project participation. We provide recommendations for those who work in public involvement and organize meetings on how resources, such as time and financial support, should be allocated. These are based on the preferences of existing public contributors who have been involved in health and social care research.

Patient or Public Contribution

We had a public contributor (Naheed Tahir) as a funded coapplicant on the UKRI ESRC application and involved members of the North West Coast Applied Research Collaboration (NWC ARC) Public Advisor Forum at every stage of the project. The survey design was informed from three focus groups held with NWC ARC public contributors. The survey was further edited and improved based on the results of six one-to-one meetings with public contributors.

FULL TEXT

INTRODUCTION

Covid-19 prevention measures, which started in the United Kingdom in March 2020, forced a shift to remote forms of working in patient and public involvement and engagement (PPIE) in health and social care research. Due to shielding and social distancing, the usual ways of involving the public (such as face-to-face meetings and events) were not possible during the pandemic. Even though at the time of writing, Covid-19 restrictions have largely been removed or substantially eased, remote working will continue to be used alongside face-to-face meetings and as part of 'hybrid' working. Remote working has provided a valuable way of continuing to do PPIE during the pandemic and ameliorating some of the isolation felt by public contributors during the periods of lockdown in 2020.[1] However, remote working with its dependence on the Internet and communication equipment, needs to be carefully considered in light of socioeconomic and health inequalities. There is a digital divide that maps onto existing socioeconomic inequalities, with those in lower socioeconomic groups and older communities having less access to and opportunities to use remote working technologies.[2] Areas of high deprivation and ethnic minority communities bear the burden of poor health and access to health care and these communities have experienced disproportional harmful effects of the pandemic.[3,4] As a consequence, health inequalities are increasing.[5] Therefore, PPIE conducted remotely has the potential to further disenfranchise already disadvantaged groups and attention needs to be paid to ensuring diversity and inclusion in PPIE remote working.

The likelihood that remote working will continue alongside face-to-face meetings means that disenfranchisement due to the digital divide is added to concerns that PPIE was insufficiently diverse before the pandemic.[6] A recent National

Institute of Health &Care Research (NIHR) (a UK-based funder of health and social care research) survey of public contributors found a lack of diversity in the public contributor community in terms of age and socioeconomic status and addressing this is an NIHR priority.[7] This paper reports on a discrete choice experiment (DCE) survey that is part of a larger UK-based study that explored remote working in PPIE in health and social care research during the Covid-19 pandemic in 2020–2021.[8]

PPIE in health and social care research

PPIE has become a widespread phenomenon in health and social care research. The NIHR state: 'Public involvement is at the centre of NIHR health and social care research, and the public has a right to have a say in what and how publicly funded research is undertaken'.[9] The terms 'patient and public involvement and engagement' (PPIE) or public and patient involvement (PPI) are commonly used to capture a broad range of activities that aim to develop effective links between researchers and the general public. We will use a broad definition of PPIE for the purposes of this paper, 'research being carried out 'with' or 'by' contributors of the public rather than 'to', 'about' or 'for' them.' PPIE includes notions of active contribution,[10] and 'good' PPIE is more about coproduction than just involvement.[11] 'Coproducing a research project is an approach in which researchers, practitioners and the public work together, sharing power and responsibility from the start to the end of the project, including the generation of knowledge'.[11]

We use the term 'remote working' to cover meetings and interactions held without face-to-face contact that use communication technologies such as telephones (landlines, mobiles, smartphones), computers, tablets, online conferencing/meetings software, social media, and apps. Hybrid meetings are where a meeting is held with some participants face-to-face and other participants joining remotely, such as via a video conferencing tool such as Zoom.[12]

There is limited research on the feasibility and assessment of remote working quality in PPIE. However, since the start of the Covid-19 pandemic, an increasing number of guidelines and recommendations have been produced on how to undertake remote working in PPIE. These include: 'Top tips for carrying out PPI activities during Covid-19' by the NIHR Research Design Service[13]; 'How do I hold a PPI meeting using virtual tools?' by the NIHR School for Primary Care Research[14] and 'Carry on coproducing: handy hints and tips to help you out', by the University College London Public Engagement Blog.[15] This paper contributes to this growing literature. In one of the few published articles, Lampa et al.[16] reported observations of digital PPIE meetings during the pandemic. They found that meeting organizers need to be committed to solving practical issues and it is important to coproduce the meeting structure and format with public contributors. Adeyemi et al.[17] discussed three case studies of remote PPIE with marginalized groups and concluded that it is possible to do remote work with such groups, but it also presents some challenges, predominately the challenge of digital poverty and lack of access to equipment and data/WIFI.

This study aims to provide evidence on what good practice in remote working in PPIE might look like. We used a DCE to elicit public contributors' preferences for different features of remote communication and working, such as investment in technology, time commitment, training, and support needs. The DCE aimed to find out:

- 1. how much time and resources public contributors would be able and willing to invest in remote communication,
- 2. what features of remote communication are the most important to maximize public contributors' participation, and
- 3. how public contributors trade off the different features when deciding whether to participate.

A better understanding of how to organize and support public contributors with remote working can help engage public contributors and allow teams to design remote working practices that are inclusive and encourage, rather than limit, diversity.

METHODS

DCEs are a survey-based method grounded in economic theory that assumes the value of a service (in this instance PPIE meetings) comprises the value of the different attributes that describe it.[18] DCEs are a widely used method to elicit preferences from the public, patients, and healthcare professionals.[19] Respondents in a DCE are asked to make a series of choices between two or more hypothetical alternatives describing different types of meeting packages. These packages are further described by different features (herein referred to as attributes) and a corresponding value (herein referred to as levels). For example, a meeting attribute could be the 'time of day the meeting takes place' and the levels could be 'between working hours' and 'between working hours and evenings'. When respondents make choices, they are implicitly trading the attributes and levels that describe the alternatives. This trade-off information can be used to estimate the relative importance of one attribute over another and predict participation in a defined meeting package.

In this study, public contributors were asked to imagine they were invited to take part in a new project. This would mean that they had to join regular project meetings using video calls. In the DCE, public contributors were then presented with a series of choices. In each choice, they were shown two different ways in which project meetings could be organized. These meetings differed in seven attributes. Contributors were asked to choose to take part in one type of meeting or not take part in the project (e.g., opt-out alternative). The DCE was designed using a state-of-practice sequential, mixed methods approach.[20,21]

The attributes and levels describing the remote meetings were identified and refined from the previous phases of this study (see Figure 1). This comprised two surveys, one with public contributors (n = 244) and one with those who worked in PPIE (n = 65) and subsequent qualitative interviews with public contributors (n = 22). The surveys asked general questions about the role and PPIE experience, digital literacy and different aspects of remote working. After analyzing the survey data, we conducted qualitative interviews to further probe and explore the themes (results of the previous phases are reported in Frith et al.[22] and Jones et al.[23]). This ensured that the included attributes of remote meetings are those which are both most important to PPIE contributors and amenable to change or under the control of meeting organizers.

Figure 1. Flowchart of the remote working in PPIE study. PPIE, patient and public involvement and engagement.

The data from the previous phases (Phase 1 and Phase 2) of our study identified three stages that influenced how PPIE contributors felt about participating in meetings: what happens before, during and after the meetings. Based on these findings, seven attributes, grouped into these three stages, were used to describe the remote meetings (see Figure 2). Four attributes described features of a meeting's organization that happen *before*: the length of the meeting, the time of day when the meeting is held, the type of connectivity support that is provided and the technical support provided to help participants to join and contribute during video calls. Two attributes described features that occur *during* meetings: the etiquette during a remote meeting and the role of the moderator. One attribute described whether any feedback on the contributors' contributions was provided *after* the meeting. The rationale for the selection of these attributes and levels, based on the previous phases' findings is described below (Table 1).

Table 1 Attributes and levels used in the choice experiment

Attribute	Levels
1. Length of meeting	1.5 h without comfort break.

	2 h with comfort break.
	2.5 h with comfort break and a socializing opportunity.
2. Time of day	Working hours.
	Working hours and/or Evenings/Weekends.
3. Connectivity	Use own device/Internet/electricity.
	Everything you need is provided (devices, Internet and electricity).
	Use own device. Internet and electricity expenses provided.
	Use own device and Internet. Electricity expenses provided.
4. Support during project (…on how to attend/participate)	Instructions only
	Instructions + online training
	Instructions + online training + one-to-one IT support
5. Etiquette during meetings	Expected to have camera on and be present throughout meeting.
	You can have camera off and you can step away when/if needed.
6. Role of moderator (Moderator focuses on…)	*(Standard moderator)* Only on ensuring meetings run smoothly.
	(Great moderator) On ensuring meetings run smoothly and makes an effort to make you feel comfortable and confident about contributing to meeting.
7. Meeting feedback (e.g., sense of contribution)	No follow up.
	General follow up that tells how broad contributions from the meeting were included.
	Personalized follow up that tells how individual contributions were included.

Figure 2. Example DCE choice task as seen by respondent. DCE, discrete choice experiment.

Length of the meeting was deemed a key feature that public contributors would want to know ahead of any remote meeting, with the data suggesting participants generally preferring shorter meetings, while some enjoyed meetings with an icebreaker and/or social activity that allowed them to interact with and get to know the other participants, albeit that is likely to lengthen the meeting time overall. Given the potential impersonal nature of remote meetings, we included the possibility of having a longer meeting with a social activity as one level in the DCE. The meeting's time of day was also important. Some contributors preferred the flexibility of meetings outside working hours and others preferred meetings during working hours, especially those with caring responsibilities. We include the provision of connectivity tools as an attribute that contributors would want to know before, as this allowed us to test whether providing web-enabled devices and reimbursing Internet and electricity costs is a way to overcome the digital divide and increase participation. Similarly, we included the provision of technical support as this was identified to be a potential driver of public contributor disenfranchisement, especially amongst contributors with limited experience and Internet literacy.

Previous phases identified meeting etiquette as a potential key driver of meeting and project uptake. Public contributors in the interviews discussed the difficulty of balancing long video calls from home with their caring responsibilities, with some describing the difficulties they experienced when they have distractions at home. Some public contributors expressed a preference for having the flexibility to attend meetings anywhere and/or have the possibility to manage other things suddenly, such as attending to family members. The meeting etiquette attribute thus described whether contributors had to keep their cameras on and be ready to contribute during the whole meeting, or whether it was possible to turn them off and step away when needed. The role of the meeting's chair or moderator was another important aspect identified in the interviews and surveys in the previous phases (for our purposes we are using them interchangeably to mean ‘the person who is running the meeting’ or organizing the meeting, as these are not formal decision making meetings where the chair has a formal role). Contributors were able to distinguish between a good and a sub-par moderator, with the majority agreeing on the importance this can have to the success of a meeting. We described this attribute in terms of a *standard moderator* who only ensures meetings run smoothly and a *good moderator* who also makes sure participants feel comfortable and confident to contribute.

A recurring theme in most of the interviews and survey data from the previous phases was the uncertainty of what happens after the meetings and, specifically, whether the public contributors had been listened to and their suggestions are taken on board. We, therefore, included the provision of feedback, either as a personalized report that details how each individual's contribution was used or a general report that explained how the group's contributions were taken onboard, as an attribute that can be both influenced by the meeting organizers and speaks to addressing this uncertainty. While not being an issue exclusive to remote meetings, this feature was deemed key given the nature of remote meetings and the way they can limit nonverbal communications between participants and moderators.

Based on the attributes and levels, there are 273,248 possible unique choice tasks (pairs of meeting descriptions). We used experimental design techniques to reduce these to a more manageable number. Specifically, we created a D-efficient experiment design with vague informative priors and allowing for estimation of nonlinear effects of attributes using Ngene software to reduce the number of choice tasks to 24.[24,25] The aim of this design was to create realistic choice tasks with statistical properties that facilitate the estimation of the effect of each feature.[26] To reduce respondent burden, the resulting design was blocked into three sets to each respondent was asked to complete eight choice questions.[27] Based on this design, a minimum of 49 respondents were required for each analysis block

to ensure the estimation of all attribute effects.[28] Respondents were randomly assigned to one block and the order of the choice tasks within each block was also randomized to minimize ordering effects.[29]

The DCE online survey was comprised of three sections (see Supporting Information 1). Section 1 asked about respondents' experience as PPIE contributors. Section 2 contained the attributes and levels. Section 3 included demographic questions to characterize the sample (age, number of children, self-perceived health and education level). The survey was tested in $n = 6$ think-aloud interviews with public contributors.

The DCE was administered as a self-complete online survey of UK residents who had been involved in at least one PPIE project as a public contributor. Survey recruitment used a combination of a targeted and opportunistic sampling amongst existing UK PPIE networks and colleagues. Data were collected between 6 September and 1 November 2021. Participants were asked to provide informed consent before the start of the survey and participants were not given any financial incentives for completing the questionnaire. The participant information sheet provided participants with details on how to access the study results. The University of Liverpool, Institute of Population Health Ethics Committee granted ethical approval (REF: 7636).

Data analysisDCE analysis

The DCE response data indicates which one of the three alternatives a respondent selects in each choice task. The data were analysed using a mixed logit (MXL) model.[30] We assume that respondents (n) choose the alternative (j) that provides them with the highest utility in each of the choice tasks (t). Following random utility theory,[31] utility can be decomposed into a deterministic part, V, which is observable and based on the attributes included in the DCE, and a random component, ε, which is unobservable. The observable component is specified as a linear and additive function of the attributes and levels describing the meeting types, where [Image Omitted. See PDF]

The $ASC_{opt\text{-}in}$ is an Alternative Specific Constant which takes a value of one for alternatives which have the participant opting into the project. This can be interpreted as the general preference to choose to take part in a project with remote meetings compared to opting out. The β parameters reflect the observed change caused by each of the meeting attributes/levels to the overall utility (e.g., benefit) derived from taking part in a project involving remote meetings. To allow for preferences to vary across the sample, the β_1–β_{12} parameters are assumed to be normally distributed. We estimate the mean and a standard deviation for each parameter, where the latter's statistically significant would indicate if the attribute's preference varied across the sample. Positive mean coefficients represent increases in utility, and negative coefficients as a loss in utility from the corresponding base level, which can be interpreted as whether they increase or decrease the likelihood of choosing to take part in a project. β_1–β_{12} attributes are effects coded, thus allowing the postestimation of mean estimates for all attribute levels.[32] The model is estimated using simulated maximum likelihood with 500 Halton Draws.

We then use the parameters to estimate participation in different remote meeting configurations. Participation probability for different scenarios h is estimated using [Image Omitted. See PDF]where β denotes the parameter of attribute k and x_{jk} is the level that the attribute takes in the scenario h.

We estimate the participation of different remote meeting configurators described in Table 2. For example, Scenario 1 describes meetings that are 2 h long, organized during working hours, where contributors can step away if needed, with a good moderator who provides general feedback. Scenario 4 describes a similar meeting organization but is less resource intensive as it does not provide any feedback. By comparing Scenarios 1 and 4, it is possible to calculate the effect of providing general feedback on participation. The chosen configurations in Table 2 describe different ways a meeting can be organized, and each involves a different allocation of resources available to meeting organizers. All analysis was done using the statistical software R. Confidence intervals were computed using the delta method.

Table 2 Features for scenarios used in participation rate analysis

Scenario	Length	Time of day	Etiquette	Moderator	Feedback
1	2 h with break	Working hours	Camera off and can step away	Run smoothly and confident participation	General feedback
2	2 h with break	Working hours	Camera on and ready at all times	Run smoothly and confident participation	Personalized feedback
3	1.5 h	Working hours and weekends/evenings	Camera on and ready at all times	Only ensures meetings run smoothly	General feedback
4	2 h with break	Working hours	Camera off and can step away	Run smoothly and confident participation	No feedback
5	1.5 h	Working hours and weekends/evenings	Camera off and can step away	Only ensures meetings run smoothly	No feedback
6	2.5 h with break and activity	Working hours	Camera off and can step away	Run smoothly and confident participation	No feedback
7	1.5 h	Working hours and weekends/evenings	Camera on and ready at all times	Only ensures meetings run smoothly	No feedback
8	2.5 h with break and activity	Working hours and weekends/evenings	Camera on and ready at all times	Only ensures meetings run smoothly	No feedback

Affordance theory

We drew on the concept of affordances to further analyse our data. The features of remote meetings can be conceptualized as furthering particular affordances. Building on Gibson's work, Norman defines affordance as 'the relationship between a physical object and a person…. [the] relationship between the properties of an object and the capabilities of the agent that determine just how the object could be possibly used'.[33] He gives the example of a chair, a chair affords—is for—sitting. There are many different potential affordances when actors use an object or artefact (such as remote meetings), there are 'bundles' of affordances. These bundles are not independent but interact, and this was captured by the attributes and levels used in this DCE.

RESULTS

Two-hundred and nine respondents completed the survey. Respondents were evenly split across the three analysis block sets of eight-choice questions. The median completion time was 14 min 29 s. The sample characteristics are described in Table 3. The modal respondent was an experienced PPIE contributor (e.g., involved in three PPIE projects), had taken part in remote meetings as part of their role, had access to devices and an Internet connection to enable joining remote meetings and was able to take part in remote meetings uninterrupted. Only 22% of respondents were completely certain that past contributions to other projects had been taken onboard (64% were at least very certain). Respondents were more likely to be female, over the age of 45, highly educated (at least

University or equivalent), living alone or with no more than one person, and having no caring responsibilities.

Table 3 Characteristics of respondents

Sociodemographic characteristics		
	Age	
	18–24	1
0.5%	25–34	5
2.4%	35–44	10
4.8%	45–54	28
13.4%	55–64	55
26.3%	65+	109
52.2%	Prefer not to say	1
0.4%	Sex	
	Female	133
63.6%	Male	76
36.4%	…Is it the same as gender you identify with	
	Yes	193
92.3%	No	2
1.0%	Prefer not to say	14

6.7%	Ethnicity	
	White	185
88.5%	Mixed or multiple ethnic groups	6
2.9%	Asian or Asian British	4
1.9%	Black, Black British, Caribbean or African	5
2.4%	Other	5
2.4%	Prefer not to say	4
1.9%	Marital status	
	Single	40
19.1%	Married, civil partnership or cohabiting	126
60.3%	Separated	2
1.0%	Divorced	11

5.3%	Widowed	25
12.0%	Prefer not to say	5
2.4%	Caring responsibilities	
	Yes	68
32.5%	No	139
66.5%	Prefer not to say	2
1.0%	Highest level of education	
	No qualifications	6
2.9%	GCSE or equivalent	17
8.1%	A levels or equivalent	20
9.6%	Apprenticeship or equivalent	33

15.8%	University or equivalent	125
59.8%	Other	8
3.8%	English first language	
	Yes	202
96.7%	No	7
3.3%	Employment	
	Full time employment	29
13.9%	Part time employment	21
10.0%	Retired	110
52.6%	Student	6
2.9%	Carer	8
3.8%	Unemployed	3
1.4%	Adults in household	
	1	55
21.9%	2	119

47.4%	3	27
10.8%	4	7
2.8%	More than 4	1
0.4%	Children in household	
	0	190
90.9%	1	13
6.2%	2	5
2.4%	3	1
0.5%	More than 3	0
0.0%	Household income	
	£0–£10,400	20
9.6%	£10,400–£20,800	31
14.8%	£20,800–£31,200	47
22.5%	£31,200–£52,000	34
16.3%	£5200–	30

14.4%	Prefer not to say	47
22.5%	*Experience as a contributor*	
		Involved in how many projects?
None	9	4.3%
One	26	12.4%
Two	38	18.2%
Three	36	17.2%
Four	11	5.3%
More than four	89	42.6%
…Out of the those involved in at least one:		
Is any project doing remote meetings?		
Yes	190	95.0%
No	10	5.0%
Involved in how many organizations as PPIE advisor		
One	79	37.8%
Two	70	33.5%
Three	43	20.6%

More than 3	17	8.1%
…which organizations?		
National Institute of Health Research (NIHR) organization or other government-funded research (MRC, ESRC, etc.)	166	79.4%
Third sector organization or charity (e.g., Alzheimer's Society, Cancer Research)	81	38.8%
The NHS or social care organization (e.g., a hospital trust, Clinical Commissioning Group, local authority)	123	58.9%
Other	46	22.0%
Involved in what capacity		
Carer	57	27.3%
Patient/service user	171	81.8%
Member of public/neighbourhood/community	128	61.2%
Other	21	10.0%
Currently has access to:		
Computer/laptop with webcam	191	91.4%
Tablet (or iPad)	110	52.6%
Mobile phone with camera	141	67.5%
Stable Internet connection (home broadband or mobile network)	179	85.6%
Headset/headphones with microphone	79	37.8%
In the past, has received payment to cover:		
Internet access	50	23.9%
Electricity bills	15	7.2%
Is able to take part in video calls without interruptions		
Yes	175	83.7%

Certainty past contributions have been taken on board		
Not at all certain	10	4.8%
Somewhat certain	20	9.6%
Moderately certain	45	21.5%
Very certain	88	42.1%
Completely certain	46	22.0%
Agree with given definition of a great moderator		
Strongly disagree	21	10.0%
Somewhat disagree	6	2.9%
Neither agree nor disagree	9	4.3%
Somewhat agree	45	21.5%
Strongly agree	128	61.2%

We found most respondents were willing to take part in a project that involved remote meetings and were willing to make trade-offs across the remote meeting attributes (in the DCE, *Meeting Package A* [first displayed alternative], *Meeting Package B* [second displayed] and *Not taking part* [third displayed] were chosen 43.8%, 47.0% and 9.2% of the time, respectively. Three respondents [1.9% of the sample] always chose the 'not to take part' option in all choice tasks). The DCE results are shown in Table 4. The ASC has a statistically significant positive parameter which suggests, on average, respondents are more likely to take part in a project involving remote meetings compared to not taking part. Statistically significant parameter estimates indicate that respondents prefer meetings which: are shorter (less than 2.5 h without a social activity); scheduled during working hours; permit them to have their cameras off and step away if needed; have a moderator that ensures participants are comfortable and confident, and provide feedback about how contributions were taken on board. There was no statistical difference between types of feedback, which suggests respondents do not distinguish between general or personalized feedback. The provision of devices or reimbursement of costs and receiving additional support such as training videos or one-to-one support to connect to meetings were not statistically significant and thus did not have an effect on the likelihood that respondents chose a type of meeting package.

Table 4 Parameter results from DCE choice questions

	Mean		Standard deviation	
Attribute	Estimate.	*p* Value	Estimate	*p* Value

Alternative Specific Constants (ASC)				
Opting in (e.g., choosing a meeting type)	1.140	<.001	-	-
Length				
1.5 h with no comfort break	0.247	.006	-	-
2 h with comfort break	0.381	<.001	0.250	.056
2.5 h with comfort break and social activity	−0.628	<.001	0.801	<.001
Time of day				
Working hours only	0.160	.008	-	-
Working hours, weekends and evenings	−0.160	.008	0.526	<.001
Connectivity tools (provide with…)				
Nothing	−0.084	.389	-	-
Devices, Internet and electricity costs	−0.064	.459	0.073	.883
Internet and electricity costs	0.042	.617	0.219	.268
Electricity costs	0.106	.256	0.104	.711
Support to connect (provide with…)				
Instructions	0.008	.907	-	-
Instructions and training videos	−0.051	.509	0.134	.504
Instructions and training videos and one-to-one support	0.043	.548	0.046	.815
Remote meeting etiquette				
Camera on and ready to take part	−0.390	<.001	-	-
Camera off and can step away when/if needed	0.390	<.001	0.677	<.001
Moderator				
Ensures meetings run smoothly	−0.091	.084	-	-

Also ensure comfortable and confident about contributing	0.091	.084	0.091	.493
Follow up (i.e., sense of contribution)				
No follow up	−1.058	<.001	-	-
General follow up document	0.565	<.001	0.785	<.001
Personalized follow up document	0.494	<.001	0.389	.033

Note: Log-likelihood = −1355.708. Number of observations = 1672. Akaike information criterion = 2761.415.
Abbreviation: DCE, discrete choice experiment.

Figure 3 shows the contributions to the overall utility and illustrates the trade-offs between meeting features respondents were willing to make. For example, the positive effect on the participation of being able to have the camera off and step away from the meeting if needed is not statistically different from the negative effect of having a 2.5-h long meeting. This suggests respondents could be compensated for taking part in longer meetings as long as they are able to have their cameras off. Similarly, meetings with a great moderator/chair can compensate for meetings that are organized outside working hours (e.g., weekends and evenings).

Figure 3. Contribution of parameter estimates to utility

Figure 4 shows the estimated participation for the different meeting configurations. Potential participation ranges from 23% to 94% depending on the remote meeting features. A project with the most desirable meeting features (e.g., Scenario 1: meetings that take 2 h with a comfort break, organized during working hours, with a great moderator, where participants can step away if needed and for which they received personalized feedback) has an estimated participation rate of 94%. Conversely, a project with the least desirable features (e.g., Scenario 8: meetings that take 2.5-h, organized outside working hours, with a standard moderator, where participants are expected to have the camera on and ready to contribute at all times, and for which they receive no feedback) would have a predicted participation of 23%.

Figure 4. Participation in different meeting configuration scenarios

Overall, the biggest effect on participation is the provision of feedback, followed by the length of the meeting (with shorter meetings being preferred) and whether respondents can turn their cameras off can step away if needed. For example, in a meeting with all the most desirable features, except the provision of feedback (Scenario 4) participation would be reduced by 19% (from 94% to 75%). As expected, features with nonstatistically significant parameters (e.g., providing devices and costs or extra support to connect to meetings) had a limited effect on participation and were not considered in the scenario analysis.

DISCUSSION

To our knowledge, this is the first DCE to investigate public contributors' preferences for how remote meetings in PPIE are organized. While remote working has the potential to limit the ability to participate for some public contributors, for others it can increase their participation.[23] People in our sample were generally willing to take part in projects even if this involves remote meetings. However, project participation can vary significantly depending on certain features of the meetings. Our findings suggest how project resources, such as time and financial support, can be best allocated to increase meeting participation by public contributors. Giving participants feedback about how their contributions to meetings are taken onboard by the organizers—how their contribution has made a difference was important to our participants. This has been found in other research in this area[34] and providing

feedback to public contributors has been described as an important, but often overlooked, part of PPIE leads' work.[35] Respondents seemed indifferent to whether this is general or personalized feedback, so there is little benefit from the additional resource cost of providing individual reports compared to a general one. We are not able to conclude if the importance of providing feedback is exclusive to remote meeting settings. However, our data suggest that this feature is important regardless of whether the meeting is remote or face-to-face. Furthermore, most respondents in our survey stated that they were not certain that their contributions had been taken on board in past projects. Given remote meeting settings can limit the nonverbal interactions and communication between all members of the team, it is likely that providing feedback that directly signals how their contributions were taken onboard is even more important in remote working.

We found that a meeting feature that is not resource intensive such as having remote meeting etiquette that permits participants to have their cameras off and step away is very important for increasing project participation. It is likely people value the flexibility to attend to other things, such as caring responsibilities while taking part in meetings. While having a moderator who ensures participants are comfortable to contribute was deemed less important, it is probable that resources should still be invested towards training or having experienced moderators/chairs. In the context of remote meetings where people might not be able to take part at all times, the role of the moderator to ensure that such flexible approaches result in meetings that run smoothly is key. Finally, we also found that long remote meetings should be avoided. Contributors are willing to forego the inclusion of social activities if the meetings are shorter. Resources allocated to arranging longer meetings with social activities should rather be focused on other features, such as moderator training and/or the provision of some type of postmeeting feedback to participants.

The features of remote meetings can be conceptualized as furthering particular affordances. Affordance theory has been used extensively in information technology and information systems research, to theorize the relationships between people and digital technologies. Thus, this theory is useful for understanding how public contributors made use of and interacted with remote working technologies. Volkoff and Strong[36] apply affordance theory to information systems research, for them, 'The power of the Affordance lens is that it helps to pinpoint the actors involved and the variety of potential actions they might engage in as they use the technology' (p. 5).

This DCE experiment shows the relative importance of the different means of bringing about, what has been found to be, key affordances in remote working. From our data, we developed three affordances: Affordance 1: reducing the burden of remote meetings; Affordance 2: involving everyone in the meeting; Affordance 3: influencing and improving research. For example, an important affordance for public contributors was 'making remote meetings less burdensome'. This DCE showed which features, such as length of meeting, camera use, and time of day of meetings were most important to our participants in terms of furthering this affordance (see Table 5). Bringing an affordance lens to our data enabled us to see how different features and elements of remote meetings interacted to understand how these different features afforded specific types of benefits to public contributors.[37]

Table 5 Elements giving rise to an affordance

Remote meeting features	Characteristics of actors
Affordance 1: Reducing the burden of remote meetings	
Camera	Public contributors can have their camera on or off

Length of meeting	Public contributors know they have time for meetings
Flexibility of attendance	Ability to step away, makes meetings less intense, public contributors can-do other things if needed
During working hours	Convenient for public contributors
Affordance 2: Involving everyone in the meeting	
Moderator	Everyone is given an opportunity to be involved
Etiquette	Everyone knows how to get involved
Affordance 3: Influencing and improving research	
Feedback	Public contributors feel valued and that their contribution is important

Source: Adapted from Strong et al.[37]

Strengths and limitations

The strength of this study is how we generated the attributes and levels to include in the DCE. This DCE was nested in a larger study that included two surveys, with public contributors and public involvement professionals, qualitative interviews and focus groups with public contributors. Therefore, the attributes and levels that were used had a firm evidence base. There are two limitations of our study. Firstly, our online survey administration will have impacted on the sample size and composition. The ongoing Covid-19 pandemic meant that we had to focus on online data collection and use existing contributor networks to distribute the survey link. We aimed to produce a short and well-presented survey that was easily and widely accessible so that it minimized data attrition and respondent drop-out rates, but future research could explore other sampling strategies. The Covid-19 pandemic also meant that many ongoing projects were using remote meetings, and therefore many people in existing contributor networks are now experienced in joining remote meetings. This may explain why we found, on average, that providing contributors with connectivity tools (e.g., devices or covering costs) and ongoing technical support to connect to meetings had no impact on project participation. Second, respondents had both technological literacy skills and experience. Ideally, we would have compared the preferences of experienced and inexperienced respondents. We did not have enough respondents who were inexperienced or had low technological literacy skills to perform subgroup analysis. This means that we cannot explore how a digital divide may affect preferences, not least as the survey was completed online. In the case of technological support to connect to meetings, while it is likely that if the public contributor has no experience some training/support is needed at the beginning of the project, our results show that once the person gains experience there is no need to allocate resources to provide ongoing support or training.

CONCLUSION

Our results provide important insights for researchers involved in the design and organization of meetings that include public contributors. The shift to remote meetings with public contributors caused by Covid-19 is likely to become a feature of PPIE. It is key we understand preferences and key drivers of project uptake to ensure remote meetings are designed so that potential public contributors are not disenfranchised. Hybrid meetings are also

becoming popular, and further research is needed on these types of meetings, as public contributors' preferences may be different in a hybrid meeting format, than when working solely online. We found that particular features of remote meetings can have a significant impact on project uptake, in our case ranging from 23% to 93% uptake. We identified features such as the provision of feedback, the role of the moderator, whether contributors need to have their cameras off and can step away, and whether the meeting length can have an impact on potential project uptake. We also found that features such as the provision of connectivity tools and support to connect to meetings did not have a significant effect, although this could be due to our sample having significant experience in remote meetings. Resources would be best allocated to moderator training and the provision of postmeeting feedback instead of arranging long meetings with socializing activities and providing ongoing technical support. These findings are useful for researchers, project managers and PPIE leads to inform the allocation of resources when designing remote meetings with public contributors. An allocation of resources that responds to contributors' preferences will likely result in higher uptake of public involvement in projects.

AUTHOR CONTRIBUTIONS

Shaima Hassan, Mark Gabbay, Naheed Tahir, Muhammad Hossain, Mark Goodall and Lucy Frith were involved in the conception of the study. Luis E. Loria-Rebolledo, Verity Watson, Shaima Hassan, Mark Gabbay, Naheed Tahir, Muhammad Hossain, Mark Goodall and Lucy Frith were involved in the design of the study. Muhammad Hossain, Shaima Hassan, Lucy Frith carried out the focus group discussions with public contributors, and Lucy Frith, Muhammad Hossain and Shaima Hassan performed the qualitative analyses. Luis E. Loria-Rebolledo carried out the survey development think-aloud interviews and led the quantitative analysis. Verity Watson reviewed the statistical model and was involved in the data analysis. Luis E. Loria-Rebolledo, Verity Watson, Shaima Hassan and Lucy Frith were involved in the original draft preparation. All authors helped shape the overall interpretation of the findings, and critically revised, edited and approved the final version of the manuscript.

ACKNOWLEDGEMENTS

We want to thank the public contributors who helped design this study and took part in the DCE survey. This research is supported by the National Institute for Health Research Applied Research Collaboration North West Coast (ARC NWC). The Health Economics Research Unit is supported by the Chief Scientist Office (CSO), which is part of the Scottish Government Health and Social Care Directorates. The views expressed in this publication are those of the author(s) and not necessarily those of the National Institute for Health Research, the Department of Health and Social Care or the CSO.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

DETAILS

Subject:	Digital divide; Polls &surveys; Socioeconomic factors; Video teleconferencing; Communication; Feedback; Patients; Financial support; Meetings; Applied research; Health disparities; Elicitation; COVID-19; Social behavior; Citizen participation; Public involvement; Resource allocation; Working hours; Moderators; Social care; Pandemics; Medical research; Health professional-Patient communication; Surveys; Multiculturalism &pluralism; Coronaviruses; Public participation; Discrete choice
Business indexing term:	Subject: Working hours
Location:	United Kingdom--UK
Identifier / keyword:	covid-19; preference; public contributors; public participation; remote
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	146-159
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-06

Milestone dates:	2022-09-05 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-06-30 (manuscriptReceived); 2022-11-06 (publishedOnlineEarlyUnpaginated); 2022-10-16 (manuscriptAccepted)
Publication history :	
First posting date:	06 Nov 2022
DOI:	https://doi.org/10.1111/hex.13641
ProQuest document ID:	2767101868
Document URL:	https://www.proquest.com/scholarly-journals/public-contributors-preferences-organization/docview/2767101868/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the " License"). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-04-24
Database:	Public Health Database,Publicly Available Content Database

Barriers and enabling factors in weight management of patients with nonalcoholic fatty liver disease: A qualitative study using the COM-B model of behaviour

Gu, Yunpeng [1] ; Zhou, Run [2] ; Kong, Tingting [2] ; Zhang, Wei [3] ; Chen, Yutong [2] ; Wang, Chunmei [4] ; Shi, Junping [5] ; Hu, Yanli [6]

[1] School of Public Health, Division of Health Sciences, Hangzhou Normal University, Zhejiang, Hangzhou, China [2] School of Nursing, Division of Health Sciences, Hangzhou Normal University, Zhejiang, Hangzhou, China [3] School of Public Health, Division of Health Sciences, Hangzhou Normal University, Zhejiang, Hangzhou, China; The Department of Metabolic Diseases Center, the Affiliated Hospital of Hangzhou Normal University, Zhejiang, Hangzhou, China [4] The Department of Metabolic Diseases Center, the Affiliated Hospital of Hangzhou Normal University, Zhejiang, Hangzhou, China [5] The Department of Hepatology, The Affiliated Hospital of Hangzhou Normal University, Hangzhou, Zhejiang, China [6] School of Nursing, Division of Medicine, Jinan University, Guangdong, Guangzhou, China

ABSTRACT (ENGLISH)

Background

Nonalcoholic fatty liver disease (NAFLD) is a global public health problem. Lifestyle modifications aimed at promoting weight loss and weight maintenance remain the current first-line treatments for NAFLD.

Objective

We aim to identify barriers and enabling factors in weight management among patients with NAFLD using the capability, opportunity, motivation, behaviour (COM-B) model of behaviour.

Design

This study adopted a qualitative design using semistructured interviews analysed with content analysis and the COM-B framework.

Setting and Participants

Interviews were conducted with 27 patients with NAFLD who experienced successful or unsuccessful weight reduction.

Results

Our study included 27 participants: 15 participants with successful weight loss (successful weight loss refers to a decrease in body weight ≥7% of the initial body weight for patients with NAFLD) and 12 participants with unsuccessful weight loss. Thirty-five themes (19 barriers and 16 facilitators) were mapped onto the COM-B model as barriers and facilitators to weight management among patients with NAFLD. The key barriers were lack of time and energy, lack of awareness of weight, lack of attention to NAFLD, treating food as a reward or compensation and social entertainment. The key facilitators were having basic weight loss knowledge and skills, strong motivation, attention to NAFLD, unsuccessful weight loss experiences and positive feedback from phased success.

Conclusion

In addition to identifying factors consistent with existing studies, this study identified factors that influence weight management in NAFLD patients, such as basic weight loss skills and rational thinking before weight loss, which were not previously reported. This has clinical implications for clinical healthcare providers and health management services for the improvement of education and support regarding lifestyle improvement and weight management in patients with NAFLD.

Patient or Public Contribution

We recruited potential participants from the Bariatric Clinic, Hepatology Clinic and Physical Examination Center of hospitals between March 2021 and October 2021. Twenty-seven patients with NAFLD who had successful or unsuccessful weight loss experiences participated in the study and responded to questions on weight management.

FULL TEXT

INTRODUCTION

Nonalcoholic fatty liver disease (NAFLD) is a progressive disease that includes a spectrum of histopathology ranging from steatosis to nonalcoholic steatohepatitis (NASH), with a risk of progressive fibrosis that may lead to cirrhosis and hepatocellular carcinoma.[1] As the incidence of obesity increases, NAFLD has become the primary cause of chronic liver disease, with a high incidence rate of up to 25% worldwide.[2] Strikingly, the prevalence of NAFLD in China has increased substantially from 18% to 29.2% within a decade.[3,4] China is predicted to have the fastest-growing NAFLD epidemic in the world and will reach 314.58 million patients by 2030, accompanied by a substantial economic burden and represents a growing public health problem.[5]

The increased global prevalence of NAFLD is particularly worrisome. Although no single recognized treatment is available, progress has been achieved in this field, and weight loss and weight maintenance remain the mainstay of treatment.[6,7] Paired liver biopsy studies have revealed that body weight loss ≥5% in patients with NAFLD is associated with a significant reduction in the incidence of hepatic steatosis (HS), weight loss ≥7% decreases the risk of hepatic inflammation and weight loss ≥10% decreases the risk of liver fibrosis.[8] Unfortunately, although the significance of weight management for patients with NAFLD has been recognized by experts and doctors, the management of patients' weights is still not adequately implemented.[9–12] Possible explanations include the lack of

implementation of behavioural programmes, lack of funds, clinicians' hesitancy to offer support and other factors. Guidance from theory might contribute to addressing these issues.

The capability, opportunity, motivation, behaviour (COM-B) model is one theory of behaviour change proposed in 2011 by Michie et al.[13] Capability is defined as an individual's psychological and physical capacity to engage in the targeted activity. Motivation is defined as the brain processes that energize and direct behaviour. Opportunity is defined as the factors external to the individual that enable or prompt the behaviour.[13] These components interact to generate behaviour that in turn influences these components. The advantage of the COM-B model is that it provides a useful framework to explain the barriers and enabling factors of behaviour change and provide a basis for the design of behavioural interventions. Therefore, this model has been applied to a number of clinical problems,[14–17] but it has not yet been applied to weight management in NAFLD patients. The aim of this study was to use the COM-B model to describe barriers and enabling factors to weight loss from the perspective of patients with NAFLD who have experienced weight loss.

METHODS

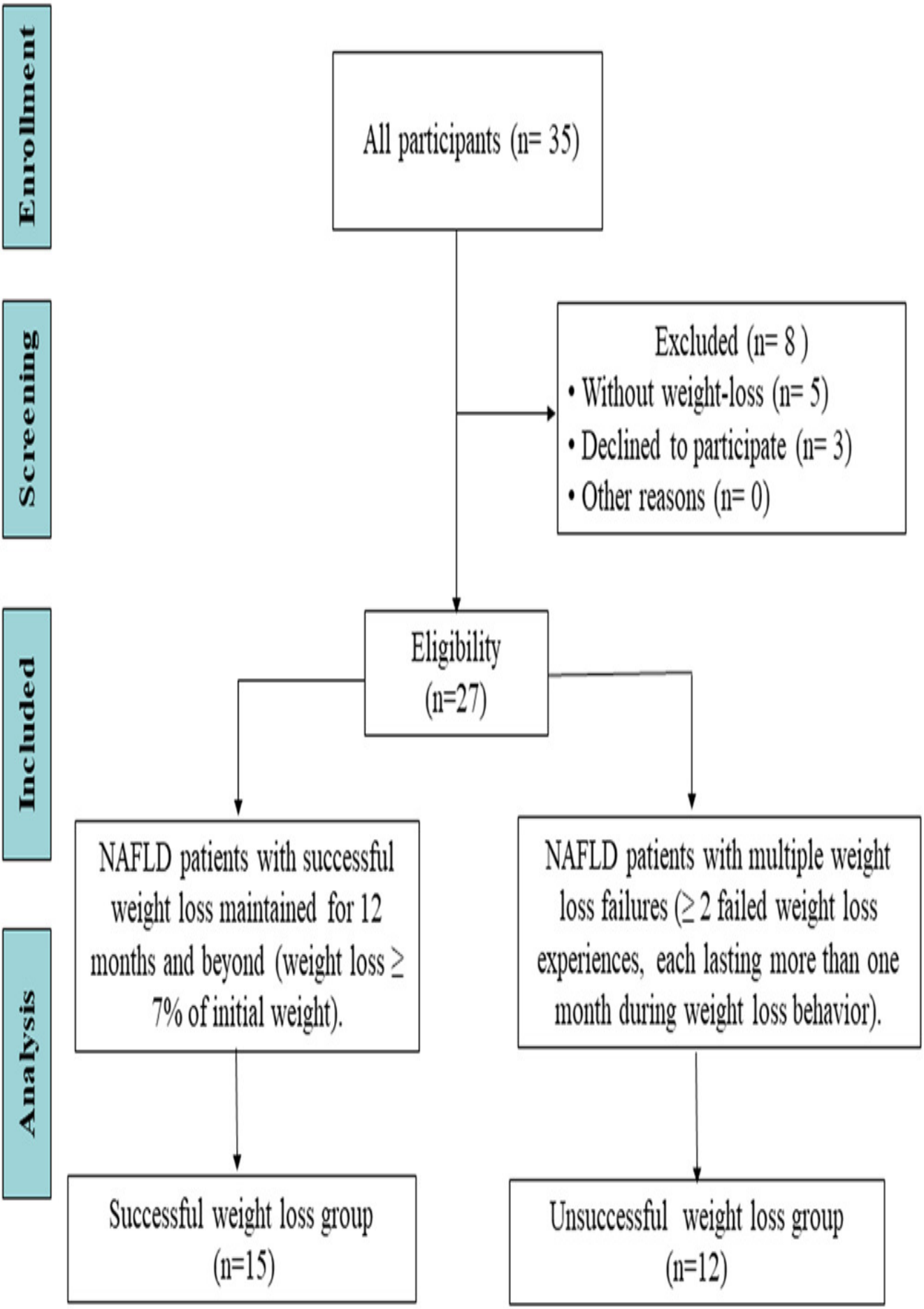
We report this study following the Consolidated Criteria for Reporting Qualitative Research guidelines.[18]

Study design

We used a purposeful sampling approach to include participants of different genders, ages, cultures and economic conditions. We performed a qualitative study that included semistructured interviews with patients with NAFLD who experienced successful or unsuccessful weight reduction. The study was approved by the Ethics Committee of the Affiliated Hospital of Hangzhou Normal University (2022E2-HS-033).

Participants

Two researchers (Y. G. and J. S.) recruited potential participants from the Bariatric Clinic, Hepatology Clinic and Physical Examination Center of the Affiliated Hospital of Hangzhou Normal University between March 2021 and October 2021. Researcher Y. G. conducted a simple interview with all the patients that included explaining the purpose, significance, content and informed consent of the study to the patients. The patients were asked whether they had planned weight loss activities or programmes. If they answered ‘yes’, they were then asked if their weight loss experience met the following inclusion criteria: (1) NAFLD patients with successful weight loss maintained for 12 months and beyond (weight loss ≥7% of initial weight); (2) NAFLD patients with multiple weight loss failures (≥2 failed weight loss experiences, each lasting more than 1 month during weight loss behaviour, including both those with repeated weight loss failures and those with successful but unmaintained weight loss). Patients with NAFLD without weight loss experience were excluded. Figure 1 shows the participant screening process. The sample size required was determined when saturation of themes was achieved.[19]



Enlarge this image.

Data collection

Eligible participants were asked to participate in a 60-min semistructured interview. The COM-B model was used as

a topic guide for interviews. All interviews were conducted by one of two chief researchers (Y. G., a practising physician and PhD candidate, and Y. H., a registered nurse with a PhD working in the university sector). Both were clinical fellows with qualitative research training. Written consent was obtained from participants who were able to complete an in-person interview, and verbal consent was obtained for telephone interviews from participants who were unable to complete an in-person interview due to logistics or distance. Patients were assured that they were free to withdraw at any time during the interview without any consequences. In addition, the researchers who conducted the interviews were not involved in providing care to the patients, so the patients' decision to participate did not affect their treatment. Interviews were conducted at a time and venue convenient to the patient. Participants were asked to describe their weight loss experiences. Upon completion of their interviews, participants were asked to complete a demographic survey, which included questions about their age, sex and socioeconomic status. The interviews were audio-recorded, transcribed verbatim and returned to the participants for verification.

Data analysis

A combination of inductive content and deductive framework analysis using the COM-B model was conducted.[20] In the first stage of analysis, data were uploaded to NVivo v.11 software to facilitate data management, and then all transcripts were read repeatedly by the first author to become familiar with the whole data set. All transcripts were reviewed, and meaningful text was inductively coded by two authors. Five transcripts were read and coded by the other two authors to ensure reliability. Regular meetings among all authors occurred in which the independent coding was compared and discussed (the conference was moderated by the two main interviewers). This procedure continued until the interviewers agreed on a set of established codes. Once all codes were determined, the two main authors discussed and allocated each code to the appropriate component of the COM-B model. Regular discussions took place during this process to ensure that all codes were allocated to the most appropriate COM-B domain. Disagreements were formally resolved at each step by discussion and in consultation with two other investigators.

RESULTSStudy participant characteristics

Table 1 shows the characteristics of the participants. We obtained informed consent from and interviewed 27 participants: 15 participants who successfully achieved weight loss (SPs) and 12 participants who were unsuccessful in achieving weight loss (UPs).

Table 1 Participant characteristics (*n* = 27)

	SLWG (*n* = 15)	ULWG (*n* = 12)
Age (mean, SD)	39.27 (13.07)	39.50 (13.46)
Sex (*n*, %)		
Male	9 (60.0)	5 (41.67)
Female	6 (40.0)	7 (58.33)
Highest level of education (*n*, %)		
Graduate or postgraduate	9 (60.0)	3 (25)
Bachelor's degree (BA or BS)	3 (20.0)	6 (50)
High school graduate	3 (20.0)	3 (25)

Current work status (*n*, %)		
Working full-time or part-time	11 (73.33)	10 (83.33)
Student	2 (13.33)	1 (8.33)
Retired	2 (13.33)	1 (8.33)
Annual household income (*n*, %)		
High-income	9 (60.0)	3 (25)
Middle-income	4 (26.67)	6 (50)
Low-income	2 (13.3)	3 (25)
Time of NAFLD diagnosis (years) (mean, SD)	3.4 (4.0)	6 (4.59)
Weight loss experiences (*n*) (mean, SD)	1.8 (0.56)	1.33 (1.16)
Baseline weight (kg) (mean, SD)	75.76 (12.69)	81.18 (17.12)
Current weight (kg) (mean, SD)	67.19 (10.02)	
Baseline BMI (kg/m^2) (mean, SD)	26.88 (2.11)	28.62 (3.96)
Current BMI (kg/m^2) (mean, SD)	23.89 (1.59)	
Body weight change (kg) (mean, SD)	8.57 (4.95)	
Weight loss time (months) (mean, SD)	3.6 (1.81)	
Weight maintenance time (years) (mean, SD)	3.73 (4.64)	

Abbreviations: BA, bachelor of Arts; BS, bachelor of Science; kg, kilogram; n, number; SLWG, successful weight loss group; ULWG, unsuccessful weight loss group.

Barriers and facilitators

Thirty-five themes were mapped onto the COM-B model as barriers to and facilitators of weight management, as described in full below. These barriers are summarized in Table 2.

Table 2 Identified barriers and facilitators in the various domains of the COM-B

Item	Domain	Definition	Barrier	*N*	Facilitators	*N*

Capability	Psychological capability	Any mental process or skill that is required for the person to perform the behaviour	Lack of correct weight loss knowledge	4	Basic weight loss knowledge	9
			Lack of rational thinking	3	Rational thinking before taking weight loss action	5
				Autonomous and active learning ability	4	

	Steady mindset	3		Physical capability	Any set of physical actions that require an ability or proficiency learned through practice	Unbearable exercise-related exhaustion
4	Basic weight loss skills	9				Unsustainable starvation diets

3	Weight loss methods suitable for individuals	4				Withdrawal reaction
3	Previous exercise habits	3	Motivation	Automatic motivation	Emotional responses, desires and habits resulting from associative learning and physiological states	Lack of awareness of weight

8	Focus on personal image	4		Reflective motivation	Beliefs about what is good and bad, conscious intentions, decisions and plans	Lack of strong motivation for weight management
5	Strong motivation for weight management	9				Lack of attention to NAFLD

6	Attention to NAFLD	6		Treating food as a reward or compensation
6	Crisis awareness	4		Unsatisfactory results after effort

4	Positive feedback from phased success	6		

Unsuccessful weight loss experiences	6	Opportunity	Social opportunity	Influences of friends, family, colleagues and other influential people that supp	Social entertainment	6

				or the implementation or lack of implementation of a behaviour		

Healthy eating habits of family members	3		Lack of social support	3
Family support	4		Other people do not pay attention to NAFLD	3
			Surrounding attitudes to weight loss	4
			Unhealthy eating habits of family members	4
			Attitudes and advice from medical personnel	4

			Physical opportunity	Anything in the physical environment that discourages or encourages the per for	Lack of time and energy	12

				maintenance of the behaviour		
Sufficient time and energy	5				Easy takeaway fast food	4
					Lack of available healthy meals at work	4

Abbreviations: COM-B, capability, opportunity, motivation, behaviour; NAFLD, nonalcoholic fatty liver disease.

Key barriers and their associated COM-B domainsCapabilityPsychological capabilityLack of correct weight loss knowledge

Lack of correct weight loss knowledge, including diet and exercise, was a common hindering factor: ‘Due to the lack of correct knowledge, I have blindly tried some very popular weight loss methods, such as trying not to eat dinner and even extreme dieting, so it often ends in failure’ (UP7, female, aged 40 years).

Lack of rational thinking

Rational thinking before taking weight loss action includes a rational examination of the participants' motivation to lose weight, an expectation that difficulties may occur in the weight loss process and awareness of the time and energy needed. Many participants who failed to successfully lose weight showed a lack of rational thinking, and their weight loss actions tended to blindly follow trends rather than following objective thinking. They, therefore, tended to underestimate the difficulty of losing weight, and they easily gave up when they encountered unanticipated difficulties during weight loss: ‘There are tons of videos on Tik Tok or other social media that show how easy it is to lose weight …they claim that you can easily lose weight just by doing a few movements or eating certain foods, as they say …some of them are actually disguised advertisements for health products …It's easy to follow suit blindly if you don't have the ability to think rationally…’ (SP5, male, aged 30 years).

Physical capabilityUnbearable exercise-related exhaustion

Some participants said they did not like the weight loss method of exercise because exercise was too difficult: ‘Exercise is really too tiring. Every time I think of the feeling of being tired, I don't want to do it…. It is really too difficult for me’ (UP4, female, aged 34 years).

Unsustainable starvation diets

Many participants thought that a starvation diet may have an effect in the short term, but it was detrimental in the long term: ‘A starvation diet is an anti-human instinct …and you need to spend a lot of energy to fight this feeling, which may make you unhappy…’ (SP4, female, aged 28 years).

Withdrawal reaction

Some participants said they faced a withdrawal reaction when they changed their dietary habits at the initial stage. ‘In the first two weeks of weight loss, I always want to eat something, even when I'm not hungry, I always feel like eating something…. I don't think this feeling is caused by hunger …I think it is more of an inertia…. Because of the habit of eating at any time and any where, it is uncomfortable to suddenly stop this behaviour’ (UP6, male, aged 50 years).

MotivationAutomatic motivationLack of awareness of weight

Most unsuccessful participants showed a lack of awareness of weight. Although many of them were already overweight, they still believed that they were ‘not so fat’ and their weight was ‘tolerable’. Even some middle-aged and elderly people thought that losing too much weight was not conducive to health: ‘I am 173 cm and 83 kg weight. For me, too thin is not necessarily a good thing at my age. I think 80 kg is completely fine for me’ (UP2, male, aged 58 years).

Reflective motivationLack of strong motivation for weight management

Those who were unsuccessful in losing weight showed weaker motivation than those who lost weight successfully. Even if doctors told them they needed to lose weight, they had many reasons not to follow this advice: ‘Ultimately, I don't really think I need to lose weight’ (UP2, male, aged 58 years).

Lack of attention to NAFLD

The participants generally lacked an in-depth understanding of NAFLD. In particular, a common belief among these UPs was that NAFLD has little effect on health, and some did not even consider NAFLD a disease: ‘I have diabetes and NAFLD. If you say that I need to lose weight because of NAFLD, I may not heed it, but if you tell me that I need to lose weight because of diabetes, I will certainly heed it because it (diabetes) is really a very serious disease…. I don't think NAFLD has any serious impact’ (UP2, male, aged 58 years).

Treating food as a reward or compensation

In both the successful and the unsuccessful weight loss groups, many participants mentioned that they treated food as a form of self-reward or compensation: ‘After a tiring day of work, I want to eat some food to compensate myself. In fact, I am not so hungry but still want to eat a good meal’ (UP5, female, aged 30 years).

Unsatisfactory results after effort

‘If I don't see any changes in the first few weeks, I would get frustrated and even want to give up’ (UP7, female, aged 40 years). Even those who lost weight successfully reported that ‘it is a blow if the weight has not been reduced in the initial stage of weight loss’ (SP4, female, aged 28 years).

OpportunitySocial opportunitySocial entertainment

Most participants mentioned that social entertainment was not conducive to weight management. In further interviews, we received the following feedback. First, the time spent eating during social entertainment is significantly increased compared with normal eating. During that time, people unconsciously eat more food. Second, social entertainment dinners tend to be varied and high in calories, which can easily lead diners to eat more food than they really need. Third, in a social situation, it seems inappropriate not to eat when everyone else is eating. In addition, social meals are often accompanied by drinking behaviour, which might also lead to overeating. ‘We often eat barbecue or hot pot at parties, which are quite caloric …and it is easy to eat more while everyone eats’ (UP1, male, aged 32 years).

Lack of social support

Some unsuccessful participants indicated that they encountered external adverse evaluations when they took weight loss actions: ‘I just started the job, and I am still a novice. I ate working meals with superior physicians after surgery, and sometimes the director or head nurse said that I wasted food when I left too much food’ (UP11, male, aged 28 years). ‘When I was trying to lose weight, I really hated hearing people say, “Wow, you're eating so little…. Can you

eat enough with such a small amount of food?" …These words always made me feel that I was weird and a misfit compared to other people' (UP8, female, aged 28 years).

Other people do not pay attention to NAFLD

'Ultimately, they still don't think NAFLD is dangerous …for example, when we drink at dinner, if you say you have high blood pressure and the doctor forbids you to drink, everyone will understand, but if you say you have fatty liver and cannot drink, everyone will laugh at you and make you drink more' (UP2, male, aged 58 years).

Surrounding attitudes to weight loss

Many participants said that the attitude of the people around them affected them. In particular, when people around them did not think that it was necessary for them to lose weight, they tended to give up on weight loss: 'I always complained to my husband that losing weight is so painful. He would tell me, "You are not so fat, stop losing weight, why make life so hard, enjoy food"…' (UP3, female, aged 34 years).

Unhealthy eating habits of family members

'My mother is in charge of cooking for the family every day. She has a strong taste and always cooks food that is too oily and salty. We've complained about it, but she wasn't happy, so no one ever mentioned it again' (UP5, female, aged 30 years).

Attitudes and advice from medical personnel

Many participants identified attitudes and advice of medical staff as directly influencing their perceptions of NAFLD. Some participants said their doctors told them that NAFLD was not as severe as diabetes or hypertension, so they did not pay attention to NAFLD and did not urgently take weight loss actions. Others said their doctor's advice to lose weight was too general to act: 'Every doctor will say that you have to lose weight and eat less and so on, but I think they just say it casually because their attitude does not seem to make me feel that the disease is serious' (UP8, female, aged 28 years), or 'Doctors told me that NAFLD would be better if I lost weight, but no one told me more details …such as how much diet to lose or how long to exercise' (UP2, male, aged 58 years).

Physical opportunityLack of time and energy

Almost all participants indicated that lack of time and energy limited their weight loss and even led people who had already lost weight successfully to regain the weight: 'I can maintain that (good) figure, but the premise of maintaining this figure is that you have enough time and energy to do this. In fact, I am now fat because I am too tired, and I have no time. If I had a month to spare, I could still lose weight successfully at the rate of one-half kilogram a day' (SP2, male, aged 31 years).

Easy takeaway fast food

Many participants said that convenient takeaway resources can affect weight loss: 'Because no one limits my eating, I can eat what I want, I can eat when I want, and there are no restrictions…. Even late at night, I can still get delicious fast food if I want…' (SP4, female, aged 28 years).

Lack of available healthy meals at work

For work reasons, many participants needed to eat in the workplace canteen or go to restaurants near their workplace for lunch, but most of these foods have 'excessive carbohydrates, heavy oil and heavy salt, and are not healthy enough' (UP6, male, aged 50 years).

Key facilitators and their associated COM-B domainsCapabilityPsychological capabilityBasic weight loss knowledge

Most successful participants had a correct basic knowledge of diet and exercise that they used in weight management, such as 'eating more (low-calorie) vegetables or fruits and less fat'. They emphasized gradually controlling and maintaining their weight by changing their dietary structure instead of 'starvation': 'Mindless starvation is never a good idea …my experience of losing weight has taught me that changes in diet and necessary physical activity is the best way' (SP2, male, aged 31 years).

Rational thinking before taking weight loss action

Most successful participants mentioned that 'really wanting' to lose weight was important when they talked about their motivation. After further questioning, they said that this 'want' involved not only 'hope' but also a rational examination of their motivation to lose weight, an expectation that difficulties may occur in the weight loss process,

and an awareness of the time and energy needed: ‘Everyone will say that they want to lose weight …but many people jump into action without thinking clearly, so their insistence does not always last long …people would have a more rational attitude after mature thinking’ (SP4, female, aged 28 years).

Autonomous and active learning ability

Some successful participants showed high autonomous learning and active learning ability: ‘I browse the internet for experience and knowledge shared by professionals …sometimes I ask people who are successful in losing weight how they do it…. Active learning is necessary’ (SP2, male, aged 31 years).

Steady mindset

Most of the participants stated that when they accepted the mentality that ‘weight loss is not so fast’, they were less disappointed during weight loss. This mentality also made it easier for them to persist until they saw an effect rather than giving up because they did not see an effect at the initial stage of weight loss.

Physical capabilityBasic weight loss skills

Basic weight loss skills are the simple basic skills needed to facilitate long-term weight management, such as calorie counting, diet matching and exercise skills. Many participants who successfully lost weight had one or several basic weight loss skills; for example, they could make simple nutritional combinations and healthy meals for themselves. Significantly, those who maintained a good weight for a long time seemed to be good at applying these skills to their daily lives: ‘I find it useful to have a simple nutritious meal cooking skills or effective exercise skills …for example, know how to prevent sports injuries, how to reasonably arrange exercise time and type, how to make healthy weight loss meals quickly…’ (SP6, female, aged 26 years).

Weight loss methods suitable for individuals

Many successful participants emphasized the importance of weight loss methods that were suitable for themselves: ‘I have no way to decide what the cook does when I eat in the staff canteen, but I can rinse the oil away with warm water every time before I eat it’ (SP13, male, aged 64 years), or ‘I don't like running …so I replaced it with brisk walking …for example, I walk to take express deliveries …I make a conscious effort to increase my walking time every day’ (SP3, female, aged 34 years).

Previous exercise habits

A small number of successful participants felt that exercise was interesting because they had exercise habits in their childhood: ‘A lot of people I knew couldn't hold to a workout. I am very different from them because of my family education …my father often took me to participate in various sports exercises when I was young …such as playing basketball or swimming …so …I feel like this is relaxing for me’ (SP2, male, aged 31 years).

MotivationAutomatic motivationFocus on personal image

Among the participants who maintained a good weight, many showed concern about their physical appearance. They could not accept their weight gain and wanted to maintain their ideal body shape rather than endure it: ‘A good appearance is not just a good social card, it also pleases me …I can wear all kinds of beautiful clothes and show my beauty …that makes me feel good’ (SP15, female, aged 35 years).

Reflective motivationStrong motivation for weight management

Almost all successful participants had clear and strong motivation. A common motivation among the younger group was the pursuit of a beautiful appearance, while the common motivation among the older group was the pursuit of health. In addition, there were short-term motivations or goals, such as work needs, marriage and fertility needs: ‘I didn't get pregnant for two years after my marriage because I was diagnosed with polycystic ovarian syndrome…. The doctor said my obesity and fatty liver would make it worse…. So, I had to lose weight’ (SP8, female, aged 31 years).

Attention to NAFLD

Compared with those who failed to lose weight, those who succeeded showed more attention to NAFLD: ‘My doctor told me that I had NAFLD and also told me that my situation would improve if I could lose some weight …I felt that it was necessary to pay attention to it, so I took 2 months to lose 3 kg’ (SP11, female, aged 40 years).

Crisis awareness

Some participants indicated that it was not difficult to lose weight because of their crisis awareness. They preferred to take early weight loss actions when they were not very fat (or had not been fat for very long): ‘I gained 5 kg a year after I got married, but when I realized that I was fat, I immediately realized that I couldn't put it off any longer …losing 5 kg was not particularly difficult for me’ (SP3, female, aged 34 years).

Positive feedback from phased success

Many participants mentioned the importance of positive feedback, especially achieving the desired results in the early stages of weight loss: ‘My blood pressure used to be very high and was not well controlled by taking antihypertensive drugs. Now (after losing weight successfully), my blood pressure is under control, and I feel relaxed. I really like this feeling’ (SP12, male, aged 59 years).

Unsuccessful weight loss experiences

Most of the participants had unsuccessful weight loss experiences, and many of the SPs mentioned unhealthy weight loss as a contributing factor: ‘I have tried many unhealthy methods and it hasn't worked …this has taught me that it is important to choose a healthy weight loss method that can be adhered to for a long time’ (SP7, male, aged 27 years).

OpportunitySocial opportunityHealthy eating habits of family members

Some participants said that the eating habits of the person who cooked at home affected their diet: ‘My parents eat very light food. They love vegetables and eat less oil. It is much healthier for me to eat at home than outside, and it's easier for me to control my weight when I eat at home than at a restaurant’ (SP7, male, aged 27 years).

Family support

Many people who live with their families say that family attitudes are important. If their family members support their behaviour rather than discourage or even oppose it, they are more likely to carry out weight-loss actions: ‘A lot of (Chinese) parents think it's ok for their children to be a little bit fat and even oppose their children losing weight. Fortunately, when I tell my parents I want to lose weight, they are very supportive. They agree with me that it is not good to be fat, so they support me in controlling my diet at home’ (SP4, female, aged 28 years).

Physical opportunitySufficient time and energy

Almost all participants, whether successful or not, mentioned the importance of sufficient time and energy. Most of their successful weight loss activities were completed in a relatively generous period of time: ‘My plan was carried out during summer vacation. I would like to say that weight loss absolutely needs time. You have to consider a lot of content, such as how to match your diet, how to perform exercise …there are so many details. You need enough time and energy to learn and to try to find suitable methods for yourself…’ (SP3, female, aged 34 years).

DISCUSSION

We applied the COM-B model to improve our understanding of the barriers and enabling factors in the weight management of patients with NAFLD. This study shows that 19 barriers and 16 enabling factors contributed to the weight loss process among patients with NAFLD. We plan to use the identified barriers and enablers to promote weight management in healthcare among patients with NAFLD and to inform future interventions aimed at optimizing evidence-based practice in NAFLD management since weight management is critical for the improvement of disease conditions.[8,21]

Comparison with existing literature

Most of the findings of this study are consistent with the existing literature in confirming that patients with NAFLD/obesity experience a range of factors related to capabilities, opportunities and motivation that impact weight management during weight loss.[9,12,22–26] Previous research has shown that patients often have an insufficient understanding of the disease, its progression, and its management.[22] This is consistent with the ability factors (lack of knowledge, lack of attention to NAFLD) identified in this study as influencing weight loss by participants who failed to successfully lose weight. Strong personal motivation to lose weight (especially the pursuit of beauty and health) and positive feedback from phased success were also identified by participants in this study. This is consistent with the existing literature, which has found that the desire to achieve rapid weight loss to improve health and early and significant weight loss are both facilitators of engagement and adherence.[12,23] Participants in this study identified

their psychological status as influencing their weight management. Many participants stated that stable mood states helped them control their weight, while others stated that negative psychological states (stress, anxiety, etc.) led to weight gain. This is consistent with existing research, which has identified psychological/emotional well-being as an important factor in weight gain.[24,25] The participants in this study also identified social networks and support as an important influencing factor throughout weight reduction. This is consistent with prior research that identified social support as the most consistent facilitator of and barrier to lifestyle change.[12] Participants in this study emphasized the importance of adequate time and energy for weight loss, especially in the initial stages. Many participants stated that their successful weight loss occurred over a sufficient period of time (e.g., winter or summer holidays), while others stated that working too much and being tired (lack of time and energy) made it difficult for them to adhere to lifestyle changes (e.g., adherence to exercise and a healthy diet). This is consistent with prior research that identified limited time and resources to support behaviour change.[26] In addition, previous studies have shown that a lack of correct guidance and support from physicians is an important factor that affects lifestyle management in patients with NAFLD.[9] This is consistent with factors related to medical staff that were identified in this study as influencing weight loss. Some participants stated they were told after their diagnosis of NAFLD that there was nothing concerning about NAFLD compared to other health conditions (e.g., diabetes or hypertension), and others said they lacked support to manage their condition effectively.

There are some differences between this study and the existing literature. Most strikingly, the participants in this study identified weight-loss skills (e.g., diet matching, calorie counting, exercise skills) as important enabling factors for weight management in patients with NAFLD. Many participants stated that basic weight loss skills made it easier for them to lose weight successfully and maintain an ideal weight for a long period of time, while others stated that a lack of these basic skills made them unsure of how to do the right thing. Second, rational thinking before weight loss action was a highlighted factor identified in this study that has not been mentioned in previous literature. Many participants in this study who succeeded in losing weight stated that rational thinking was essential, including a rational examination of their motivation to lose weight, the expectation that difficulties may occur in the weight loss process, and an awareness of the time and energy required. This rational thinking helped them develop appropriate weight management goals and select appropriate weight loss methods to facilitate long-term weight management. In addition, a focus on personal image and crisis awareness were identified in this study as important enabling factors by participants who successfully lost weight. They stated that they were unable to tolerate weight gain and tended to initiate weight loss activities early.

Clinical implications

NAFLD is not a sudden disease, and it often does not seem dangerous in the short term, which is why patients with NAFLD have a low willingness to receive advice from doctors on lifestyle management or weight control. This was reflected in our study. Among the patients in this study who successfully lost weight, only a few participants lost weight simply because of the emphasis on NAFLD; more participants adopted weight loss recommendations and took action because of multiple factors. This may suggest the following strategies for weight management recommendations to NAFLD patients. First, for patients with simple fatty liver, it is not sufficient to explain the long-term harm of the disease. Many patients will not adopt advice to change their lifestyle, but they can be advised to lose weight from the perspective of personal image or general health. Second, for patients with metabolic diseases such as diabetes or hypertension, the harm of the disease should be emphasized because NAFLD often exacerbates their primary disease. This aggravation theory is more likely to arouse the attention of patients. Third, weight loss for NAFLD patients involves physical, psychological, nutritional, exercise and other aspects, which calls for multidisciplinary clinical approaches to help patients solve their problems. Finally, education on NAFLD should not be provided only to patients; because it is a lifestyle disease, family members living together should also understand its harm. The appropriate participation of family members can be considered in future education or interventions.

Strengths and limitations

The advantage of our research is that we interviewed successful and unsuccessful weight loss groups to increase

the depth and credibility of the research. Furthermore, we used the purposive sampling method to include as many people as possible of different ages, genders, cultures and income levels so that the researchers could explore whether different themes appeared in these groups. However, our study also has some limitations. First, as a qualitative study, we could only preliminarily explore the problem and could not reveal causal relationships, which means that our results need to be confirmed by further quantitative studies with large samples. Second, some topics were not clearly classified; for example, social entertainment topics can be classified as both social opportunity and physical opportunity. To address this issue, we resorted to experts and reached an agreement through discussion among multiple authors. Third, we acknowledge that the volunteers who participated in the semistructured interviews were likely to be passionate about the topic. This is a common limitation of qualitative interviews that cannot be avoided.

CONCLUSION

This study identified a range of factors related to capability, opportunity and motivation, consistent with the existing literature. This study also identified factors such as basic weight loss skills and rational thinking before weight loss that influence weight management in patients with NAFLD that were not previously reported. This has clinical implications for clinical healthcare providers and health management services to improve education and support regarding lifestyle improvement and weight management in patients with NAFLD.

AUTHOR CONTRIBUTIONS

Yunpeng Gu, Yanli Hu and Junping Shi conceived and designed the study. Yunpeng Gu and Yanli Hu developed study instruments, led the analysis and wrote the first draft of the manuscript. Yunpeng Gu, Yanli Hu, Wei Zhang, Yutong Chen contributed to data collection. Yunpeng Gu, Yanli Hu, Run Zhou and Tingting Kong coded the data, and Yunpeng Gu, Yanli Hu, Run Zhou and Tingting Kong contributed to the thematic analysis. Chunmei Wang contributed to the second round of manuscript revisions and a partial collection of materials. All authors had full access to all the study data and take responsibility for the data integrity and reliability of the analysis. All authors had final responsibility for the decision to submit for publication.

ACKNOWLEDGEMENTS

The authors would like to thank all participants who encouraged the production of this article. This study was supported by grants from Guangdong Medical Research Foundation (A2022055), Natural Science Foundation of Zhejiang Province (LQ19G030003) and the Open Project Program Foundation of Key Laboratory of Liver and Kidney Diseases (Shanghai University of Traditional Chinese Medicine), Ministry of Education (No. GS2022-003).

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

DETAILS

Subject:	Feedback; Liver; Public health; Healthy food; Fatty liver; Patients; Skills; Health problems; Entertainment; Weight loss; Attention; Liver diseases; Lifestyles; Capabilities; Motivation; Qualitative analysis; Positive feedback; Body weight; Compensation; Behavior; Management; Body weight loss; Physical examinations; Barriers; Health promotion; Facilitators; Content analysis; Disease management; Codes; Management services; Interviews; Reinforcement; Consent; Hospitals; Weight control; Weight reduction; Qualitative research
Location:	China

Identifier / keyword:	COM-B model; health care; nonalcoholic fatty liver disease; qualitative study; weight management
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	355-365
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-16
Milestone dates:	2022-11-02 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-06-10 (manuscriptReceived); 2022-11-16 (publishedOnlineEarlyUnpaginated); 2022-11-03 (manuscriptAccepted)
Publication history :	
First posting date:	16 Nov 2022
DOI:	https://doi.org/10.1111/hex.13665
ProQuest document ID:	2767101867

Parental COVID-19–related health information practises, sources, evaluations and needs: A qualitative interview study

Altawil, Hala [1]
; Klawunn, Ronny [1] ; Dierks, Marie-Luise [1] ; Lander, Jonas [1]
[1] Deparment for Patient Orientation and Health Education, Institute for Epidemiology, Social Medicine and Health System Research, Hannover Medical School (MHH), Hannover, Germany

ABSTRACT (ENGLISH)

Background
Parents of infants and young children may have specific health information needs and preferences, as they are responsible for their children's health. COVID-19 posed many challenges for families, not least in terms of the constantly updated disease-prevention guidelines. However, little is known about parents' experiences with this unprecedented situation, that is, how and where they seek, use and evaluate COVID-19 (child)-specific health information. We aimed to find out more about this to provide insights to health (information) providers when communicating pandemic information to parents.
Methods
We conducted semistructured telephone interviews (August to October 2020) with a purposively selected sample of 20 German-speaking and 10 Arabic-speaking parents of children up to 4 years old. Recruitment occurred through multiple channels, including childcare institutions and social media. Qualitative content analysis of the interview transcripts illustrates the main differences between the two groups.
Results
By the time the interviews were conducted (mid-2020), some parents reported to seek information less actively or not at all, compared to the beginning of COVID-19. German speakers frequently used Google to obtain information, whereas Arabic speakers mentioned social media (particularly Facebook) as a central source. However, medical providers were the most trusted source for child health. Though determining the credibility of online information was difficult for some parents, others, mostly German speakers (middle–high education), were aware of some author-related criteria. When deciding on information use, parents often rely on their own judgement and gut instinct. Besides the necessity to disseminate information via multiple outlets to reach all parents, Arabic speakers desired

audio-visual and translation tools to facilitate understanding.

Discussion and Public Conclusion

Apart from education, language and knowledge of the health system and of the attributes of credible information may determine its quality and consequent decisions. There seems to be a considerable need to foster knowledge about reliable information sources, a greater understanding of the range of quality criteria and specific support for nonnative speakers, not least to better inform parents' decision-making.

Patient and Public Contribution

A parent panel ($n=7$) contributed to gathering ideas regarding recruitment, discussing initial results and the choice of topics and questions for a second interview phase.

FULL TEXT

INTRODUCTION

COVID-19 has illuminated the role and relevance of health information (HI) more than ever before.[1–3] While the situation affected everyone, the situation of, for instance, parents of infants and young children is special given their responsibility towards children when estimating the risks and impacts of everyday-life activities on health.[4–6]

Given this, and also since many core information and consultation services were substantially burdened, seeking, understanding and applying HI to respond rapidly and properly may be more challenging.[7] Though the number of parents searching the web for child health-related information is high and rising,[8,9] the ability to handle (digital) HI depends, on the one hand, on individual health literacy (HL)—which has further worsened according to recent representative statistics for Germany, showing a particular deficit for digital HL, and variations among population groups.[10] On the other hand, healthcare organizations have a key role in actively supporting individuals' information and decision processes,[11] particularly amid the torrent of information including inaccurate and false ones.[12] Further, (digital) HI often disregards its target populations' specific needs[13] and instead, parents may draw their information from various sources,[14] such as health professionals (HPs) and peers.

In addition, at least four aspects specific to parental COVID-19 information behaviour (IB) can be identified. First, empirical insights into parental HI behaviour—for instance, regarding prevention measures—would reveal whether parents sought information generally in terms of ‘COVID-19’ or specifically regarding infection prevention for the child. Second, aspects related to (mis)trust, acceptance of behavioural advice and handling uncertainty are decisive for health-related decision-making.[15] Respective factors, however, still need to be understood, as few comparable situations have been studied. This is particularly true for parents, as child health is a highly emotional topic.[12] Understanding parents' IB may be relevant for regular, postpandemic HI issues, as the amount of available digital information is constantly growing.

Third, prior research suggests that user perspectives are often inadequately considered for digital HI.[16,17] Parents' information requires particular attention, as they need to make decisions on behalf of their children (and family).[18] Last, it can be vital to clarify possible differences among culturally and linguistically diverse user groups: While all societal groups should have equal information access, insufficient language proficiency in the host country may limit information access for migrant groups.[19] Moreover, sociocultural backgrounds may substantially affect how individuals apply advice[20]; the case of parents may be an exemplar.

A study on the differences in the perceived risk perceptions between nine ethnic minority groups of young adults aged 24–26 in Germany found, contrary to expectations, a higher increase in their COVID-19-related health risk perceptions in comparison with the general population and could not explain many of the few ethnic differences discovered.[21] Given the need for a deeper understanding about potential differences for parents with distinct social and cultural backgrounds, the target group of this study is split up into (a) parents with German as native or second language, affected and not affected by COVID-19, and (b) migrant parents, affected and not-affected by COVID-19, and represented by those who migrated from the Arab region recently. The latter is characterized by a small but growing population in Germany, particularly since 2015.

The specific objectives of this study were to explore how parents:

•(1)
access and search for COVID-19–related (child) HI,

•(2)
understand and appraise COVID-19–related (child) HI to make decisions, handle challenges and how they trust/distrust certain sources,

•(3)
apply COVID-19–related (child) HI in daily life,

•(4)
express needs and preferences regarding the provision and communication of (digital) HI, and

•(5)
differs due to cultural and/or linguistic backgrounds regarding objectives 1–4.

MATERIALS AND METHODS

This study employed an exploratory, qualitative interview design. Its development, conduct, analysis and reporting were performed in line with the Consolidated Criteria for Reporting Qualitative Research (COREQ) Checklist[22] (Supporting Information: Appendix 1).

Sampling and target group

To gather a broad spectrum of parental views, we aimed to increase the diversity of the ex-ante identified sample via purposive and snowball sampling, according to gender, number and age of children and education status for two groups: (native) German-speaking residents (hereafter referred to as German speakers, $n = 20$), and native Arabic-speaking migrants—particularly those who migrated to Germany during the last few years (hereafter referred to as Arabic speakers, $n = 10$). We inserted these attributes into a self-developed sampling matrix to constantly review which participants were still missing. Besides the fact that Arabic speakers represent a large and a growing proportion of recent migrants to Germany, we concentrated on this group to enable a more detailed analysis of potential differences due to sociocultural backgrounds for one specific, exemplary subgroup.

Recruitment process and channels

As COVID-19 limited the opportunities for using ‘classic’ recruitment channels such as doctor's offices, we concentrated on alternatives. A focus here was on contacting multipliers, that is, facilities, institutions and individuals with regular and trustworthy contact with our target group, particularly (public and private) family centres, municipal facilities and kindergartens. For most of these, we forwarded the study call electronically and asked those contacts to pass it on. In addition, we used our own and partner projects' websites and social media accounts, mostly Twitter, and asked each recruited parent to forward the call to their peers. To enrol Arabic speakers, we additionally used in-person recruitment combined with a written study invitation in specific settings. There was no affiliation with any of the research participants before this study, with no personal relationship before or during the study. And interaction before the study commencement was used to clarify the purpose of the study and participation requirements.

Data collection

Qualitative data were gathered via individual telephone interviews to allow participation from different regions and despite the lasting contact restrictions. This also seemed appropriate for those who may not feel confident speaking in a group discussion. A male researcher (PhD) (J. L.), and a female Arabic-speaking researcher (Master's degree) (H. A.) experienced in qualitative research methods conducted a total of 30 interviews from August to October 2020.

The average interview lasted 43 min, each one only ended once participants stated they had sufficient opportunity to elaborate on each question and could also address any further issues in a final open question. No interviews were repeated and all participants completed the interview. We offered several options to participants to avoid any interview fatigue, for example, scheduling the interview to each individual's time preferences, pausing the interview for a break to take care of the child, and conducting the interview from home via telephone or video. Participants received a 30 Euro honorarium. We developed a semistructured interview guide based on (a) the research themes and objectives, (b) the main project underlying this study[23] and (c) core dimensions of the concept of HL. A draft version was revised by four project staff members and pilot tested with parents (n = 3) (Supporting Information: Appendix 2). The researchers explored participants' experiences without advanced fixed assumptions and made notes of relevant responses when possible.

We also set up a short online survey (SocSciSurvey GmbH, Germany) to further characterize the participants in terms of sociodemographic characteristics and HL (HLS-EU-Q16).[24] The interview guide and the online survey were translated into Arabic (H. A.) for respective participants. To follow up on parents' IB and needs as the pandemic continued—particularly to explore changes compared to parents' statements reported here, follow-up interviews took place from late 2021 to early 2022 and will be reported at a later stage.

Data analysis

All interviews were audio-recorded, pseudonymized and transcribed verbatim (in both German and Arabic) by all project staff members using MS Word and MAXQDA (VERBI, Version 2020), without a subsequent review by the participants. Arabic transcripts were translated into English by the researcher who conducted the interviews with Arabic speakers (H. A.). We applied established principles of structuring qualitative content analysis (QCA)[25] and used MAXQDA. QCA is common for the analysis of qualitative data[26] and fits the purpose of a simple, in-depth description of both variation and significant common data patterns.[27,28] Structuring QCA is considered the core of QCA,[29] in which the material is structured based on two dimensions: cases ‘interviewees’ and categories ‘themes’, and a category system is developed.[25] In Step 1 (pre-analysis), we deductively coded 10 random interviews (J. L.), using the research objectives and interview guide to develop a first, broad structure of main and subcategories. In Step 2 (test phase), we applied the deductively formed Level 1 and Level 2 categories to another three interviews, analysed in full and independently by two researchers (J. L. and H. A.), inductively added Level 3 categories, and in parallel confirmed the definition of each Levels 1 and 2 categories. In Step 3 (test phase), we compared the results from Step 2, discussed the definitions and flagged up principal dissimilarities. In Step 4 (test phase), we adapted the coding scheme based on the discussions from Step 3, and agreed a final version with a third female researcher (PhD) (M.-L. D.). In Step 5 (analysis), each researcher (J. L. and H. A.) analysed 15 interviews independently using the final categories from the test phase; additional codes were occasionally added. In Step 6 (integration), we integrated all interviews (n = 30) into one coding scheme, discussed new questions and unclear items and combined some similar (mostly Level 3) codes. Finally, we performed a category-based analysis, describing and summarizing the Levels 1 and 2 categories.[26] The results of the analysis were discussed with the author team, but not with the study participants. Initial results were, however, discussed with the study's parent panel.

To analyse the short survey, we entered n = 30 data sets into SPSS for a descriptive portrayal of interviewee characteristics (sociodemographic data). Using the HLS-EU-Q16, study participants indicated their ability to find, understand, evaluate and apply HI on a four-point response scale (1 = very difficult, 2 = fairly difficult, 3 = fairly easy, 4 = very easy), in addition to a ‘don't know’ item. A total HL score was calculated to build three levels of HL (inadequate HL: 0–8 points, problematic HL: 9–12 points, sufficient HL: 13–16 points).[24,30]

RESULTSParticipant characteristics

We interviewed *n* = 30 mothers and fathers, of whom *n* = 20 were German speakers and *n* = 10 were Arabic speakers. The sociodemographic characteristics and the levels of HL of the sample are illustrated in Table 1.

Table 1 Participant characteristics (*n* = 30)

Characteristic	*N*	%
Gender		
Male	8	26.7
Female	22	73.3
Age of parents, mean (SD) = 34.23 (5.75), range = 22–48		
18–29	5	16.7
30–39	21	70
40–50	4	13.3
No. of children, mean (SD) = 2.03 (1.098), range = 1–6		
1	11	36.7
2	10	33.3
3	8	26.7
>3	1	3.3
Age of children		
<1	3	10
1	8	26.7
2	11	36.7
3	10	33.3
≥4	14	46.7
Mother language		

German	13	43.3
Other than German	17	56.6
(School) Educationa		
Low	5	16.7
Middle	13	43.3
High	12	30
Health literacy levels		
Inadequate (0–8)	4	13.3
Problematic (9–12)	12	40
Sufficient (13–16)	14	46.7

a

Education level: low: the completion of the Volks/Hauptschulabschluss/8th/9th in the German system (GE) or the elementary school education (6th class) in the Arab system (AR); middle: mittlere Reife/10th class (GE) or secondary school/9th class (AR); high: all other degrees from Fachhochschulreife, Abitur/≥12 class (GE) or high school degree/≥12 class (AR).

QCA

Parents' perspectives are presented here according to the main themes and subcategories derived inductively and deductively from the interviews (Table 2).

Table 2 Main themes and subcategories

Main theme	Subcategories
• (1) Accessing and obtaining information	
Information Gathering	Information behaviour Information sources Information reasons
• (2) Understanding and appraising information	

Information Handling	Positive perception of information Negative perception of information
Trust	General aspects Family, friends Classic, mass media Online media Medical personnel Other actors
• (3) Applying information	
Information Handling	Processing, using of Information
Rules and Recommendations	Applying rules and recommendations
• (4) Information needs and preferences	
Future Information Needs	Needs for digital offer Other needs

Accessing (and obtaining) informationIB

While some parents indicated actively searching for COVID-19–related (child) HI, others received information passively.[31] Some did both. Over the course of the pandemic, some parents with different educational levels changed from active to passive searching or even gave up consuming information, primarily because of feeling overwhelmed, or because they did not expect new insights.

In the meantime, I take no more information, because of course I have now taken what I could take, and my head is also totally full of it. (GE, middle edu., P6:62)

Information sources

The most frequently used source was Google (*n* = 19), whereas Arabic speakers mentioned Facebook most frequently (*n* = 8). The latter included nonofficial sources (e.g., YouTubers, community groups, people's stories) and official, that is, public sources such as the German Ministry of Health, or HPs (doctors, scientists). The latter, however, were mostly mentioned by those with higher education. Public institutions' websites, such as the local health agency ‘Gesundheitsamt’ were the third most popular source by German speakers (*n* = 9, middle–high education). Additionally, parents frequently received information from family members, friends and acquaintances (*n* = 18), of which some had a professional healthcare background. Regarding online sources, Arabic speakers mentioned YouTube in particular, whereas German speakers referred to Podcasts (e.g., NDR Corona update). Other popular sources for Arabic speakers included childcare facilities (e.g., kindergartens), whereas German speakers rather cited medical experts, including relatives and friends. The former were mostly seeking, reading or

hearing information in Arabic (online and offline), due to their limited German language proficiency. They preferred information in their native language, to ease understanding and save time.

For me here in Germany, these YouTubers were coming out (mentions names) I was following them, they were saying daily the recovered and infected cases and advising […], and they are not a public source, I mean a private source […]. (AR, middle edu., P3:51–52)

Information reasons

Most parents (*n* = 22) reported searching for general COVID-19 information, that is, about the transmission pathways. This contrasts with child-specific information such as infection risks, but also nonhealth-related aspects such as contact restrictions imposed protectively by childcare facilities. Child-related information was of particular interest to pregnant women, parents with a chronically ill child or visiting a childcare facility. Other important topics included infection and safety measures, the number of infected and death cases, and high-risk groups. Parents also reported seeking others' opinions about and experiences of the disease. When seeking additional information, German speakers primarily referred to federal state requirements, hospital measures and a future COVID-19 vaccine, whereas Arabic speakers rather referred to dietary intake and information concerning countries in the Arab region.

I informed myself about the children at the very beginning and it was said that things would be milder and I didn't think about it that much then. I've been looking more for the numbers, how many people are getting infected, is it going up or down and so on and what the signs are, how it's happening […]. (GE, low edu., P13:22)

Understanding and appraising informationInformation perception

Some parents found the available information helpful and sufficient to protect their families, and referred to it as being consistent, accurate, detailed and easily accessible, particularly online. However, many expressed facing uncertain, confusing or contradictory information, and reported being exposed to a great deal of false information. Some parents repeatedly considered information to be dramatizing and inaccurate—particularly information found via Google—whereas some found it to be scientific in nature, leading to difficulties in comprehension.

Yes, even now I have seen a few reports in YouTube. There are virologists who say something and then there are virologists who say something else […] What is true now? You're really confused again because when you hear opinions from experts, you can't really believe them either. (GE-middle edu., P18:44)

Trust

While few parents indicated a general level of trust, many reported that, first, a clear decision on trust is difficult to make, particularly because of the variety of (online) sources. To determine trustworthiness, parents repeatedly mentioned checking and comparing multiple sources with their previous knowledge or personal perception. Others, mostly German speakers (middle–high education) differentiated between the sources and specified several criteria for trust, particularly regarding the author (identity, qualifications, seriousness, neutrality).

Second, trust depends on the source: It was mostly ascribed to nondigital sources, particularly medical personnel, given their professional knowledge and expertise. Though parents used a range of information sources, they stressed that paediatricians and family physicians are the first and most reliable references, and said they followed their advice. Only a very few mentions relate to not relying on a paediatrician for pandemic-related information and to the necessity for a second, specialist opinion.

The primary 100% trusted source is the doctor. (AR, low edu., P9, Pos. 42)

So I would say that if it was about my children, if I noticed that they had any symptoms or pain, then I would honestly say that I would not trust anyone except the doctor. (GE, low edu., P20:56)

Trust also seemed to be comparatively high for Arabic speakers in kindergartens and schools, as they apply the

rules and recommendations to protect children. In comparison, German speakers rather referred to governmental public health (RKI, Gesundheitsamt), and scientific, that is, medical institutions (medical schools, health experts, journals' studies, health magazines (e.g., Apothekenumschau). Few reported scepticism of public institutions (e.g., World Health Organization), the pharmaceutical industry, health experts and funded scientific studies, for example, due to inconsistency.

Regarding family members and friends, parents varied in their appraisal of trust, and ascribed most trust to those relatives and friends who had medical knowledge or experience of the disease. In cases of distrust, this was due to perceiving advice as subjective, influenced by emotions or nontransferable to one's own situation.

Varying perceptions of trust were also found for online sources, which were distrusted by Arabic and German speakers because of its multiple conflicting opinions. Parents also stated that the internet is an open place, where those with no knowledge or expertise share information, including nonfactual, and some parents found it difficult to identify the information producer or its reliability. In addition, parents criticized using Google for disease diagnosis, as this causes fear from search results. However, Arabic speakers especially trusted online sources (*n* = 7) when this was provided by doctors on social media, or Google, or by previously known YouTubers. These convey or translate information in Arabic, which guarantees understanding, in addition to providing references for delivered information, which allows parents to check its validity. Arabic speakers more often ascribed distrust to classic media, for example, given a lack of transparency about infection statistics.

Applying informationInformation processing and use

While some parents were overwhelmed and affected by false and negative information, many expressed that it did not affect them. Few said it was easy to identify fake news based on personal judgement, common sense or replicability of information from multiple sources. A few argued with others about veracity (e.g., of conspiracy theories) and tried to convey correct information to them.

When deciding which information to use, parents implemented different approaches: deliberately selecting the quantity and/or kind of relevant information (for child health); matching it to their current situation; weighing multiple sources against each other or against previous knowledge; discussing information with others (e.g., family, peers, colleagues), and exchanging opinions, experiences and knowledge; applying reasoning, common sense or relying on gut instinct or personal judgement; collecting information from (multiple) digital sources to get an initial idea about the subject and, following this, consulting a doctor to avoid having to make their own decision.

So it's always been different and then I was just a little bit like what am I doing now and then somehow you acted according to your gut feeling. (GE, middle edu., P9:50)

Few parents reported looking for more, detailed information, for example, to gain more relevant, specific or additional knowledge. Regarding others' opinions and experiences, some indicated that these influenced their own opinion and information decisions. Others found such experiences inapplicable. Furthermore, some parents reported educating children about the disease.

Applying rules and recommendations

In terms of committing to public health safeguarding measures, most parents (*n* = 27) reported following and applying these in their daily life, especially at the beginning of the outbreak. As the pandemic continued, nearly a third reported to still adhere strictly, whereas others reported lower adherence and easing, for example, in settings where in-person contact with relatives occurred as restrictions were eased. Additionally, some indicated that their own or others' (family or friends) disease experience affected their adherence and precaution, as they undergo or learn about the, most often, mild course of the disease. Parents also stressed the difficulty of strictly following the restrictions regarding social contacts and safe physical distancing, for example, in playgrounds, where there is

inevitable interaction. Sometimes, this led to not continuing or becoming tired of applying some guidelines in daily situations with the child.

[…] there are many things not like before […] one wears a mask, I told you there is no shaking hands, we try to keep distance but there are things involuntary for example, we want to eat together at the same table, what can you do? Nothing. So, it is not negligence but there is easing. (AR, high edu., P8:79–80)

Information needs and preferences

First, parents referred to how HI is communicated (communicating, messaging), pointing out that dissemination should happen through kindergartens and schools, particularly to reach migrant parents, besides general public information campaigns via classic broadcast and print media.

[…] I think most families are actually reached via television and radio and especially posters. […] just pictorial and large and appeals to everyone […]. (GE, high edu., P7:96–98)

Parents stressed the role of frequently accessing HPs, particularly paediatricians (midwives were less relevant for Arabic speakers), regarding emerging disease knowledge and its impact on children's health, receiving instructions and advice in daycare centres and finding and applying child health-specific information. While parents did want to understand whether COVID-19 is a threat for children and if this is based on evidence, they also desired COVID-19 to be treated as a ‘normal’ disease to avoid further panic, particularly for new parents, and instead focus on advice that is helpful for dealing with the crisis situation more generally, for example, regarding nutrition, mental hygiene and social contact.

Compared to nondigital sources, it seemed rather difficult for parents to state clear preferences for digital information; they repeatedly mentioned not feeling the need for specific changes, or feeling well-informed. A few mentions related to increasing the transparency and up-to-dateness of digital information and its respective sources—preferably public sources—and adding options for direct, personal interaction with HPs in case of specific questions. Arabic speakers desired audio-visual and translation tools to facilitate understanding, as this would be beneficial for saving time.

[…] When the information is issued from an official authority, it is more reliable than the doctor because it is issued by an official body, meaning it targets all people. Yes, sure it is much better to be in Arabic but I tell you again, even when the information is issued by a responsible authority and only in German, [it is important that] there is someone who translates the information and passes it on to us […]. (AR, high edu., P1:130–132)

Parents also referred to the communication of public health messages, for which a few Arabic speakers mentioned the need to raise awareness about the necessity of adhering to guidelines. Further, they urged the need for better disease control and management of rules and guidelines, particularly in private settings and for regularly experienced situations, for example, (crowded) childcare facilities. In that sense, the need for better coordination among parents and childcare facilities (here: kindergartens) by educating staff to deal wisely with COVID-19–related information and making child-related decisions, such as deciding whether a child with symptoms should stay at home, was also mentioned. German speakers called for rules and regulations to be issued and applied uniformly nationwide.

DISCUSSIONInformation sources

Parents use both formal and informal sources for pandemic-related information, predominantly online. Existing research outlined the increasing use of online sources for health and medical information[32] and its importance during the COVID-19 outbreak.[33] In Germany, the internet is the fourth most important HI source, in general, after mass media, HPs and family members.[34] In our study, parents frequently googled COVID-19, whereas migrant parents more often relied on social media platforms (SMPs), finding them easier to access, more up-to-date[35] and available

in their native language, which helped them to understand and practice preventive measures. The reliance of migrants with limited local language skills on (informal) media channels has been reported in Oktavianus et al.[19] Recent research highlights the reach of information delivered through SMPs to diverse population groups and its role in promoting health prevention behaviour. Nevertheless, it underscores the vast spread of mis- and disinformation and its potential harm to health.[32] Official information sources were more used by German speakers, mostly those with middle–high education, while only few, high-educated Arabic speakers named (inter-)national institutions, almost without referring to other institutional information channels responsible for crisis communication.[36] This is probably due to limited language proficiency and lack of knowledge of the German health systems' communication channels, according to Finell et al.[37] As Arab parents often relied on accessible and familiar information sources, there could be a focus on engaging more informal information mediators for these groups, as suggested by Mason et al.[38]

IB

Over the course of the pandemic, many changed their IB, and only a few remained active information seekers. Griebler et al.[36] show a reduction of interest in and need for COVID-19-related information and/or 'selective usage behaviour' among the Austrian population. At the pandemic's onset, uncertainty and perceived seriousness of the disease for their children's health triggered more active IB, which may be due to risk perception and uncertainty as drivers for seeking COVID-19-related information, as expressed by Huang and Yang.[39] However, the evolving knowledge, its effect on children and the stream of ample information led to a change in the state of emergency and the need to constantly seek related information, particularly when either feeling overwhelmed or satisfied.[19] Although almost all parents sought information on infection prevention, our study highlights a slight difference between the two groups regarding other issues of interest (e.g., requirements of federal states vs. state of infection in (Arab) home countries). Here, Oktavianus et al. point at migrants' information due to concerns about the safety of family members in their home country, not only for their own safety. Hence, parents may be in a dual or even triple role of seeking information for themselves, for their child, and for further family members.[19]

Information handling

Though parents were aware of the infodemic, the overflow of (false and misleading) information still led to confusion and uncertainty. The susceptibility to the infodemic might be partly explained by reliance on, for example, SMPs, especially by Arabic speakers, facilitating the dissemination of misinformation.[40] However, the results indicate that it did not constitute a problem for some parents who reported not encountering, ignoring or avoiding such information. Tandoc et al.[41] stated that readers often ignore fake news, but in some cases may act on it. Moreover, some parents felt able to distinguish correct from incorrect information; though to do that they relied on their own judgement.[42] This is in line with the internal and external authentication of Oktavianus et al.[19] Prior research explained the use of nonrational factors for judging information when there is a lack of knowledge and conflict among information sources, emphasizing the need for critical thinking to handle misinformation.[42] Here, consideration should be given particularly in the case of migrant parents, as previous research points at the difficulties of knowing which (reliable) sources to turn to when not being familiar with the HI context in a different country.[37]

Okan et al.[43] outlined the continuous and broad provision of coronavirus prevention measures through various channels that were easy to understand and apply, and their positive effects on people's HL. This concurs generally with the parents' perception of the usefulness of available COVID-19 information, and is particularly obvious for migrant parents, due to the ease of access to different, largely online sources in Arabic. Additionally, this is vital for newcomers who may especially lack social interaction and support in the host community, particularly amid the imposed restrictions.[44] In addition, our study indicates that digitally seeking COVID-19-specific HI seemed helpful for

parents in making child health decisions during the outbreak. However, it shows that the physician is the main, if not the primary, reference to consult in terms of child health issues or to verify information, which resonates with previous findings by Jaks et al.[45]

Our study shows that parental adherence to PH measures is influenced by: the context and nature of the activity (e.g., indoor vs. outdoor); the compliance, acceptance and support from the social environment (private or public); the consistency of the issued rules and guidelines; the organization and management of resources (e.g., public transportation) and the personal beliefs, experiences and mentality. Oktavianus et al.[19] reported similar findings regarding the effect of external factors on adopting preventive behaviours during the outbreak. This corresponds to what King et al.[46] underlined regarding the influence of social and structural—in addition to individual determinants of health—on compliance behaviour. Moreover, Benham et al.[47] referred to the Theoretical Domains Framework of Atkins et al.[48] to explain 'the need to understand the characteristics of the people in whom a change is to be effected, their behavioural context, and the components driving change, in order to facilitate behaviour change'.

Trust

Parents trusted medical personnel the most, confirming previous research,[49] and also in terms of COVID-19 information.[36,50] In our study, they do not fully trust online sources, including SMPs; trust varied depending on the sources used. Fewer found classic media sources (highly) reliable and trustworthy for COVID-19 and rather a source of fear, which is supported by Finell et al.[37] Griebler et al.[36] observed a loss of trust in TV, radio, internet and health authorities as the pandemic evolved. Though some parents are acquainted with some 'quality criteria', we found that parents may not consciously apply them. This is in line with Slomian et al.,[51] in which very few participant women seemed to be aware of the existence of any quality standards for HI sites. Looking at the sociocultural diversity of target groups, there could be further research to understand if trust can be better established by optimizing the use of quality criteria, or whether the focus should be on engaging professionals and institutions that guide, for instance, migrant parents to the 'right', that is, high-quality sources. In particular, Bergman et al.[52] highlight the need for tailored, effective strategies as well as the importance of social (information) networks.

Further, others' opinions and experiences are not applicable for some parents to their own situation. However, some emphasized that exchanging information and knowledge with family and friends, and learning about shared stories on SMPs was a reason for trusting information obtained from peers with experience of the disease. Scholars referred to these experiences as 'testimonies', 'case reports'[34] or 'experiential knowledge' and related it to 'social support'[42] that serves to support decision-making and understandability.[34]

Needs and preferences

Parents required online sources to satisfy their information needs, though these do not replace direct personal interaction with HPs. Previous research has shown that digital media complements rather than substitutes for traditional HI sources.[45] Further, our study underscores that other means of communication are important to parents, endorsing the significance of considering multiple information channels. Our findings emphasize the importance of HI being comprehensible, navigable and official. This aligns with the suggested principles for designing HI in prior research.[32] Additionally, we shed light on the preference of participants from other cultural backgrounds to receive information in their native language. This confirms previous calls to constantly provide reliable information and help in migrants' native language, or more generally, in multiple languages.[37,53–56] Our results also support the finding of existing research that 'high-quality HI must be (…) culturally competent—the ability to interact effectively with diverse audiences by recognizing and responding to variations in social, cultural, and linguistic needs'.[32] Hence, while previous research pointed to the Internet to spread culturally sensitive HI,[52] our findings suggest that respective sources need to be provided by those deemed as trusted and familiar within a specific community, for example,

(Arabic) medical experts on YouTube or Instagram. When developing respective approaches, ‘cultural mediators’ should be included.[57] In terms of format preferences of digital information, which may affect parents' understanding and evaluation, the use of visual and interactive communication tools that may also allow (online) contact with HPs, display elements (e.g., bulleted points) and use of simple language are in line with previous findings.[34,53,58]

Study contributions and future research

The current study contributes to the understanding of IB, how parents make decisions and apply information to care for their child, and what they need to do so in a pandemic situation. For future research, it is essential to study IB and the role of trust in specific sources on information and prevention decisions at different time intervals, given the changes in IB over time Additionally, observing the actual (digital) search behaviour may yield a better understanding of parents' considerations. We also explored differences among diverse parent groups, and further assessments using quantitative or structured qualitative survey studies seem warranted, particularly to specify support needs for parents who are unfamiliar with the host countries' information channels and providers. Future research could also assess parental IB for noncrisis health situations, where information may be more difficult to access.

Limitations

The interview and survey translation was done by an Arabic-speaking researcher. While this does not guarantee comprehension and validity, research suggests that the adaptation of the English version of HLS-EU-Q16 is applicable to the Arabic language, to measure HL among native Arabic-speaking people in Sweden.[52] Likewise, the verbal translation from Arabic to English represented a challenge to ensure the meaning or stay true to the interviewee narratives. Besides that this researcher is a native Arabic speaker, hence reducing the chance of misunderstandings, the focus was on avoiding interpretations, that is, translating expressions that may contain different meanings in its most literal sense. Since our sample size was predetermined and we applied purposive sampling, we focused on ensuring sufficiently diverse participant characteristics using a sampling matrix. The nonrandomized selection implies subjectivity,[59] but qualitative research and purposive sampling do not aim generalization. The over-representation of mothers (n = 22) may imply that fathers' views were not fully captured. Furthermore, parents with a low educational level are underrepresented (n = 5), which points to the difficulty of accessing this group. Lastly, while we could not (fully) achieve transcript validation through participant feedback, initial results were discussed with the established parent panel to promote research validity.

CONCLUSION

Overall, the spread of pandemic-related information, specifically on infection protection and safety measures, through a multitude of channels allowed parents to access and select sources that satisfy their information needs. However, the results reveal some differences between the two groups regarding the type of information sources preferred, and their knowledge of the health (information) system, and of the attributes of high-quality HI and credible (digital) sources. This has critical implications for the type and quality of information content, thus influencing (parents') beliefs and decisions to act preventively. It is a joint responsibility between HPs and information producers to empower parents to make informed health decisions for their children and to point at strategies for identifying dis- and misinformation. Besides acting as a primary source, especially for parents who encounter difficulties appraising and using digital information, HPs are key to give guidance about high-quality (digital) information sources. This prompts engaging HPs in crisis communication and training them on delivering culturally competent information. It is imperative for information producers to provide parents with adequate information about the current state of knowledge, use multiple communication channels and pay attention to differences among distinct groups. This requires the involvement of parents' representatives, including those from

migrant communities, in the development of communication strategies for future health crises. Public health initiatives should foster reliable information sources, build trust in these sources and educate parents, particularly (new) immigrants, to navigate the health (information) system, thereby fostering their HL.

AUTHOR CONTRIBUTIONS

Hala Altawil: Methodology; investigation; writing –original draft. **Ronny Klawunn**: Methodology; writing –review and editing. **Marie-Luise Dierks**: Conceptualization; writing –review and editing; supervision; funding acquisition. **Jonas Lander**: Conceptualization; methodology; investigation; writing –review and editing.

ACKNOWLEDGEMENTS

We would like to thank the members of the steering committee of the interdisciplinary HELICAP research group, located at Otto von Guericke University Magdeburg, University of Education Freiburg, University of Regensburg and Hanover Medical School: Christian Apfelbacher, Eva Maria Bitzer, Susanne Brandstetter, Janina Curbach, Marie-Luise Dierks and Markus Antonius Wirtz. We also thank all our interview partners for sharing their insights and experiences with us, as well as those institutions and individuals that provided invaluable support for the recruitment process. We are grateful for the language editing support provided by Roy Reuben Thomas. This study is funded by the German Research Foundation (DFG, Deutsche Forschungsgemeinschaft)—[GZ: DI-1757/2-1]—'Health literacy in early childhood allergy prevention: parental competencies and public health context in a shifting evidence landscape' [FOR 2959; GZ: 409800133]. The funder had no role in the design of the study, data collection, data analysis or interpretation of the results. Open Access funding enabled and organized by Projekt DEAL.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data presented in this study are available on request from the corresponding author. The data are not publicly available due to the need to preserve the anonymity of the interview partners.

ETHICS STATEMENT

The Ethics Committee of Hannover Medical School approved this study (ID 8161_BO_K_2018_Extension2020). Written consent was obtained from all study participants in accordance with ethics approval. Participation was voluntary. Financial incentives for participation were offered.

DETAILS

Subject:	Information sources; Infants; Communication; Childrens health; Families &family life; Child care; COVID-19; Health literacy; Qualitative analysis; Recruitment; Education; Medical research; Criteria; Health education; Access to information; Children &youth; Audio data; Health information; Coronaviruses; Decision making; Pandemics; Parents &parenting; Social networks; Information needs; Disease control; Content analysis; Mass media; Interviews; Children; Research methodology; Parents; Instinct; Social media; Information seeking behavior; Qualitative research; Digital media; Translation
Location:	Germany
Identifier / keyword:	child health; COVID-19 health information; information behaviour; information handling
Publication title:	Health Expectations; Oxford

Volume:	26
Issue:	1
Pages:	555-565
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-12-08
Milestone dates:	2022-12-02 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-09-05 (manuscriptReceived); 2022-12-08 (publishedOnlineEarlyUnpaginated); 2022-12-04 (manuscriptAccepted)
Publication history :	
First posting date:	08 Dec 2022
DOI:	https://doi.org/10.1111/hex.13688
ProQuest document ID:	2767101862
Document URL:	https://www.proquest.com/scholarly-journals/parental-covid-19-related-health-information/docview/2767101862/se-2?accountid=211160

Last updated:	2023-04-24
Database:	Public Health Database,Publicly Available Content Database

Youth engagement in mental health research: A systematic review

McCabe, Erin [1]
; Mungunzul (Megan) Amarbayan [2] ; Rabi, Sarah [3] ; Mendoza, Justino [4] ; Syeda Farwa Naqvi [3] ; Kalpana Thapa Bajgain [3] ; Zwicker, Jennifer D [5] ; Santana, Maria [6]
[1] Department of Pediatrics, School of Public Policy, University of Calgary, Calgary, Alberta, Canada [2] Department of Medicine, School of Public Policy, University of Calgary, Calgary, Alberta, Canada [3] Department of Community Health Sciences, University of Calgary, Calgary, Alberta, Canada [4] Department of Biology, Faculty of Science and Technology, Mount Royal University, Calgary, Alberta, Canada [5] Department of Social Policy and Health, School of Public Policy, University of Calgary, Calgary, Alberta, Canada [6] Department of Pediatrics, Community Health Sciences, University of Calgary, Calgary, Alberta, Canada

ABSTRACT (ENGLISH)

Introduction
Patient engagement in youth mental health research has the potential to inform research on the interventions, services and policies that will benefit youth. At present, there is little evidence to guide mental health researchers on youth engagement. This systematic review aims to describe the impacts of youth engagement on mental health research and to summarize youth engagement in mental health research.
Methods
We searched the following databases: MEDLINE, EMBASE and PsycINFO, using a combination of subject headings, keywords and synonyms for the concepts ‘patient engagement’, ‘youth’ and ‘mental health’. Articles that described engaging youth in mental health research were included. Two reviewers performed the study selection. Study characteristics, research activities performed by youth, impacts of youth engagement, challenges, and facilitators to engagement and recommendations for youth engagement described by authors were extracted. Quality appraisal involved determining the level of engagement of youth and the stage(s) of research where youth were involved.
Results
The database search returned 2836 citations, 151 full-text articles were screened and 16 articles, representing 14 studies, were selected for inclusion. Youth were involved at nearly all stages of the research cycle, in either advisory or co-production roles. Youth engagement impacts included enhancing relevant research findings, data collection and analysis and dissemination to academic and stakeholder audiences. Both youth and academic researchers reported personal development across many domains. One negative impact reported was the increase in funding and resources needed for engagement. We produced a list of 35 recommendations under the headings of training, youth researcher composition, strategy, expectations, relationships, meeting approaches and engagement conditions.

Conclusions
This study provides an understanding of the impacts and recommendations of youth engagement in mental health research. The findings from this study may encourage researchers to engage youth in their mental health research and support youth engagement in funding applications.
Patient and Public Contribution
We consulted three youths with experience being engaged in mental health research about the review findings and the discussion. One youth designed a visual representation of the results and provided feedback on the manuscript. All youth's input informed the way the findings were presented and the focus of the discussion.

FULL TEXT

INTRODUCTION

Mental health conditions affect 1.2 million children and youth in Canada and this number is increasing.[1] Five percent of Canadian children aged 5–17 years old report anxiety disorders and 2.1% reported a mood disorder in 2019.[2] This aligns with the findings of a systematic review reporting on the prevalence of these disorders in high-income countries (5.2% anxiety, 1.8% depressive disorder, 12.7% any mental health disorder).[3] Of the 12.7% of children experiencing a mental health condition, only 44.2% received any services, revealing a large gap in services for children and youth mental health.[3] Emergency department visits for paediatric mental health concerns have increased 61% from 2009 to 2019,[4] which are often the result of a lack of availability of timely appointments in the community.[5] It seems that current mental health services are not meeting the needs of children and youth, suggesting an urgent need to transform mental health services so that effective, accessible services are being provided.[3,6] As mental health services undergo a redesign, new innovative ways of implementing and delivering mental health care are being studied. It is important to involve youth in that research to ensure that practices, services, programmes and policies are appropriate, accessible and meet their needs.[7] Using patient engagement in research is one approach to ensuring the youth perspective is integrated into mental health research and innovation. The Canadian Institute for Health Research (CIHR) defines ‘patient engagement’ as the meaningful and active collaboration of individuals with personal experience of a health issue and their informal caregivers (including family and friends) in governance, priority setting, conducting research and knowledge translation activities.[8] Patient engagement is a close equivalent of the United Kingdom's concept of Patient and Public Involvement.[9] There is a growing acceptance of patient engagement as being essential in health research on the part of researchers, funders and research institutions. The arguments for patient engagement are philosophical (i.e., patients have a right to shape research about their condition), pragmatic (patient input improves the research process and relevancy of outputs) and practical (i.e., increased transparency and accountability for research that is produced by public funds).[10]

While patient engagement in adult health research is becoming well-established, the momentum for youth patient engagement (herein, youth engagement) appears to be lagging (Mawn, 2015).[11] This may be due to system-level considerations for youth engagement, such as institutional research ethic board approval, issues of consent in youth and a lack of institutional support.[12–14] It may also be due to practical issues such as researchers not feeling competent with youth-friendly engagement methods, difficulties reaching youth for recruitment and funding issues.[12] Also, the changing interests and developmental needs of youth may make it difficult to sustain engagement partnerships over the entire duration of a research project.[15] Recruiting youth for mental health research may have additional challenges, as youth may have experienced stigma related to mental health in their community or within healthcare settings which may create issues of trust between youth and health researchers, leading to youth being reluctant to engage (Knaak, 2017).[16] Youth may also be hesitant to disclose their mental health condition or may be concerned that their condition may become known to their peers as a consequence of their involvement in research. Furthermore, researchers may perceive youth with mental health conditions as vulnerable, and that research engagement activity may affect their well-being.[14]

Despite these potential barriers, youth engagement is considered a guiding principle in recent efforts to redesign

youth mental health services.[17] Youth engagement allows researchers to gain important insights into why youth may not be accessing mental health services, create relevant and responsive interventions and create the conditions that make services accessible to young people.[18] Youth engagement is also a way of recognizing youths' rights for agency and power in shaping mental health services that are for them.[19] Learning about the benefits, successes, challenges and recommendations of researchers with experience with youth engagement in mental health research could help inspire researchers to engage youth in their own mental health research. Furthermore, an understanding of the impacts of youth engagement could support mental health funding applications where youth are engaged as research partners.

To date, the impacts of youth engagement on mental health research and the researchers have not been described. As well, while some recommendations exist about engaging youth in health research, there is little guidance for researchers about youth engagement specific to mental health research. Therefore, the primary purpose of this systematic review was to synthesize the impacts of youth engagement in mental health research. A secondary aim was to describe the challenges and facilitators encountered in mental health studies with youth engagement and to summarize the recommendations for youth engagement in mental health research made by authors.

METHODSStudy design

This systematic review follows the meta-aggregative approach to qualitative synthesis outlined in the JBI Manual for Evidence Synthesis.[20] JBI meta-aggregative approach seeks to enable generalizable statements to guide practitioners and policymakers. It focuses on producing a synthesis of findings that authentically represent the aggregation of data from primary studies, rather than a more interpretive approach where authors re-interpret findings from qualitative studies. The protocol for this review was registered with PROSPERO (CRD42022319240). We used the preferred reporting items for systematic review and meta-analysis (PRISMA) guidelines to report this review.[21] In this review, we distinguish youth co-researchers from academic researchers by using the terms ‘youth researcher’ and ‘adult researcher’, respectively. We use the term ‘co-production’ when referring to activities where youth are collaborating with adults or leading the activity, for example, developing recruitment materials. We use the term ‘advise’ to mean that youth researchers provided ideas and feedback on aspects of the project but were not directly involved in those activities. Three youths with experience engaging in youth mental health research were involved in this project.

Search

We searched MEDLINE, EMBASE and PsycINFO, using a combination of subject headings, keywords and synonyms for the concepts ‘patient engagement’, ‘youth’ and ‘mental health research’. The ‘patient engagement’ concept included participatory action research approaches, which are not always included in definitions of ‘patient engagement’, but were included here because they engage people who bring the collective voice of specific, affected communities to health research.[8] We limited the search to 2000 to the present since patient engagement is a relatively new phenomenon in health research. The ‘mental health research’ concept included mental health, mental health services, as well as clinical diagnostic terms adapted from the Cochrane Common Mental Disorders Group with input from a pediatric psychiatrist. Duplicate citations were removed using automated software and manually by reviewers. Our search strategy is available online as Supporting Information: File 1.

SelectionInclusion and exclusion criteria

We included original research studies where youth were engaged as partners in the research process. We wanted to capture the variations in the approaches to including youth in mental health research, therefore we included a broad age range of youth researchers (8–25 years). To acknowledge that youth may be part of a research team over several years, we included articles where the majority of youth researchers were 25 years or younger. The age of the youth was assessed using the age at which the youth joined the team (where this information was available). Youth researchers could have lived experience with a mental health condition or not. All study contexts were included (i.e., mental health clinical research, mental health services research, community-based participatory research or health promotion/public health research) and any setting (i.e., inpatient, outpatient, community, schools, residential treatment). We included studies conducted in countries with publicly funded health systems. The study

must have described at minimum, one youth research activity and one impact of youth engagement.
We excluded articles that were not peer-reviewed (e.g., commentaries, theses), those studying youth engagement in a programme of research (rather than a specific research project) and those where youth were engaged only in the stage of developing an intervention (e.g., mental health technology or clinical pathway) but not in research or evaluation of that intervention.
Two reviewers (E. M. and M. A.) screened citations on the title and abstract. The same reviewers reviewed the full text of the articles, comparing them against the inclusion criteria. At both stages, discrepancies between reviewers were resolved through discussion. Inter-rater reliability was calculated using percent agreement and Cohen's κ. Covidence was used to manage the study selection process.

Quality appraisal

The focus of this review is on youth engagement within the research studies, and not the specific findings of each study. We felt that assessing the methodological quality of the studies themselves would be less meaningful than assessing the quality of engagement. However, to our knowledge, there are no quality assessment tools available to assess youth engagement as reported in a research article. Therefore, rather than an assessment of quality, we described youth engagement on two dimensions: level of youth engagement, and stages of the research cycle where youth were involved. The description of the level of youth engagement is based on the 'Types of youth participation' in *INNOVATE Research: Youth Engagement Guidebook for Researchers* (2019). These are *Participation* (i.e., youth are the subject of study), *Consultation* (i.e., youth provide feedback on research), *Partnership* (i.e., youth work collaboratively with researchers as equals) and *Youth-led* (where every stage of research is driven by youth). Key stages in the research lifecycle are (1) Priority setting and planning; (2) Development of the research proposal; (3) Scientific review; (4) Ethics review; (5) Oversight of a research project; (5) Recruitment of research participants (for some types of research); (6) Data collection; (7) Data analysis and interpretation; (8) Knowledge exchange; (9) Evaluation and quality assurance.[22] One reviewer (E. M.) categorized each study on these two dimensions, with a second reviewer verifying the descriptions (K. T. B.).

Data extraction and synthesis

Data extracted included study characteristics, characteristics of youth researchers, research activities of youth, as well as the findings of the study that related to youth engagement. We extracted findings about youth engagement for each of the following features: impacts of youth engagement on the research process and researchers, the facilitators and challenges to youth engagement and author recommendations for youth engagement. We used line-by-line extraction, from any location in the article, including methods, results, discussion and conclusions. Data extraction was performed by a single researcher (E. M.), with a second researcher cross-checking the extracted data (K. T. B.). Discrepancies were resolved through discussion.
The findings for each feature were reviewed and descriptively coded. Codes were grouped by similarity in concept by a single reviewer and then combined into categories. One researcher (E. M.) created category descriptions, which were reviewed by one member of the research team (S. R.) and three youth researchers who were consulted.

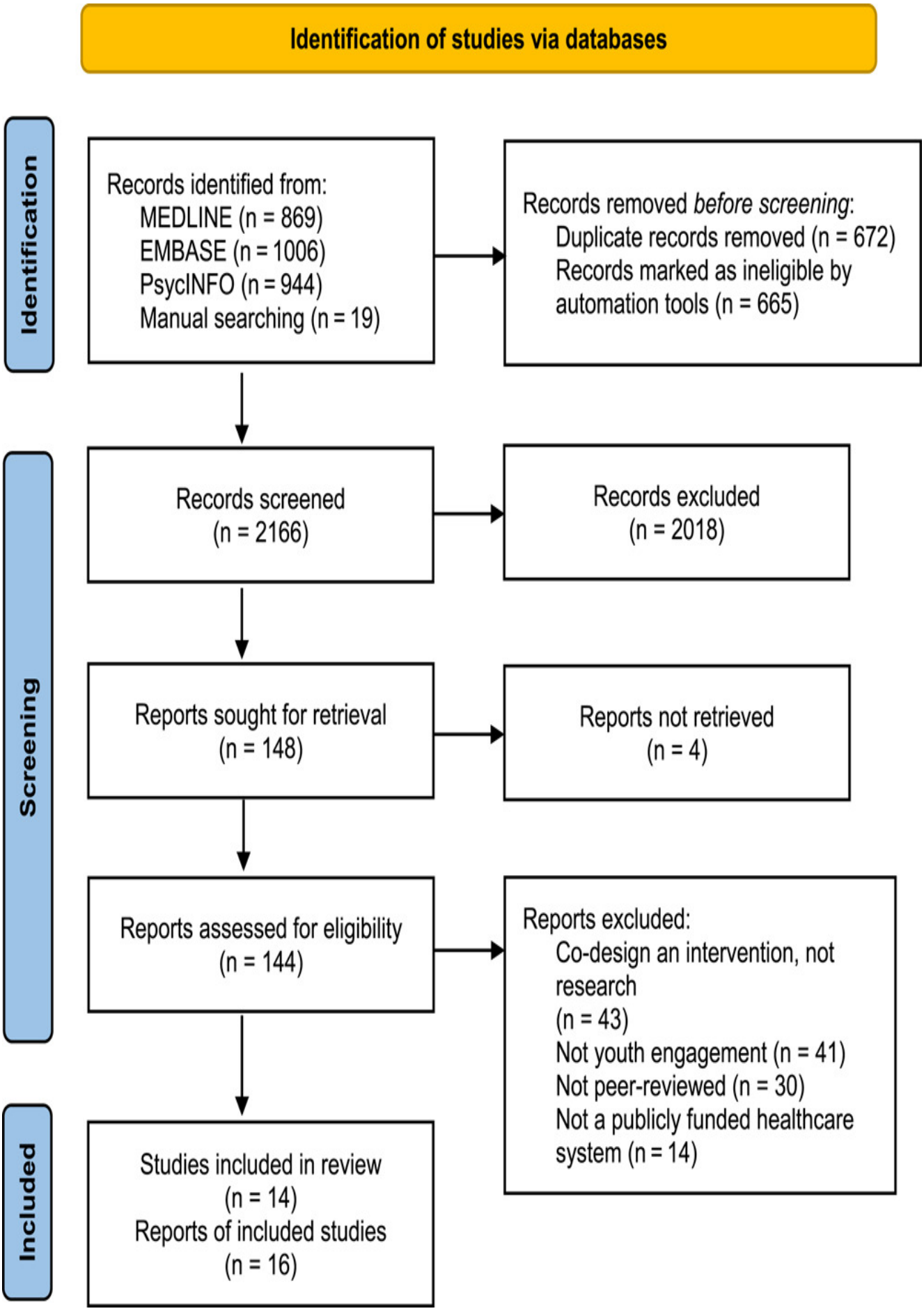
Youth engagement in this review

We held a consulting meeting with three youths (ages 19–24, all identify as cis men, all Canadian citizens, one with Chinese and one with Southeast Asian heritage), all with previous experience engaging in mental health research. The aims of the consultation were threefold: to understand whether the way we presented the findings aligned with their experiences as youth engaged in research if they had additional recommendations for youth engagement and which of the findings were most salient to youth engaged in research. The feedback from the consultation informed how we presented the study's results and structured the discussion.

RESULTSSearch and selection

Figure 1 summarizes the search and selection process. The search retrieved 2838 citations. We removed 672 duplicates and 2166 citations were screened on the title and abstract. The percent agreement between authors was 88.4% (Cohen's $\kappa = 0.52$). The full-text articles for 148 citations were reviewed, and 132 were excluded, primarily because they were describing co-design of an intervention or clinical service (43 articles), or youth were participants

in the study rather than involved as researchers (34 articles). Sixteen articles were included. The percent agreement between authors was 93.6% (Cohen's $\kappa = 0.45$). Two pairs of articles described the same study, therefore, a total of 14 studies were analysed.



Enlarge this image.

Description of studies

Table 1 contains the key characteristics of the articles. The articles were published in four countries: Canada (*n* = 6),

the United Kingdom (*n* = 8), Australia (*n* = 1) and Norway (*n* = 1). None of the articles were published before 2014 and most were published between 2020 and 2022 (*n* = 11). In nine articles, a description of youth engagement was embedded within the report on the research project, while seven articles reported directly on the youth engagement aspects of a research project.

Table 1 Characteristics of articles included in the analysis

Study ID	Year	Country	Sample characteristics	Sample age range	Sample size	Mental health research area	Study designs	Methods	Study setting	23
2020	United Kingdom	Children from a school in North West England	5-10 years old	17 in focus groups, entire school for playtime observations	Public health research	Multiple methods	Focus groups and observation of play, participatory thematic analysis	School in North-West England	24	2020
Canada	Varied in sex and gender, education, socioeconomic status, literacy, and mental health care experiences	16-25	approximately 24	Health services research	Priority-setting	Research priority setting	Community MH services, Hospital MH services	25	2020	Canada

Adolescent using child and adolescent mental health serv	16-19	21	Health services research	Qualitative study	Qualitative interviews	Child and adolescent mental health services in Ontario	26	2021	Canada	Youth who accessed CAMHS, caregivers of youth accessing CAMHS, and clinicians/administrators

i c e s c l o s e t o t r a n s i t i o n a n d 1 y e a r p o s t t r a n s i t i o n										

youth19-25	20 youth, 17 caregivers, 21 clinicians and administrators	Health services research	Priority-setting	Delphi study with qualitative analysis of comments	Child and adolescent mental health services in Ontario	27	2019	United Kingdom	Youth with severe mental illness, 81% female	18-25 years old
16	Clinical research	Qualitative study	Qualitative interviews	Patients under the care of a community mental health team	28	2020	United Kingdom	Youth and adult researchers involved in a mental health research study with youth engagement	NR	NR

Clinical research	Descriptive study	Researcher reflections	Community mental health services	29	2021	Australia	Youth living in Youth Residential Rehabilitation Services	17-25	18	Clinical research
Qualitative study	Qualitative interviews and focus groups	Youth Residential Rehabilitation Services	30	2017	United Kingdom	NA	NA	NA	Clinical research	Systematic review

Systematic review	Inpatient mental health services	31	2014	United Kingdom	Mixed ethnicity, genders, employment/education/unemployed	16-25	65	Health services research	Participatory qualitative study	Quality standard development through focus group and nominal group technique

Community MH services, primary care, hostel, drop in servi	32	2018	Canada	Youth accessing care for mental health concerns	NR	500	Health services research	Descriptive	Randomized controlled trial	Integrated community-based collaborative care team

ce										
33	2017	United Kingdom	2 male, 9 female, most in education	16-18	11	Health services research	Multiple methods	Participatory design	Community	34
2021	Canada	Black youth living in Alberta, gender diverse, predominantly Christian	16-30	30 interviews, 99 in conversation cafes	Public health research	Participatory qualitative study	Qualitative interviews and focus groups	Community	35	2021
United Kingdom	Students in 2 schools	18-Nov	115	Public health research	Priority-setting	Priority-setting	Schools	36	2021	Canada

NR	NR	28	Health services research	Qualitative study	Qualitative study	Integrated community-based collaborative care team	37	2022	Norway	1 adult researcher and 10 youth researchers involved in youth engagement in research

adolescents over 15, adult	11	Health services research	Authoethnography	Autoethnography of patient engagement in a 4 year mental health research project	University of Stavanger	15	2021	United Kingdom	NA	NA
NA	Clinical research	Systematic review	Systematic review	NA	Study ID	Study primary purpose(s)	Key findings	Study focus	Number of youth researchers	Age ranges

Ethnicity	Gender/Sex	Youth with lived experience of mental health concerns	Model of youth engagement	Specific methods	23	How do playtime experiences impact social, emotional and mental health and well-being, from the perspective of children?	Four themes: having someone to play with, games, things about playtime and how people treat each other.	Study report with engagement embedded	4	9-10 years old

2 white, 1 chinese, 1 pakistani, all British born	2 boy 2 girls	No	Research and Development in Organisations (RADIO) model	Co-produce	24	Aim to engage with youth and parents (or caregivers) in an emerging program of research aimed at understanding increasing mental health ED presentations in a Canadian paediatric tertiary health centre to support the development of effective interventions.	Research areas highlighted: (1) Access to mental health and addictions services; (2) Gaps in care; (3) Standards of care; (4) Stigma; (5) Experience of care; Also made recommendations	Study report with engagement embedded	NR	NR

							for future engagement activities			

NR	NR	Yes	IAP2 Core Values for the Practice of Public Participation	Advisory meetings	25	To qualitatively explore the experiences of youth in relation to their knowledge, expectations, and experiences transitioning out of CAMHS services at age 18.	Themes: (1) Shifting awareness of the meaning of 'transition', (2) Ready or not to transition, (3) Mixed reactions to transitional age of 18 years, (4) Lack of information, preparation and	Study report with engagement embedded	3	18+

							involvement in the transition planning process, (5) Confusion around roles and responsibilities within the transition process, (6) Concern over transition gaps leading to poor mental heal			

							th outc ome s			

NR	NR	Yes	McCain Model for Youth Engagement	NR	26	Prioritizing and refining the core components of effective transitions from child and adolescent to adult mental health services	26 core components; 3 themes: (1) need for youth and adult services to collaborate, (2) suggestions on how to operationalize core components, (3) barriers to implementation	Study report with engagement embedded	3	NR

NR	NR	Yes	NR	Advisory meetings and co-produce	27	Explore the feasibility and acceptability of technologies to detect mental health deterioration	Four main themes: (1) dealing with mental health symptoms, (2) signs of mental health deterioration, (3) technology concerns and values and (4) technological applications to identify	Study report with engagement embedded	7	18-25 years old

							worseningmentalhealth.			

5 White British, 1 British - Asian, 1 Black - British	2 male, 5 female,	Yes	NR	Advisory meetings and co-produce	28	Explore the experiences and impacts of engagement in a qualitative study	Co-producing research with youth makes a significant impact to the research, researchers and co-researchers; co-production takes time; build flexibility into budget and more intervie	Study of engagement	7	18-25 years old

							w training is recommended.			

5 White British, 1 British - Asian, 1 Black - British	2 male, 5 female,	Yes	NR		29	Explore what matters to young people living in a 12-month voluntary residential program for young people aged 16–25.	Two themes: factors that supported an environment for young people to thrive, and the 'change work' that young people undertook	Study report with engagement embedded	4	between 16-25

NR	NR	Yes	Participatory action research informed by the critical emancipatory paradigm	Advisory meetings and co-produce	30	Explore “risk” in inpatient mental healthcare with an evidence synthesis and input from stakeholders	Priority areas of “risk” of inpatient stays for MHC: Dislocation, Contagion, Harm from organisation, Institutionalisation, Self-harm, Decision-making, Suicide, Aggression, Other (in cont	Study of engagement	2	under 18

							rast to the clinical risks found in the literature ie self harm and aggression)			
NR	NR	Yes	Nominal group technique	Advisory meetings	31	To develop user-generated quality standards for young people with mental health problems in primary care using a participatory research model.	16 quality standards for youth mental health in primary care	Study report with engagement embedded	29	16-25

NR	NR	Yes	NR	Advisory meetings and co-produce	32	For youth with mental health concerns, does an integrated collaborative care model, compared to usual care, result in better outcomes? Economic evaluation of the new model	A description of facilitators, barriers and youth researcher activities	Study of engagement	30	16-26
NR	2 male, 6 female	Yes	McCain Model for Youth Engagement	Co-biuld	33	To understand the emotional support related needs of young people.	Two ways of supporting young people's mental health: provide choice, raise awaren ess	Study of engagement	12	16

	N R	2 male, 9 female	No	Nominal group technique	Advisory meetings	34	The purpose of this qualitative research study was to identify the barriers and facilitators to mental health care for Black youth in Alberta.	Barriers to mental health care access: lack of cultural safety and inclusion; lack of knowledge/information on mental health services; cost of mental health services and geographical and	Study report with engagement embedded	10	NR

							locational barriers; stigma and judgmentalism; and limits of resilience			
Black	NR	No	Youth empowerment model situated within intersectionality theory	Meetings, co-produce	35	Describes the establishment of a youth research advisory group to plan health research	Student health priorities centred on mental health and stress	Study report with engagement embedded	115	18-Nov

“cultural and linguistic diversity”	100 female, 15 male	No	NR	Advisory meetings	36	Explore the research team's experience of youth and family engagement in the design of an RCT and clinical pathway.	A description of facilitators, barriers and recommendations for youth engagement in mental health research	Study of engagement	7	NR

NR	NR	Yes	McCain Model for Youth Engagement	Co-produce	37	To understand the collaborative relationship between a lead researchers and youth researchers in a research project aiming to improving mental health services for adolescents.	6 themes: (1) Commitment motivated by altruism, personal interests and a common purpose, (2) Inclusiveness and support to reduce social uncertainty and strengthen collaboration, (3) Red	Study of engagement	10	Adolescents over 15 years of age

							uced power differentials while ensuring clarity of roles and task, (4) Diversity in representation to expand the perspectives of ‘the adolescent voice’, (5) Self-determination - supporting			

							adolescents' involvement in decision-making processes, (6) Flexible and systematic project management.			

“a variety of different ethnic backgrounds” and with different	“both genders”	Mix	NR	Advisory meetings and co-produce	15	Understand interventions for mental health in children with long term conditions.	Challenges, facilitators and recommendations for youth engagement	Study of engagement	8	17-Oct

r e n t l i f e a n d h e a l t h c a r e e x p e r i e n c e s										

The majority of studies engaged youth 16+ years old, with only one study engaging children 9–10 years old. Studies were on mental health services (*n* = 7), clinical research (*n* = 4) and public health (*n* = 3). Studies engaged between 2 and 115 youth. The studies with higher numbers of youth (*n* >30) were priority-setting and brainstorming-type engagement activities. Five studies reported on the racial/ethnic diversity of the youth researchers, while seven reported on the sex or gender of engaged youth. A focus on diversity and inclusion within the research team was present in five studies. Most studies engaged youth with lived experience of mental health conditions (12/14). Five studies used advisory meetings as their only approach to engagement, while two studies engaged youth in specific research activities without conducting formal advisory meetings. Six studies used a combination of both advisory meetings and youth researchers engaging in specific research activities. A variety of models of youth engagement were used (see Table 1). Structured research training was provided to youth in five studies.

Youth engagement

The activities of youth researchers are described in Table 1. Youth were engaged as advisors and/or actively carried out specific research activities, in some cases leading the activities. Table 2 contains a summary of youth researcher activities, divided by whether the activity was done in a co-production or advisory role. In four studies, the youth performed an advisory role only. The most common research activities were focusing on the research topic (*n* = 7), co-analysis of qualitative data (*n* = 7) and dissemination of findings (*n* = 10).

Table 2 Research activities performed by youth researchers

Co-production	Advisory role	References
Co-produce an agreement on roles and responsibilities for research team		[32, 36, 37]
Co-develop research design/protocol		[23, 27, 28, 32, 34, 36, 37]
	Advise on scope of research, research design/focusing research question	[15, 23, 30, 34, 37]
Co-develop funding proposals		[30, 34, 37]
	Advise on recruitment strategies	[26–28]
Co-develop study informational materials		[26–28, 32, 36, 37]
Recruitment of participants		[34, 37]
	Participate in advisory meeting(s)	[24, 30, 33, 35]
Advise on environment/contextual factors for participant interactions	Advise on contextual factors and ways of relating for participant interactions	[24, 32, 36]
	Advise on data collection instrument(s) (survey, interview guide)	[25, 26]
Co-develop data collection instrument(s) (survey, interview guide)		[27–29, 34, 37]
Co-facilitate focus groups/interviews/gather observational data from peers		[23, 27–29, 31, 34, 37]
	Review content/thematic analysis and interpretation of findings	[15, 25, 26, 31]
Co-analysis of qualitative data		[23, 27–29, 31, 33, 34, 37]
	Advise on dissemination strategies for stakeholders	[26]

Present findings to stakeholders		[15, 23, 29, 34, 37]
Co-present at academic conferences		[15, 27, 28, 37]
	Review journal manuscripts and final reports	[15, 33]
Co-write journal manuscripts and final reports		[25, 34, 37]
Co-produce recommendations for action based on research		[23, 31, 35]

Quality appraisal

Youth were engaged at a 'consultation' level in five studies, a 'partnership' level in eight studies and one study was 'youth-led'. In three studies at the partnership level, a hybrid model was used where they had a small number of youth researchers were involved in research activities and a larger advisory committee of youth was consulted at key stages in the research process. This model was used to increase the diversity of the youth perspectives that influenced the research project. Table 3 contains the results of the quality appraisal, that is, the level of engagement of each study, and the stages of research where youth were involved. Seven studies involved youth in almost all stages of research.[23,27–29,32–34,36,37] All studies involved youth in some form of quality assurance or evaluation of the research project, with five studies specifically involving youth in evaluating the engagement aspect of the project.

Table 3 A description of youth engagement by level of engagement and stage of research involvement

[IMAGE OMITTED. SEE PDF.]

[a]Hybrid model of primary partnership with a small number of co-researchers, with a larger advisory committee that was consulted for key stages in the research study. **Impacts of youth engagement**

No studies reported a formal impact assessment of youth engagement, although four studies explored the impacts and experiences of youth engagement in research.[15,28,36,37] Table 4 contains a list of the impacts of youth engagement.

Table 4 Impacts of youth engagement on the research process and researchers

a

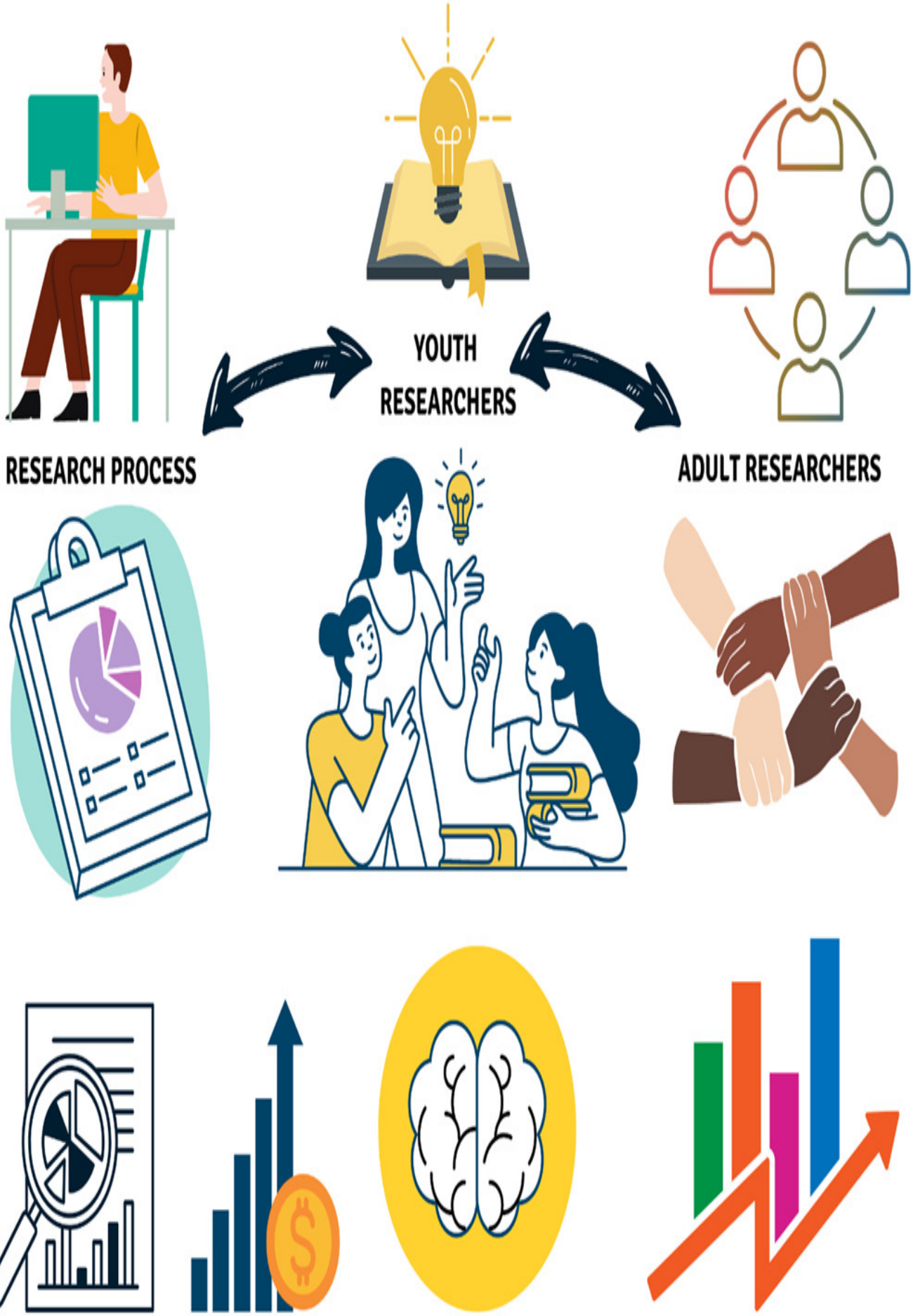
Reported as both positive and negative impacts in different articles.

The most common research process impacts of youth engagement reported by authors were (1) the data (*n* = 9), either by shaping the data collection instrument or being actively involved in data collection; (2) the findings from the study (*n* = 9), by youth involvement in the analysis; (3) enhanced knowledge dissemination (*n* = 9), by co-presenting and advising on knowledge translation strategies. Enhancing the relevancy of research topics was another common impact reported in six studies, and four studies reported that having youth on the research team enhanced the safety and comfort of their research participants.[24,27,28,32,36,37] One study reported that youth engagement made decision-making more efficient because youth provided perspectives that made the decision clearer.[32,36] Another study reported the opposite, that decision-making was less efficient, but this was attributed to the adult research team members' intention to create an inclusive environment.[37] Besides the efficiency of decision-making, other negative impacts included the increased resources required for youth engagement (*n* = 6), and that youth may have unintentionally influenced data collection by asking leading questions or reassuring participants and sharing their own experiences.[27,28]

Adult researchers reported increasing their knowledge of youth engagement strategies,[15,27,28,32,36,37] stating that youth engagement broadened their networks and enhanced their understanding of the research findings.[27,28] A sense of

pride in the youth researchers' development over the course of the project was mentioned in two studies.[15,37] In one study, authors reported a greater sense of accountability for their research and thus more motivation to perform high-quality research, which was described as positive.[15] Related to this, in two studies, a greater sense of responsibility for youth researchers was reported as having a negative impact on adult researchers.[27,28,31]

Youth researchers reported positive findings, feeling empowered and respected, particularly when witnessing their input being acted upon[15,23,30] and increased confidence in their abilities.[27,27] They reported that they gained knowledge about research and mental health, and developed research, project management and communication skills.[15,23,27,28,37] A sense of social connectedness and expanded networks were mentioned[15,27,28,37] as well as the research experience being a benefit for their job resumes and applications for postsecondary education and generating income.[37] Figure 2 illustrates the impacts of youth engagement in research.



Enlarge this image.

Facilitators and challenges to youth engagement

Table 5 describes the challenges and facilitators to meaningful youth engagement reported by the authors. One

challenge reported in three studies was the time and effort for relationship-building within the research team, and this was considered especially important in a mental health context.[15,27,28,37] There were challenges related to the recruitment and retention of youth researchers, and one study mentioned that as youth researchers become more skilled and acculturated to academic research environments, there was a need to monitor whether they were still representing the youth voice.[32,36] A final area of challenge related to navigating diverse perspectives and priorities of the research team. For example, adult researchers prioritize rigour versus youth wanting to reassure participants,[27,28] managing divergent youth and caregiver perspectives,[32,36] and perspectives of youth from different cultural backgrounds.[24,32,36,37]

Table 5 A description of the facilitators and challenges to youth engagement

	Facilitators of youth engagement	Challenges of youth engagement
Relational	Create safe spaces Reflexivity in adult researchers (i.e., an awareness of power dynamics, how they are relating with youth) Efforts to build relationships (genuine, trusting) between youth and adult researchers Power-sharing with youth (i.e., empowered in decision-making, treating youth as equals) Using accessible language	More time/effort to build relationships, especially in mental health which can be a sensitive issue Power imbalance between youth and adults Communication barriers between adult and youth researchers Navigating diverse perspectives/conflicting priorities (adult vs. youth, youth vs. parents) Managing youth expectations (e.g., about the impact of the project)

Process	Using youth-friendly communication tools (e.g., text messaging) Having a dedicated youth engagement coordinator Building relationships with community organizations Refreshments/ice-breaking activities Flexibility with degree of involvement and scheduling Use of pre- and debriefs for large meetings Having diversity among youth voices Clear expectations for youth about engagement	More work to set up engagement (as a new process) More work to support (e.g., training, accommodating needs) and coordinate youth engagement More funding, time, work Recruitment of youth researchers (finding appropriate youth, representing diversity) Monitoring whether youth are remaining representative (as they become more involved in the project, youth researchers may begin to think more like adult researchers) Sustaining engagement over the course of the project Research ethics board Balancing bringing together a diversity of backgrounds and perspectives versus efficiency in decision making Not involving youth early enough to influence project Potential for youth engagement to affect research rigour

Relational facilitators of engagement included creating a safe, inclusive space for youth to share perspectives, adult researchers having an awareness of power dynamics and how they are relating with youth, and efforts to build genuine and trusting relationships. Process facilitators included having a dedicated youth engagement coordinator and providing refreshments and compensation for youth researchers.

Recommendations for youth engagement

Four articles contained recommendations for youth engagement in mental health research,[15,27,28,36,37] while other articles contained recommendations embedded within Section 4. Table 6 contains a summary of recommendations for youth engagement. Recommendations were around training for both youth and adult researchers, the composition of the youth on the research team, processes for engagement, approaches to consultation meetings, agreement between youth and adult researchers about expectations, roles and responsibilities, elements of the relationship between youth and adult researchers and the conditions in which engagement occurs.

Table 6 Summary of recommendations for youth engagement in mental health research

Area	Recommendations
Training	Training should include education about the research topic, the research process and the opportunity to practice skills before project start. Training on communication and leadership skills should also be included.

	Training and support should be more intense early in the project, with a gradual reduction of support as youth competency increased.
	When transitioning youth into a project already in progress, be mindful that they are adequately prepared and have the same opportunity for training as youth who begin at the start of the project.
	Experienced youth researchers can lead youth research training.
	Enhance academic researchers' knowledge of youth engagement, for example, include patient engagement as part of a research Masters and PhD curriculum, provide additional training for established researchers.
Youth researcher composition	Consider recruiting several youth at the outset of the project due to difficulty sustaining youth involvement over time.
	Ensure diversity in youth representation when appropriate for the project, including diversity in research experience (include youth naïve to research).
Processes	Engage youth early in the research process to optimize their impact on the project.
	Have a dedicated engagement facilitator or share engagement coordination responsibilities with youth researchers.
	Be strategic about youth engagement activities, plan ahead for engagement during key transitions in research project when decisions will be made.
	Have a flexible budget with a contingency fund for unexpected research activities suggested by youth researchers.
	Build in a mechanism for asking for feedback from youth about the engagement process and how you will incorporate feedback into the process.
Meeting approaches	Provide opportunity for both written and verbal participation in the research process (e.g., nominal group technique, opportunities for written feedback if a youth cannot attend a meeting).
	Use age-appropriate and engaging activities during consultation meetings.
	Consider having youth co-facilitate meetings.
	When seeking feedback, use case scenarios and examples to make abstract concepts concrete.
	Use warm-up activities before consultation meetings.

	Provide small group prebriefs for youth before meetings, explaining meeting objectives, key terms and an opportunity to ask questions.
	Hold small group debriefs after meetings, giving an opportunity to ask questions and provide feedback that youth were perhaps reluctant to share with a larger group of research team members.
	Provide refreshments.
Agreement on expectations	Be clear with youth about the objectives of the project and its expected impact.
	Establish clear role expectations, including the responsibilities of both the youth and adult researchers. This includes an agreement about the degree of control that youth have over the project.
Relational elements	To reduce power differential between youth and adults, establish a collaborative relationship between adult and youth researchers, on a foundation of trust, respect and rapport.
	Create a safe space for open discussion (e.g., include social identity in introductions, adult researchers being transparent and genuine).
	Dedicate time and funding for relationship building.
	Demonstrate respect for youth and their impact on the project by following through on their decisions and recommendations and sharing final results.
Engagement conditions	Consider ways of minimizing the potential for distress in youth (e.g., hold sessions at community agencies they are familiar with, provide peer and/or professional support, seek feedback from youth).
	Include caregivers but use separate forums to encourage youth's voice and unique opinions.
	Use youth-friendly meeting spaces and communication tools (e.g., group messaging apps).
	Flexibility with meeting times and venues to accommodate youth schedules.
	Be flexible about the degree of involvement of youth.
	Be aware of and accommodate physical, mental and emotional needs of youth.
	Share power and leadership responsibilities with youth.

Incentives	Include incentives like course credits and certificates of completion where possible.
	Provide compensation for youth's time and travel for meeting and research activities.

Youth engagement in this review

Overall, the youth agreed with the findings of this review. They emphasized that overcoming the power differential between youth and adult researchers, as well as the representation of diverse youth voices was important. Their input resulted in the addition of one new impact, two new challenges, the reorganization of the recommendations section and the addition of concrete examples to some of the recommendations. We also revised the wording of some of the recommendations based on their feedback. One youth (J. M.) produced the visual of the impacts and also contributed to the writing of the manuscript, he is included as a co-author on this paper.

DISCUSSION

Patient engagement research impacts have been conceptualized as both positive or negative, short or long-term, and are either related to the research process (e.g., research instruments, outcomes measure choice, data collection design, delivery, time, dissemination) or impacts to the people involved (e.g., youth and adult researchers' experiences).[38] Documented impacts of youth engagement on the research process include a positive influence on research design, recruitment, data collection and analysis and dissemination.[39] It has also been reported to increase the youth friendliness and validity of research, the usability of practical tools, accessibility of consent forms and questionnaires and increase media attention.[7,39] There were few negative impacts reported, but inexperienced youth facilitators can negatively impact the quality of focus group data, and youth may interpret findings in relation to their own experiences impacting generalizability.[39] Skill development, feeling empowered, confident and valued, as well as enhanced social connectedness, are positive impacts reported by youth engaged in research.[7,39] Academic researchers report an increased feeling of commitment to their project, inspiration and pride in their work.[39] In this review, the impacts of youth engagement ranged from enhancing the relevancy of research topics to enhancing dissemination and impact on the health system. This aligns with what has been found in other reviews of youth engagement.[39,40] An impact unique to mental health research engagement was the enhanced comfort and emotional safety of research participants resulting from the involvement of youth. In one study, researchers used a pre-engagement consultation with youth and caregivers to design a distress-sensitive approach to their recruitment and data collection process, which included holding data collection sessions at community agencies with peer and professional support, providing written materials, giving participants the option of providing written feedback and to separate youth and caregivers.[24] Another study reported that youth completing interviews were able to quickly develop rapport with participants and humanize the interview process for them. This was felt to enhance the emotional safety of participants, for whom talking about mental health may be uncomfortable or stressful.[28]

We found that youth researchers reported many personal benefits to being engaged in mental health research, including feeling empowered, a sense of social connectedness, gaining knowledge and skills and enhancing career and education opportunities.[15,23,28,30,37] Youth researchers felt that research engagement expanded their professional networks, which was also reported by adult researchers.[28,37] The impact on adult researchers of engaging with youth was less often the focus of the studies, however, some impacts were reported such as gaining an appreciation for engagement, increased accountability for their research products and a sense of pride in youth researchers' development.[15,28,36,37] Adult researchers report that youth engagement added more to their responsibilities during research, because of their desire to foster positive engagement experiences for youth, which was viewed as both a positive and a negative impact.[15,28,31]

The negative impacts of youth engagement include the increased time and resources needed for engagement, which is commonly reported across all types of patient engagement studies.[39–43] Researchers have reported concerns that youth with some mental health conditions could be vulnerable and engagement could potentially

negatively impact their well-being, whether from experiencing the power imbalance between adults and youths, or perhaps embedding the mental health condition as a part of a youth's identity.[14,43] We did not find evidence of these potentially negative impacts in our review, which may be reassuring for mental health researchers. Another potentially negative impact on the research relates to the methodological rigour of the research. Through their involvement in data collection and analysis, youth very commonly impacted data collection and analysis. This was viewed as positive in most cases, though there was some concern expressed about youth introducing bias into data collection and analysis through, for example, asking leading questions or incorporating their own experiences into data analysis.[28] This was viewed by some as a negative impact, but one that could be overcome through training and close supervision.[28] We also found that only one of the studies in this review used quantitative methods,[32,36] which could suggest that researchers believe quantitative studies are not suited to engagement or that youth engagement could limit the researchers' choice of methods to answer a particular research question. This was an issue that was also brought up by our youth researchers during the consultation meeting. However, outside of mental health research, youth have been engaged in quantitative research, for example, randomized controlled trials, comparative effectiveness research and measurement instrument development studies, which suggests that youth can be engaged in quantitative mental health research.[40]

There were practical challenges encountered by researchers engaging youth in mental health research. The increased resources that are needed for setting up and supporting engagement, recruiting and sustaining youth researchers throughout a project were mentioned across almost all studies Adult researchers also grappled with ethical considerations as well as navigating conflicting priorities of different groups, such as the youth and adult researchers, within youth researchers with different backgrounds and experiences, or between youth and caregivers.[24,27,28,32,36] There were also challenges related to the relationship between the adult and youth researchers that needed to be overcome for productive working relationships to develop between youth and adult researchers. These included the inherent power imbalance between youth (as younger, novice researchers) and adults (as older, established researchers) and communication barriers between youth and adults. While these challenges are not unique to youth engagement in mental health research, authors felt that their importance was heightened in a mental health research context, which is a potentially sensitive subject.[15,28,36,37] Authors reported that putting in the time and effort to build trusting and genuine relationships was a successful way to overcome this challenge, as well as the adult researchers practising reflexivity (i.e., being self-aware, reflecting on the way they relate to youth researchers). This finding aligns with the recent interest in the importance of relationships in patient engagement work.[44,45]

The findings of this review support the idea that youth are willing and capable of being involved in research activities across the research cycle. Youth were involved, either in an advisory role or performing research activities, at all stages of CIHR's research cycle (i.e., from developing topics to disseminating findings). Studies reported successful youth engagement across all levels of engagement (Collaboration, Partnership, Youth-led), which differs from some visions of patient engagement, where a partnership or complete control over research is considered the gold standard. This supports the idea put forth by Greenhalgh et al.[10] that a more flexible approach to youth engagement, where the desired outcomes of engagement for the project and the motivations and capabilities of the individuals involved drive the engagement approach, rather than a single framework informing all patient engagement activities.

The recommendations contained in this article will be useful to researchers planning youth engagement in mental health research. They align well with the practical recommendations for youth engagement in health research put forth by Hawke et al.[7] The recommendations from our review that might be considered unique to a mental health research context, such as creating a safe space for open discussion, accommodating emotional and mental needs, are incorporated in Hawke and colleagues' recommendations. The youth researchers we consulted in this review agreed with all the recommendations in the review. They emphasized the importance of overcoming power imbalances, which was a common theme among the articles in our review. They also felt that representation of diverse youth voices, in terms of ethnicity, race, gender and sexual identity and degree of experience in research was important. Related to this, they felt that adult researchers engaging with youth in a mental health context should have training in trauma-informed approaches, as well as cultural competence. Although this was not a

recommendation in any of the articles in this review study, it is supported by Shimmin and colleagues' argument that patient engagement should be underpinned by trauma-informed approaches, as well as a recommendation in *INNOVATE Research*.[46,47] This may be especially true in a mental health context, where typically youth researchers are seeking to help shape a research project because of their experiential knowledge of mental health or mental health services. These experiences may co-occur with traumatic experiences and asking the youth to share their experiences may be retraumatizing or cause them significant distress.[47]

Strengths, limitations and future directions

A strength of this review is the rigorous study search and selection strategy, and our focus on describing patient engagement in lieu of a traditional quality appraisal, which would have been less informative for this study. Also, we used an established method for aggregating qualitative findings.

A limitation of this review is the degree of youth engagement in the project. Youth were involved at the later stages of the review but were not involved in the conception or design of the review, which may limit the relevancy of this review for youth involved in research.

Also, as this is a relatively new field, the terminology used in the field of patient engagement varies across geographic settings. Though we made an effort to be comprehensive in our search strategy, there is the possibility that we missed some studies due to variability in terminology. As well, since this review relied upon authors' reporting on engagement activities, it is likely that some activities and impacts were missed, especially in studies where engagement was not the focus of the article.

One final limitation in this review is the possibility of a bias in our findings towards more positive engagement impacts. This could be due to adult researchers' position of power exerting control (intentionally or unintentionally) over what is reported in the manuscript leading to underreporting of negative experiences or impacts of youth engagement. Also, the inclusion criteria for this review included a requirement that authors reported on at least one activity and one impact on youth engagement. This may have created led to a positive bias in our findings because researchers who report more extensively about engagement may also have been more measured in their approaches to youth engagement, leading to positive engagement experiences for the research team. Similarly, due to the power imbalance between adult and youth researchers, youth researchers may be reluctant to report the negative impacts or experiences during the project. Finally, youth researchers could have experienced negative impacts in studies where youth engagement was minimally reported or where youth engagement was not evaluated. Therefore, our findings should be interpreted with some caution.

The impacts described in the articles were mostly proximal (e.g., effects of youth engagement on the research process), with some intermediate (e.g., skill development of researchers). However, the long-term impacts of youth engagement, such as impacts on patient outcomes, were not reported. As previously discussed, none of the studies described a formal assessment of the impacts of youth engagement. This unfortunately limits the extent of the evidence for youth engagement in mental health research and also suggests a need for more formal evaluations of youth engagement in future projects. While impact assessment is complex and requires more resources, it is nevertheless important to lend credibility to the argument that patient engagement in research is worth the return on investment. To overcome the positive bias described above, these evaluations could be led by youth, giving them more power to openly report on engagement impacts.

CONCLUSION

The overall purpose of this systematic review was to synthesize the impacts of youth engagement on mental health research. We aggregated the reported impacts of youth engagement across research studies and described how youth were being engaged in research, challenges and facilitators to engagement. The recommendations for youth engagement in mental health research contained in this article can be applied by researchers who are planning to engage youth in mental health research. This study provides an understanding of youth engagement in mental health research that may encourage researchers to engage youth in their mental health research. It will also be useful in supporting requests for funding for youth engagement.

AUTHOR CONTRIBUTIONS

Erin McCabe conceptualized and designed the study, search and selected articles performed data extraction and analysis and wrote most of the manuscript. Mungunzul (Megan) Amarbayan contributed to the study design, and article selection, and critically reviewed the manuscript. Sarah Rabi assisted with youth researcher consultations, and wrote parts of and critically reviewed the manuscript. Justino Mendoza contributed to the analysis and interpretation of results, developed a figure and critically reviewed the manuscript. Syeda Farwa Naqvi assisted with youth researcher consultations and critically reviewed the manuscript. Kalpana Thapa Bajgain performed data extraction and critically reviewed the manuscript. Jennifer D. Zwicker contributed to the study design, and data interpretation and critically reviewed the manuscript. Maria Santana conceptualized the study, contributed to study design and data interpretation and critically reviewed the manuscript. All authors reviewed and approved the final manuscript.

ACKNOWLEDGEMENTS

We would like to acknowledge the support of Heather Ganshorn, Health Sciences Librarian and our youth researchers Eric Au, Thomas Tri and Justino Mendoza for their insights into this project. This project was supported by the Alberta Children's Hospital Foundation and Kids Brain Health Network. Jennifer D. Zwicker was supported by a Tier II Canada Research Chair in Disability Policy for Children and Youth.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data are available on request from the corresponding author.

DETAILS

Subject:	Databases; Mental health; Composition; Personal development; Systematic review; Subject headings; Facilitators; Mental health care; Synonyms; Mental disorders; Dissemination; Data collection; Medical research; Researchers; Health professional-Patient communication; Visual representation; Children &youth; Citations; Health services; Funding; Youth
Identifier / keyword:	co-design; community-based research; mental health; mental health services; patient and public involvement; patient engagement; youth engagement
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	30-50
Publication year:	2023
Publication date:	Feb 2023
Section:	REVIEW ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford

Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Evidence Based Healthcare, Journal Article
Publication history :	
Online publication date:	2022-11-16
Milestone dates:	2022-10-21 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-07-29 (manuscriptReceived); 2022-11-16 (publishedOnlineEarlyUnpaginated); 2022-10-22 (manuscriptAccepted)
Publication history :	
First posting date:	16 Nov 2022
DOI:	https://doi.org/10.1111/hex.13650
ProQuest document ID:	2767101860
Document URL:	https://www.proquest.com/scholarly-journals/youth-engagement-mental-health-research/docview/2767101860/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the “License”). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-08-22
Database:	Public Health Database,Publicly Available Content Database

Development of an inflammatory bowel disease (IBD) Patient-Reported Experience Measure (PREM): A patient-led consensus work and ‘think aloud’ study for a quality improvement programme

Sheldon, Elena M [1]
; Lillington, George [1] ; Simpson, Kati [1] ; Gibson, Kirsty [1] ; Chambers, Lucy [1] ; Manfredi D'Afflitto [1] ; Greig, Nancy [1] ; Stearn, Theresa [1] ; Hind, Daniel [1] ; Ainley, Rachel [2] ; Winsor, Gemma [2] ; Ridsdale, Katie [1] ; Totton, Nikki [1] ; Lobo, Alan [3 1] Sheffield Health and Related Research, The University of Sheffield, Sheffield, UK [2] Crohn's &Colitis UK, Hatfield, UK [3] Gastroenterology and Liver Unit, Royal Hallamshire Hospital, Sheffield Teaching Hospitals NHS Foundation Trust, Sheffield, UK

ABSTRACT (ENGLISH)

Background
Patient-Reported Experience Measures (PREMs) are key in improving healthcare quality, but no PREM exists for inflammatory bowel disease (IBD). This study aimed to co-produce a PREM with IBD service users for IBD service evaluation and quality improvement programme.
Methods
A pool of 75 items was drawn from published survey instruments covering interactions with services and aspects of living with IBD. In Stage 1, during two workshops, eight expert service users reduced candidate items through a ranked-choice voting exercise and suggested further items. During Stage 2, 18 previously uninvolved people with IBD assessed the face and content validity of the candidate items in 'Think Aloud' interviews. During two final workshops (Stage 3), the expert service users removed, modified and added items based on the interview findings to produce a final version of the PREM.
Results
Stage 1 generated a draft working PREM mapped to the following four domains: Patient-Centred Care; Quality; Accessibility; Communication and Involvement. The PREM included a set of nine items created by the expert group which shifted the emphasis from 'self-management' to 'living with IBD'. Stage 2 interviews showed that comprehension of the PREM was very good, although there were concerns about the wording, IBD-relevance and ambiguity of some items. During the final two workshops in Stage 3, the expert service users removed 7 items, modified 15 items and added seven new ones based on the interview findings, resulting in a 38-item PREM.
Conclusions
This study demonstrates how extensive service user involvement can inform PREM development.
Patient or Public Contribution
Patients were involved as active members of the research team and as research participants to co-produce and validate a PREM for IBD services. In Stage 1, eight expert service users ('the expert group') reduced candidate items for the PREM through a voting exercise and suggested new items. During Stage 2, 18 previously uninvolved people with IBD (the 'think aloud' *participants*) assessed the validity of the candidate items in 'Think Aloud' interviews as research participants. In Stage 3, *the expert group* removed, changed and added items based on the interview findings to produce a final version of the 38-item PREM. This study shows how service user involvement can meaningfully inform PREM development.

FULL TEXT

BACKGROUND

Crohn's disease (CD) and ulcerative colitis (UC), the main forms of inflammatory bowel disease (IBD), are lifelong debilitating conditions. Symptoms often follow an unpredictable trajectory between active disease and remission, which significantly affects the quality of life and psychosocial functioning.[1] People living with IBD have heterogeneous needs which are often unmet by healthcare services. The views of healthcare professionals and patients differ concerning care priorities and quality.[2] In 2021, IBD UK published a UK-wide survey of 10,222 people with IBD, in which 28% rated their quality of care as only fair or poor.[3] The report identified four areas for change:

improvements in diagnosis and information provision; personalized care and support for self-management; faster access to specialist advice and treatment and effective multidisciplinary team working.

Many IBD quality improvement initiatives take it for granted that organizations must learn from patients.[4] Self-report survey instruments are increasingly used as quality indicators,[5] including in IBD,[6] but not all measures are considered useful or effective.[7] Patient satisfaction measures, which capture whether a patient received care that met their expectations are biased by previous experiences.[8] Patient expectations are influenced by health status, frequency of service interaction and level of dependency on healthcare providers.[7,9] Satisfaction measures lack sensitivity, fail to distinguish between good and bad care and often overrate satisfaction due to gratitude bias.[10] Simply put, high self-reported satisfaction may not correlate with a positive healthcare experience.[9] Patient-Reported Experience Measures (PREMs) represent healthcare quality more accurately.[11] PREMs capture 'what' happened in the care process, 'how' and 'how often'.[8,12] Aspects of patient experience can become targets for service development,[5] and are essential to quality improvement as they provide actionable data based on what matters to patients.[5,7–9,12,13]

Survey instrument development requires a conceptual framework—a set of interlinked ideas that provide an understanding of, or are used to represent a phenomenon.[14,15] Thematic analysis of PREMs from a recently published conceptual framework maps out eight domains of patient experience of services: patient-centred care; quality; integration; accessibility; involvement; communication; discomfort and environment and facilities.[16] These domains strongly align with NHS England, National Clinical Guidelines Centre (NICE) and Institute of Medicine (IOM) definitions of quality in health care, which advocate that care should be patient-centric, safe, effective, efficient and equitable.[17,18] A scoping review identified a range of IBD-specific instruments that measure experience-related concepts, such as patient satisfaction,[6] patient knowledge,[19] patient concerns,[20] self-efficacy[21] and quality of care,[22] but no validated PREM. To fill this knowledge gap, this study aimed to develop a PREM for people with IBDs to support IBD service evaluation.

Patient involvement in the development of survey instruments is recommended by regulators[23]; however, it is rarely well-evidenced, except in a 'cursory and poorly reported' fashion,[24] leading to differences in the understanding of survey items.[25] Patient-led approaches make the instrument development process more accountable and ensure that instruments are relevant, transparent and less subject to ambiguity.[26] We combined patient leadership and qualitative research to ensure patients felt the PREM covered the most important issues (content validity)[27] with a meaningful relationship between the items and what matters to them (face validity).[28]

The PREM was intended for use in a service evaluation alongside the Patient-Activation Measure (PAM) of knowledge, skills and confidence in self-management.[29] Expert patients expressed concern that—for newly diagnosed patients, those on surgical pathways and those in a flare—some of the PAM's items inappropriately implied that disease management was wholly the patient's responsibility. Their response echoed the previous research[30] and policy[31] flagging that some conditions and cases require higher proportions of professional care to self-management, that self-management should be a choice and that poor self-management often arises from low health literacy or overwhelming circumstances. Consequently, *the expert group* developed items which referred to behavioural determinants of 'living well with IBD' instead of 'self-management'. This broadened the instrument's scope to experiences beyond interactions with services. For this reason, in addition to Bull's experience framework,[8,16] we guided questionnaire development using two related conceptual frameworks for understanding how patient experiences might illuminate problems involving behavioural determinants. The COM-B system—which understands behaviour as determined by capability, opportunity and motivation—is a synthesis of 19 behaviour change frameworks,[32] the Theoretical Domains Framework (TDF), a synthesis of 33 theories of behaviour and behaviour change.[33] Its developers describe the TDF as 'an elaboration of the COM-B model' with 'domains of theoretical constructs that map onto the COM-B components and allow for a more detailed understanding of behaviour'.[34] In line with the behaviour change wheel system, we use the COM-B model to talk in broad terms, and the TDF to talk in more narrow terms about behavioural determinants addressed by different PREM items. The COM-B and TDF are relevant because a large part of the experience of living with IBD involves the adoption and maintenance of what

clinical academics would call 'self-management' behaviours,[19–21] although this term is not preferred by *the expert group*, and the TDF is often used to identify barriers to, and facilitators of, desirable self-management behaviour.[35] Our scoping review indicated that 339 items on 20 existing IBD measurement instruments were not symptom measures, nor were they measured constructs to do with capability (*n* = 213 items), opportunity (*n* = 99) and motivation (*n* = 87). The TDF includes a wider range of determinants for successful self-management than the PAM and is often used to identify targets for the improvement of supportive services.[36] Both patients and clinicians have therefore recognized the utility of the COM-B and TDF for areas of living well with IBD where the patient can have more agency in managing the condition.

METHODSOverview

Service users led the development and validation of the PREM across a three-stage process: Stage 1—theme selection and item generation; Stage 2—face and content validity testing and Stage 3—item reduction and scale generation. A group of seven expert service users (*the* 'expert group') led Stages 1 and 3, supported by a *project team* (E. M. S., D. H., A. L.), partners at Crohn's &Colitis UK (R. A., G. W.) and a statistician (N. T.). Crohn's &Colitis UK selected *the expert group* of seven people with IBD (co-authors G. L., K. S., K. G., L. C., M. D., N. G., T. S.) from a range of professional backgrounds using online methods. *The expert group* were recruited via the Crohn's &Colitis UK website, social media and with key contacts who had relevant disease experience using a REC-approved advert. The group were selected based on their previous research experience, professional background and a range of geographic locations. This included, but is not limited to, an editor in survey research; a Crohn's &Colitis UK Health Service Project Manager for Scotland; an IBD UK patient representative; a medical student; a self-employed Organisational Development Coach, Facilitator and Leader and a lay member of the Research Strategy and Funding Committee for Crohn's and Colitis UK. All individuals were known to Crohn's &Colitis UK as having experience in IBD advocacy and had previous voluntary work experience, for example in the readers' panel, implementing self-management projects or as lay members of the charity's committees. Successful applicants were contacted via email to make introductions to *the project team*. Informed consent was received to record all workshops.

In Stage 2, the *project team* interviewed other service users ('the think-aloud group') as research participants to test the face and content validity of the instrument with independent patients.

Stage 1: Theme selection and item generation

The project team identified domains of the patient experience from three sources: (1) PREMs from analogous contexts[8]; (2) survey findings, policy documents and IBD UK standards identified by a Crohn's &Colitis UK exercise summarizing what matters to people with IBD and (3) principles for patient-centred care.[11] *The expert group* considered the appropriateness of the following patient experience domains at an online workshop: patient-centred care; quality; integration; accessibility; involvement; communication; discomfort and environment and facilities.[16] These candidate items and domains were used only as stimuli for discussion.

The expert group recommended the inclusion of items in the PREM about determinants (barriers and facilitators) of service user behaviour. *The expert group* did not have to accept any item, its wording, or any theme. In the event, through discussion amongst themselves, patients re-categorized items in domains most meaningful to them. The academic frameworks were retained to allow for comparison with patient-derived themes and their own 'second-degree' constructs.[37] Based on individuals' availability, four *expert group* members participated in individual one-to-one sessions with the project team to adapt candidate PREM items or create new ones that mapped to the TDF,[33] ensuring relevance to their lived experience while avoiding implications that barriers derived from the patient rather than the service (see Supporting Information: 1). An applied health service researcher with experience in using the TDF (D. H.) and a graduate psychologist (E. M. S.) trained and assisted the four *expert group* members with this mapping exercise. New items mapped to the following TDF domains: Knowledge, Skills and Memory (Psychological Capability); Social Role and Identity; Beliefs about Consequences, and Goals (Reflective Motivation); Emotion (Automatic Motivation) and Social influences and Support (Social Opportunity). *The project team* developed sub-themes, both positive and negative, for each TDF domain, generating a pool of 75 candidate items that reflected good or poor patient experience.

After the initial meeting, *the expert group* completed a ranked-choice voting exercise by allocating points to each of the eight experience domains and 75 candidate items on a spreadsheet. The purpose of the ranking was to allow *the expert group* to anonymously choose candidate domains and items in order of preference, where those receiving the fewest or no votes were eliminated. Individuals from *the expert group* completed the voting spreadsheet independently and the project team collated the results to present at the subsequent workshop. Based on their experience, *the expert group* modified existing items or wrote new ones to reflect anything they considered imprecise, in error or absent. These items were mapped to the top five rated PREM domains: Patient-Centred Care; Quality; Accessibility; Communication and Involvement. *The project team* presented a working PREM based on the results of the voting exercise and the new items based on the TDF. Items were added, improved or removed with reference to Streiner and Norman's criteria: too complex; ambiguous; double-barrelled; jargon; value-laden; negatively worded or too lengthy.[38] *The expert group* chose a Likert scale ranging from 'Not At All' to 'To a Very Large Extent' and wrote a definition for how the term 'Care Team' should be used and understood. The co-produced pool of candidate items was combined to represent a draft working PREM for use in Stage 2.

Stage 2: Face and content validity testing of shortlisted items with a 'think aloud' group

Stage 2 work was conducted by E. M. S. (BSc), a female psychologist with qualitative research experience. To understand face validity[28] (whether items were acceptable to people with IBD), the study used the 'think aloud' protocol,[39] in which participants were asked to say what came into their mind as they completed the survey instrument. A brief unstructured interview followed in which participants were asked to clarify any matters arising during the 'think aloud' interview and to evaluate content validity (the extent to which candidate items cover aspects of care that are important to people with IBD).[27] This included items about how participants found the overall length of the questionnaire, the Likert scale and general formatting. Crohn's &Colitis UK identified a purposive sample of previously uninvolved people with IBD via social media and the charity's website using REC-approved standard advertisement text, inviting patients to opt-in by email. Eligible participants were adults (aged 16 years or over) with IBD (CD or UC) and the capacity to give fully informed consent. Interviews of 30–40 min were conducted by telephone or videoconference. Participants were provided with a £20 shopping voucher as compensation for their time.

Interview transcripts from encrypted recordings were analysed by E. M. S., D. H. and K. R., in NVivo (QSR International) version 12, using the National Centre for Social Research 'Framework' qualitative data analysis method, which involves five stages: familiarization, identifying a thematic framework, indexing, mapping and interpretation.[40] Following Morgan, we understand codes as a system for marking up 'parts of the text that are of special interest' and themes as converting 'codes into core concepts that represent the most important aspects of the results'.[41] In this case, the aim of the study is to ensure that items on the final PREM would reflect the range and content of deductively and inductively derived themes. Our analysis of think-aloud data combined deductive coding (based on Streiner and Norman's criteria[38] for face validity and the conceptual frameworks[8,16,32,33] for content validity) with the inductive development of codes for 'parts of the text of special interest',[41] with content not already covered by the frameworks. In general, these new inductively derived codes were developed during the closing brief unstructured interview (see above) and involved IBD context-specific responses to the face validity of items drawn from other survey instruments. For instance, interviewees felt that some questions presupposed a more predictable disease course than was typical with IBD.

Following Francis et al.,[42] we specified a priori that 12 interviews would be considered analysed before considering saturation, allowing for stopping after every two further interviews if two coders agreed that no new themes were identified. Interviewing ran ahead of analysis which, retrospectively, showed that data saturation was achieved in the first 14 interviews with no substantial different suggestions for question modification or new items thereafter. The final four interviews were included and the sample size (n = 18) was in line with methodological research that shows that 9–17 interviews are generally sufficient for saturation with a fairly homogenous study sample and narrowly defined items.[42,43]

Stage 3: Item reduction and scale generation

Interview findings from Stage 2 were summarised and presented to *the expert* group during two online workshops (see Supporting Information: 7 and 8 for the workshop slides). The *expert group* assessed the importance of each interview finding, agreed on the formatting of the PREM, including item order, and considered new items for inclusion. Where conflicting views between 'think aloud' *participants* and *the expert group* were identified, *the project team* proposed different solutions for each item. Where verbal agreement was not reached during the workshop, *the expert group* independently voted ad hoc by email on their preferred solution and suggested new items where required by interview findings (Supporting Information: 9). Gunning Fog index scores[44] operationalized Streiner and Norman's 'readability' criterion.[38] Items with scores of nine or over were rewritten where possible. The Gunning Fog index is widely used in health research[45] and provides an easily available, free-to-use web tool which *the project team* and *expert patient group* used to experiment with alternative wordings and sentence lengths. The ideal score for readability with the Fog index is 7 or 8; which is the equivalent of Years 8-9 in the UK schooling system and the seventh and eighth grades in the US education system.[44] The project team allowed scores of 9 with exceptions that allowed for contextually specific words with which patients were likely to be comfortable, for example 'colitis' and 'hospital'. At the final workshop, voting exercise results and revisions were presented. Outstanding issues, for example, where items received no majority vote, were resolved through discussion.

RESULTSStage 1: Theme selection and item generation

Of 75 candidate items in the ranked-choice voting exercise, 36 were selected for inclusion, along with three new items (Supporting Information: 2): 'I know how to contact the Care Team between appointments if I need to'; 'It is easy to get the help I need from a member of the Care Team when I need it' and 'I feel able to discuss my mental health with the Care Team if I want to'. In one-to-one sessions, *the expert group* added 14 self-management-based items, to replace the PAM-13. After the addition, removal or modification of items at the second workshop, a 35-item survey draft instrument was developed for use in Stage 2.

Stage 2: Face and content validity

Eighteen participants took part in 'think aloud' interviews, with a median age of 32 (range 26–82) years. Demographics are shown in Table 1. The average length of interviews was 40 min, with a range of 46 min. The shortest interview was 17 min and 47 s; the longest was 1 h and 3 min. Participants highlighted problems with 10 items (Table 2), including items that were too ambiguous, too value-laden, contained jargon or were negatively worded. This later resulted in the removal of two items and the rewording of five items by the expert group.

Table 1 Respondent demographics for PREM Stage 2 'think aloud' interviews (*n* = 18)

Characteristic	Number of respondents (%)
Gender	
Female	12 (66.7)
Male	6 (33.3)
Age	
20–29	4 (22.2)
30–39	9 (50)
40–49	3 (16.7)

50–59	0 (0)
60–69	1 (5.6)
70–79	0 (0)
80–89	1 (5.6)
IBD diagnosis	
Crohn's	14 (77.8)
Ulcerative colitis	4 (22.2)
Ethnicity	
White British	16 (88.9)
Tamil	1 (5.6)
Black Caribbean/British	1 (5.6)
Employment status	
Full-time	9 (50)
Part-time	4 (22.2)
Self-employed	3 (16.7)
Retired	2 (11.1)
Education level	
Postgraduate degree	5 (27.8)
Degree	9 (50)
Secondary education	4 (22.2)
Region in England	
Yorkshire and the Humber	9 (50)
South East	3 (16.7)

North East	2 (11.1)
South Central	1 (5.6)
London	1 (5.6)
South West	1 (5.6)
East Midlands	1 (5.6)

Abbreviations: IBD, inflammatory bowel disease; PREM, Patient-Reported Experience Measure.

Table 2 Interview participant quotes about items in the Stage 2 version of the PREM

Streiner and Norman criteria	Item	Quote	Final outcome for item
Ambiguity	The Care Team knows how I feel emotionally while they are treating me	Well I've never had an emotional sort of complaint talking with the Care Team (Participant 14)	Reword item
	I feel that I have the emotional strength to live with IBD on a day-to-day basis	Day to day [I'm] not sure about that, it depends on what day of the week it is (Participant 2)	Reword item
	My mental health and well-being affects my ability to live with Crohn's or Colitis	It just seems a bit open ended […] I don't really know where you're driving on that one (Participant 6)	Remove item
Value-laden terms	I am able to access sufficient support from the wider IBD community to help me live with Crohn's or Colitis	I felt like it was saying I should be active in the IBD community, putting a little bit of pressure on, when that's not for me, I've tried it and didn't want that contact reminder. (Participant 17)	Reword item
	My Care Team understands what's important to me as an individual (my preferences and priorities in healthcare and beyond)	…but I don't think that's a bad thing. I think they're focused on my disease as they should be as they're experts and they want to get some treatment going to make you feel better, and whatever's important to me in my life doesn't really matter	Reworded
Jargon	I believe that my care and treatment plan will have beneficial effects	Well how do I know that, because I'm not a medical practitioner? So I can only relate that to how I feel, I guess, and my hope (Participant 13)	Reword item

Negatively worded items	My mental health and well-being affect my ability to live with my Crohn's or Colitis	It was at the wrong end of the scale, you expect the 5 s to be the positives and the 1 s to be the negatives, whereas that one was switched round (Participant 1)	Remove item

Abbreviations: IBD, inflammatory bowel disease; PREM, Patient-Reported Experience Measure.

Some items had few objections, particularly those related to mental health and other nonmedical aspects of living with IBD. Participants liked that the PREM covered broader aspects of living with a chronic condition, which is often neglected in clinical encounters.

The length of the PREM was acceptable to participants and most items were considered clear, relevant and easy to understand. Some participants suggested that the term 'Care Team' could be confusing, given the multidisciplinary nature of IBD care. For instance, while some service users are on a surgical pathway and regularly interact with surgeons, dieticians and gastroenterologists, others who are in remission might only see the IBD nurse specialist on an annual basis. Participants recommended removing items where they perceived overlap or repetition, and suggested 11 new items based on aspects of their experience that they felt were missing.

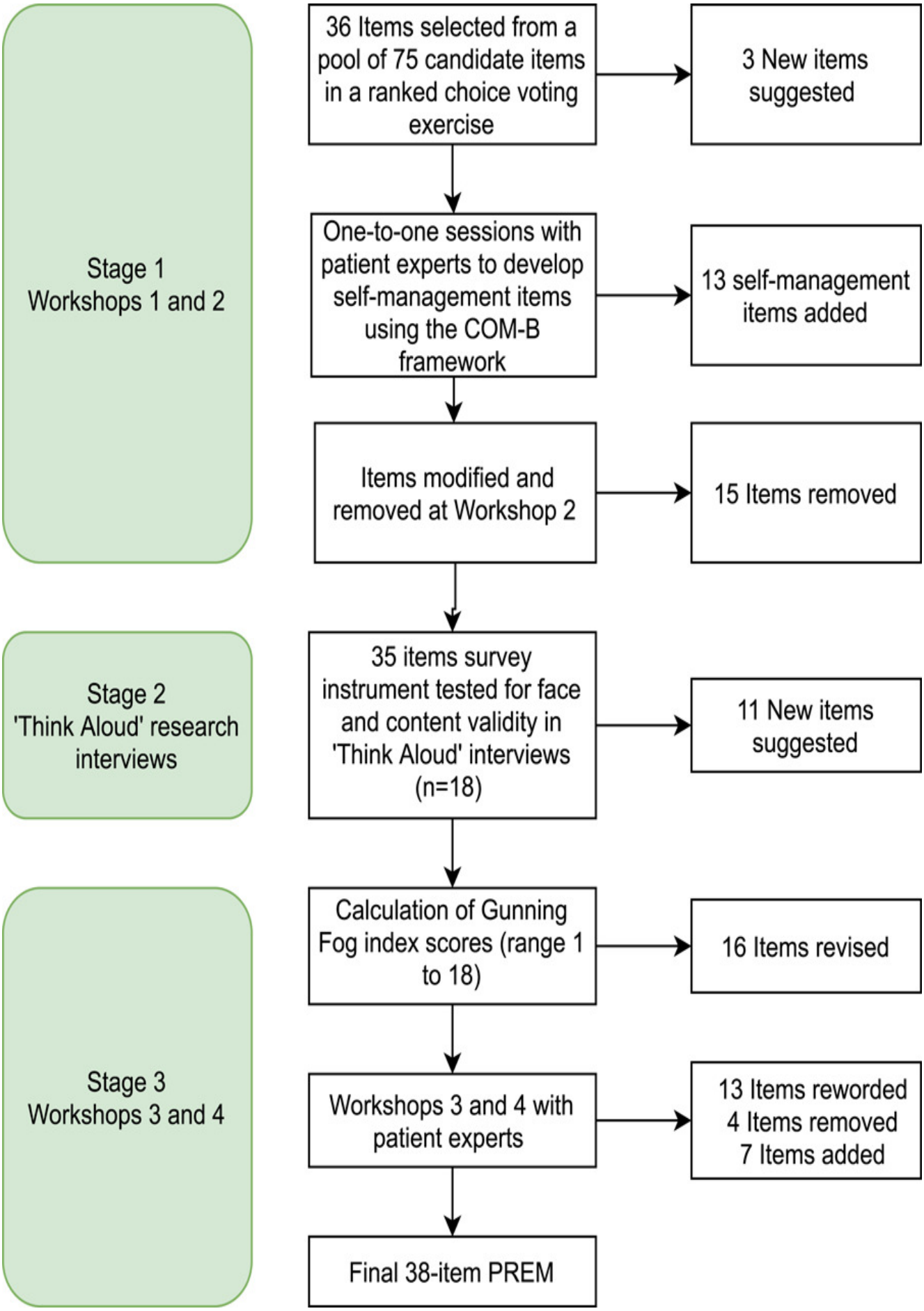
Participants described some items as inappropriate because of the unpredictability of IBD. With reference to Item 34 ('I have a clear picture of where I want to be in terms of my Crohn's or Colitis'), one participant (P04) explained: 'You just never know because all of a sudden you can have a flare out of absolutely nowhere so it's hard to have a clear picture. …I don't think a Crohn's or Colitis journey is a clear one for anyone'.

Some items were considered ambiguous, for example, whether mental well-being items referred to mental health conditions or the impact of living with IBD, as well as difficulties with how emotions fluctuate alongside symptom severity:

With diseases like Crohn's and Colitis, because it can go up and down so much, the ebb and flow of that changes the other stuff around it. […] I definitely know my emotions and mental health change depending on the activity of my disease. (P18)

Stage 3: Item reduction and scale generation

Initial Gunning Fog index scores ranged from 1 to 18 (median 10; see Supporting Information: 3). Sixteen items were revised to improve readability (recalculated score range 11; median 8), allowing for three-syllable words with which IBD service users are familiar, such as 'hospital'. For example, 'My Care Team understands what's important to me as an individual (my preferences and priorities in healthcare and beyond' was reworded to 'The Care Team understands what matters to me (in healthcare and beyond)', reducing the Gunning Fog index score from 18 to 8. At the third workshop, based on the interview findings and Gunning Fog index scores, *the expert group* removed three and reworded seven items (Supporting Information: 4). The Likert scale, layout and item order were finalized. *The expert group* voted to reword five, remove one and include seven new items suggested by the 'think aloud' *participants* (Supporting Information: 5). *The expert group* reworded each of the included items, resulting in the final 38-item PREM (Figure 1). The length of the PREM was deemed appropriate by *the expert group*. The PREM was re-structured by *the expert group* using the following three headings: 'The Care Team'; 'What Matters to Me' and 'Living with Crohn's and Colitis'. Supporting Information: 6 shows how individual items map to the conceptual frameworks which informed the PREM's development. To ensure relevance to decision-makers, we mapped 27 of these items to policy imperatives from the IBD UK standards (Table 3; see Section 4). For example, item 38 'I have a personalised written care plan' was mapped to Statement 7.1 ('A personalised care plan should be in place for every IBD patient, with access to an IBD nurse specialist and telephone/email advice line') as per the IBD UK standards.



Enlarge this image.

Table 3 PREM mapped to IBD UK standards where applicable

Item no.	AWARE-IBD PREM	IBD standards
The Care Team		
1	I know who the different people are in the Care Team looking after me	**Statement 1.1** Patients should be cared for by a defined IBD multidisciplinary team led by a named consultant adult or paediatric gastroenterologist. **Statement 1.9** Clear information about IBD, the local IBD service and patient organizations should be accessible in outpatient clinics, wards, endoscopy and day-care areas
2	I know how to contact the Care Team between appointments if I need to	**Statement 2.4** All patients should be provided with a point of contact and clear information about pathways and timescales while awaiting the outcome of tests and investigations. **Statement 4.2** All patients with IBD should be provided with clear information to support self-management and early intervention in the case of a flare. **Statement 7.1** A personalized care plan should be in place for every IBD patient, with access to an IBD nurse specialist and telephone/email advice line.
3	I can get a response from the Care Team by the end of the next working day when experiencing a flare	**Statement 4.3** Rapid access to specialist advice should be available to patients to guide early flare intervention, including access to a telephone/email advice line with response by the end of the next working day.
4	I feel that the Care Team has enough time for me when I talk to them	
What Matters to Me		

5	I know the person on the Care Team who coordinates my care	**Statement 1.1** Patients should be cared for by a defined IBD multidisciplinary team led by a named consultant adult or paediatric gastroenterologist. **Statement 1.9** Clear information about IBD, the local IBD service and patient organizations should be accessible in outpatient clinics, wards, endoscopy and day-care areas. **Statement 7.1** A personalized care plan should be in place for every IBD patient, with access to an IBD nurse specialist and telephone/email advice line.
6	The Care Team understands the impact my Crohn's or Colitis has on my life	**Statement 3.2** After diagnosis, all patients should have full assessment of their disease, nutritional status, bone health and mental health, with baseline infection screen, to develop a personalized care plan. **Statement 3.3** Patients should be supported to make informed, shared decisions about their treatment and care to ensure these take their preferences and goals fully into account.
7	My concerns are taken seriously when I talk to the Care Team	
8	The Care Team ask how I feel while they are treating me	
9	I feel I can approach the Care Team to discuss any concerns about my treatment and its effects on my life	**Statement 3.3** Patients should be supported to make informed, shared decisions about their treatment and care to ensure these take their preferences and goals fully into account. **Statement 5.4** Patients with IBD being considered for surgery should be provided with information in a format and language they can easily understand to support shared decision-making and informed consent and offered psychological support. **Statement 7.1** A personalized care plan should be in place for every IBD patient, with access to an IBD nurse specialist and telephone/email advice line.

10	The Care Team understands what matters to me (in healthcare and beyond)	**Statement 7.1** A personalized care plan should be in place for every IBD patient, with access to an IBD nurse specialist and telephone/email advice line.
11	I have the confidence to express my needs and concerns with the Care Team	
12	I feel that the Care Team do their best to give me the care I need	**Statement 1.2** Multidisciplinary team meetings should take place regularly to discuss appropriate patients.
13	I am involved in decisions about my care and treatment	**Statement 3.3** Patients should be supported to make informed, shared decisions about their treatment and care to ensure these take their preferences and goals fully into account. **Statement 7.1** A personalized care plan should be in place for every IBD patient, with access to an IBD nurse specialist and telephone/email advice line
14	I feel I have a good relationship with my Care Team	
15	I am treated with dignity and respect by the Care Team	
16	I usually see the same person from the Care Team at each appointment (either face-to-face, telephone or online).	
17	The Care Team offers me appointments in a format that suits me, such as face to face, by telephone or video call	
18	There is good coordination between the different people involved in my care and treatment: (1) Within my Care Team (e.g., doctors, IBD nurse specialists, surgeons, dietitians)	**Statement 5.1** Patients should have access to coordinated surgical and medical clinical expertise, including regular combined or parallel clinics with a specialist colorectal surgeon (paediatric colorectal surgeon where appropriate) and IBD gastroenterologist. **Statement 1.2** Multidisciplinary team meetings should take place regularly to discuss appropriate patients.

	(2) Between my Care Team and other teams in the hospital that I may be in contact with (e.g., rheumatology, dermatology, obstetrics)	**Statement 1.1** Patients should be cared for by a defined IBD multidisciplinary team led by a named consultant adult or paediatric gastroenterologist.
	(3) Between my Care Team and my GP Practice	**Statement 3.6** GPs should be informed of new diagnoses and the care plan that has been agreed within 48 h. **Statement 7.3** Clear protocols should be in place for the supply, monitoring and review of medication across primary and secondary care settings. **Statement 7.5** Any reviews and changes of treatment in primary or secondary care should be clearly recorded and communicated to all relevant parties within 48 h.
	(4) Between my Care Team and other healthcare professionals	
19	The Care Team will refer me to other services if needed (e.g., mental health services)	**Statement 3.2** After diagnosis, all patients should have full assessment of their disease, nutritional status, bone health and mental health, with baseline infection screen, to develop a personalized care plan. **Statement 5.7** Patients and parents/carers should be provided with information about postoperative care before discharge, including wound and stoma care, and offered psychological support. **Statement 6.8** On admission, patients with IBD should have an assessment of nutritional status, mental health and pain management using validated tools and be referred to services and support as appropriate.
20	In general, I am able to understand all the information the Care Team gives me	

21	Thinking about the last time I was given information by the Care Team about my care and treatment: (1) It was given in a way that was easy to understand (2) It met my needs (3) It was relevant to me and my needs (4) I had the opportunity to discuss and ask questions about it (5) I liked the way it was given (e.g., verbal or on paper)	**Statement 1.9** Clear information about IBD, the local IBD service and patient organizations should be accessible in outpatient clinics, wards, endoscopy and day care areas. **Statement 3.3** Patients should be supported to make informed, shared decisions about their treatment and care to ensure these take their preferences and goals fully into account. **Statement 7.1** A personalized care plan should be in place for every IBD patient, with access to an IBD nurse specialist and telephone/email advice line.
22	The Care Team has recommended or directed me to good, reliable information resources, such as charities and the NHS website	**Statement 3.5** Patients should be signposted to information and support from patient organizations. **Statement 7.2** Patients should be supported in self-management, as appropriate, through referral or signposting to education, groups and support.
23	The Care Team informs me about opportunities to take part in research studies and clinical trials	**Statement 1.17** IBD services should encourage and facilitate involvement in multidisciplinary research through national or international IBD research projects and registries.
24	The frequency of my routine appointments is acceptable	**Statement 7.7** All IBD patients should be reviewed at agreed intervals by an appropriate healthcare professional and relevant disease information recorded. **Statement 7.8** A mechanism should be in place to ensure that colorectal cancer surveillance is carried out in line with national guidance and that patients and parents/carers are aware of the process.
25	I am able to easily access toilet facilities at the hospital	**Statement 6.2** Where en suite rooms are not available, inpatients with IBD should have a minimum of one easily accessible toilet per three beds on a ward.

26	I know how to provide feedback on the service, should I want to	**Statement 1.7** Patients and parents/carers should have a voice and direct involvement in the development of the service.
Living with Crohn's or Colitis		
27	I know what care and treatment options are available for my Crohn's or Colitis	**Statement 1.13** Patients should be fully informed about the benefits and risks of, and the alternatives to, immunomodulator and biological therapies, including surgery. **Statement 3.3** Patients should be supported to make informed, shared decisions about their treatment and care to ensure these take their preferences and goals fully into account.
28	I understand how Crohn's or Colitis affects me physically	**Statement 1.9** Clear information about IBD, the local IBD service and patient organizations should be accessible in outpatient clinics, wards, endoscopy and day care areas. **Statement 3.5** Patients should be signposted to information and support from patient organizations.
29	In general, I feel that I can mentally cope with my Crohn's or Colitis	
30	I feel able to discuss my mental health with the Care Team if I want to	
31	I *can do* all the tasks that my care team ask me to do at home (such as manage my diet, lifestyle, treatment)	**Statement 7.1** A personalized care plan should be in place for every IBD patient, with access to an IBD nurse specialist and telephone/email advice line. **Statement 7.2** Patients should be supported in self-management, as appropriate, through referral or signposting to education, groups and support.

32	I *remember to do* all of the tasks that my care team ask me to do (such as take tablets, keep a food diary, etc.)	**Statement 7.1** A personalized care plan should be in place for every IBD patient, with access to an IBD nurse specialist and telephone/email advice line. **Statement 7.2** Patients should be supported in self-management, as appropriate, through referral or signposting to education, groups and support.
33	I am able to keep track of my symptoms	**Statement 7.2** Patients should be supported in self-management, as appropriate, through referral or signposting to education, groups and support.
34	I feel it is important to take an active role in my own healthcare	**Statement 7.2** Patients should be supported in self-management, as appropriate, through referral or signposting to education, groups and support.
35	I get enough support from the people around me to help me live with Crohn's or Colitis (such as friends, family or people at work)	
36	I can access support from the IBD community to help me live with Crohn's or Colitis, if I want to (such as charities, online groups, support groups)	**Statement 1.9** Clear information about IBD, the local IBD service and patient organizations should be accessible in outpatient clinics, wards, endoscopy and day care areas. **Statement 7.1** A personalized care plan should be in place for every IBD patient, with access to an IBD nurse specialist and telephone/email advice line. **Statement 7.2** Patients should be supported in self-management, as appropriate, through referral or signposting to education, groups and support.
37	I believe that my care and treatment will benefit me	**Statement 3.3** Patients should be supported to make informed, shared decisions about their treatment and care to ensure these take their preferences and goals fully into account.
38	I have a personalized written care plan	**Statement 7.1** A personalized care plan should be in place for every IBD patient, with access to an IBD nurse specialist and telephone/email advice line.

Abbreviations: IBD, inflammatory bowel disease; PREM, Patient-Reported Experience Measure.

DISCUSSIONPrincipal findings

IBD service users developed a 38-item survey instrument to capture the experience of healthcare delivery and living with IBD. They selected and rephrased items from other instruments and proposed new items based on their experience and on the interviews with their peers. Their feedback on the PAM-13 resulted in the formulation of new items, based on a more robust framework, reflecting how people with IBD view and manage their condition. This rigorous patient-led process should ensure the PREM's relevance, acceptability and validity and keep service users at the centre of an initiative designed to improve the person-centredness of care.[46] To our knowledge, this is the first and only PREM for IBD healthcare settings, based on Bull et al.s'[8] definition—'what' happened during an episode of care from the patient perspective. Other IBD-specific tools which discuss experience, such as the WE-CARE IBD Score,[6] contain Likert scales which focus on satisfaction rather than the extent to which a phenomenon occurred.

The use of more than one conceptual framework is often relevant in complex situations, where a high-level abstraction stands in for multiple entities that can be understood in multiple ways or approached with different interests and purposes in mind because frameworks never deal with phenomena in their entirety.[15] Our PREM maps IBD-specific experiences to valid constructs representing broader social scientific processes (the TDF), as well as policy imperatives (UK IBD standards) and broader constructs for understanding experience (Bull's framework). Mapping the PREM items to the TDF, IBD UK standards and Bull's framework invites other researchers to use the PREM in IBD quality improvement exercises where a health psychology perspective is desirable. As such, it has wide application in research and service improvement contexts.

Positive feedback from service users from different areas of the United Kingdom, and user-testing with numbers adequate for saturation,[47] provides confidence that this instrument has relevance and utility. Purposive sampling methods from social media and the internet contributed to the homogenous sample in this study in terms of age and ethnicity. Service users were all aged over 25 years, warranting investigation as to how developmentally appropriate [48] its content and language are for younger adults. When translated from English, the cultural appropriateness of the wording and concepts should be assessed for similarity to the source language and how meaningful they are to the speakers in the target population. A further limitation is that the health literacy levels of the 'think aloud' *participants* were not assessed. Future research will assess different forms of reliability and validity in more representative quality improvement cohorts of people with IBD, and investigate the face and content validity of the instrument in young adults.

IBD UK standards provide a consensus of how high-quality care is defined.[3] We have related experience to such quality standards in the mapping exercise, with 28 of the PREM items defined by patients mapping to one or more standards. As such, the PREM can give a clear description, from a patient's perspective, of the extent to which they are actually experiencing these standards in their care. Services might use the responses as robust, patient-reported evidence of meeting the quality standards. Ten items in the PREM are not represented in the IBD UK standards. These include items which cover important issues, including the ability of an individual to mentally cope with their IBD; that they are treated with dignity and respect; that they understand the information given to them; that their concerns are taken seriously; that they have the confidence to express their needs and that appointments are in a format that suits them. Future research and iterations of the IBD standards should consider whether such items should be included within the overall standards of care.

Positive patient experience is associated with higher levels of care quality and clinical effectiveness.[39] Experience measures are increasingly used to complement process, clinical and cost data as evidence of a service's compliance with top-down policy,[8] and used as a bottom-up method of identifying targets for improvement.[9] As such, PREMs have the potential to benefit patients as well as to provide system-wide benefits. However, clinical teams can find experience data removed from day-to-day concerns or difficult to translate into actionable improvements and the use of PREMs without structured staff training is not recommended.[5]

The AWARE-IBD collaboration (doi:10.17605/OSF.IO/H7FCP) is currently collecting PREM data using a co-produced web-based application, allowing service-user completion from home. The purpose of using the instrument

is to make the patient experience visible to healthcare professionals so that they can optimize care at an individual and service level. Future evaluations will look at how PREM data are used to structure clinical encounters. PREM data will also be used in time series analyses to understand the success of patient-led quality improvement efforts.

CONCLUSIONS

This paper describes a patient-led process for the development and validation of a 38-item IBD PREM. We are confident that our sample was adequate to explore content and face validity across two major subpopulations of IBD given the strength of complementary public involvement. However, further validation is required to test the psychometric properties of the PREM and to determine how patient experience data can evaluate the effects of changes in service delivery, particularly for underrepresented patient groups in IBD.

AUTHOR CONTRIBUTIONS

Design, conduct, analysis, interpretation and drafting of this article: Elena M. Sheldon. *Design, analysis and interpretation for this article*: Daniel Hind, Katie Ridsdale, Nikki Totton and Alan Lobo. *Design, conduct and review this article*: Rachel Ainley and Gemma Winsor. *Conduct and interpret the study (as patient experts) and review the article*: George Lillington, Kati Simpson, Kirsty Gibson, Lucy Chambers, Manfredi D'Afflitto, Nancy Greig and Theresa Stearn. All authors read and approved the final manuscript.

ACKNOWLEDGEMENTS

This project is part of the Health Foundation's Common Ambition programme. The Health Foundation is an independent charity committed to bringing about better health and health care for people in the UK. For the purpose of open access, the author has applied a Creative Commons Attribution (CC BY) licence to any Author Accepted Manuscript version arising. This research was funded by The Health Foundation as part of the Common Ambition Programme (FR-000002444).

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data sets used and/or analysed during the current study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

London—Riverside Research Ethics Committee (REC) reference granted a favourable opinion (20/PR/0974). *The expert group members* were collaborators (not research participants), and worked in a service improvement (not research) paradigm, but gave informed consent for workshops to be recorded and preferred wording accurately captured.

DETAILS

Subject:	Quality management; Patient-centered care; User behavior; Patients; Selfmanagement; Voting; Inflammatory bowel disease; Ambiguity; Quality of care; Teams; Workshops; Access; Patient satisfaction; Intestine; Health care; Wording; Validity; Interviews; Quality control; User involvement; Thinking aloud; Quality improvement; Inflammatory bowel diseases; Research &development--R &D; Candidates; Qualitative research; Health services
Business indexing term:	Subject: Quality improvement
Location:	United Kingdom--UK
Identifier / keyword:	inflammatory bowel disease; patient experience; quality improvement

Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	213-225
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-06
Milestone dates:	2022-10-19 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-05-26 (manuscriptReceived); 2022-11-06 (publishedOnlineEarlyUnpaginated); 2022-10-20 (manuscriptAccepted)
Publication history :	
First posting date:	06 Nov 2022
DOI:	https://doi.org/10.1111/hex.13647
ProQuest document ID:	2767101856
Document URL:	https://www.proquest.com/scholarly-journals/development-inflammatory-bowel-disease-ibd/docview/2767101856/se-2?accountid=211160

Comparing an in-person workshop and a postal Delphi survey for involving health service users in health care and health research prioritization

Baumann, Lisa Ann [1]
; Brütt, Anna L [1]
[1] Department of Health Services Research, Carl von Ossietzky University of Oldenburg, Oldenburg, Germany

ABSTRACT (ENGLISH)

Introduction
To involve health service users in health care and health research priority setting, different methods exist. Which method is most suitable under which circumstances is unknown. We compared a postal Delphi survey and an in-person workshop to involve health service users in priority settings for rehabilitative care and research in Germany.
Methods
One hundred and eighty-four former rehabilitants were randomly assigned to a postal Delphi survey (*n*=152) or an in-person workshop (*n*=32). Two hundred and seventy-six employees in rehabilitation were also invited to the Delphi Survey. The methodological comparison refers only to the sample of rehabilitants. Within each method, the participants agreed on the top 10 priorities for practice improvement and research in rehabilitative care. The priorities were compared descriptively. Participants' satisfaction was measured with the Public and Patient Engagement Evaluation Tool. The usability of both methods was compared based on the effort, time and material costs required for implementation.
Results
Seventy-five former rehabilitants and 41 employees in rehabilitation completed both Delphi survey rounds. Eleven former rehabilitants participated in the in-person workshop. Priorities for practice improvement showed a high degree of overlap between both methods whereas research priorities differed greatly. Participants of the in-person workshop felt significantly better prepared, more listened to and more likely to feel that different views on the topics were discussed. Participants of the Delphi survey expressed difficulties in understanding all survey questions. The Delphi survey was more elaborate in preparation and implementation but caused lower material costs.
Conclusion
The differences in research priorities between the two methods could be due to the different samples, differences in the individual interests of participants or differences in the prioritization process. In-person workshops seem to be more appropriate for complex topics, where clarifications of questions and deeper discussions are needed. Delphi

surveys seem to be more suitable for easily understandable topics, larger sample sizes and when fewer financial resources are available.

Patient or Public Contribution

The different study phases were supported by employees in rehabilitation and former rehabilitants (e.g., developing study documents, and interpreting results).

FULL TEXT

INTRODUCTION

There is consensus among politicians and researchers, that health service users need to be more involved in future healthcare decision-making and health research.[1–7] Health service users are either patients, who currently use health services or citizens, who used or will use health services in the future.[6,8,9] Involving both groups in healthcare decision-making and research has been shown to improve the quality and patient-centeredness of the healthcare system.[5,6,10–14] However, public and patient involvement (PPI) is not yet the standard of practice in these areas. Health care and research agendas are often still determined without health service users and rather shaped by individual or commercial interests of healthcare providers, funders or politicians.[15–19] As a result, current health policy and research do not adequately address the interests of citizens and patients.[15,16,20,21]

To make the healthcare system more patient-centred, health service users need to be involved from the very beginning in setting priorities for healthcare and health research.[1,2] Priority setting includes the collaborative identification, prioritization and consensus building of and on the most important areas for future practice improvement and research in health care.[22] For the purposes of this study, areas for practice improvement are defined as areas where direct action is needed to improve health care. Research areas, on the other hand, are areas where unanswered questions exist and need to be answered before practical action can be taken. Only a few examples of prioritization processes for areas for practice improvement in health care are described in the scientific literature.[1,3] Methods used include surveys, focus groups, citizen juries, consensus conferences, negotiated rule-making, deliberative pooling or citizen councils.[1,3] Some countries, like the United Kingdom, have further well-developed standards and procedures for involving citizens and patients in priority settings for health care improvement. One example is the involvement of citizens and patients in the work of Health Watch. The organization operates at the national and local levels and conducts regular surveys of patients on the state of health care and the potential for improving it. The feedback is used to develop agendas for health policy. However, such priority-setting processes are usually not comprehensively documented or published as grey literature and are therefore not fully reflected in the scientific literature.[23] In contrast, several joint prioritization processes are described and published for the field of health research.[1,4,24–28] Methods used for the prioritization of research questions were, for example, group discussions, voting exercises or consensus meetings.[1,4] The most frequent method used is the *priority-setting partnership* of the James Lind Alliance (JLA).[1,4] The JLA is a nonprofit organization from the United Kingdom which brings together patients, healthcare staff and experts to identify future research priorities.[29,30] The participants of such priority-setting partnerships are first asked to name important research questions in their view. For these questions, the state of the evidence is checked. If the research questions are identified to be not sufficiently answered yet, they will be prioritized by the participants in an in-person workshop and summarized in a research agenda.[29]

So far, there is little evidence on how the method chosen for prioritization affects prioritization or which method is most appropriate under which circumstances.[3,7,24,31,32] We could not identify any comparative study of methods used for priority setting in health care and only two for health research prioritization. Elliott et al.[33] compared an in-person and a wiki-inspired nominal group technique to identify the top 10 research priorities for chronic kidney disease. They involved patients, caregivers, healthcare providers and politicians. Lavallee et al.[32] compared online crowd-voting, an in-person focus group using the nominal group technique and a two-round postal Delphi survey to identify research priorities for lower back pain. Only patients were involved. In both studies, no standardized evaluation tool was used for assessing the satisfaction and experience of participants. The ranking of the identified research

questions varied between the different methods compared in both studies. Participants were most satisfied with the in-person format, as they could better express their opinions and contribute meaningfully.

Due to the limited evidence regarding the appropriate use of different prioritization methods, more comparative studies are needed. We, therefore, compared a postal Delphi survey and an in-person workshop (based on the nominal group technique) to involve health service users in setting priorities for rehabilitative care and research. The two methods represent two different and commonly used approaches for health care and health research prioritization. We compared the methods regarding the identified priorities, participants' satisfaction and the implementation effort.

METHOD

Within the study, areas for practice improvement and research questions were first identified and afterwards prioritized.[34] The focus of this paper is on the prioritization phase. For prioritization, participants were randomly assigned to either a postal Delphi survey or an in-person workshop. From each of the two methods, a top 10 list of priorities for practice improvement and research was created. Afterwards, both methods were evaluated and compared.

Before the study was carried out, it was approved by the responsible Ethics Board (number 2019-150). The reporting of this study was guided by the reporting guideline for priority setting of health research (REPRISE).[22]

Setting

The study was conducted in the setting of inpatient orthopaedic and psychosomatic medical rehabilitation in Germany. In Germany, patients receive rehabilitative care financed by the Pension Insurance Fund to maintain or restore their social participation and to prevent early retirement. Insured persons can apply for rehabilitation every 4 years to the responsible Pension Insurance Fund, which decides on the approval. If the patient's employment is at considerable risk, the health insurance fund can oblige the patient to apply for rehabilitation (§51 SGB V). After successful approval, rehabilitants stay for approximately 3 weeks (orthopaedic rehabilitation) or 6 weeks (psychosomatic rehabilitation) in an inpatient rehabilitation centre. In our study, we involved rehabilitants, who attended a rehabilitation at one of the three inpatient rehabilitation centres of the German Pension Insurance Oldenburg-Bremen (DRV OL-HB). The DRV OL-HB is a regional institution of German Pension Insurance in the north of Germany. The rehabilitation centres of the DRV OL-HB have a capacity of about 150 beds each and are focused on the treatment of burnout, depression, personality and behavioural disorders or anxiety disorders for psychosomatic rehabilitation and diseases of the musculoskeletal system, related chronic pain and psychosomatic comorbidities for orthopaedic rehabilitation. After rehabilitation, the patient can receive follow-up care if needed (e.g., outpatient physiotherapy or psychotherapy).[35]

The German rehabilitation sector is already further advanced in terms of patient participation when compared to other healthcare settings.[36,37] The rehabilitation setting is, for example, the only care setting, where patient participation in rehabilitative care is required by law (German Social Code IX). Rehabilitants must be involved in the planning of their own rehabilitation stay or in the evaluation and quality assurance of rehabilitative services. However, it is not standard yet to involve rehabilitants in priority-setting processes for rehabilitative care. To our knowledge, this is the first study in Germany, where different approaches to engaging rehabilitants have been used to identify and prioritize areas for practice improvement and research questions in rehabilitation.

Study designIdentifying themes for prioritization

Areas for practice improvement and research questions were identified through a qualitative cross-sectional survey (paper and online format) between August and November 2020. Former rehabilitants (n = 3872) and employees from rehabilitation centres (n = 235) and the sociomedical and administration departments of the DRV OL-HB (n = 31) were invited to name relevant areas for practice improvement and research questions for orthopaedic and psychosomatic rehabilitation from their perspective. The response rate for the former rehabilitants was 5.7% (n = 217), for the clinicians 13.6% (n = 32) and for the employees of the DRV OL-HB 41.9% (n = 13). Survey responses were analysed according to Mayrings qualitative content analysis using an inductively developed coding system.[38] Because participant responses were rarely related to indication-specific topics, but primarily to topics that affect

rehabilitation as a whole, the responses were analysed collectively. A total of 20 areas for practice improvement and 30 research questions were derived from the responses of the study participants (see Supporting Information: Appendix 1).

PrioritizationDelphi survey

The Delphi survey[39] consisted of two rounds of postal surveys. Participants were asked to assess the importance of each area for practice improvement and each research question on a 5-point Likert-type scale (*very important* to *unimportant*). Participant responses from the first Delphi round (June to July 2021) were descriptively analysed. For each topic, the frequency distribution was calculated across the individual response categories and reflected back to the participants in the second Delphi round by using a bar chart. In the second Delphi round (August to September 2021), participants were asked to assess the same areas for practice improvement and research questions again, considering the average rating of all participants. Finally, the areas for practice improvement and research questions were ranked according to their mean score and standard deviation by the research team.

In-person workshop

The 5-h in-person workshop was based on the nominal group technique[39] and included group discussions on the ranking of the areas for practice improvement and research questions. Two weeks before the workshop started, participants received written information material.

The workshop was conducted on a Saturday in September 2021 and started with a short introduction to the process and clarification of questions. Afterwards, the participants were divided into two teams, one dealing with the areas for practice improvement and one with the research questions. The number of areas for practice improvement and research questions was limited to 20 topics each, as the ranking of more topics in the allotted time was considered impractical. If more topics were available from the identification phase, the respective teams made a preselection of the 20 topics to be discussed and ranked at the workshop.

Within the teams, two small groups were formed to rank the topics. Each small group was supervised by a member of the research team. The participants were asked to first rank the topics by their own. Afterwards, the participants discussed their individual rankings with their group. Each small group was given cards with the corresponding topics, which they were to put in order together. In the case that no consent could be achieved on the ranking, adhesive dots were used. Each participant could distribute his points among the topics (more points meant more relevance). The ranks of the topics from the small group ranking were then summarized into an overall team ranking. The results were discussed by the team members and adjusted in case of discrepancies (consent was thought, if this was not possible, dots were used).

The team rankings were finally presented to all workshop participants. All participants had the opportunity to question the team rankings in case of strong discrepancies and to revise it together until all participants agreed on the final rankings.

Study sample and randomization

Former rehabilitants, who already participated in the identification phase and agreed to participate in the prioritization phase as well (n = 184) were randomly assigned to one of the two prioritization methods by using the statistical programme R. Thirty-two former rehabilitants were invited to the in-person workshop and 152 to the Delphi survey. In addition, 239 employees from the three rehabilitation centres and 37 employees from the DRV OL-HB were invited to participate in the Delphi survey. We could not involve the rehabilitation employees in the workshop because of ethical considerations. Rehabilitants could have felt uncomfortable with meeting their administrators or formal caregivers. Further, due to the dependency of rehabilitants on their administrators (employees of the DRV OL-HB decide on the approval of the rehabilitation applications), rehabilitants could also have felt restricted in their participation. In the context of this study, the comparison of the two prioritization methods refers only to the sample of former rehabilitants to rule out any difference between the methods due to the different stakeholders involved.

EvaluationDifferences in prioritization

Differences in the prioritization of areas for practice improvement and research questions by former rehabilitants are described and compared descriptively between the two prioritization methods. We compared the top 10 priorities

and recorded the degree of overlap.

Participant satisfaction

To evaluate the experiences of the former rehabilitants, we used the German version of the Public and Patient Engagement Evaluation Tool.[40] The questionnaire consists of several 5-point Likert-type items (1 = *don't agree at all* , 5 = *strongly agree*) and free-text fields, asking participants about their general participation experience, their satisfaction with their participation and their opinion on the future use of the results. Additionally, we added questions on the agreement with the final rankings. All former rehabilitants, who participated in the prioritization phase were invited to participate in the evaluation survey. The participants of the in-person workshop filled out the questionnaire directly after the workshop while the participants of the Delphi survey received the evaluation questionnaire together with the ranking results of the second Delphi survey round by post.

Method usability

We assessed the usability of both methods by comparing the implementation effort (necessary steps for preparation), the time and personal resources required for implementation (time of preparation, execution and evaluation) and the approximate material costs.

Statistical analyses

To assess possible differences between the characteristics of the former rehabilitants in both prioritization methods, we used descriptive statistics and calculated a χ^2 test (or Fisher's exact test, if the expected frequency in the cells was less than 5). To test whether the former rehabilitants rated their satisfaction and experience with the process and the results of the prioritization process significantly different between the two methods, we used the Mann–Whitney *U*-test. Written comments of the evaluation survey were qualitatively analysed.

The significance level was set on a two-sided *p*-value <.05 for all statistical tests. Because we performed multiple tests to identify differences between the groups (for participants' characteristics and participants' satisfaction/experience), we adjusted the *p*-value using the Bonferroni correction (*p*-value/number of tests). By this, we accounted for what is called Alpha error accumulation. If multiple tests are performed on the same hypotheses, the greater the probability of obtaining a *p*-value <.05 and thus obtaining a result that is falsely significant. All statistical analyses were carried out in SPSS Version 26.

Patient and public involvement

The project was accompanied by a project advisory board which was involved in the planning and implementation of the study, the development of study material and the interpretation of the results. The board consisted of three clinicians from the participating rehabilitation centres and three employees from the DRV OL-HB. We further aimed to include one rehabilitant, but this did not succeed over the entire duration of the project so that a rehabilitant was present in the phases of interpreting the results from the identification phase and in the preparation of the study materials for the prioritization phase. Four rehabilitants were further involved in the pretesting of the questionnaire for the identification phase.

RESULTS

Of the 152 former rehabilitants who were randomly assigned to the Delphi survey, 92 (response rate: 63%) participated in the first round of the Delphi survey (some letters could not be delivered and are therefore not included in the calculation of the response rates). Seventy-five (81.5%) of these also participated in the second round. Eleven of the 32 invited former rehabilitants (35.5%) participated in the in-person workshop. The sociodemographic data of the former rehabilitants who participated in the Delphi survey and the in-person workshop and the calculation of significant differences between both groups are shown in Table 1. Besides the former rehabilitants, 46 of the 239 invited clinicians (19.3%) and 9 of the 37 invited employees from the DRV OL-HB (24.3%) participated in the first Delphi round. Of those, 33 clinicians (82.5%) and 8 employees of the DRV OL-HB (88.9%) participated also in the second survey round.

Table 1 Characteristics of former rehabilitants who participated in the Delphi survey and the in-person workshop

Note: If the expected frequency in the cells was less than 5, Fisher's exact test was calculated instead of χ^2. The value in bold are those where a significant difference was found between the groups compared in the study. *

p <.007 (Bonferroni adjusted p-value [p-value of .05/7]).

Comparison of the top 10 priorities for practice improvement and research

The identified top 10 priorities for practice improvement and research for orthopaedic and psychosomatic rehabilitation by former rehabilitants for each method are shown in Tables 2 and 3.

Table 2 Comparison of top 10 areas for practice improvement identified by former rehabilitants between the in-person workshop and Delphi survey

Top 10 priorities for practice improvement			
Top 10 in-person workshop	Rank Delphi survey	Top 10 Delphi survey	Rank in-person workshop
Implementation of a holistic therapy approach	1	Implementation of a holistic therapy approach	1
Thorough initial examination	3	Individualize rehabilitative treatment	8
Participation of rehabilitants in rehabilitation	5	Thorough initial examination	2
Quality assurance of rehabilitation treatment	9	Preparation of rehabilitation stay	17
Application process for rehabilitation	7	Participation of rehabilitants in rehabilitation	3
Support of rehabilitants following rehabilitation	11	Discharge management	15
Making rehabilitation sustainable	20	Application process for rehabilitation	5
Individualize rehabilitative treatment	2	Patient education	9
Patient education	8	Quality assurance of rehabilitation treatment	4
Re-organization of the rehabilitation stay	12	Re-organization of already offered therapies and measures in rehabilitation centres	13

Table 3 Comparison of top 10 research questions identified by former rehabilitants between in-person workshop and Delphi survey

Top 10 priorities for research

Top 10 in-person workshop	Rank Delphi survey	Top 10 Delphi survey	Rank in-person workshop
How do insureds rate access to rehabilitation measures?	24	What influence does the rehabilitant's motivation have on the treatment outcome?	6
What tasks can case managers take on to support the rehabilitant?	26	How can the cooperation between the pension insurance, the healthcare insurance and the employment agency be improved to avoid waiting times and gaps in care?	3
How can the cooperation between the pension insurance, the healthcare insurance and the employment agency be improved to avoid waiting times and gaps in care?	2	How often should rehabilitation be performed to achieve sustainable treatment success?	13
How much time should optimally elapse between an inpatient hospital stay and the subsequent rehabilitation?	12	Which factors are relevant for rehabilitant's satisfaction in rehabilitation?	Excluded during preselection
How do one-on-one therapy and group therapy differ in terms of treatment outcome?	22	What influence does the removal of rehabilitants from their home environment have on the treatment outcome?	9
What influence does the rehabilitant's motivation have on the treatment outcome?	1	What are the main factors that determine the success of treatment in rehabilitation?	Excluded during preselection
How can rehabilitation be designed to meet the individual needs of rehabilitants?	20	What influence does the obligation to file an application for rehabilitation in the case of a significant risk to gainful employment (allocation via §51 SGB V) have on the treatment outcome and rehabilitants satisfaction?	Excluded during preselection
How can early steering, especially into psychosomatic rehabilitation, be achieved?	12	How do an endurance-oriented and a strength-oriented training programme differ in terms of treatment outcome for back pain?	Excluded during preselection
What influence does the removal of rehabilitants from their home environment have on the treatment outcome?	5	What is the effect of regular rehabilitation (every 4 years) on the course of illness and ability to work?	16

How can the effectiveness of follow-up services following rehabilitation be increased?	19	How long should rehabilitation ideally last?	18

A comparison of the ranks for the areas for practice improvement between both methods revealed a large proportion of overlap. Seven areas were present in both top 10. The first rank was the same for both methods ('Implementation of a holistic therapy approach') and only a few areas differed considerably in their ranks. In both rankings, it gets clear that participants saw a need for more individualized rehabilitative interventions as well as increased education and participation of rehabilitants about and in their rehabilitation.

In contrast, the priorities for research differed clearly between the two methods. Only three research questions were present in both top 10 and the difference between the ranks was quite high for most of the questions. The former rehabilitants in the in-person workshop saw a priority need for research in the analysis and further development of the rehabilitation system with a focus on access to rehabilitation and the support of rehabilitants during and after rehabilitation. Former rehabilitants in the Delphi survey also saw a need for research regarding the analysis and further development of the rehabilitation system, but with a focus on structural issues such as the duration and frequency of a rehabilitation stay. Further, questions regarding the theoretical and methodological foundation of rehabilitation (e.g., relevant factors for patient satisfaction) were more relevant to the former rehabilitants in the Delphi survey.

Participants satisfaction

Fifty-four former rehabilitants from the Delphi survey (response rate: 72%) and all former rehabilitants from the in-person workshop (n = 11) participated in the evaluation survey. The sociodemographic characteristics of the participants can be found in Table 4.

Table 4 Sociodemographic data of participants in evaluation survey

	Participants of Delphi survey (n = 54)	Participants of in-person workshop (n = 11)
Gender		
Male	46.3%	54.5%
Female	51.9%	45.5%
Missing	1.9%	0%
Age		
18–29	0%	0%
30–39	3.7%	0%
40–49	20.4%	18.2%
50–59	53.7%	45.5%

60–69	22.2%	36.4%
Missing	0%	0%
Education		
Without school-leaving qualification	1.9%	0%
Secondary school diploma	75.9%	63.7%
Technical baccalaureate/high school diploma	16.7%	36.4%
University of applied sciences/university	5.6%	0%
Missing	0%	0%
Indication		
Orthopaedic	44.4%	36.4%
Psychosomatic	33.3%	45.5%
Both	22.2%	18.2%
Missing	0%	0%
Years with disease		
<1	0%	0%
1–5	24.1%	27.3%
6–10	16.7%	54.5%
11–15	16.7%	0%
>15	42.6%	18.2%
Missing	0%	0%
Number of rehabilitations attended		
1–2	55.6%	63.6%

3–4	33.3%	18.2%
5–6	5.6%	0%
>6	5.6%	18.2%
Missing	0%	0%
Satisfaction with own rehabilitation		
Very satisfied	37%	27.3%
Satisfied	48.1%	27.3%
Neither nor	11.1%	9.1%
Not satisfied	0%	36.4%
Not satisfied at all	0%	0%
Missing	3.7%	0%

In both groups, most of the former rehabilitants (strongly) agreed with all statements. The statistical results of the pairwise comparisons between both groups are summarized in Table 5. There was no significant difference in the overall satisfaction with the priority-setting exercise as well as the satisfaction with the final results. However, former rehabilitants from the in-person workshop significantly felt better prepared for the discussion on the topics (*Median workshop* = 5, *Median Delphi* = 4, *U* = 163.5, *p* = .002, *moderate effect* [*r* = .38]) and more heard in their views (*Median workshop* = 5, *Median Delphi* = 4, *U* = 155.5, *p* = .001, *moderate effect* [*r* = .40]).[41] The workshop participants further significantly more agreed, that the participants represented different perspectives on the topics discussed (*Median workshop* = 5, *Median Delphi* = 4, *U* = 125.5, *p* = .001, *moderate effect* [*r* = .44]) and that a range of views on the topics discussed were shared (*Median workshop* = 5, *Median Delphi* = 4, *U* = 132.5, *p* = .001, *moderate effect* [*r* = .41]).

Table 5 Statistical results of the evaluation study

Statements in evaluation survey	Median Delphi (IQR)	Median workshop (IQR)	Mann–Whitney *U* (*p*-value)	*z*	*r*
I had a clear understanding of the purpose of the Delphi survey/workshop.	4 (0)	4 (1)	258.0 (*p* = .385)	−0.869	
The supports I needed to participate were available.	4 (0)	4 (1)	271.5 (*p* = .562)	−0.580	

I had enough information to contribute to the topic being discussed.	4 (0)	5 (1)	**163.5*** (***p* = .002**)	−3.042	.38
I was able to express my views freely.	4 (1)	5 (1)	197.5 (*p* = .059)	−1.886	
I feel that my views were heard.	4 (0)	5 (1)	**155.5*** (***p* = .001)**	−3.220	.40
A wide range of views on the topic discussed was shared.	4 (0)	5 (1)	**132.5*** (***p* = .001)**	−3.341	.41
The individuals participating in the Delphi survey/workshop represented a broad range of perspectives on the topic.	4 (0)	5 (1)	**128.5*** (***p* = .001)**	−3.457	.44
I think that the Delphi survey/workshop achieved its objectives.	4 (0)	5 (1)	167.5 (*p* = .012)	−2.516	.32
I am confident the input provided through the Delphi survey/workshop will be used by the German Pension Insurance Oldenburg-Bremen.	4 (0)	4 (1)	272.0 (*p* = .753)	−0.315	
I think the input provided through the Delphi survey/workshop will make a difference in the work of the research group and the German Pension Insurance Oldenburg-Bremen.	4 (0)	4 (1)	250.0 (*p* = .427)	−0.794	
As a result of my participation in the Delphi survey/workshop, I am better informed about the need for practice improvement and research in rehabilitation.	4 (0)	4 (1)	224.5 (*p* = .098)	−1.652	
Overall, I was satisfied with this Delphi survey/workshop.	4 (0)	4 (1)	190.5 (*p* = .024)	−2.260	.28
This Delphi survey/workshop was a good use of my time.	4 (1)	5 (1)	216.0 (*p* = .123)	−1.541	
I agree with the list created for the priority areas for practice improvement.	4 (1)	4 (1)	207.0 (*p* = .088)	−1.704	
I agree with the list created for the priority areas for research.	4 (1)	5 (1)	187.0 (*p* = .044)	−2.011	.25

I am satisfied with the results of the Delphi survey/workshop.	4 (0)	5 (1)	175.5 (p = .014)	−2.458	.30

Note: Effect size was only calculated for significant differences. The values in bold are those where a significant difference was found between the groups compared in the study. Abbreviations: IQR, interquartile range; *r*, Pearson correlation coefficient. *

p <.003 (Bonferroni adjusted p-value [p-value of .05/16]).

The analysis of the written comments showed, that some Delphi survey participants had difficulties in understanding and answering the survey questions, especially in the first Delphi round.

Methods usability

Detailed information on the usability of both methods is summarized in Table 6. The effort for the preparation, implementation and analysis of the results was higher for the Delphi survey. The in-person workshop was also time-consuming in the preparatory phase, but directly usable results were available afterwards.

Table 6 Compare of usability for in-person workshop and Delphi survey

	In-person workshop	Delphi survey
Effort for preparation and implementation	Development and dispatch of information material Organization of catering Development of hygiene concept (necessary because of Covid-19 pandemic) Organization of materials (cards with research questions and areas for practical improvement for sorting, moderation materials like colourful cates, glue dots)	Development and dispatch of three questionnaires Manual transfer of participants responses into a digital data format by one person and controlled by a second person Initial analysis of first-round responses and analysis of final results after second round
Time frame	2 months (planning and preparation in August, Implementation and analysis of results in September)	8 months (preparation from April to June, first survey round from June to July, analysis of initial analysis in July, second survey round from August to September, final analysis in October, evaluation survey in November)
Approximate personal resources	One person (research associate) with 30 h/week for 2 months and four persons (one research associate, one academic researcher, two student assistants) for 9 h each at the workshop day	One person (research associate) with 30 h/week for 8 months and one person (student assistant) with 9 h/week for 1 month

Approximate material costs	Compensation for participants: 100€ per person, in total 1100€ Catering for 11 participants: 561€ Printing information material: 50€ Moderation material: 32€ Letter postage for sending and return for date request and workshop information material: 155€ Envelopes for sending study material to participants: 11€ Printed return envelopes: 5€	Compensation for participants: none Printing questionnaires (2 Delphi rounds and evaluation survey): 513€ Letter postage for sending and return of study documents to former rehabilitants: 671€ Envelopes for sending study material to former rehabilitants: 48€ Printed return envelopes: 22€
	Total: 1898€	*Total*: 1254€

Note: Only the largest cost items are included in the costs, smaller amounts are not listed, costs were rounded on full euro amounts.

From the point of view of the participants, however, the effort was reversed. Here, the Delphi survey took significantly less time than the in-person workshop, for which, including travel to and from the workshop, more than half a day had to be taken. The Delphi survey, on the other hand, took about 30–40 min per survey round and could also be completed flexibly in terms of time (within the specified return period).

The in-person workshop further incurred higher material costs due to the expense of catering and compensation for participants.

DISCUSSION

We conducted a comparative study of two methods used in health care and health research prioritization. Our study provides important insights into how different prioritization methods work and when to use which method. In the following, we first look at the differences in question prioritization between both methods. Afterwards, participants' satisfaction and experience and the usability of both methods are discussed. Finally, further research need is outlined.

Comparison of the top 10 priorities for practice improvement and research

The differences we observed in the ranking of research questions between different prioritization methods confirm the results identified in the studies of Elliott et al.[33] and Lavallee et al.[32] for the setting of rehabilitative care. The first, apparent explanation for the prioritization differences between the two prioritization methods in our study is the different samples in the methods. Besides former rehabilitants, also rehabilitation employees participated in the Delphi survey (with a share of the total Delphi sample of 37% in the first survey round and 35% in the second survey round). Even when we only compared the final rankings of the former rehabilitants, the prioritization of the former rehabilitants, who participated in the Delphi survey, could have been influenced by the opinions of the clinicians and employees of the DRV OL-HB (after the first Delphi survey round, participants could change their first assessment of each topic after considering the average assessment of all participants).

In the studies of Elliott et al. and Lavallee et al., it was further unclear whether the differences in prioritization were due to the different priority-setting procedures or to the different characteristics of study participants. By randomly assigning the former rehabilitants to one of the two prioritization methods, we tried to control for sociodemographic differences between the two study samples we compared in our study. However, we identified that our study participants differed significantly in their satisfaction with the rehabilitation they received, with workshop participants less satisfied. This could be due to the randomization process itself, which may have randomly resulted in unequal

samples. It could also be due to self-selection by participants. Individuals, who were more dissatisfied with their rehabilitation stay, might have been more likely to attend the in-person workshop to take advantage of the opportunity to share personal experiences with other rehabilitants and researchers in the field face-to-face. The difference between the groups in satisfaction with their own rehabilitation may have led to the different research priorities in the two methods.

However, because only the prioritization of research questions differed greatly between the two methods (if the differences were due to differences between the study groups, one would expect this to be the case for both areas, research and practice) and previous study results also found differences in the prioritization of research questions between different methods (despite more comparable study samples), we do not believe that the prioritization differences we observed were only due to differences in our study samples. What might also be important are other individual beliefs and interests that we did not collect within our survey. Compared to areas for practice improvement, where former rehabilitants are likely to draw on similar experiences through their rehabilitation stay, research questions might be more driven by individual interests which rehabilitants do not share. It is equally likely that the process for prioritization mostly influenced the decisions of the former rehabilitants. Previous studies suggest, that individuals are more likely to change their opinions on the relevance of different healthcare topics after face-to-face discussions.[32,33,42] Especially in the case of research questions, which are initially more difficult to understand than areas for practice improvement, a face-to-face conversation might have had a major impact on ranking decisions. Therefore, the method for prioritization must be carefully selected for the appropriate purpose.

Comparison of participants' satisfaction

As part of our study, we compared participant satisfaction between two prioritization methods using a standardized assessment tool for the first time. We identified that average participant satisfaction was high in both groups (no significant differences), differing only in the range of *satisfied* and *very satisfied*. However, workshop participants rated their experience in some points significantly better. They felt better prepared and listened to, and were more likely to feel that different opinions on the issues were discussed. These results are in line with previous study findings.[32,33] It might reflect that participants of face-to-face methods value the personal exchange of different views with other participants and the opportunity for a direct and deeper discussion on the issue. Participants of the in-person workshop were able to respond directly to what other participants were saying, which could have led to the feeling of being heard more and hearing different opinions on the topics. The workshop participants further received an introduction to the research project and the identified priorities for practice improvement and research. In the course of this, the opportunity of clarifying questions on the course and content of the workshop existed. This may have resulted in workshop participants feeling more informed about the topics discussed, while former rehabilitants who participated in the Delphi survey expressed some difficulties in understanding the questions. Even when the possibility to contact the research team in case of upcoming questions during answering the Delphi survey existed—this was associated with effort and time delays in filling in the survey for the former rehabilitants and was not comparable with a real-time face-to-face discussion.

In conclusion, in-person workshops seem to be preferred by participants and might also lead to more reliable results as it can be ensured that the task and discussed topics are understood correctly by the participants.[33] Considering this, an in-person workshop might be more suitable for complicated topics where the opportunity for direct clarification of questions is needed whether a Delphi survey could be used when the topics for discussion are already comprehensively known by participants.

Comparison of method usability

Advantages in the usability of the in-person workshop were the lower effort, fewer personal resources and shorter time needed for preparation and implementation when compared to the Delphi survey. Therefore, an in-person workshop could be especially suitable, when results need to be available quickly (e.g., in health policy decision-making) and when few personal resources are available.

The material costs for the in-person workshop were higher than for the Delphi survey. In the literature review of Mitton et al.,[3] face-to-face interventions were also associated with higher costs. Material costs certainly depend on

the number of participants involved. But if the number of participants in both methods would have been the same, the material costs for the in-person workshop would still have been higher due to the costs for catering and participant compensation. A Delphi survey might therefore be more suitable when fewer financial resources are available.

Besides the higher resource need, the in-person workshop we conducted had further disadvantages. It was held at a specific time and place and was therefore not feasible for some individuals due to work or personal commitments or for individuals with limited mobility. Some of the invited former rehabilitants couldn't participate because the date or time did not suit them. This could lead to a selection bias of participants and might limit the generalizability and expressiveness of the results. Such problems may be solved by conducting the workshop as an online meeting or by conducting an asynchronous discussion among participants on an online platform. However, when implementing the workshop as an online meeting, this could cause other barriers to participation, as access to the internet and a computer is required. An asynchronous discussion, which allows more time flexibility for the participants, on the one hand, makes a moderated and controlled discussion on the other hand more difficult and requires more time.

Another problem of our in-person workshop might be the ranking procedure, which was mainly based on face-to-face discussions between participants. This carries the risk that a few individuals dominate the discussion, which would lead to the unequal representation of individual opinions in the results. The nominal group technique, which guided the methodology of our workshop, has been designed to minimize this risk by having individual and group work phases and involving each participant in the discussion. Within our evaluation survey, we could not find evidence that some individuals felt, that their opinion was insufficiently considered in the final ranking. However, it still remains a risk when a discussion between participants is used for setting priorities.

Where the workshop showed problems, the strengths of the postal Delphi survey became clear. The Delphi survey could be filled in by participants at a convenient time for them within the survey phase. It further saved travel time, which might be especially important when participants from different geographical regions or nonmobile individuals should be involved (this might also be solved by the implementation of the workshop as an online format). This might lead to higher response rates like was the case in our study. Another strength of the Delphi survey is the possibility to involve a larger sample.

Further research need

As there are still very few studies comparing the influence of different methods on prioritization, more research needs to be conducted on this issue. To assess, what difference in prioritization can be attributed to the method itself and what different characteristics of study participants, the same method can be conducted for the same topics but with different participants. To assess the influence of relevant contextual factors (e.g., country, health care setting), (quasi-)experimental study designs are necessary, where the same method is implemented under different circumstances.[1,33,43]

LIMITATIONS

Our results should be interpreted considering some limitations. First, we were not able to involve clinicians and employees from the DRV OL-HB in the in-person workshop due to ethical considerations. This limited the comparison between both methods since it is more difficult to understand to what extent differences in prioritization are due to differences in the methods or the samples.

Second, our results are limited in their generalization due to the small number of participants. Especially at the in-person workshop, we had fewer participants than we would have liked. The date of the workshop was not suitable for everyone and there were still contact restrictions because of the Covid-19 pandemic, so it was also not possible to involve a much larger sample. However, as our results are mostly in line with previous study findings, we are confident that they are at least partly generalizable.

Third, we were not able to fully assess the representativeness of our study sample in the prioritization phase. Ideally, the characteristics of the study participants could have been compared to the entire sample to better assess, if differences in the participant characteristics are caused by participants' self-selection. However, because the first study phase was conducted anonymously, it was not possible to link the sociodemographic data to the contact

information of those participants, who agreed to participate in the second phase of our study.
Fourth, since some Delphi survey participants expressed difficulties in understanding the questions/tasks in the first survey round, it would have been useful to pilot the questionnaire before the survey. We involved only one rehabilitant in the preparation of the study material for the prioritization phase, which may not have been sufficient retrospectively. This should also be considered when assessing the usability of both methods, as an extensive pilot phase of the questionnaire would increase the effort required for a Delphi survey.
Finally, the method we used to rank the topics within the Delphi survey had an influence on our results. We used the mean and standard deviation calculated from participants' assessments, as this is the most commonly used method in the literature.[44–50] However, a standardized approach for ranking is missing and the topics could have also been ranked based on the median or percentage of people who rated the topic as very important or important.[51,52] We tested different methods to rank the topics before we decided on the mean and standard deviation as the decisive factors. The methods resulted in approximately the same top 10, but different subject rankings. Thus, for selecting future priorities and for comparing priorities between different methods, it seems important not to focus too much on the exact ranking, but to consider all of the top 10 issues.

CONCLUSION

Priorities for practice improvement differed slightly between the in-person workshop and the Delphi survey. Large differences in the prioritization were observed for research questions. The former rehabilitants who participated in the in-person workshop rated their experience significantly better regarding their preparation, the feeling of having been heard and the opinion that different views were expressed. Former rehabilitants who participated in the Delphi survey mentioned difficulties in understanding the survey questions. While considering this and the comparison of the usability of both methods, in-person workshops seem to be more appropriate for complex topics and issues on which results need to be available quickly and when few personal resources are available. Delphi surveys instead are more suitable for easily understandable topics, for involving a larger sample and when fewer financial resources are available.
Our results can help organizers of prioritization events with health service users to decide, which method is most appropriate for their purposes and circumstances. More studies comparing different methods for priority setting in health care and health research are needed to develop clear recommendations on when to use which method.[3,7,24,31,32]

AUTHOR CONTRIBUTIONS

Anna L. Brütt contributed to the development of the study design. Lisa Ann Baumann and Anna L. Brütt contributed to the development of the Delphi survey questionnaire and planned and conducted the in-person workshop. Lisa Ann Baumann did the statistical analyses and wrote the manuscript. Anna L. Brütt supervised the process of manuscript preparation and edited the manuscript. Both authors approved the final version of the manuscript.

ACKNOWLEDGEMENTS

We want to thank PD Dr. med. Aike Hessel and Dr. Natalie Schüz for their support in the study design and the preparation of the questionnaire. The study was embedded in a project funded by the German Pension Insurance Oldenburg-Bremen.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

DETAILS

Subject:	Health care policy; Priorities; Health care; Rehabilitation; Health services; Orthopedics; Workshops; Politicians; Conferences; Patients; Questions; Appropriateness; Decision making; Medical research; Citizens; Surveys; Methods; Polls &surveys; Ratings &rankings; Topics
Location:	United Kingdom--UK; Germany
Identifier / keyword:	participation; PPI; priority setting; rehabilitation
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	199-212
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-08
Milestone dates:	2022-09-13 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-07-15 (manuscriptReceived); 2022-11-08 (publishedOnlineEarlyUnpaginated); 2022-10-19 (manuscriptAccepted)
Publication history :	
First posting date:	08 Nov 2022

Embedding health literacy research and best practice within a socioeconomically and culturally diverse health service: A narrative case study and revised model of co-creation

Muscat, Danielle M [1] ; Mouwad, Dana [2] ; McCaffery, Kirsten [1] ; Zachariah, Dipti [3] ; Tunchon, Lyn [4] ; Ayre, Julie [1] ; Nutbeam, Don [5] [1] Sydney Health Literacy Lab, School of Public Health, Faculty of Medicine and Health, The University of Sydney, Sydney, New South Wales, Australia [2] Western Sydney Local Health District, Integrated and Community Health, Health Literacy Hub, Sydney, New South Wales, Australia [3] Western Sydney Local Health District, Integrated and Community Health, Multicultural Health, Sydney, New South Wales, Australia [4] Western Sydney Local Health District, Integrated and Community Health, Child and Family Health, Sydney, New South Wales, Australia [5] School of Public Health, Faculty of Medicine and Health, The University of Sydney, Sydney, New South Wales, Australia

ABSTRACT (ENGLISH)

Background

Health literacy interventions and research outcomes are not routinely or systematically implemented within healthcare systems. Co-creation with stakeholders is a potential vehicle through which to accelerate and scale up the implementation of innovation from research.

Methods

This narrative case study describes an example of the application of a co-creation approach to improve health literacy in an Australian public health system that provides hospital and community health services to one million people from socioeconomically and culturally diverse backgrounds. We provide a detailed overview of the value co-creation stages and strategies used to build a practical and sustainable working relationship between a University-

based academic research group and the local health district focussed on improving health literacy.

Results

Insights from our experience over a 5-year period informed the development of a revised model of co-creation. The model incorporates a practical focus on the structural enablers of co-creation, including the development of a Community of Practice, co-created strategic direction and shared management systems. The model also includes a spectrum of partnership modalities (spanning relationship-building, partnering and co-creating), acknowledging the evolving nature of research partnerships and reinforcing the flexibility and commitment required to achieve meaningful co-creation in research. Four key facilitators of health literacy co-creation are identified: (i) local champions, (ii) co-generated resources, (iii) evolving capability and understanding and (iv) increasing trust and partnership synergy.

Conclusion

Our case study and co-creation model provide insights into mechanisms to create effective and collaborative ways of working in health literacy which may be transferable to other health fields in Australia and beyond.

Patient and Public Contribution

Our co-creation approach brought together a community of practice of consumers, healthcare professionals and researchers as equal partners.

FULL TEXT

INTRODUCTION

Improving health literacy is an international policy priority, grounded in evidence that links lower health literacy with poorer health outcomes.[1] Most health literacy policies recognize that the responsiveness of the health system needs to be improved[2] and many identify the need for greater implementation of evidence-based practices and research.[3] However, research outcomes are not routinely or systematically implemented within healthcare systems.[4–7] This is true across all health research domains, including for research on health literacy.[8,9] This failure to systematically translate, implement and deliver evidence-based improvements in outcomes for patients and the community has attracted attention to the way we conduct health services research, its perceived relevance and the potential for practical implementation by health professionals and health organizations. This, in turn, has led to advocacy for more effective and consultative partnerships at every stage of the research process, from initial idea generation to implementation.[10,11]

This approach to the collaborative generation of knowledge by academics working alongside stakeholders from other sectors is often referred to as co-creation.[12] The concept of co-creation is grounded in the belief that proactive linkage and exchange builds bridges between researchers and the intended users of research (health professionals, patient and community members), and develops the mutual trust on which successful collaboration depends.[12] Answering research questions that have been generated through partnership can lead to interventions that are closer to consumer needs and preferences, are ‘owned’ by health professionals and more likely to be sustained.[12] In this way, co-creation is a potential vehicle through which to accelerate and scale-up the implementation of innovative research and support longer-term sustainability of a change in practice.

Although there is a growing literature about co-creation and its contribution to research translation, there remain relatively few working examples. In the domain of health literacy, a small number of studies report on the ‘co-creation’ of solutions to improve the design and navigation of health services and written materials with patients and consumers.[13–15] However, these examples are narrowly focused on one aspect of health service delivery, and often fail to engage the full spectrum of end users of the research including clinicians, health service managers and other key stakeholders within healthcare systems. These existing studies also appear to be researcher-driven with consumer involvement often limited to market research and testing. Few exhibit the key features of co-creation including involvement and input from the strategic partners throughout the entire research journey (i.e., from the development of the research questions to the implementation and evaluation phases).[16]

This paper presents a model developed over a 5-year period to build a practical and sustainable working relationship between a University research group and a local health district working across clinical and community services and

focussed on improving health literacy (i.e., the ability of individuals to gain access to, understand, appraise and use information in ways which promote and maintain good health[17]). Previous research has consistently shown that low health literacy has a negative impact on healthcare access,[18] physician–patient communication,[19] medication adherence[20] and effective healthcare use[18]; and that organizational factors including clinical communication have a major role in easing or complicating health for people with limited health literacy. Communication between clinicians, patients and carers is a core business in healthcare systems but is often done poorly.[21,22] To address this, we have sought to develop a working relationship to support enhanced clinician communication, organizational health literacy responsiveness and improved consumer health literacy that is led by health system priorities, engages patients, consumers and health staff, and is based on high-quality research.

METHODSSetting and context

Here we describe an example of the application of a co-creation approach to improve health literacy in a local health district that provides hospital and community health services to one million people in a culturally and economically diverse community in Sydney, Australia. Following the case study approach of Greenhalgh et al.,[12] we present this case in narrative form. We provide a detailed overview of the value co-creation stages and strategies to allow others to consider and apply our learnings in different health system contexts. This narrative case study also provides a basis from which we can compare existing conceptual models with practical experience in a real-world context.[23]

Policy context

Over the past 5 years in Australia, there have been consistent policy statements advocating a more systematic approach to embedding interventions to improve health literacy within the healthcare system. This has been justified as a priority for health, social and economic reasons.[24,25] The Australian Commission on Safety and Quality in Health Care mandates improvements in health literacy for clinical safety and social justice reasons.[26] To this end, the Australian government has implemented new regulatory requirements for health literacy through prescribed criteria and actions for health literacy in the revised National Standards (2019) for health organizational accreditation. All public and private hospitals are required to be accredited to the Standards, with a 3–4 year accreditation cycle.[27] The 2020–2025 National Health Reform Agreement similarly focuses on ‘empowering people through health literacy’ with an emphasis on person-centred health information and support to enable consumers to manage their own health and engage effectively with health services.[28] Given this context, strategic priorities for local health districts often reflect the need to improve communication and help people to better understand their health and manage their care.[29]

Geographical and social context

Western Sydney Local Health District (WSLHD) is 1 of 15 local health districts in the New South Wales (NSW) health system. WSLHD has approximately 12,000 staff members and delivers services to almost one million residents in Sydney's west. It has the highest urban indigenous population in Australia, 47% of residents were overseas-born, and one in two speak a language other than English at home.[30] WSLHD is one of the state's fastest growing areas with more than 1.3 million residents estimated by 2031, with a disproportionate increase in people aged 70 years or over.[30]

Co-creation steps

Given the increasing policy emphasis and growing national interest in health literacy and the diverse needs of western Sydney, the idea of developing a ‘Health Literacy Hub’ to provide a consolidated support service for staff emerged in mid-2017. A senior health manager identified the need for a more systematic approach to addressing health literacy in the health district and reached out to established contacts at a local university. The initial team was attracted to a co-creation approach which brought together a community of practice of healthcare professionals and consumers with an interest in improving health literacy in western Sydney to form the Health Literacy Hub alongside an established academic health literacy team (the Sydney Health Literacy Lab; https://sydneyhealthliteracylab.org.au/). This was seen as a mechanism by which many of the local priorities and national strategic imperatives could be met. Although it was recognized that health literacy cannot compensate for health inequities created by the unequal distribution of opportunity and resources in societies, we were motivated by

the belief that it is possible to optimize the contribution health literacy makes in mediating the causes and effects of established social determinants of health.[31]

The partnership also provided a focus and dedicated resource for testing, adaptation and implementation of health literacy interventions. By bringing people with a common interest in health literacy together, we hoped to move from a previously siloed approach in addressing the issue, to a multidisciplinary, collaborative model of working. This approach could control variability and improve service delivery effectiveness and research output by leveraging the combined resources of a university and a local health district. Co-creation was seen as a mechanism to create a 'win more–win more' environment for health literacy research and practice.[16]

Establishing the strategic direction for the Hub

We invited internal and external stakeholders (including service users, primary and secondary care providers, health services managers and health department policy-makers) to determine the strategy and priorities for the Health Literacy Hub. Meetings involved a structured workshop format, drawing on elements of the Nominal Group Technique.[32] Each stakeholder was invited to state their priorities for the Hub, which were each recorded and then discussed as a group. The purpose of this discussion was to allow stakeholders to clarify, elaborate, defend or dispute the items and to add any new priorities that emerged from the discussion. Priorities were grouped into three broad themes presented in Box 1. It was agreed that all Hub activities would be anchored in these priorities and aligned to the local health district's priorities, as determined jointly with our stakeholders.

1 **Box Health Literacy Hub priorities**

- (1)

  Build staff capacity: Provide practical assistance to clinicians to better understand the communication needs of their patients; and equip them with evidence-based methods and tools to optimize the impact and effectiveness of communications with patients and consumers.

- (2)

  Create a health literate organization: Establishing systems and organizational structures that enable and reinforce effective patient and public communication, including health services navigation and physical wayfinding.

- (3)

  Provide public resources, tools, support and advice to assist patients, their carers and families to communicate and connect in a meaningful way to the broader health system; specifically supporting them to understand and utilize the information provided and make informed decisions.

Developing shared management systems: Governance, leadership, resourcing

Successful and sustainable partnerships require resources.[33] Having identified priorities for the Hub, we were better placed to secure funding to support our partnership. We were able to successfully position the Hub as an important resource supporting core Local Health District objectives in improving clinical quality and safety, and enabling it to meet current and future requirements for institutional accreditation. This alignment with the core purpose was important in securing executive support and subsequent resource allocation.

Initial funding was provided for 4 years to support a 'Director of Strategy and Operations' position for the Hub and a Senior Academic Advisor. The Academic Advisor was a senior University academic embedded in the local health system, with previous experience working in both health and academic sectors. We recognized that partnerships need boundary-spanning leaders who understand and appreciate partners' different perspectives, can bridge their diverse cultures and are comfortable sharing ideas, resources and power.[33] Initially, the Academic Advisor prioritized building good working relationships, trust and openness among partners; ensuring that our health services partners had access to the best available evidence to support them in thinking and working differently and mobilizing the

resources needed to support the development of the partnership of the university, Local Health District and external sources. These early actions provided a shared sense of purpose on what the Health Literacy Hub partners could accomplish together, and how their joint work would benefit not only the community but also each of them individually.

The Director of Strategy and Operations' role was to work on transformational change and to support awareness, engagement and increased capacity of healthcare staff to improve health literacy. They too acted as a boundary-spanner working with university colleagues in the development of a supporting programme of health literacy research. The Director had connections to people, organizations and groups—including target populations, political decision-makers, government agencies, private sector funders and other partnerships in the community—as well as 'convening power' to bring people together for meetings and other activities.[33] Unlike more bureaucratic forms of management, which are often rigid and structured to control what people do, we endeavoured to have a management approach that was more flexible and supportive particularly given that we were engaging with health staff in established roles who were employed centrally rather than through Hub funding.[33]

These two positions were bolstered by early success in attracting funding for a health literacy Postdoctoral Research Fellow who would act as an important day-to-day point of connection between the health services Hub and the university Lab. The Research Fellow played a strategic role in brokering academic evidence and knowledge related to health literacy and bringing it into the Hub and working directly with health district staff and consumers to enable health literacy research.

Building a community of practice through a network of engagement platforms

To bring health staff, consumers and researchers together, we sought to develop a Community of Practice. Wenger [34] described Communities of Practice as building blocks of a collective learning system. They are dynamic social groups bound by a common concern or passion and a desire to learn how to improve their practice. Communities of practice differ from other forms of organization in several ways. They are not designed to deliver a specific product or service or to complete specific projects or tasks in the same way that a formal work group or department would be.[35] Communities of practice also differ in that membership is self-selected, and that passion, commitment and identification with the group's expertise holds the group together rather than specific project milestones.[35] In this way, the Health Literacy Hub was developed to be a point of connection for researchers, health staff and consumers to share information, solve problems and drive innovation in health literacy.[34] It was a way of aligning people with shared values and commitment to health literacy.

To build the Community of Practice, we strategically designed a network of engagement platforms. See Table 1. These included the development of a Health Literacy Hub website, seminar series and Community of Practice mailing list. Hub staff and university academics were also involved in a number of one-on-one consultations and targeted health literacy training initiatives with Local Health District staff. The goal of this broad engagement strategy was to build interest in health literacy, support continuing professional development for health staff and iteratively develop and expand the circle of stakeholders engaged with the Health Literacy Hub.

Table 1 Engagement platforms used to build and engage a community of practice

Initiative	Description

Health Literacy Hub website	The Health Literacy Hub website built during 2018 is designed to largely support staff development, exchange of ideas and information, and to facilitate access to useful, best practice health literacy tools and resources. To this end, the website is organized on three levels:
	Level 1—Publicly accessible, including information about health literacy and the Health Literacy Hub, and external links to support consumers to find health information, access health services etc.
	Level 2—Accessible through registration to health professionals and the academic community, providing access a wide range of educational materials, practical tools and advice on health literacy.
	Level 3—Accessible to Western Sydney Local Health District staff only, including access to an online system to support staff in developing health literate consumer information.
Health literacy seminar series	The bi-monthly seminar series introduces health literacy concepts (e.g., e-health literacy), evidence-based health literacy interventions and practices (e.g., teachback), and relevant policy (e.g., the Australian National Standards) in an accessible manner. The annual programme is formulated collaboratively by researchers, health staff and consumers, and seminars are generally co-presented by an academic and healthcare professional.
Community of practice mailing list	An electronic mailing list was developed to facilitate the distribution of information to Community of Practice members, such as information about upcoming seminars and available health literacy resources. The mailing list is also intended to foster interactivity between members, such as through moderated problem solving.
Meetings and consultations	The Hub has strategically engaged with people embarking or already undertaking health literacy initiatives for consultation, advice, the shaping of ideas or proposals and determining ways of engaging more directly with researchers. This has been facilitated through co-location of university and health staff within the Local Health District.
Targeted training	The Hub has also led formal, targeted training in health literacy with over 190 clinical (Allied Health; Child and Family Health Nursing) and preclinical (Pharmacy; General Practice) staff and students to date. Building ‘capability ecosystems’[12] in this way is intended to improve access to research and evidence-based health literacy practices, as well as expand the circle of stakeholders engaged with research.

Co-creation in research practice

Having in place jointly determined priorities, continuous learning opportunities and an active Community of Practice provided a platform to facilitate the implementation of evidence-based health literacy practices in the local health district as well as to bring together a range of health staff, consumers and researchers to co-create research projects together from the outset. Early research partnerships were often consultative, with healthcare staff seeking feedback

and advice on decisions or analyses related to health literacy research which had already been conducted (see Box 2 Case Study 1, e.g.). While these early partnerships were important for relationship building, they were ultimately missing core elements of co-creation—namely, collaboration from the outset to develop research questions, co-design research activities and plan and implement evaluation frameworks.

2.**BoxCase studies**

Case study 1—'Relationship building' Integrated and Community Health	Case study 2—'Partnering' Allied Health
Over a 4-year period, Integrated and Community Health in Western Sydney Local Health District delivered the Stanford Chronic Disease Self-Management Program (CDSMP[36]) to 486 people living with one or more chronic disease, and assessed health literacy pre- and postintervention. The Integrated Chronic Care Program Manager partnered with the Health Literacy Hub to analyse the data from this project. Outcomes of value were achieved through partnership; our analysis identified statistically significant improvements across all domains of health literacy,[37] and provided evidence of programme effectiveness to support continuation of its application for patients with chronic conditions across the Local Health District. Healthcare staff, students and researchers also co-authored a research publication in a journal special issue.	Before the development of the Health Literacy Hub, the WSLHD Allied Health Research Group conducted a cross-sectional survey of health literacy in outpatient allied health clinics. Employing a strategic approach, the Hub was able to partner with the Allied Health Research Group in the analysis of the data from their survey.[38] Building on this initial collaboration, allied health staff became integral members of the Hub, and continued to work with researchers to develop, implement and evaluate a targeted health literacy training programme for allied health professionals in western Sydney.[39] Allied health staff in this partnership co-presented a Hub seminar on health literacy measurement in 2018.

Over time, research collaborations have moved away from consultation towards models of partnering and co-creation where healthcare staff and researchers have worked together from the outset to frame locally relevant research questions, create research designs that reflect 'real-world' environments and commit to both implementing research as well as utilizing and embedding findings in the broader health service delivery community. For example, Case Study 2 (Box 2) reflects an evolving research partnership in which early consultation with members of the Community of Practice built interest, awareness and knowledge of health literacy and opened up future possibilities for more integrated research partnerships with this group.

Facilitators of co-creation

As research partnerships have evolved over time, we have identified key facilitating factors for co-creation including identification of local champions, co-generated resources, increasing trust and partnership synergy, and evolving capability and understanding. These are discussed in turn below and supplemented by Table 2 through the example of the Parenting Plus project that has brought together researchers, health staff and consumers to embed health literacy training into child and family health services in western Sydney.

•(1)

Local champions—The activities of the Health Literacy Hub have been strengthened by the identification of local champions who have been proactive in advocating for cultural change and facilitating partnership projects across the District—both directly as partners themselves and indirectly through outreach activities which they have mediated.[33] In the early stages of developing the Health Literacy Hub, the Director of Strategy and Operations identified staff with natural leadership characteristics and prior commitment to improving health literacy, and

facilitated meetings and engagement.

•(2)
Co-generated resources—Financial and in-kind resources are the basic building blocks of co-creative interaction and research. Ongoing research collaborations have been facilitated by co-generated research funding—allowing dedicated human and material resources to drive specific research projects. Funding applications have necessarily involved both health staff and academics (with alternating leads based on the funding scheme), and have strategically included both direct research costs and budget to build capacity for research within the district through participatory approaches (e.g., clinical staff secondments).[41]

•(3)
Evolving capability and understanding—As researchers have become increasingly engaged with the Local Health District, we have seen bidirectional learning and knowledge gain for different stakeholders, including an evolving understanding of different values, needs, and ways of working in research and clinical practice. This has been facilitated through deliberate actions such as the co-location of research and clinical staff and the prioritization of clinical staff secondments to the Hub.

•(4)
Increasing trust and partnership synergy—There has been a sustained commitment to building relationships of trust between researchers and communities engaged with the Health Literacy Hub. Both within and across projects, we have seen increasing partnership synergy (i.e., synergy that arises from collaboration among members of diverse knowledge, perspectives and cultures) as researchers and health professionals have worked together over time. The synergy of collaboration is manifested in the increasing number of co-created research projects across the District, as well as a shift in the point of engagement; rather than working with university academics at the analysis stage, health staff and researchers have increasingly come together at the earliest stages to identify problems, generate solutions and consider practical, culturally appropriate methodological approaches.

Table 2 Four key facilitating factors of co-created research in the Parenting Plus project

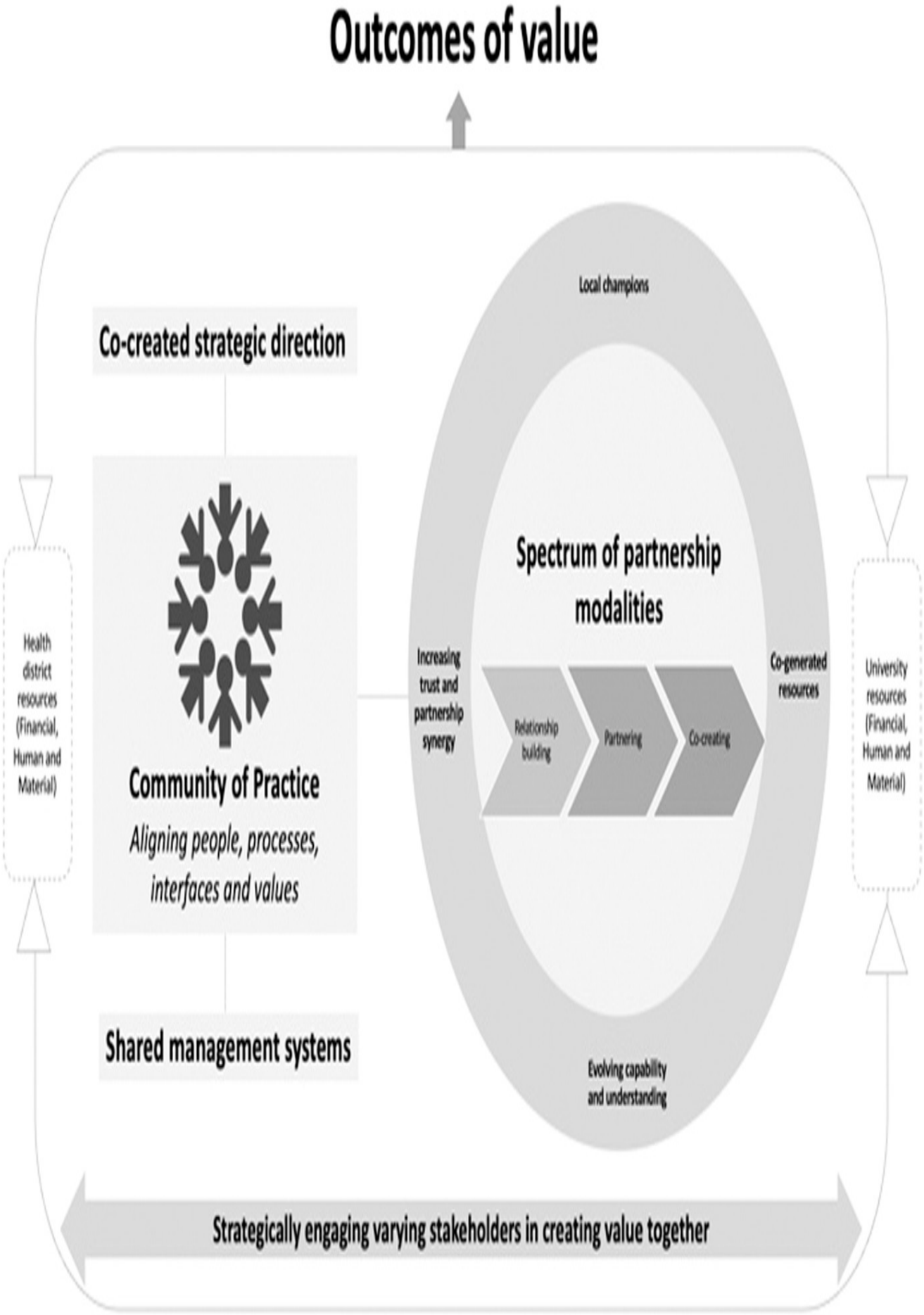
Key facilitating factor	Demonstration in the Parenting Plus project
Local champions	Researchers and Western Sydney Local Health District Child and Family Health staff first came together in 2018 in an initial meeting facilitated by the Hub Director. The Program Lead of Child and Family Health had previous experience working on health literacy projects, advocated for health literacy and agreed to partner in pilot testing the programme across six sites. Initial stages of the project were also enabled through strategic engagement with the Program Lead of Multicultural Health who championed health literacy, the Parenting Plus programme and the co-creation approach across the District. Our local champions connected us with consumers (new parents) who also became partners in the development and adaptation of the Parenting + materials, ensuring that the programme was developed in consultation with multicultural communities from the health district.

Co-generated resources	Initial funding for the piloting of the Parenting Plus project in western Sydney was awarded—on a competitive basis—from the Local Health District's Research and Education Network and the Primary Health Network. This funding was strategically allocated to direct research costs related to roll-out of the pilot programme and the secondment of a Child and Family Health Nurse to work directly in the Health Literacy Hub for the duration of the pilot. Successful piloting informed a larger funding application for a randomized trial of Parenting Plus, awarded in 2021.
Evolving capability and understanding	Evolving capability of researchers and health staff was achieved through the secondment process which enabled health staff and researchers to work directly together on the Parenting Plus project over a 10-month period.[40] Formal and informal interactions between team members during this time helped us to appreciate one another's worldviews, priorities and ways of working. Health staff were also involved in research capacity building and transferable skills training in data collection and analysis.
Increasing trust and partnership synergy	Increasing partnership synergy was evidenced by modifications to the Parenting Plus programme made postpilot which better reflected the perspectives and priorities of community stakeholders, including the target population (new parents) and health staff. Researchers (n = 2) and health staff (n = 2) worked together to analyse and interpret feasibility study data and identify necessary modifications to programme content which was iteratively reviewed by managerial health staff (n = 3) and patient partners (new parents; n = 3) in a series of workshops and follow-up correspondence.

Increasing trust and partnership synergy between researchers and health district staff has also served to strengthen collaborations with consumers and the broader community. The District's—and, in particular, our local champions'—strong ties to the community have strengthened the capacity of the Hub to access and involve community members in the co-creation of health literacy research. In addition to Parenting Plus, another recent example of this was the rapid mobilization of staff and consumers in Western Sydney and two adjoining health districts to co-design and conduct the largest Australian COVID-19 survey of people who speak a language other than English at home.[42,43]

RESULTS: A (REVISED) MODEL OF CO-CREATION

Insights from our experience over a 5-year period have led to a refined understanding of how co-creation is facilitated in practice, as summarized in Figure 1. At the core of the partnership has been a common vision oriented to improving health literacy and delivering outcomes of value for academics, health staff and consumers combined with the engagement of each stakeholder group at every stage.



Enlarge this image.

Our revised model for research co-creation builds on previous work, but advances from this foundation by focussing

on practical stages, strategies and structures to build research partnerships and work towards co-creation in real-world health services and health systems. Three key elements of differentiation from existing conceptual models of co-creation[12,16] are outlined below.

•(1)

Our revised model incorporates a practical focus on the structural enablers of co-creation. The centrality of the Community of Practice, for example, reflects the key role that the iterative expansion of the Community of Practice played in enabling us to reach a broad range of consumers and health providers and to grow the Hub's presence within the district. This was key to facilitating co-creation across multiple settings and projects.

•(2)

By including a spectrum of partnership modalities (spanning relationship-building, partnering and co-creating), our model acknowledges the evolving nature of research partnerships and seeks to realistically reinforce the flexibility and commitment required to achieve meaningful co-creation in research. We employed a range of connected strategies and invested the necessary time and resources to develop capability, trust and understanding between researchers, frontline health workers and consumers. Importantly, this foundational work enabled research partnerships to evolve over time.

•(3)

Finally, we have brought together literature related to co-creation[12,16] and partnership synergy[33,41,44] to depict key facilitators of research and collaboration including (i) local champions who advocated for health literacy and institutional collaborations and had the ability to influence change regardless of organizational position, (ii) co-generated resources, particularly from external sources, (iii) evolving capability ecosystems and understanding and (iv) increasing trust and partnership synergy.

Barriers and challenges

While the process of developing the Health Literacy Hub has informed the above model of co-creation, we have also faced challenges and barriers in the establishment and maintenance of this collaborative partnership which warrant attention. Foremost, the experience of bringing together the Hub and the Lab has highlighted the flexibility and commitment required to achieve meaningful co-creation in research. Processes for partnering in the Health Literacy Hub have necessarily been dynamic and evolving, and this has required significant investments of time over and above other models for conducting health research. Given the inherent time commitments, one of the greatest threats to the maintenance of Hub relationships has been the turnover in staff, senior managers and executives in the health district. For example, the Lead of Child and Family Health who co-led the development and feasibility testing of the Parenting Plus programme retired, as has the Child and Family Health nurse seconded to co-design programme content. There has also been a turnover of Chief Executives and several Executive Directors since the establishment of the Hub. To maintain the momentum and stability of the Health Literacy Hub despite such turnover, we continue to engage broadly with staff at all levels of the organization and externally to sustain relationships and continually build new ones.[45] To date, we have had over 70 consultations with different clinical services in the Local Health District, members of senior management and district executives and external stakeholders including state health services, other local health districts, councils and consumer organizations. An additional challenge has related to ongoing funding and capacity. While we have been successful in obtaining project-specific funding through grant applications, there is an ongoing need for designated administrative and support staff to maintain engagement platforms (e.g., the Hub website) and capacity-building initiatives within the District. Funding for such roles has been

harder to secure on a sustainable basis.

Outcomes and future directions

To date, key outcomes and achievements relate to reach and the scope of collaborative activities. There are currently over 1300 members of the Community of Practice and 11 completed or ongoing research projects which have quite literally ranged across the lifespan from early childhood/parenting education, through chronic disease management, to end-of-life decision-making. The Lab/Hub collaboration has generated $1.9 million in research project funding, >15 jointly authored research outputs and has been linked to organizational-level improvements in health communication.[46] Moving forward, the monitoring and evaluation of Hub research outcomes is an intentional focus, to ensure that efforts are recorded and recognized for their value to both the academic, health and broader communities and consumers. We are also seeking to develop more comprehensive and systematic models for engaging with consumers, patients and carers across all collaborative projects.

DISCUSSION

The Health Literacy Hub represents a rare form of collaboration between hospitals, healthcare services, communities and health literacy researchers, which has evolved to develop innovative, practical and scalable health literacy interventions. This research 'laboratory' has enabled us to develop relevant, contextualized research questions and undertake applied research with clear pathways to research translation and practical implementation to benefit communities with significant social disadvantages.

Our revised model for research co-creation complements and builds on previous research related to co-creation, cross-sectoral collaboration and translational research. Components of our revised model are supported by both theoretical and empirical literature which highlights the importance of building and maintaining relationships of trust and 'partnership synergy' in collaborative research,[33,41,44] the key role of local champions[47,48] and the need for resources to sustain such initiatives.[41] Our model also advances from this foundation by focussing on practical stages, strategies and structures to build research partnerships and work towards co-creation in real-world health services and health systems. This manuscript is also one of few to report on collaborations specifically focused on health literacy. Another example is the Health Literacy Initiative involving Keele University and Stoke-on-Trent City Council Public Health.[44] In describing their collaborative health literacy work, Estacio et al., for example, similarly noted the importance of trust to ensure that the partnership was sustainable and able to achieve systemic transformations. In their case, and our own experience, the growth and development of health literacy collaborations was based on mutual trust from individual members and the understanding that the partners were contributing to the achievement of a common goal.[44]

Strengths and limitations

We have developed a revised model of co-creation based on our experience in establishing the Health Literacy Hub in western Sydney, Australia. Without testing in other settings, it is not yet clear whether this model is replicable or which components are entirely necessary for similar success. In addition, this model and manuscript may not capture the perceptions of all partners engaged with the Hub. Going forward, a more formalized evaluation including all partners will be valuable. This could, for example, replicate the evaluation of a UK public health collaborative, AVONet, which used a convergent parallel mixed-methods design with quantitative surveys and qualitative semistructured interviews to capture the experiences of all partners involved in the collaboration in some way.[49]

CONCLUSIONS

A co-creation approach—with its necessary time and resource commitments—has not always been rewarded in research. However, as researchers are pressed to highlight 'impact' by a growing number of funding bodies, co-creation becomes more attractive. For the past 5 years, we have worked to build a practical and sustainable working

relationship between an academic research lab and a local health system and its community focussed on improving health literacy. The goal was to improve both service delivery effectiveness and research output by leveraging combined resources, with involvement and input from all partners throughout the entire research journey. Our conceptual model reinforces core learnings from this process. We engaged broadly through the strategic development of a community of practice, with extensive commitments to build capability and relationships of trust and to progress research partnerships from relationship-building activities to co-creation. Partnership with local 'champions' and co-generated resources helped to maintain momentum. Our co-creation model can provide useful insight into mechanisms that have created an effective, collaborative and practical approach to researching and solving locally based problems working alongside healthcare staff and consumers.

AUTHOR CONTRIBUTIONS

Danielle M. Muscat developed the revised conceptual model of co-creation and drafted the manuscript. Don Nutbeam was a major contributor in writing the manuscript and revising the conceptual model. All authors played a key role in the establishment of the Health Literacy Hub and read and approved the final manuscript.

ACKNOWLEDGEMENTS

The Health Literacy Hub was funded by Western Sydney Local Health District. D. M. M. is supported by a Westmead Fellowship (Early Career Researcher) from Western Sydney Local Health District. The listed funding bodies had no role in the design of the study and collection, analysis and interpretation of data and in writing the manuscript.

CONFLICT OF INTEREST

D. M. M., K. M. and J. A. are joint Directors of Health Literacy Solutions Pty Ltd.; a health literacy consultancy company. The remaining authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no data sets were generated or analysed during the current study.

DETAILS

Subject:	Narratives; Collaboration; Public health; Health care policy; Communication; Literacy; Health care; Community health services; Health research; Capabilities; Health care industry; Consumers; Health literacy; Health education; Innovations; Research partnerships; Best practice; Research; Evolution; Facilitators; Accreditation; Medical personnel; Case studies; Community; Health services; Management systems; Patients; Flexibility; Multiculturalism &pluralism; Partnerships; Researcher subject relations; Strategic management; Embedding
Business indexing term:	Subject: Consumers
Location:	Australia
Identifier / keyword:	co-creation; health literacy; health research systems; knowledge production; research translation
Publication title:	Health Expectations; Oxford
Volume:	26

Issue:	1
Pages:	452-462
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-29
Milestone dates:	2022-09-12 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-05-24 (manuscriptReceived); 2022-11-29 (publishedOnlineEarlyUnpaginated); 2022-10-31 (manuscriptAccepted)
Publication history :	
First posting date:	29 Nov 2022
DOI:	https://doi.org/10.1111/hex.13678
ProQuest document ID:	2767101851
Document URL:	https://www.proquest.com/scholarly-journals/embedding-health-literacy-research-best-practice/docview/2767101851/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the “License”). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-07-10

Trauma-informed patient and public-engaged research: Development and evaluation of an online training programme

Gum, Amber M [1] ; Goldsworthy, Mary [2] ; Guerra, Lucy [3] ; Salloum, Alison [4] ; Grau, Meredith [5] ; Gottstein, Sheri [6] ; Horvath, Carol [6] ; Fields, Annanora [6] ; Crowder, Johnny [6] ; Holley, Robb [2] ; Ruth, Leigh J [7] ; Hanna, Karim [8] [1] myPATH Partnership, University of South Florida, Tampa, Florida, USA; Department of Mental Health Law &Policy, University of South Florida, Tampa, Florida, USA; Department of Internal Medicine, University of South Florida, Tampa, Florida, USA [2] myPATH Partnership, University of South Florida, Tampa, Florida, USA; Department of Mental Health Law &Policy, University of South Florida, Tampa, Florida, USA [3] myPATH Partnership, University of South Florida, Tampa, Florida, USA; Department of Internal Medicine, University of South Florida, Tampa, Florida, USA [4] myPATH Partnership, University of South Florida, Tampa, Florida, USA; School of Social Work, University of South Florida, Tampa, Florida, USA [5] myPATH Partnership, University of South Florida, Tampa, Florida, USA; Crisis Center of Tampa Bay, Tampa, Florida, USA [6] myPATH Partnership, University of South Florida, Tampa, Florida, USA [7] myPATH Partnership, University of South Florida, Tampa, Florida, USA; Department of Psychiatry and Behavioral Neurosciences, University of South Florida, Tampa, Florida, USA [8] myPATH Partnership, University of South Florida, Tampa, Florida, USA; Department of Family Medicine, University of South Florida, Tampa, Florida, USA

ABSTRACT (ENGLISH)

Introduction

As patients, members of the public, and professional stakeholders engage in co-producing health-related research, an important issue to consider is trauma. Trauma is very common and associated with a wide range of physical and behavioural health conditions. Thus, it may benefit research partnerships to consider its impact on their stakeholders as well as its relevance to the health condition under study. The aims of this article are to describe the development and evaluation of a training programme that applied principles of trauma-informed care (TIC) to patient- and public-engaged research.

Methods

A research partnership focused on addressing trauma in primary care patients (‘*my*PATH’) explicitly incorporated TIC into its formation, governance document and collaborative processes, and developed and evaluated a free 3-credit continuing education online training. The training was presented by 11 partners (5 professionals, 6 patients) and included academic content and lived experiences.

Results

Training participants (*N*=46) positively rated achievement of learning objectives and speakers' performance (ranging from 4.39 to 4.74 on a 5-point scale). The most salient themes from open-ended comments were that training was informative (*n*=12) and that lived experiences shared by patient partners were impactful (*n*=10). Suggestions were

primarily technical or logistical.

Conclusion

This preliminary evaluation indicates that it is possible to incorporate TIC principles into a research partnership's collaborative processes and training about these topics is well-received. Learning about trauma and TIC may benefit research partnerships that involve patients and public stakeholders studying a wide range of health conditions, potentially improving how stakeholders engage in co-producing research as well as producing research that addresses how trauma relates to their health condition under study.

Patient or Public Contribution

The *my*PATH Partnership includes 22 individuals with professional and lived experiences related to trauma (https://www.usf.edu/cbcs/mhlp/centers/mypath/); nine partners were engaged due to personal experiences with trauma; other partners are community-based providers and researchers. All partners contributed ideas that led to trauma-informed research strategies and training. Eleven partners (5 professionals, 6 patients) presented the training, and 12 partners (8 professionals, 4 patients) contributed to this article and chose to be named as authors.

FULL TEXT

INTRODUCTION

Patient and public involvement and engagement (PPIE) in co-producing health-related research are growing rapidly on an international scale, with efforts across numerous countries recently described in a special issue of *The BMJ*[1] and other reviews.[2-5] In the United States, the Patient-Centered Outcomes Research Institute (PCORI)[6] was created in 2012 to build the nation's capacity, production and dissemination of stakeholder-driven research. ('Stakeholder-driven research' is a phrase commonly used by PCORI and is conceptually similar to phrases such as PPIE as applied to research, co-production of research, community-based participatory research or participatory action research.) As groups of patients, members of the public, and professional stakeholders join to co-produce research, they would likely benefit from attending to an important issue that is relevant across a wide range of health conditions—trauma. Traumatic events are very common, impacting 54%–74% of adults globally,[7,8] which has implications for how PPIE-based research partnerships function as well as the health condition they are studying. First, among patients with health conditions and members of the public who become engaged in co-producing research, many of them will have experienced one or more traumatic events, given how common these events are for individuals living with chronic health conditions and in the public. This lived experience of trauma can make it difficult for some individuals to feel safe and empowered to fully engage as research partners. Second, trauma complicates the identification, course and treatment for many health conditions,[9-11] so research groups may want to incorporate trauma into their research agendas.

To address these challenges, our stakeholder-driven research partnership aimed to develop and evaluate training about the application of trauma-informed principles to stakeholder-driven research, as part of a PCORI-funded capacity-building project.

The relevance of trauma to stakeholder-driven research partnerships

Trauma is highly prevalent in the general population[7,8] and places individuals at risk of a wide range of health conditions (e.g., cardiovascular disease, diabetes, cancer, respiratory disease, substance misuse, depression, anxiety),[7,12-22] which suggests that research partnerships will engage many people impacted by trauma, whether intending to or not. Trauma is defined as 'an event, series of events, or set of circumstances that is experienced by an individual as physically or emotionally harmful or life-threatening and that has lasting adverse effects on the individual's functioning and mental, physical, social, emotional, or spiritual well-being' (p. 7).[23] Trauma includes a range of experiences, such as child abuse (i.e., physical, emotional or sexual) and neglect, potentially other adverse childhood experiences (e.g., unstable home environment),[13] domestic violence, physical or sexual assault, combat or war, serious accidents and injuries, natural disasters, witnessing trauma or cultural trauma.[23]

Globally, most adults have experienced at least one traumatic event, with estimates across countries ranging from 54% (Spain) to 74% (South Africa),[7] for a global estimate of 70.4% across 24 countries.[7] Over half (60%) of American adults have experienced at least one traumatic event.[8] Among people with at least one traumatic

exposure, the mean number of traumatic events is 4.6 (chronic traumas such as repeated child abuse were counted as one event),[24] and individuals exposed to childhood trauma (e.g., abuse, neglect) are at increased risk of further trauma exposure as adults.[24,25]

Trauma exposure is a potent risk factor for many deleterious effects. Globally, of those exposed to traumatic events, 5.6% develop posttraumatic stress disorder (PTSD), which is persistent for approximately half.[26] Beyond PTSD, trauma exposure increases the risk of many other outcomes—poor health behaviours (e.g., smoking, not eating well or exercising, risky sexual behaviours); numerous physical health problems and physical disability (e.g., cardiovascular disease, diabetes, respiratory disease, cancer, autoimmune conditions); other behavioural health conditions (e.g., anxiety, depression, substance misuse); relationship problems and death, including suicide and all-cause mortality.[7,12-22] In the general population, death by suicide has been found to be over five times higher for individuals with PTSD compared to those without PTSD.[27] Individuals with trauma histories use more healthcare services,[28] and trauma is estimated to cost $748 billion annually in health-related outcomes in North America.[19] Patients with posttraumatic stress symptoms tend to show poorer adherence to medical regimens and worse physical health outcomes.[9-11] This situation has worsened since the onset of the COVID-19 pandemic, with traumatic events increasing for the public[29-32] and healthcare workers.[33] Addressing and treating posttraumatic effects has mental, as well as physical, benefits; a systematic review has shown that treating PTSD improves not only mental health, but also physical health, including cardiovascular, diabetic and metabolic outcomes.[34]

Therefore, it will likely benefit many research partnerships to learn more about trauma and consider its impact on their partners and relevance to the health condition under study, although some preparation of the partnership is warranted when beginning this process. Some trauma survivors often feel unsafe, disempowered and dysregulated (i.e., hyperarousal or hypoarousal).[36] These characteristics could make it difficult for some partners to discuss or learn about trauma; these characteristics also could exacerbate power differentials that already occur between patient and family stakeholders with professional stakeholders. Trauma-informed care (TIC) is an organizational model designed to address these types of challenges across the diverse community, educational, health and social service settings that could be applied to research partnerships.

Overview of TIC

TIC aims to promote a sense of safety, collaboration and empowerment for trauma survivors and all stakeholders in an organization to promote healing. It '*realizes* the widespread impact of trauma and understands potential paths for recovery; *recognizes* the signs and symptoms of trauma in clients, families, staff, and others involved with the system; and *responds* by fully integrating knowledge about trauma into policies, procedures, and practices, and seeks to actively *resist* re-traumatization' (p. 9).[23] TIC involves six principles: safety, trustworthiness and transparency, peer support, collaboration, empowerment and humility and responsiveness.[23] A recent systematic review of 23 studies across settings (e.g., schools, behavioural health service settings) found that trauma-informed staff training improved staff knowledge, attitudes and behaviours in some studies, and five studies found positive impacts on student or patient outcomes.[36]

Application and research on TIC in healthcare settings is in the early stage. A general model of trauma-informed healthcare involves a foundation of TIC knowledge among all personnel, a calm and empowering environment, educating patients and personnel about trauma and its relationships with health, inquiring about patients' trauma histories and responding appropriately to patients' disclosures of trauma.[12] In addition to its hypothesized benefits for patients, TIC promotes awareness of trauma among personnel and encourages self-care; as such, TIC is thought to benefit staff well-being,[24] although empirical evidence is lacking. TIC principles and practices have also been applied to community outreach, policy,[38,39] research participants[40,41] and a research advisory board,[42] but no publications were identified that developed and evaluated training applying TIC principles to PPIE-based research partnerships.

myPATH partnership and aims

Because our research partnership explicitly addresses the topic of trauma, we applied the principles and practices of TIC to its initial development and continuation over the past 4 years. Our partnership is called *my*PATH—a *patient-centered* Partnership Addressing Trauma and Healing. *my*PATH's mission is to 'sustain a partnership of patients,

providers, researchers, and other stakeholders, that will collaboratively develop and conduct research to: (1) integrate principles of trauma-informed care into primary care settings; and (2) deliver personalized interventions to patients with trauma histories that will impact outcomes meaningful to patients'.[42] *my*PATH partners have met monthly since November 2017, during which time we engaged in co-learning about trauma, developed a governance document, developed and implemented trauma-informed research practices, developed live and online training applying TIC to research partnerships, conducted research related to the COVID-19 pandemic, conducted surveyed 249 stakeholders to plan research and submitted a grant proposal involving interventions to address trauma in primary care settings. These efforts have been supported by two PCORI contracts. For this article, we aimed to describe the development and evaluation of the training.

METHODSDevelopment of training content, format and processesPartnership development and capacity-building

The training was based on the partnership's experience forming and sustaining the partnership over the first 2 years. The partnership was formed through networking, beginning with an academic psychologist with expertise in behavioural health integration in primary care and an academic internal medicine physician, adding other professionals with expertise in trauma and healthcare and selecting and inviting individuals who had engaged in healthcare or social services related to traumatic experiences. Care was taken to engage individuals from various professional disciplines and patient representatives with diverse traumatic experiences. The application of TIC principles began in the initial discussions inviting patient representatives to join the partnership, by (a) discussing its purpose; (b) conveying the importance of learning from each person's perspective; (c) assuring they would not be required to disclose personal experiences, they could do whatever was needed for self-care if any discussions became difficult, and they could resign at any time and (d) encouraging them to take as much time as needed and ask questions to facilitate an informed, uncoerced decision regarding whether to join the partnership.

The initial partnership included 10 founding members. The five professionals' disciplines included psychology, internal medicine, nursing and mental health counselling. The five patient partners had experienced various types of traumas, including child abuse, workplace trauma and physical and sexual assault. It was soon recognized that patient partners also contributed professional expertise, including childcare, healthcare, social services and entrepreneurship. Nine partners were female, and one was male; partners represented a range of ages (from early 20s to 70s) and racial and ethnic groups (e.g., white, Latina/Latino, Black/African-American, Asian, American Indian). (Partners have not been formally surveyed regarding their demographic characteristics or trauma experiences, given the partnership's decision, as documented in the governance document, to not require partners to disclose personal or traumatic experiences. Thus, the descriptions of partners' characteristics in this manuscript are based on public information and partners' voluntary statements during meetings.)

These initial partners secured funding from a PCORI Pipeline to Proposal award (October 2017–2018), which provided compensation for all professional and patient partners to meet monthly and develop infrastructure. During the first year of meetings, partners focused on rapport-building; co-learning about trauma, TIC and stakeholder-driven research and developing a governance document. Partners learned about trauma and TIC through brief presentations by partners; review and discussion of readings and other resources regarding trauma and TIC as presented by the US Substance Abuse and Mental Health Services Administration (SAMHSA), Trauma-Informed Care Implementation Resource Center and experts in trauma in primary care[23,43-45] and general discussion among partners. Incorporating elements of TIC, initial meetings also included discussions about the nature of trauma, difficulties discussing trauma, limitations of what would or would not be discussed and strategies for managing difficult emotions that might arise during meetings or when reviewing materials. Partners also reviewed resources related to community-based participatory research[46,47] and stakeholder-driven research.[48] These resources and discussions led to the governance document, which included elements suggested by PCORI, including vision and mission, membership requirements and expectations, communication, decision-making and values. The governance document also included the results of our discussions about trauma. As summarized in the governance document (available from the first author), it was determined that 'trauma' has a subjective component, partners would not be required to disclose their personal experiences, no subjects were off-limits if discussions remained respectful and

considerate, and partners could engage in various self-care behaviours if they became distressed during a discussion.

Partners also identified additional capacity-building needs, which resulted in a second PCORI proposal being submitted and awarded (January 2020 to December 2021). Its first aim was to expand the partnership and network connections (22 partners currently), by intentionally considering professional disciplines, networks, trauma-related and demographic characteristics that were not represented in the original 10-member partnership. New professional partners were added from psychiatry, family medicine, social work, substance use treatment and healthcare administration; and they represented multiple networks across Florida, including the state unit of a mental health advocacy organization, a regional agency that contracts with over 100 behavioural health agencies and another state university's academic medical centre. New patient partners were added by connecting with other organizations (e.g., healthcare organizations serving veterans and LGBTQ+). New patient partners also added new areas of professional expertise, including television production, music, marketing, peer counselling and veterans services. The current partnership includes 16 females and 6 males with a similar range in age, race and ethnicity as the founding group.

Training development and pilot-testing

The second aim of the second PCORI contract was to develop, deliver and evaluate training on TIC and trauma-informed, stakeholder-driven research. The initial training content was developed collaboratively by the partnership through discussion across nine monthly meetings, by discussing potential topics and learning objectives, reviewing websites and resources, reviewing presentation platforms and discussing format and speakers. The training content was divided into three components: (a) an overview of trauma and TIC; (b) an overview of stakeholder-driven research and (c) an application of TIC to stakeholder-driven research. Patient partners noted that personal stories can be more compelling than academic, research-based content alone. Thus, partners were invited to share personal experiences, but it was emphasized that no partner should feel obligated to present, or if they chose to present, that they should not feel obligated to present details they would prefer to omit. Eleven partners (five professionals and six patients) volunteered to present a portion of the training content. Those who presented personal statements elected to prerecord these statements; one patient partner who was a television news executive producer arranged for a professional videographer to video-record these statements.

Regarding format, partners planned to offer the training in two phases: a two-part live training workshop that would involve the partners and a small number of stakeholders, followed by enduring online training. Due to the COVID-19 pandemic, partners changed the format of the live training workshops from in-person to synchronous videoconference. Partners elected to use Prezi as the presentation software, given its features that promote engagement with online content. Partners also selected Moodle for housing the online training, given that one goal of the training was to build sustained relationships with more stakeholders, and Moodle requires a login and email address to access the training. These decisions were led by patient partners after academic partners presented options.

Partners established processes by which several types of professionals (i.e., physicians, advanced practice providers, nurses, social workers, psychologists, mental health counsellors and peer specialists) could earn free continuing education units (CEUs). This process was initiated after a patient partner inquired whether offering free CEUs would be possible; this patient partner had observed at a professional conference that sessions offering CEUs were particularly well-attended. Research partners worked with two groups of university CEU providers (one for health professionals and one for social service and behavioural health professionals), reallocated the budget for CEUs and completed all required CEU forms (e.g., learning objectives), obtaining feedback and approvals from all partners at each monthly meeting (e.g., reviewing proposed budget reallocations before implementing changes).

Twenty-one individuals (including partners who presented a portion of the training) participated in the live videoconference training in November 2020 (21 for Part 1, 17 for Part 2), and 13 completed anonymous online evaluations. At the monthly meeting following the live training, the results of these evaluations were reviewed, and revisions were planned. The feedback was largely positive, and suggestions primarily focused on improving the

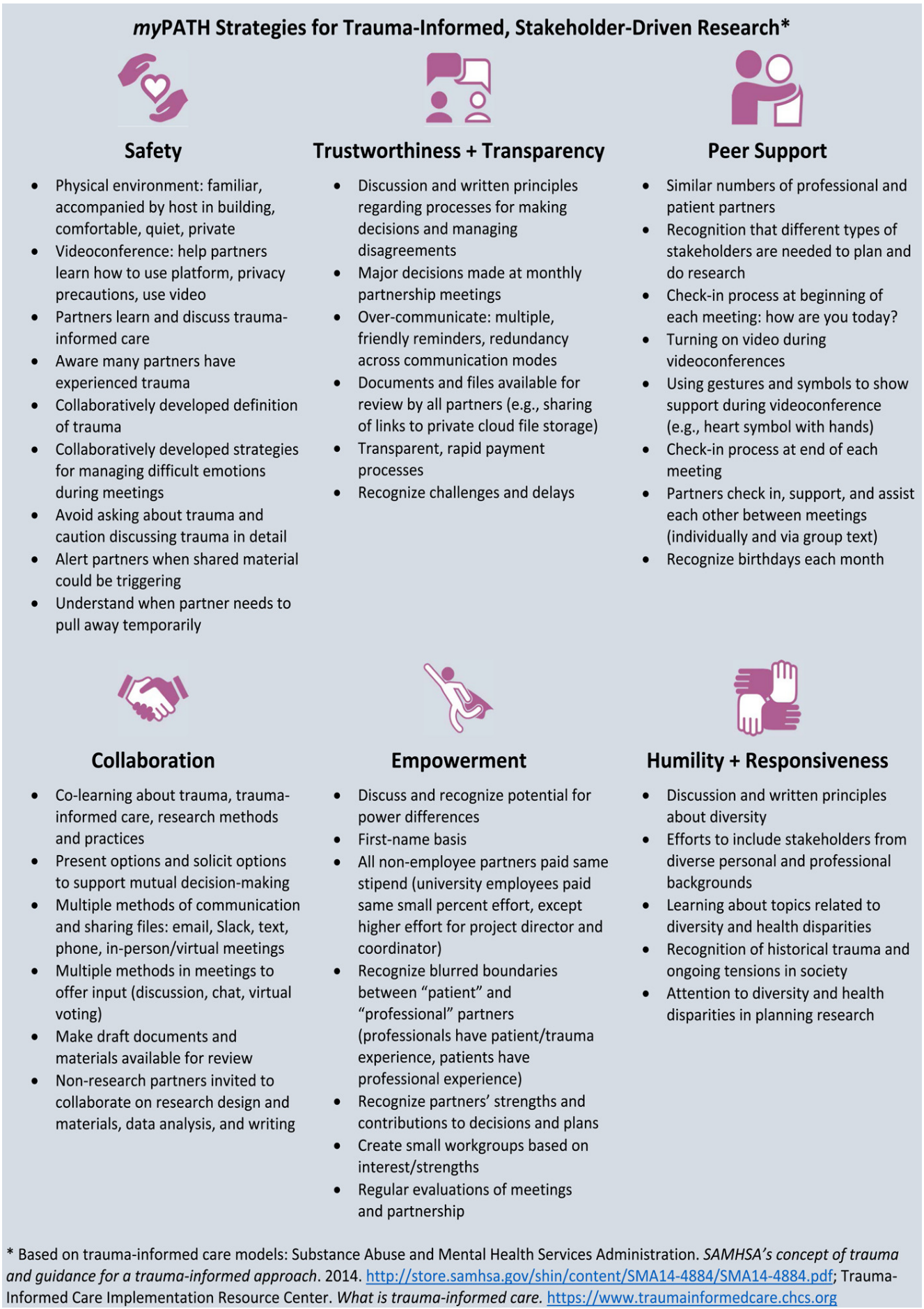
audiovisual quality of the training.[50] After changes were made to the presentation materials, the academic presenters rerecorded their video presentations for the final online training.

Final online enduring trainingTraining content

As stated previously, the training content was divided into three components, all available on a Moodle website. Part 1 involved watching a 60-min video, in which partners provided an overview of trauma, the prevalence of trauma in the United States, the impacts of trauma on physical and behavioural health, the importance of resilience and the principles and practices of TIC. This video summarized: (a) research findings from the seminal ACE Study,[13,14] updated with recent research on physical and behavioural health impacts of trauma and resilience[15-22,29]; (b) SAMHSA's TIC model[24] and (c) Machtinger et al.s'[12,44] model of TIC in healthcare. This information was presented by four different academic partners. This video also incorporated personal statements from patient partners about aspects of their trauma and healthcare experiences they wished to convey.

Part 2 utilized PCORI resources about stakeholder-driven research, which learners read and interacted with at their own pace. PCORI has developed a Research Fundamentals Learning Package, with two introductory sections and five modules that help stakeholders new to research learn about the health research process and how to become fully involved as a stakeholder. Two sections were included as part of our training package: ‘Engaging in Stakeholder-Driven Research’[51] and ‘Developing Research Questions: Module 1’.[52] The first of these sections is an interactive video that includes different types of stakeholders, including patient representatives, that provides an introduction to stakeholder-driven research. The second section provides an overview of how to begin developing research questions and designing a research study. While not directly related to trauma, this segment of the training was included to provide all participants with encouragement, basic concepts and strategies for becoming engaged in research. One aim of our partnership was to build a larger collaboration including members of the public and community-based professionals, who would then have opportunities to provide input regarding research being planned by the partnership.

Part 3 involved viewing a 60-min video, in which *my*PATH partners described strategies for applying TIC principles to stakeholder-driven research partnerships, organized by the six TIC principles. These strategies are summarized in Figure 1 and below. This section was presented by the first and second authors (the Project Director and Coordinator, respectively), incorporating video-recorded statements by patient partners regarding their experiences as research collaborators.



Enlarge this image.

As described in Part 3, to foster *safety*, strategies involve creating a comfortable physical environment, privacy precautions for videoconference meetings (e.g., private link to enter, verifying the identity of participants, using video

as much as possible) and agreed-upon guidelines for discussing the trauma and managing distress. Partners attempt to avoid trauma triggers by not asking other partners detailed personal questions, not discussing traumatic topics in graphic detail and presenting warnings when sharing resources that might be distressing. Partners are encouraged to engage in self-care as needed during meetings, such as stepping away or 'zoning out' temporarily if a difficult topic arises.

Many of the strategies related to other TIC principles are consistent with general principles and strategies for stakeholder-driven research,[53] particularly strategies for *trustworthiness and transparency, collaboration, empowerment, humility, and cultural responsiveness* (Figure 1). Given the overlap between these TIC principles and stakeholder-driven research principles, many research partnerships may already be implementing some strategies that foster TIC, without realizing it. Strategies that other research partnerships may already be using include developing communication and decision-making guidelines, sharing information and documents, budget transparency, first-name basis and other strategies to minimize power differentials and attending to diversity and health disparities.

Peer support is a value emphasized in TIC, and several strategies were included to build and sustain relationships among partners, including a brief check-in at the beginning and end of each meeting, which is a common practice in trauma-informed organizations. Another noteworthy strategy has been the recognition of the blurred boundaries between 'patient' and 'professional' partners—many 'professional' partners have had personal (and work-related) traumatic experiences, and 'patient' partners also advance our work through their professional experiences and skills. We have found small workgroups and projects to be an effective way to actively engage patient partners, such as in the video-recorded statements for the training and a branding workgroup that developed the *my*PATH name and logo.

Regarding *humility and responsiveness*, the strategies applied to our research partnership included a statement about diversity across a variety of domains in the governance document; intentional discussions about stakeholder perspectives that needed to be added to the partnership; learning about diversity, health disparities and cultural trauma and attending to diversity in planning research (Figure 1). The training included a brief overview of these strategies as well as research findings regarding trauma for diverse populations (including race/ethnicity, gender, age).

Evaluation survey

To obtain the CEU certificate, individuals were required to access each section of the training content, and then they were required to complete a brief, anonymous online evaluation survey. The survey included 23 questions (Table 1): (a) how well each learning objective was met (12 items; ranging from 1 = not well at all to 5 = extremely well); (b) how well the speakers did (5 items; ranging from 1 = not well at all to 5 = extremely well); (c) detection of bias (yes/no); (d) how the participant heard of training and (e) four open-ended items (what the participant liked best, how to improve the training, other topics of interest and additional comments).

Table 1 Survey evaluation results (*N* = 46)

	M	SD
Part 1 Objective 1 (impacts of trauma)	4.52	0.81
Part 1 Objective 2 (principles of trauma-informed care)	4.57	0.58
Part 1 Objective 3 (trauma-informed care strategies)	4.48	0.69
Part 2 Objective 1 (define patient-centred outcomes research)	4.50	0.72

Part 2 Objective 2 (PCORI: types of stakeholders)	4.46	0.75
Part 2 Objective 3 (PCORI: benefits of engaging in PCOR)	4.41	0.72
Part 3 Objective 1 (engaging in trauma-informed PCOR)	4.39	0.71
Part 3 Objective 2 (collaborating on trauma-informed PCOR)	4.57	0.62
Speakers performed well	4.57	0.58
Speakers used effective methods	4.48	0.69
Speakers were clear and understandable	4.63	0.57
Speakers provided accurate information	4.74	0.49
Speakers provided current information	4.74	0.49
Speakers used time wisely	4.67	0.60
	N	%
Bias detected (no)	45	97.83
How learn about training		
Organization where I work	22	47.83
Community/advocacy organization	7	15.22
Colleague or friend	6	13.04
Professional organization of which I am a member	3	6.52
Other	8	17.39

Note: Means are based on a 5-point scale, ranging from 1 = not well at all to 5 = extremely well.
Abbreviations: PCOR, patient-centred outcomes research; PCORI, Patient-Centered Outcomes Research Institute.

Dissemination

Brief emails and a flyer were disseminated by all partners to their contacts in Florida, such as by email, listservs and social media. Partners had contacts with a variety of networks, including the academic units of all faculty partners, regional crisis centre, statewide network of primary care providers, regional network of over 100 behavioural health provider agencies, National Alliance on Mental Illness (NAMI) Florida and other mental health advocacy contacts. A news story was aired in which the first author was interviewed. These dissemination efforts took place periodically from spring to fall of 2021, resulting in 46 individuals who completed the online training and evaluation survey between March and December 2021. The university's IRB provided approval for the dissemination of the

anonymous evaluation results reported herein.

Data analyses

Descriptive statistics were calculated for all closed-ended survey items. For the open-ended questions, the first two authors used an inductive method to determine themes.[54] Independently, the two authors analysed and categorized responses from the open-ended responses. Next, the two authors discussed, reviewed and refined the categories into final themes, discussing until they resolved discrepancies. Regarding reflection activities, the quantitative and qualitative results were presented at a monthly partnership meeting, with designated time for discussion, after which a draft of this manuscript was distributed to all partners with 1 week to review and submit written feedback or discuss with the first author. Following the submission of the manuscript to this journal, the process was repeated: journal reviewers' feedback was reviewed and discussed at a monthly partnership meeting, the first author revised the manuscript and cover letter and all partners had 1 week to review and submit feedback.

RESULTS

Forty-six individuals completed the online training and evaluation survey. As shown in Table 1, participants' ratings for the achievement of the learning objectives and speakers' performance were very positive, with average ratings ranging from 4.39 to 4.74 on the 5-point scale. One individual detected bias; 45 did not. Most participants learned about the training from their work setting (*n* = 22, 47.8%).

The main themes of the open-ended comments (Table 2) were largely positive, with the most salient theme being that it was informative (*n* = 12; e.g., 'It was quite informative and delivered in an easy to absorb manner. Plus it applies real-world examples to the concepts'). Another salient theme involved the impact of the lived experiences shared by patient partners (*n* = 10; e.g., 'What I liked best was the patient feedback and sharing of experiences'.). Most participants commented that they did not have suggestions for improvements (*n* = 15), with the most common suggestions involving technical improvements (*n* = 8; e.g., 'The sound was not consistent and I had to keep making it louder or softer depending on the video'.), desiring more detailed information (*n* = 6; e.g., 'Suggestion of more tools that can be used with patient care') and suggesting more interactive exercises (*n* = 5; e.g., 'more activities to complete after each section'). Similarly, most participants commented that they did not have other suggestions for topics (*n* = 17); the most common suggestion related to wanting more details regarding trauma interventions (*n* = 10; e.g., 'I would like more practice-based examples of how to work with patients who have experienced trauma'). Last, most participants commented that they did not have additional input (*n* = 22), with the most common final comments involving general positive feedback (*n* = 16; e.g., 'Thank you for providing me with such great knowledge!').

Table 2 Main themes of open-ended responses (*N* = 46)

	n
What liked best	
Informative	12
Lived experiences	10
Trauma information	5
Easy to access	4
Visuals	4
Self-paced format	4

Trauma-informed care research	2
Applicability of information	1
Patient-centred focus	1
Different perspectives	1
Did not respond	2
How improve	
No suggestions	15
Technical improvements	8
More detailed information	6
Interactive content	5
Navigating Part 2 readings	3
Shorten	3
Other	3
Did not respond	3
Other topics	
No suggestions	17
Trauma interventions	10
Other relevant health topics	3
Involving patients in research	2
Mental health	2
Patient advocacy	2
Self-care	2
Other	3

Did not respond	5
Additional comments	
None	22
General positive comments	16
Learning how to get involved	2
Other	2
Did not respond	4

DISCUSSION

This article describes the development and evaluation of training about TIC and its application to the co-production of research by the patient and professional stakeholders. The experience of the *my*PATH partnership indicates that it is possible to integrate TIC principles into a research partnership's processes, and feedback from 46 training participants outside the partnership was largely positive. Training participants perceived that the learning objectives were met; they particularly valued the informative nature of the training and personal statements based on individuals' lived experiences. The inclusion of partners' lived experiences related to traumatic experiences and co-producing research seemed to help trainees relate better to the academic material or to see how the ideas could be realistically applied. The most salient suggestions for improvements related to technical matters and the desire for additional details, particularly related to trauma interventions. The primary limitations of the current evaluation include the relatively small number of participants, the lack of information regarding participants' backgrounds and the lack of data regarding real-world impacts, such as how the training may have impacted participants' work or collaborations. These limitations could be addressed in future research to evaluate the training with other research partnerships. It would be valuable to incorporate short-term and longer-term follow-up and to evaluate real-world impacts; hypothesized impacts of this training include improving the engagement of partners with lived experience of trauma in PPIE-based research partnerships, improving the functioning within such partnerships, increasing the number of partnerships that incorporate trauma into their research and improving how stakeholders interact with trauma survivors in their regular work.

In addition to future research on training, the *my*PATH partnership continues to apply these TIC-based research strategies to ongoing initiatives. Partners have reviewed and updated the governance document as new partners have been added, to emphasize the importance of thorough review before committing to joining the partnership. Regarding research endeavours, the partnership has conducted online surveys and qualitative interviews with professionals and patients regarding COVID-19, trauma and telehealth, and has submitted a research proposal involving TIC and other interventions for primary care patients with trauma. This proposal was informed by input from training participants and other professional and patient representatives in Florida through an online survey (N = 249). The proposed study design emphasized inclusion, such as applying broad inclusion criteria (posttraumatic stress symptoms, not only PTSD), including English- and Spanish-speaking patients, and including primary care clinics across different geographical regions that serve diverse patients (e.g., a range of racial and ethnic groups, urban/rural, LGBTQ+, older adults). We also have developed a listserv and quarterly newsletter including brief information about topics requested from the training feedback, such as trauma, trauma interventions, TIC and relevant research. The listserv and newsletter are also avenues to solicit feedback on future research products and plans.

CONCLUSION

To summarize, traumatic experiences and their impacts are very common among individuals with a wide range of physical and behavioural health conditions, and trauma is likely relevant to many partnerships that are co-producing health-related research, both in terms of the health condition being studied as well as personal relevance for many stakeholders who engage with the group. Our partnership developed training that reviews trauma, TIC and stakeholder-driven research, and that applies TIC principles to stakeholder-driven research. Training participants perceived that the learning objectives were met, and they seemed to appreciate the combination of academic and personal perspectives and desire additional training on these topics.

It is recommended that research partnerships consider learning more about trauma, how trauma relates to the health condition under study, TIC and how TIC principles could be applied to their partnership's work. Many research partnerships may already be implementing strategies that foster some TIC principles, given their overlap with general principles of research co-production. For example, PCORI has identified four categories of engagement principles: reciprocal relationships (defining roles and decision-making collaboratively); co-learning (about the research process, content and research engagement); partnerships (fairness in compensation and expectations, commitment to diversity and cultural competence) and transparency, honesty and trust.[53] Several of these principles overlap with TIC, such as fostering collaboration and empowerment of all stakeholders, building transparency and trustworthiness and attending to cultural diversity and competence. TIC principles also include explicitly attending to safety, such as discussing how trauma will be conceptualized and discussed among the partners and paying special attention to the empowerment of all partners, including recognizing the blurred boundaries between ‘patient’ and ‘professional’ partners regarding traumatic experiences and impacts.

There are numerous resources for learning more about trauma and TIC, including seminal SAMHSA publications[24] and the Center for Health Care Strategies' TIC Implementation Resource Center.[46] The TIC Implementation Resource Center is focused on implementing TIC in healthcare settings and has a wide range of resources, ranging from introductory material to detailed implementation guidance. To learn more about strategies for applying TIC principles to research co-production, the *my*PATH strategies (Figure 1) and training is the only resource of which we are aware. One article was recently published (after our training) that described the application of TIC to a research advisory board of women who had experienced intimate partner violence,[42] describing similar strategies as *my* PATH.

For interested research partnerships, it is recommended to begin discussing trauma in a general sense, and then if the partnership agrees, preview training options, select training and other resources together, participate in selected training together and then discuss how the content and strategies relate and could be incorporated into their partnership's processes and work. Potential benefits of implementing the *my*PATH strategies or learning more about trauma and TIC, in general, include a greater sense of safety, empowerment, mutual understanding and engagement among stakeholders involved in co-producing research, as well as producing research that is more relevant to patients and providers in the real world, by addressing the impacts of trauma for the health condition under study.

AUTHOR CONTRIBUTIONS

Amber M. Gum, PhD, directs the *my*PATH Partnership and led the development of the partnership, training, training evaluation and preparation of this manuscript. Mary Goldsworthy, MPH, is a *my*PATH Partner and was a Project Coordinator who facilitated partnership development, contributed significantly to the preparation of the training content and presentations, conducted qualitative analyses for this manuscript and reviewed a draft manuscript. Lucy Guerra, MD, MPH, is a *my*PATH Partner who contributed significantly to the development of the partnership, contributed significantly to the preparation of the training content and presentations and reviewed a draft manuscript. Alison Salloum, PhD, is a *my*PATH Partner who contributed significantly to the development of the partnership, contributed significantly to the preparation of the training content and presentations and reviewed a draft manuscript. Meredith Grau, MS, is a *my*PATH Partner who contributed significantly to the development of the partnership, contributed significantly to the preparation of the training content and presentations and reviewed a draft manuscript. Sheri Gottstein, BA, RN, is a *my*PATH Partner who contributed significantly to the development of the partnership,

contributed significantly to the preparation of the training content and presentations and reviewed a draft manuscript. Carol Horvath, is a *my*PATH Partner who contributed significantly to the development of the partnership, contributed significantly to the preparation of the training content and presentations and reviewed a draft manuscript. Annanora Fields, BA, is a *my*PATH Partner who contributed significantly to the development of the partnership and reviewed a draft manuscript. Johnny Crowder, BA, is a *my*PATH Partner who contributed significantly to the development of the partnership, contributed significantly to the preparation of the training content and presentations and reviewed a draft manuscript. Robb Holley, MD, is a *my*PATH Partner who worked as a Project Coordinator and facilitated partnership development and infrastructure and reviewed a draft manuscript. Leigh J. Ruth, MD, is a *my*PATH Partner who contributed significantly to the development of the partnership and reviewed a draft manuscript. Karim Hanna, MD, is a *my*PATH Partner who contributed significantly to the development of the partnership and reviewed a draft manuscript.

ACKNOWLEDGEMENTS

The *my*PATH Partnership's capacity-building efforts and training development and evaluation were supported by contracts from the Patient-Centered Outcomes Research Institute (PCORI contracts 17443, 7709601). The views expressed in this manuscript and other *my*PATH resources are solely the authors' views; they do not necessarily represent the views of PCORI or the authors' affiliated organizations.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

De-identified data from the evaluation results are available from the first author upon request.

DETAILS

Subject:	Collaboration; Computer assisted instruction--CAI; Patients; Trauma; Health status; Training; Professional training; Health research; Primary care; Post traumatic stress disorder; COVID-19; Stakeholders; Continuing education; Alliances; Credit; Health behavior; Medical research; Online instruction; Research partnerships; Child abuse &neglect; Respiratory diseases; Governance; Coronaviruses; Evaluation research; Diabetes; Personal experiences; Activities of daily living; Distance learning; Interest groups; Sex crimes; Health care; Partnerships; Pandemics; Empowerment; Cardiovascular disease; Systematic review; Principles
Business indexing term:	Subject: Stakeholders
Identifier / keyword:	co-production; participatory research; patient and public involvement and engagement; stakeholder-driven research; training; trauma; trauma-informed care
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	388-398
Publication year:	2023
Publication date:	Feb 2023

Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-16
Milestone dates:	2022-11-08 (publishedOnlineAccepted); 2022-10-17 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-07-28 (manuscriptReceived); 2022-11-16 (publishedOnlineEarlyUnpaginated); 2022-11-06 (manuscriptAccepted)
Publication history :	
First posting date:	16 Nov 2022
DOI:	https://doi.org/10.1111/hex.13668
ProQuest document ID:	2767101848
Document URL:	https://www.proquest.com/scholarly-journals/trauma-informed-patient-public-engaged-research/docview/2767101848/se-2?accountid=211160
Copyright:	
Last updated:	2023-10-17
Database:	Public Health Database,Publicly Available Content Database

Stakeholder outcome prioritization in the Biologic Abatement and Capturing Kids' Outcomes and Flare Frequency in Juvenile Spondyloarthritis (BACK-OFF JSpA) trial

Neu, Emily [1] ; Sears, Cora [1] ; Brandon, Timothy [1] ; Kohlheim, Melanie [2] ; Leal, Jenny [2] ; Kweli Archie [2] ; English Holland [2] ; Holland, Miles [2] ; Hameed, Aamena [2] ; Khan, Asad [2] ; Murphy, Lynn [2] ; Murphy, Sean [2] ; Neu, Antoinette [2] ; Neu, Jerome [2] ; Neu, Justin [2] ; Richmond, Rachel [2] ; Suplee, Dylan [2] ; Suplee, Theresa [2] ; Forrest, Christopher B [2] ; Weiss, Pamela F [3] [1] Department of Pediatrics, Division of Rheumatology, Clinical Futures: A CHOP Research Institute Center of Emphasis, Children's Hospital of Philadelphia, Philadelphia, Pennsylvania, USA [2] BACK-OFF JSpA Research Partners Group, Children's Hospital of Philadelphia, Philadelphia, Pennsylvania, USA [3] Department of Pediatrics, Division of Rheumatology, Clinical Futures: A CHOP Research Institute Center of Emphasis, Children's Hospital of Philadelphia, Philadelphia, Pennsylvania, USA; Department of Pediatrics and Epidemiology, Perelman School of Medicine, University of Pennsylvania, Philadelphia, Pennsylvania, USA

ABSTRACT (ENGLISH)

Background

The Biologic Abatement and Capturing Kids' Outcomes and Flare Frequency in Juvenile Spondyloarthritis (BACK-OFF JSpA) study is a randomized, pragmatic trial investigating different tumour necrosis factor inhibitor de-escalation strategies for children with sustained inactive disease. In this project, we elicited concept rankings that aided in the selection of the patient-reported outcome (PRO) measures that should be examined as part of the BACK-OFF JSpA trial.

Methods

We conducted a discrete choice experiment to evaluate individuals' preferences regarding PROs. Stakeholders assessed a discrete list of 21 outcome concepts, each of which had a Patient-Reported Outcome Measurement Information System (PROMIS) measure associated with it. PROMIS measures are self- or proxy-reported instruments that are universally applicable to the general population and all chronic conditions. Stakeholders were required to make choices instead of expressing the strength of a preference.

Results

Fourteen caregivers, 12 patients (9–22 years old), 16 rheumatologists and three executives from health insurance companies completed the exercise, which took approximately 10 min. The discrete choice experiment resulted in an estimate of the relative importance of each outcome and rank. All stakeholder groups agreed that the primary PRO should be 'Pain Interference', a measure that evaluates the effect of pain on a child's everyday activities, including its impact on social, emotional, mental and physical functioning. Patients and caregivers were mostly aligned in their top priorities, with patients valuing physical health (50% of the top 10) whereas caregivers were more interested in mental health (60% of the top 10). Rheumatologists and health insurance executives were most interested in physical health outcomes, which were ranked 80% and 60% of their top 10 PROs, respectively. Overall, the patients had the most diverse set of prioritized outcomes, including at least one of each category in their top 10 rank order of importance. Patients were also the only stakeholders to prioritize 'social' health.

Conclusions

Patients and caregivers were mostly aligned in their outcome priority rankings. The rank-order list directly informed

the creation of a profile of PRO measures for our upcoming trial.

Patient or Public Contribution

Stakeholder partners helped with acquisition of data and lead parent partners helped interpret data.

FULL TEXT

BACKGROUND

In 2018, an international task force of pediatric rheumatologists developed recommendations for treating juvenile arthritis to target.[1] The primary treatment target was the inactive disease, defined as the absence of all clinical signs and patient-experienced symptoms of inflammatory disease activity. Additionally, the international task force specified several overarching principles for the management of juvenile arthritis which included not only controlling signs and symptoms of disease but also avoidance of drug toxicities and optimization of personal well-being. Since the introduction of biologic disease-modifying agents such as tumour necrosis factor inhibitors (TNFi), the inactive disease is a feasible target for children with spondyloarthritis, which accounts for up to 30% of juvenile arthritis. In fact, current treatment approaches for children with spondyloarthritis have resulted in up to 60% attaining inactive disease while on therapy.[2–4]

However, there is no information to inform decisions regarding tapering (increasing the time between doses) or stopping TNFi after the inactive disease is achieved. The Biologic Abatement and Capturing Kids' Outcomes and Flare Frequency in Juvenile Spondyloarthritis (BACK-OFF JSpA) Trial is a randomized pragmatic trial that will improve the evidence base that patients, caregivers and rheumatologists use to make shared decisions about continued treatment versus de-escalation of therapy in children with spondyloarthritis who have the inactive disease. Unless there is high-quality and unbiased evidence on TNFi de-escalation experiences, pediatric patients with spondyloarthritis and their caregivers will not be able to make decisions that take into account the outcomes that are most important to them. In fact, the Outcome Measures in Rheumatology (OMERACT) updated core domain set for studies in juvenile idiopathic arthritis includes patient-reported outcomes (PROs) like pain, physical function and patient perception of disease activity.[5] Timing and risk of flare is critical for patients and caregivers to know when making informed decisions about potential de-escalation of TNFi therapy. The BACK-OFF JSpA trial will not only determine the timing and risk of disease flare but also the lived experiences of patients undergoing the various treatment strategies. The growing importance of the patients' lived experiences in clinical research such as the BACK-OFF JSpA trial is underscored by the US government's Patient Protection and Affordable Care Act that created the Patient-Centered Outcomes Research Institute,[6] the creation of the NIH Patient-Reported Outcomes Measurement Information System® (PROMIS®) initiative,[7] and the Food and Drug Administration's mandate to use these assessments for medical product labelling claims.[8]

Prior mixed-methods work from Horton and colleagues has shown that when making decisions about stopping medication, caregivers and patients with juvenile arthritis consider the risk from both the disease and treatment.[9] Participants emphasize the importance of how their underlying arthritis and treatments aided or hindered with a sense of 'normalcy' and safety and also the uncertainty regarding risk of future treatment effects/harms and risk of flare. Ultimately, participants' decisions were informed by trust in their physician and alternate sources of information including social media. In another study consisting of web-based surveys and focus groups, research themes prioritized by patients and caregivers of children with rheumatic disease included disease flare and medication side effects.[10] A study by Moser and colleagues underscored the importance of considering relevant PRO measures over the course of juvenile arthritis.[11] Additionally, several key concepts were elucidated: in this study (1) youth did not feel adequately informed about the purpose of collecting or value of patient-reported outcomes, (2) assessments used during routine care—in particular those related to function—were outdated and not pertinent to current issues and (3) youth should be involved in the development/selection of instruments to ensure relevance.

The BACK-OFF JSpA trial team includes a Research Partners Group consisting of members from across the United States who are JSpA patients, caregivers, foundation representatives, payor partners and an adult and pediatric rheumatologist. The Research Partners Group participated in a family studio to help the investigator team

understand the trial design preferences of patients and parents within the juvenile SpA population. The family studio provided Research Partners Group members with an opportunity to offer suggestions and voice concerns based on their own experiences. The exercise reported herein aimed to capture the PROs most important to the patient and caregiver stakeholders for inclusion as the primary and secondary outcomes of the second aim of the BACK-OFF JSpA trial.

METHODSSubjects

This was a prospective cross-sectional study of juvenile SpA stakeholders. Subjects were a convenience sample including the BACK-OFF JSpA Research Partner Group members (patients, caregivers, foundation and payor representatives), patients and caregivers of children with SpA being treated at the Children's Hospital of Philadelphia, and site investigators for the BACK-OFF JSpA trial. Eligibility requirements for patient and caregiver stakeholders included either membership on the BACK-OFF JSpA Research Partners Group or a patient or caregiver of a patient fulfilling all of the following: (1) Diagnosis of juvenile SpA, (2) age 7 years or above, (3) current treatment with a TNFi and (4) evaluated in a rheumatology clinic at the Children's Hospital of Philadelphia in January 2021. Rheumatologists were either members of the BACK-OFF JSpA Research Partners Group or a Site Investigator for the BACK-OFF JSpA trial who treats patients with JSpA. National organization (foundation) stakeholders were active organization members that advocate and support individuals living with JSpA. Similarly, payor partner stakeholders were insurance company representatives that provide medical coverage for patients with JSpA. There was no compensation for participation in the survey. The Children's Hospital of Philadelphia Committee for the Protection of Human Subjects approved the protocol for the conduct of this study (IRB 20-018224) and consent or assent, as appropriate, was obtained from all participants.

Patient-reported outcomes

We aimed to have participants choose the primary and secondary PROs of most importance from a list of 21 health and wellbeing concepts, each of which had an associated PROMIS tool (https://www.healthmeasures.net/explore-measurement-systems/promis/intro-to-promis/list-of-pediatric-measures). PROMIS measures are self- or proxy-reported instruments designed for use in the general population and across all chronic conditions.[12] Each PROMIS domain is composed of a collection of items called an ‘item bank’ which encompasses the full range of the latent variable being evaluated. A ‘short form’ is a selection of items that represent the item bank with fewer questions, typically four to eight items. Short forms can be easily administered on mobile devices and are easy to complete. Youths 8–17 years of age can complete self-report instruments and caregivers can complete parent proxy-report instruments. The PROMIS short-forms have been validated in children with juvenile arthritis.[13]

Discrete choice experiment

The discrete choice experiment was pilot tested with local pediatric rheumatologists and research assistants and several stakeholder parent partners. The survey was administered electronically (iPad, smartphone, or computer) using Sawtooth Software either in the office at the time of rheumatology assessment or remotely through emailed invitation.[8] The experiment was a user-friendly, quantitative, choice-based approach to evaluate individuals’ preferences regarding the importance of potential PROs. Sawtooth Software uses empirical Bayes to conduct relative comparisons among the items in the study and provide individual-level score estimates.[14] With this software, stakeholders assessed 21 outcomes. If both a caregiver and patient were completing the survey, they were instructed to complete it independently. In preparation for the exercise caregivers and patients were provided with a list of the outcomes under consideration and an explanation of what each measure.

Outcome concepts were drawn from physical, mental, social and global health dimensions (Table 1).

Table 1 Pediatric outcomes by category

Dimension	Outcome
Global	Global health

Mental	Life satisfaction Cognitive function Sense of life's meaning Depressive symptoms Stress Anxiety symptoms Positive mood Angry mood or irritability
Physical	Pain interference Mobility Pain behaviours Physical activity Upper extremity function Fatigue Impact on strength activities Sleep-disturbance Sleep-related daytime impairment Physical responses to stress
Social	Family relationships Peer-relationships

Note: All domains are measurable by PROMIS pediatric instruments.

Rather than asking stakeholders to rate all items at once, three outcome concepts were presented at a time (Figure 1). Within each set, stakeholders were instructed to 'Please choose what you feel are the most and least important outcomes to consider for youth with spondyloarthritis as medication is managed and potentially changed'. This process was repeated for 21 unique outcome combinations, with each outcome being shown three times. Stakeholders were required to make choices instead of expressing the strength of a preference (as would be done with a Likert scale or Delphi rating process). Using this process left no opportunity for scale use bias, where respondents often rate different attributes similarly.

Enlarge this image.

RESULTS

Fourteen caregivers, 11 patients (ages 9–22 years old), 16 rheumatologists and three executives from health

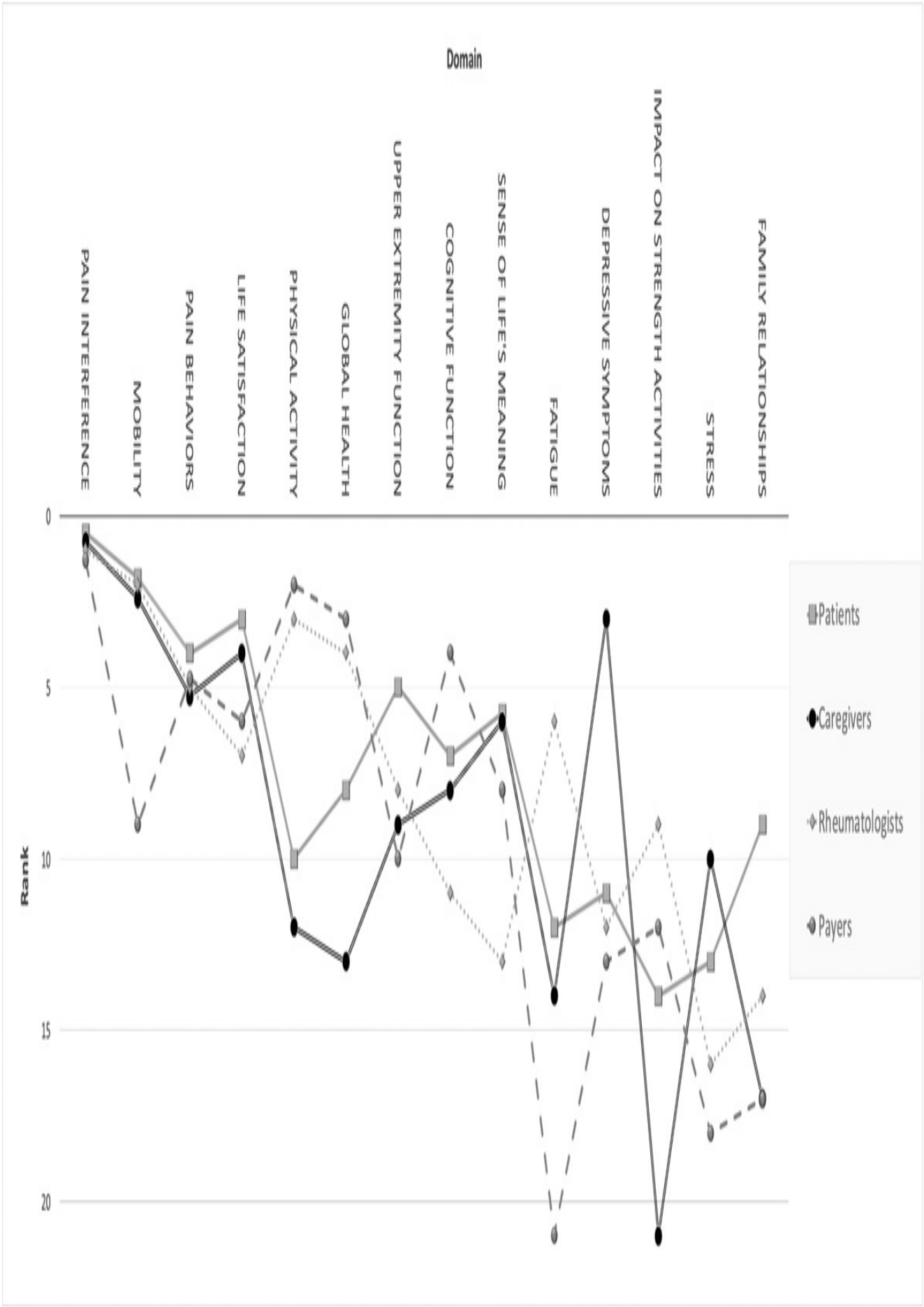
insurance companies completed the exercise in a median time of 10.2 min (interquartile range: 7.3–14.0 min). For patient stakeholders (*n* = 11), the average age was 16 years, 91% were White and seven identified as male and four as female. Half of the rheumatologists identified as female, while all of the payor representatives (*N* = 3) identified as male. Caregivers were predominately female (13 females, one male) and were the most racially diverse group of stakeholders with 21% of respondents reporting their race as non-White.

The discrete choice experiment resulted in an estimate of the relative importance of each outcome and rank (Table 2 , Figure 2). All stakeholder groups agreed that the PROMIS Pain Interference measure was the most important to consider during TNFi therapy de-escalation. This measure evaluates the effect of pain on a child or adolescent's everyday activities, including its impact on social, psychological and physical functioning. All groups except for payors ranked Mobility as the second most important outcome. Payors ranked Mobility ninth.

Table 2 Stakeholder estimates of relative importance

Pediatric domains	Relative importance				
	All	Patients	Caregivers	Rheumatologists	Payors
Pain interference	10.9	9.8	11.2	11.4	10.9
Mobility	9.2	8.9	8.4	10.6	5.8
Pain behaviours	7.2	7.1	6.4	8.0	7.1
Life satisfaction	6.9	8.3	6.5	6.1	7.1
Physical activity	6.5	4.8	4.3	8.7	10.5
Global health	6.5	5.8	4.1	8.6	7.7
Upper extremity function	5.6	6.1	4.9	6.0	5.3
Cognitive function	4.8	5.8	5.2	3.4	7.4
Sense of life's meaning and purpose	4.8	5.9	6.4	2.5	5.9
Fatigue	4.8	3.9	4.1	6.7	0.3
Depressive symptoms	4.6	4.1	6.7	3.3	3.8
Impact on strength activities	3.4	3.2	1.8	4.7	3.9
Stress	3.4	3.4	4.9	2.5	1.6
Family relationships	3.2	5.5	2.6	2.5	1.8
Anxiety symptoms	3.0	3.2	5.4	1.4	1.1

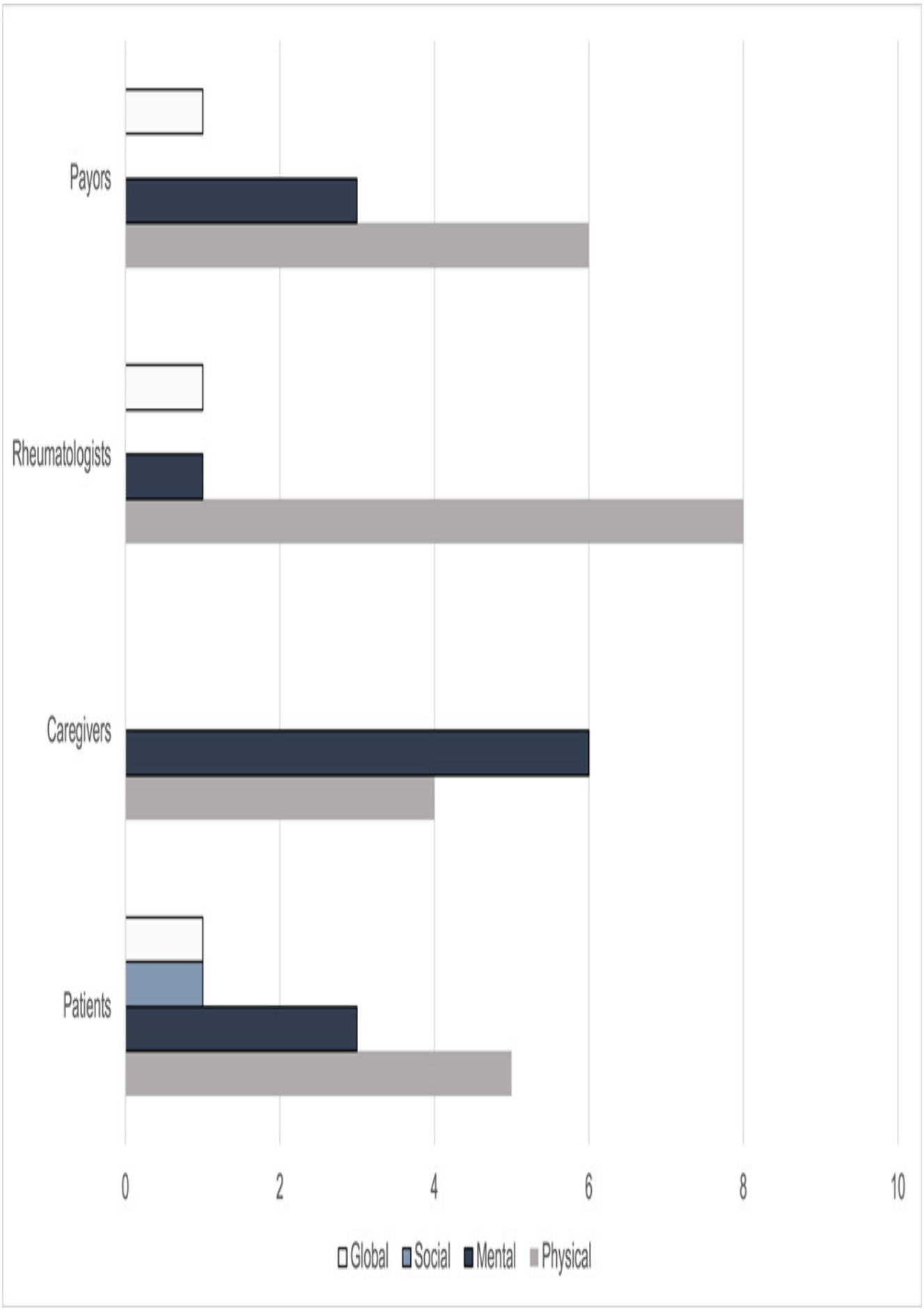
Positive mood	3.0	3.0	4.3	2.0	2.0
Sleep-disturbance	2.7	1.5	2.5	3.6	4.0
Peer-relationships	2.6	2.9	3.0	1.8	3.4
Sleep-related daytime impairment	2.5	1.9	2.1	2.5	6.5
Physical responses to stress	2.5	2.5	3.0	1.8	3.2
Angry mood or irritability	2.0	2.4	2.2	1.8	0.6



Enlarge this image.

After Pain Interference and Mobility, patients' next priority was Life Satisfaction. The top 10 pediatric domain categories by stakeholder type are shown in Figure 3. Overall, patients highly valued physical health, with 50% of

their top 10 categorized as physical health outcomes. Of all stakeholder groups, the patients selected the most diverse set of outcomes, which included at least one outcome from global, mental, social and physical health in their top 10 rank order of importance. Patients were the only stakeholder group to prioritize an outcome (family relationships) from the social health category.



Enlarge this image.

Caregivers highly valued mental health, with six mental health outcomes occupying the top 10. After Pain Interference and Mobility, the next outcomes prioritized by this group were Depressive Symptoms, Life satisfaction

and a Sense of Life's Meaning. Rheumatologists and payors valued physical health the most, which included 80% and 60% of their top 10, respectively. Following Pain Behaviours and Mobility, rheumatologists ranked Physical Activity and Global Health as the most important.

DISCUSSION

In this study, we asked patients, caregivers, rheumatologists and payor partners to rate the importance of specific patient-reported outcomes that were being considered for an upcoming trial evaluating three therapy de-escalation strategies for children with SpA who have achieved sustained inactive disease for at least 6 months. We found that the top priority for patients, caregivers and rheumatologists was Pain Interference, which measures the impact of pain on multiple dimensions of functioning and as per PROMIS classification, it was assigned to physical health. Of the stakeholder groups, patients were the only ones to include at least one outcome from each of the four dimensions in their top 10 rank order of importance. This underscores the diversity of outcome preferences among patients. Conversely, the rheumatologists were fairly narrow in their view, rating multiple physical health outcomes highly. These results emphasize the importance of including multiple perspectives in outcome prioritization and ultimately assessment, which aligns with the updated JIA Core Domain Set following the OMERACT methodology.[5] Our results also underscore findings from a recent qualitative study of youth with juvenile arthritis that concluded that youth must be involved in outcome choice to ensure relevance.[11]

This study has both strengths and limitations. The strengths include stakeholders from pediatric academic centres from across the United States, a relatively efficient data collection process that allowed respondents to complete the survey in about 10 min, and very few missing data points. A limitation of the study is the relatively small sample size, especially when stratified by stakeholders which limits the generalizability of these findings. It is possible that patients and parents who are not members of the BACK-OFF JSpA Research Partners Group or survey nonrespondents have different values and perspectives on what PROs are most important to consider in a de-escalation trial. However, in the updated JIA Core Domain Set[5] which included a large international sample of stakeholders, pain, physical health and overall well-being was voted as mandatory outcomes in all trials and consistent with our findings. Finally, most but not all BACK-OFF JSpA site investigators or Research Partner Group members completed the survey. It is unknown what differences, if any, exist between survey respondents versus nonrespondents.

With these strengths and limitations in mind, our findings underscore the importance of stakeholder involvement in study design. Patient and caregiver stakeholders are an integral part of the investigative team for the BACK-OFF JSpA trial. If patients and caregiver stakeholders had not done this exercise, the PRO measures that would have been investigated, based upon physician prioritization, would not have been in direct alignment with what the patients and caregivers are truly most interested in. Specifically, if the PRO profile was developed with input only from clinicians the profile would still have included pain interference, mobility and pain behaviours—albeit in a slightly different priority order- however global health would have been included rather than life satisfaction. Since PROs are now part of the updated OMERACT JIA core set,[5] a strong argument could be made that all trials in JIA should incorporate input from patients, caregivers and clinicians into the design and/or conduct of the study. Depending upon the question being studied in each trial or study, the PROs prioritized by alternative stakeholder groups are likely to differ. As it relates to the BACK-OFF JSpA trial if the risk of disease flare is only marginally different between the treatment strategies being studied, differences in PROs could be tremendously informative for shared decision-making regarding which strategy patients and caregivers will ultimately prefer.

Our findings highlight the importance of collecting patient and caregiver preferences on study questions during the planning stages of a trial. The rank-order list from the patient and parent caregiver stakeholders from this exercise directly informed the primary and secondary PROs for the upcoming BACK-OFF JSpA trial with the primary patient-reported outcome being Pain Interference and the secondary outcomes being Mobility, Life Satisfaction and Pain Behaviours. Further, we need to learn how these outcomes ultimately influence stakeholders' interpretation of the results of the upcoming trial and their subsequent point-of-care therapy de-escalation preferences. Our study was designed specifically for the JSpA population being treated with a TNFi who would be potentially eligible for trial

enrolment to evaluate therapy de-escalation. Therefore, our results may not generalize to all juvenile arthritis trials. However, the ease with which this exercise was conducted and our results underscore that similar exercise(s) can, and should be completed for trials of patients with juvenile arthritis at the design phase so that measures and outcomes that are both relevant and highly valued to the patient population under study are included.

AUTHOR CONTRIBUTIONS

Pamela F. Weiss, Cora Sears and Timothy Brandon helped with the design of work, analysis, interpretation of data and drafting of the manuscript. All authors helped with the acquisition of data. Emily Neu, Melanie Kohlheim and Jenny Leal helped with interpretation of the data. Christopher B. Forrest helped with the design of work. All authors approved the submitted version of the manuscript.

ACKNOWLEDGEMENTS

This study was funded through a Patient Centered Outcomes Research Institute® (PCORI®) Award (CER-2020C1-19212). The study design, analysis and interpretation of results presented in this publication are solely the responsibility of the authors and do not necessarily represent the views of the Patient-Centered Outcomes Research Institute® (PCORI®), its Board of Governors or Methodology Committee.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

This study's protocol was reviewed and approved by the Children's Hospital of Philadelphia's (IRB 21-018442) Committees for the Protection of Human Subjects.

DETAILS

Subject:	Exercise; Mental health; Physical fitness; Task forces; Inactive; Chronic conditions; Hospitals; Health status; Families &family life; Health insurance; Tumors; Arthritis; Physical ability; Pain; Tumor necrosis factor; Pediatrics; Evaluation; Rheumatology; Measurement; Stakeholders; Experiments; Decision making; Chronic pain; Children &youth; Rheumatic diseases; Caregivers; Children; Software; De-escalation; Patient Protection &Affordable Care Act 2010-US; Rheumatologists; Functional impairment; Clinical outcomes; Inflammatory diseases; Patients; Chronic illnesses; Insurance; Females; Discrete choice; Ratings &rankings; Necrosis
Business indexing term:	Subject: Stakeholders Patient Protection &Affordable Care Act 2010-US
Location:	United States--US
Company / organization:	Name: Partners Group; NAICS: 523920; Name: Childrens Hospital-Philadelphia PA; NAICS: 622310; Name: Sawtooth Software; NAICS: 513210
Identifier / keyword:	patient-reported outcomes; spondyloarthritis; stakeholder; trial; tumour necrosis factor inhibitors
Publication title:	Health Expectations; Oxford
Volume:	26

Issue:	1
Pages:	290-296
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-17
Milestone dates:	2022-06-27 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-02-04 (manuscriptReceived); 2022-11-17 (publishedOnlineEarlyUnpaginated); 2022-10-21 (manuscriptAccepted)
Publication history :	
First posting date:	17 Nov 2022
DOI:	https://doi.org/10.1111/hex.13655
ProQuest document ID:	2767101841
Document URL:	https://www.proquest.com/scholarly-journals/stakeholder-outcome-prioritization-biologic/docview/2767101841/se-2?accountid=211160

| Last updated: | 2023-07-11 |

Women's experiences along the ovarian cancer diagnostic pathway in Catalonia: A qualitative study

Vela-Vallespín, Carmen [1]
; Medina-Perucha, Laura [2]
; Jacques-Aviñó, Constanza [2]
; Codern-Bové, Núria [3]
; Harris, Meggan [4]
; Borras, Josep M [5]
; Marzo-Castillejo, Mercè [6]

[1] Primary Health Care Center Riu Nord i Riu Sud, Catalan Health Institut, Barcelona, Spain; Research Support Unit Metropolitana Nord, University Institute for Primary Health Care Research (IDIAP) Jordi Gol, Catalan Health Institute, Barcelona, Spain [2] Unitat Transversal de la Recerca, Fundació Institut Universitari per a la Recerca a l'Atenció Primària de Salut Jordi Gol i Gurina (IDIAPJGol), Barcelona, Spain; Unitat Transversal de la Recerca, Universitat Autònoma de Barcelona, Barcelona, Spain [3] Department of Nursing, Escola Universitària d'Infermeria i Teràpia Ocupacional de Terrassa, Universitat Autònoma Barcelona, Barcelona, Spain; Evaluation and Qualitative Research, ÀreaQ, Barcelona, Spain [4] Independent Researcher, Valencia, Spain [5] Department Clinical Sciences, Bellvitge Biomedical Research Institute (IDIBELL), University of Barcelona, Barcelona, Spain [6] Research Support Unit Metropolitana Sud, University Institute for Primary Health Care Research IDIAPJordi Gol, Barcelona, Spain

ABSTRACT (ENGLISH)

Background

Early detection of symptoms and prompt diagnosis of ovarian cancer are considered important avenues for improving patient experiences and outcomes.

Methods

This qualitative study used a phenomenological approach to perform patient interviews, collecting individual accounts of the prediagnostic phase in women diagnosed and treated for ovarian cancer in 2016–2017. Purposive sampling was used to obtain a diverse sample of 24 participants, while thematic content analysis was used to extract themes and subthemes from interview data.

Results

Three themes and nine subthemes were identified. The first theme was women's delay in recognizing symptoms and seeking care, with subthemes on the lack of knowledge about early signs of ovarian cancer, gender-related barriers and false reassurance from negative test results. A second theme was missed opportunities during healthcare encounters, due to misattribution of women's symptoms by their physicians, underestimation of symptom severity and need for mediation and inadequate tests and/or false negative results. Finally, interviews highlighted the use of resources and alternative healthcare pathways, including complementary/alternative medicines, access to

private health care and women's capacity for action and decision-making (agency) about their health.

Conclusion

Delayed diagnosis of ovarian cancer is rooted in both individual factors (lack of health literacy, reluctance to seek care) and systemic issues (missed opportunities in healthcare encounters, access to timely specialist care). Further research is needed to investigate the extent to which traditional gender roles and socioeconomic inequalities condition women's ability to manage their own health and to interact with health professionals and the health system.

Patient and Public Contribution

In addition to the patient participation during the interviews, one author was a representative of a patient association.

FULL TEXT

INTRODUCTION

Ovarian cancer is the eighth most common tumour in Europe and the gynaecological tumour with the highest mortality.[1] Estimates of age-standardized 5-year net survival generally range from 30% to 50%, figures that have held steady over the past two decades.[2] Tumour stage at diagnosis is an important factor determining the patients' survival, which is threefold higher in women diagnosed at Stage I compared to Stages III–IV. Unfortunately, most women and other people with ovaries are diagnosed with Stage III or Stage IV cancer.

Ovarian cancer develops mainly in women aged 55 years or older. Genetic factors (BRCA mutations) greatly increase the risk,[3] while other determinants include age, obesity, first pregnancy after age 35 and nulliparity. In contrast, breastfeeding and oral contraceptives have a protective effect, especially the longer the pills are used.[3] Given the survival benefits of early diagnosis and the absence of any effective screening test for ovarian cancer,[4] focusing on detecting symptomatic cases as soon as possible may improve the odds of early diagnosis and successful treatment. However, the symptoms of ovarian cancer can vary from person to person, and these can be decisive for diagnosis.[5] Ovarian cancer most commonly presents as vague and nonspecific abdominopelvic and urinary symptoms, and women often interpret these as normal changes associated with ageing, menopause or stress.[6–8]

The Model of Pathways to Treatment is a conceptual framework for understanding diagnostic and treatment pathways in people with symptomatic cancer.[9] It identifies five key events in the pathway to care: detection of bodily changes, perceived reasons to discuss symptoms with a health care provider, first consultation with a health care provider, diagnosis and start of treatment. The four intervals between these events are defined as the appraisal, help-seeking, diagnostic and pretreatment intervals. The patient interval, encompassing the appraisal and help-seeking intervals, is one of the most important sources of diagnostic delay.[10]

Systematic reviews identify symptom knowledge, interpretation of symptoms as cancer-related, and beliefs about cancer as three (likely universal) predictors of help-seeking.[10,11] Individuals with lower literacy and socioeconomic levels often have lower symptom knowledge and more fatalistic beliefs about cancer.[11] Additionally, gender appears to be an important barrier to help-seeking and delayed cancer presentation.[11,12] The World Health Organization (WHO)[13] points out that gender norms, socialization, roles and differences in power relations contribute to differences in perceiving diseases, in health behaviours and in access to health services. However, the available systematic reviews show that most studies focus on breast cancer, while the evidence for ovarian cancer remains relatively sparse.[10]

The estimated interval from first noticing ovarian cancer symptoms to receiving a diagnosis varies widely by country.[14] Delays between the first consultation with symptoms and the diagnostic confirmation and treatment initiation are broadly attributed to the general practitioner (GP) and the healthcare system.[15] The cancer diagnostic process is often complex, involving different levels of care and it varies significantly with different healthcare models.[15] Gatekeeper systems have been associated with better quality of care but also with longer diagnostic intervals.[16,17] An audit of 513 women diagnosed with ovarian cancer in 2013–2014 in Catalonia confirms that long diagnostic intervals are also the norm in this setting, but it did not show an impact on 5-year survival.[18] Nevertheless, shortening the

interval in ovarian cancer diagnosis remains a key goal for improving quality of care, women's experiences and psychological well-being[19] and cancer outcomes.[20,21]

In recent years, qualitative research has emerged as a useful method for an in-depth exploration of the cancer diagnostic pathway. In our setting, few studies have assessed how women experience ovarian cancer before diagnosis. A phenomenological approach offers the opportunity to effectively capture patterns of meaning from their accounts. The aim of our study is to understand women's experiences of ovarian cancer diagnosis and their interactions with the healthcare system to identify avenues for improving care at the prediagnostic stage in people with ovarian cancer in Catalonia.

METHODSStudy design and setting

To gain a comprehensive insight into women's experiences of the ovarian cancer diagnostic process, a descriptive qualitative exploratory study was conducted using in-depth, semistructured, individual interviews, underpinned by a phenomenological approach. Phenomenology aims to explain how individuals give meaning to social phenomena through their lived experience, using a rigorous description of experiences and their detailed analysis to understand how these meanings are created.[22] The present study was conducted according to the criteria for reporting qualitative research (COREQ).[23]

This study was carried out in public-sector primary health care in Catalonia. The Catalan Health Service is a national health system model. Primary health care is the gatekeeper to specialist services; however, users may directly present to the emergency department and to sexual health and reproductive care centres (known as ASSIR clinics according to the Catalan acronym). The ASSIR clinics, usually located within primary healthcare centres, follow a one-stop-shop approach, bringing together family planning, prenatal care and preventive and health promotion activities, as well as diagnosis, treatment and follow-up of gynaecological pathologies, including cancer. Around 25% of Catalan public health care users also have private health insurance.[24]

Study participants and recruitment

The sampling frame for patients consisted of women diagnosed with primary ovarian cancer in 2016–2017 who had completed the first phase of treatment with a curative intent (cytoreduction plus chemotherapy) in the Catalan public healthcare system. Participation was on a voluntary basis. Purposive sampling was used to ensure discursive diversity of the participants' characteristics: age, educational level, occupation, geographical residence and hospital level.[25] These characteristics were used to construct 12 participant profiles, and the sample size was estimated at 24 participants, 2 for each discourse profile. A total of 29 women were recruited by general gynaecologists, oncologists and GPs based on their perceived interest. The interviewer called the women, explained the study objectives and researchers' role and set an interview date. Twenty-four agreed to participate, four did not meet inclusion criteria and one refused due to scheduling conflicts. Data saturation was reached with a sample size of 24 participants.

Women's age ranged from 40 to 77 years. Five had university studies, and 13 had stopped their schooling at the primary level. Fourteen lived in urban areas, while five were from rural areas.

As for their medical history, nine women had a family history of cancer, including one who carried the BRCA mutation. Two thirds of the women had regular gynaecological check-ups (ASSIR or private) for routine preventive care or for benign pathologies like ovarian cysts, myomas or endometriosis. The diagnostic intervals ranged from 10 days to 12 months. Most were diagnosed in the private setting, seven through their GP and six in the emergency department. See Table 1 for further details on participant characteristics.

Table 1 Patient characteristics

Case	Age	Highest education obtained	Residencea	Gynaecological history	Gynaecological history	Health service entry point	Months to diagnosis

1	51	University	City	No	No	GP	3
2	51	University	City	Myomas, HPV, regular check-ups	Sister—ovarian cancer	Gyno (pub.)	3
3	75	University	City	Ovary removal, 1996; check-ups every 2–3 years	No	GP	2
4	77	Primary	Village	No	Mother died of cancer; sister—ovarian cancer; sister and son—brain cancer	GP	12
5	70	Primary	Village	No	Maternal grandmother—ovarian cancer; paternal side—several cancers	ED	10–12
6	50	Secondary	Village	Ovarian cyst; check-ups in public healthcare every 6 months	No	Gyno (pub.)	8
7	59	University	Town	Endometriosis, 1990; check-ups every 2–3 years	Father and brother—died of cancer	ED	2
8	68	Secondary (Year 10)b	Town	Myomas, hysterectomy (age 37); priv. check-ups	No	Gyno (priv.)	2
9	61	Primary	Village	Annual check-up (priv.)	No	Gyno (priv.)	3
10	62	Primary	City	Check-up every 3-4 years	No	GP	9
11	58	Primary	City	No	No	Gyno (priv.)	3
12	40	Primary	Village	Breast cancer	No	GP	1
13	53	Primary	City	Annual check-up (priv.)	No	Gyno (priv.)	2
14	76	Primary	Town	No	Sister—died breast cancer	ED	3

15	61	Primary	Town	Bi-annual check-up	No	ED	4
16	58	Primary	City	Ovarian cysts; annual check-up (priv.)	No	Gyno (priv.)	1
17	49	Secondary (Year 10)b	City	Annual check-up (priv.); BRCA gene	Paternal grandmother and aunt—ovarian cancer	Gyno (priv.)	7
18	54	Secondary	Town	Annual check-up (priv.)	No	Gyno (priv.)	5
19	53	Primary	City	No	No	ED	1
20	62	Secondary (Year 10)b	City	No	Grandmother—leukaemia; grandmother—breast cancer	GP	2
21	69	Primary	City	Breast cysts, revision every 6 months	Mother—biliary tract cancer	ED	5.5
22	65	Primary	City	No	No	GP	0.5
23	45	Secondary (Year 12)b	City	Ovarian cysts, myomas (2003); private check-ups	No	Gyno (priv.)	0.33
24	59	University	City	Fibrocystic breasts; myomas; private check-ups; annual ultrasound	Father—died prostate cancer; maternal aunt—breast cancer	Gyno (priv.)	1

Abbreviations: ED, emergency department; GP, general practitioner; HPV, papillomavirus. a
City: pop. >50,000; town: pop. 10,000–50,000; villages: pop. <10,000 (2017 census data.[26]
b
Mandatory secondary education is to Year 10 (age 16), followed by 2 years of preuniversity studies (to age 18).

Data collection

A semistructured interview guide was developed, comprising an initial section to elicit women's narrative experiences followed by a set of semistructured questions to ensure the collection of basic data around the key points and time intervals defined in the Aarhus Declaration for Early Cancer Diagnosis Research[15] (Supporting Information: Box 1). The interview questions were discussed within the multidisciplinary research group, which included professionals from primary care, nursing, political science, sociology and epidemiology, plus a patient from the Association of People Affected by Ovarian Cancer (ASACO).

Sociodemographic data, gynaecological history and family history of cancer were collected on recruitment. Two experienced female qualitative methodologists conducted the interviews (N. C. B. and A. C. C.), which took place in early 2017. They were usually in the woman's home to favour a more personal and in-depth response, with no supervision by clinicians, and they lasted approximately 60 min and were audio recorded.

Data analysis

All interviews were transcribed verbatim and anonymized (N. C. B.). Thematic content analysis was performed to identify, analyse, organize and report the preliminary themes across the data.[27,28] Interviewing continued until no new themes were identified, and data were considered rich and saturated. One researcher (C. V. V.) verified transcripts against original audio data, and several authors closely examined the data to identify and agree on the key themes (C. V. V., M. M. C., L. M. P., C. J. A.).

Atlas ti software. 7.5.18 was used to import the text file into the software and analyse the data. All other co-investigators sense-checked the transcripts to ensure they reflected the research objectives, and the research team discussed the data to develop an initial coding scheme. Through an iterative process and frequent discussions, the research group identified three key themes that addressed women's experiences, staying as close as possible to the source material. The main findings are described and presented along these lines.

Informed consent statement

Before beginning the interviews, participants were given the opportunity to ask questions or voice concerns, and all signed informed consent.

RESULTS

Three key themes were identified in the analysis: (1) delay in recognizing bodily symptoms as serious and in seeking timely care; (2) missed opportunities for women during healthcare encounters and (3) use of resources and alternative healthcare pathways. These themes encompassed nine subthemes.

Delay in recognizing bodily symptoms as serious and in seeking timely careLack of knowledge about early signs and symptoms of ovarian cancer

Most women were unaware of or disregarded the symptoms associated with ovarian cancer, such as abdominal distension, bloating and pressure in the abdomen and pelvis. Only some women with a family history of ovarian cancer were particularly concerned about their symptoms in relation to ovarian cancer.

…and only later did I realise I had the typical symptoms, which is that you eat and feel full right away. (P9)

Gender-related barriers

Women tended to normalize their symptoms or attribute them to their gender and age or to natural processes such as menopause. Consequently, the response to symptoms, in some cases, included self-management and or self-medication, which delayed consultation with health professionals. Some women attributed the symptoms to psychological causes, such as the stress of caring for a sick child or elderly parents, or to the psychological impact of retiring from work.

I thought it was gas and started taking Aerored [a gas remedy]. My belly swelled a little bit, at that time I was very nervous, I was taking care of my mother with Alzheimer's, maybe it was the nerves. (P36)

I started spotting a little, as if it were a period. I didn't think much of it and blamed it on an argument I'd had with my son. (P26)

In many cases, women were used to having abdomino-pelvic discomfort and tolerated it without going to the doctor, either because they suffered or had suffered from menstrual cramps or in some cases because they had been diagnosed with fibromyalgia. The symptoms that caused the most alarm among women were progressive abdominal distention and postmenopausal bleeding.

I had painful menstrual cramps …I was wearing an intrauterine device, and the periods are very painful and I didn't insist. (P21)

In some cases, women reported waiting a year or more to go to the doctor's office, prioritizing their work activity, presenting to health services only when their symptoms worsened and were severe enough to interfere with daily life.

I went to Portugal for work, when I arrived, I said to myself: you should have gone to the emergency room instead of going on a trip. (P1)

One of the participants, who had suffered from breast cancer and had a young daughter and a sick father, was told by her gynaecologist that there was a high suspicion of malignant ovarian tumour. The patient refused to undergo surgery because she prioritized having another child over confirming the cancer. Despite her doctors' opposition, the

patient did not change her mind until her father died and the symptoms became unbearable.

They decided to perform surgery, but I was not ready, and I refused the operation, I said that I wanted to be a mother again and I stayed like that for almost two years. (P15)

Friends and family members of some interviewees advocated for their well-being and convinced them to seek medical care. The support of friends and family was crucial in validating women's concerns about their symptoms and overcoming their fears, especially embarrassment and fear of cancer.

When the spotting didn't stop, my friends said I had to go to the doctor. (P26)

False reassurance because of negative check-up

Many women reported undergoing gynaecological examinations through their private health insurance or ASSIR, in some cases to monitor benign gynaecological pathologies (e.g., myomas, endometriosis) and in others for annual or biannual preventive check-ups. Receiving a negative result in periodic follow-up tests or a normal result on cervical screening reassured women that they were free of gynaecological disease, and this led them to disregard symptoms and forego consultations with other specialists.

In May, I had an ultrasound and an annual Pap smear …I had an episode of more severe menstrual pain …as if I had a stone in the lower part, and I decided to go to the private urologist. (P21)

Missed opportunities for women during healthcare encountersMisattribution of women's symptoms by their physicians

Some participants, once they recognized the bodily changes and the need to seek medical help, reported inadequate diagnostic guidance from their primary care physician, who did not even suspect a gynaecological pathology. Several women were repeatedly treated for urinary tract infections. In one case, a woman consulted the ASSIR about her symptoms, and the attending physician considered that the symptoms were due to a yeast infection brought on by antibiotics prescribed for cystitis.

My GP always treated me with antibiotics and never sent me to a specialist, even when I asked for it. At the same time, the reproductive health clinic kept treating me for a yeast infection. (P4)

In one case, the woman's discomfort was even attributed to a depressive disorder, and her doctor prescribed psychotropic drugs.

I couldn't even stand up, couldn't walk, and I went to the GP, and I said, ‘Send me someplace, I'm so sick it's depressing me!’ And he goes and says, ‘Take this for the depression and you'll see how you feel better’. (P4)

Underestimation of severity of symptoms and need for medication

Some women repeatedly consulted their primary care physician for persistent symptoms. They agreed that their GPs did not have time for them or did not take their concerns seriously enough.

I started to swell…. But it didn't hurt, I was just bearing weight, walking and holding on. I went to the doctor and he said I had nothing: ‘Nothing, nothing, you have nothing, it's perfect…’. (P18)

Some women, especially those who were older and less educated, needed their social network's support for health professionals to validate their symptoms and agree to investigate them. In some cases, a family member (especially adult sons) intervened directly, accompanying the women to the health centre, validating their discomfort and insisting on the seriousness of their condition to obtain a referral to secondary care or hospital emergency departments.

My son and daughter came with me and said: ‘Hey, do me a favour and give us a referral to take my mother to the emergency department [to the hospital]’. ‘Ah, but your mother is fine, her belly is fine, blah, blah, blah’. ‘I don't care, I know my mother, and something is wrong’. They gave us the paper and we went to the hospital. (P18)

Inadequate tests and/or false negative results

In one case, a colonoscopy was requested due to recurrent abdominal pain, which of course did not lead to a diagnosis of ovarian cancer. In another case, although a transvaginal ultrasound was requested, the result was interpreted as negative. Such circumstances can clearly prolong diagnostic intervals by providing (temporary) false reassurance despite the persistence of the symptoms.

In May, I had an ultrasound and an annual Pap smear …and I told him [the gynaecologist] again that I had

discomfort …he said that everything was fine and that I should calm down. (P21)

Use of resources and alternative healthcare pathwaysUse of complementary/alternative medicines

Some young women interpreted their symptoms as ‘normal’, choosing to self-manage using naturopathic treatments and alternative medicines.

Over the last month I've had a feeling of being full, and I used alternative medicine treatments to clean out my body. (P9)

In one case, a woman with a previous history of cancer, unable to cope with a second neoplasm, and against the advice of health professionals, resorted to alternative medicines to avoid biomedical therapies.

They decided to operate but …I wanted to fight to be a mother. I took other ways, I took alternative therapies, and so I was holding on for two years. (P15)

Access to private health care

Women with private health insurance had regular gynaecological check-ups, and if any worrisome symptoms appeared, they had direct and rapid access to their usual private specialists. In some cases where the suspected diagnosis was confirmed in private practice, gynaecologists (many of whom combine public and private practice) used their professional networks to streamline referral to a tertiary public hospital for treatment of ovarian cancer.

My son and daughter-in-law went to a gynaecologist we know in Barcelona …three days later we went to the hospital and there were three doctors waiting for me in the consultation room. (P4)

In contrast, some women who struggled to get a diagnosis or faced long waiting lists for tests or referrals from their primary care centre opted to go to a private practice on the advice of their children, fully assuming the physicians' fees and the cost of complementary tests. Others, without the means to access private care and in the absence of a response to their health problems from primary care physicians, used the hospital emergency department as a shortcut to quickly access care. On several occasions, this avenue facilitated the process for diagnosing ovarian cancer, but in other cases, the fragmentation of care caused delays and made it even more difficult to suspect cancer.

I went to the doctor almost every week. He wouldn't send me to any specialist, and then I felt so bad that I went to the hospital two or three times. (P4)

Women's capacity for action and decision-making (agency) about their health

One participant was a university-educated woman who was comfortable searching for information through the Internet and finding resources through the public health network. After being discharged from the emergency department of the county hospital with a suspicion of ovarian cancer, she adopted a proactive attitude and managed to be seen at the tertiary hospital of her choice.

I found out …and I picked up the phone and made an appointment: ‘It looks like I have ovarian cancer and I would like a visit with a gynaecological oncologist’ …and they gave it to me on the same Thursday. (P9)

However, this was not a common experience. Many women reported that, beyond face-to-face consultation with their physicians, they and their families had difficulty navigating the healthcare system due to poor information, for example, in making follow-up appointments or obtaining diagnostic test results.

…I have been waiting for an ultrasound since August and they haven't called me. (P13)

DISCUSSION

This qualitative study identified three key themes and nine subthemes. The first theme was women's delay in recognizing bodily symptoms as serious and in seeking timely care, with subthemes on the lack of knowledge about early signs of ovarian cancer, gender-related barriers, and false reassurance from a negative check-up. A second theme was missed opportunities during healthcare encounters, due to misattribution of women's symptoms by their physicians, underestimation of symptom severity and need for medication and inadequate tests and/or false negative results. Finally, interviews highlighted the use of resources and alternative healthcare pathways, including the use of complementary/alternative medicines, access to private health care and women's capacity for action and decision-making (agency) about their health.

Comparison with findings from other studiesDelay in recognizing bodily symptoms as serious and in seeking timely

care

Numerous studies have examined factors affecting the length of the appraisal and help-seeking intervals for cancer in general[12,29] and ovarian cancer in particular.[30–33]Most women in our study expressed a lack of knowledge regarding the symptoms they were experiencing and shared concern that their symptoms had not aroused suspicion earlier, which is largely consistent with the literature.[30–33] The presence of abnormal vaginal bleeding is associated with prompt help-seeking,[18] while common and sometimes vague symptoms, such as bloating, pelvic or abdominal pain, difficulty eating or feeling full quickly and urgent or frequent urination, did not usually raise any red flags.[6,31,32] Some studies have highlighted the low level of awareness of ovarian cancer among the general public,[31,32] suggesting that if women were able to recognize symptoms of ovarian cancer, this might increase their own suspicion of a malignancy and shorten the help-seeking interval.[32]As in other studies, our participants struggled to balance specific bodily sensations with aspects of their life-worlds (individual, social, perceptual and practical experiences) before consulting a medical doctor.[34] The normalization of initial symptoms acted as a barrier to help-seeking[32] and may be explained, in part, by the subtlety and nonspecificity of early signs of ovarian cancer and by the fact that these often coincide with perimenopausal changes. Women would benefit from gaining more knowledge of the disease and confidence in their own observations of bodily changes[12] through the promotion of body awareness and health literacy.[35]Our participants had competing responsibilities related to work and to caring for children, grandchildren and elderly parents, which they frequently prioritized over self-care, a deeply rooted sociocultural issue among women. In this context, as in other studies,[29,30,32] women demonstrated a high capacity for disregarding bodily changes and tolerating symptoms, which kept them from seeking medical attention until symptoms become severe and impossible to ignore. Help-seeking for gynaecological cancer symptoms differs from that for other illnesses because of fears associated with embarrassment of the affected body part and with the perception of cancer itself.[12] Women of all ages often experience anxiety and fear before and during a pelvic examination due to the invasive nature of the procedure.[36] Prior experiences of gynaecological violence—situations unfortunately often normalized and rendered invisible—could help explain emotional barriers to help-seeking for some women. However, in our interviews, women did not openly express feelings of shame or embarrassment about undergoing a pelvic examination. In the interviews, only one woman acknowledged fear of cancer and the consequences of treatment, specifically in relation to loss of fertility, which led her to refuse the recommended treatment. Other fears noted in the literature (though not explicitly mentioned by our participants) include fear of change in body image and the sudden arrival of menopause, which can lead to a feeling of loss of female identity, with possible repercussions on their sexual life and that of their partners.[37]Validation and legitimization of help-seeking by the media or by friends and family is known to reduce women's concern about being labelled as time-wasters[12] and helps them overcome feelings of shame and fear around the disease and its consequences. In contrast to other studies,[30,32] in our interviews, women did not express concern that their complaints were inappropriate or trivial, suggesting that fears about wasting their doctor's time were not a barrier to seeking help. In our study, as described elsewhere,[38,39] normal test results contributed to a false sense of security and delay in seeking care. Even when patients underwent routine investigations and appropriate medical check-ups, ovarian cancer often went undetected. There is a widespread belief that a negative Pap or papillomavirus test result excludes any type of gynaecological tumour; however, screening is only effective for cervical cancer, not for other forms of gynaecological cancer.[32]

Missed opportunities for women during healthcare encounters

On a woman's first presentation with nonspecific abdomino-pelvic or urinary symptoms, primary care physicians will rarely suspect ovarian cancer because, fortunately, it rarely turns out to be cancer.[5,40] Many physicians tended to ignore or normalize the symptoms or misattribute them to urological or digestive causes. This misattribution may be explained to some extent by the low incidence of the tumour and hence the lack of previous knowledge and experience, making it imperative to train and sensitize health professionals to be able to recognize and promptly manage ovarian cancer symptoms. However, as confirmed by other studies,[41] physicians' requests for and interpretation of the information necessary for diagnosis may also be conditioned by stereotypes, prejudices and

their preconceived notions regarding women. Specifically, sexism and ageism can negatively impact how health professionals approach the diagnostic process,[41,42] normalizing symptomatology and hindering optimal assessment and clinical reasoning,[39] which partly explains the disparity in care.[41,42]As is the case with some of our interviewees, omission or delay in the diagnosis of ovarian cancer may also be due to the existence of various biases inherent to healthcare practice, for example, anchoring bias (focusing exclusively on a single piece of information), availability bias (relying too much on already known or readily available information) and confirmation bias (tendency to seek information that supports preconceived ideas).[39] As described elsewhere, these attitudes and practices, together with the lack of knowledge about this cancer and the difficulty of some physicians to overcome communication problems, could affect the initial evaluation of women with ovarian cancer and lead to misdiagnosis.[43]Some of our patients, in a situation of great vulnerability due to the persistence or recurrence of their symptoms, recounted that their GPs did not recognize or respond to their problems despite repeated care encounters. Health professionals, who have historically been attributed a role of authority within the doctor-patient relationship, may be reluctant to change their diagnostic orientation. In this situation, some patients turn to their social network, friends and children, preferably male, to validate the severity of their symptoms and obtain appropriate medical care, challenging the power dynamics established around the physician. Avoiding similar situations in daily practice would imply, as suggested by others,[35] a change in the approach to physician-patient relationships, facilitating bidirectional communication, interactions based on empathy, respect for the subjective experiences of users and shared decision-making. For some women in our study, missed opportunities were related to the performance and interpretation of diagnostic tests by practitioners.[38,39] This can occur when suspicion of cancer is correctly raised but decisions about planned investigations are suboptimal or inadequate. Such scenarios may be more likely for cancers that share common symptoms (e.g., an abdominal symptom is investigated with a colonoscopy that is negative, and this finding is initially interpreted as a ‘diagnostic closure’). This circumstance can clearly prolong the diagnostic interval and represents a missed opportunity for an accurate diagnosis. However, when the correct tests have been performed, but the results are falsely interpreted as negative without adequate backup reassurance or re-evaluation mechanisms in place, the difficulties around diagnosis are compounded.[39]

Use of resources and alternative healthcare pathways

Several studies have examined the use of alternative/complementary medicines.[44] For one woman in our study, the use of these treatments was related to the normalization of symptoms and her consequent desire to self-manage, while another questioned the appropriateness of biomedical treatments and the authority of the doctor to control her health. In the latter case, the woman's personal history of cancer and possibly a limited social network is likely to have conditioned her response. Our participants showed individual differences in their capacity and opportunity to seek alternative diagnostic pathways (mainly through the private healthcare sector), rooted in their socioeconomic conditions and social networks. Women without the means to access private care came into conflict with professionals and the health system when their problems were not addressed. Lack of trust in their referring physicians, as reflected in other studies,[35] often translates into the ‘transgression’ of established norms within health systems, for example, presenting to the emergency department without a physician's express indication or refusing the prescribed treatment. Although the present study focused on women's experiences during the prediagnostic stage, the challenges of navigating a complex healthcare system also continue through the diagnostic, treatment and survival phases. In addition to aspects related to gender,[45] we observed differences rooted in health literacy and in how women process information and make decisions about their care, with implications for the patient experience and health disparities.

Strength and limitations

This study focused on the narratives of women diagnosed with ovarian cancer. The experiences described were in a system based on the gatekeeper model, so they may not be generalizable to other populations or healthcare settings. However, the saturation of the sample data was achieved without new issues arising, and this supports the validity of our findings, which could have implications for many other cancers that affect women in settings similar to ours. In addition, the interview script was agreed upon by all members of the research team, including the

representative of a patients association (ASACO). Nevertheless, the study has some limitations. We excluded women with very advanced ovarian cancer, whose ill health would have limited their ability to contribute. Moreover, sampling was done without regard to socioeconomic status, comorbidities or race/ethnicity. However, published studies have not found important differences in marginalized people compared to dominant groups.[12]Health professionals' choices on which patients to invite for interview also introduces a risk of selection bias. Only women who had a good relationship with their current physician at the time of recruitment could participate, even if their previous experiences with other professionals had been unfortunate. Women's narratives, like any experience, are the result of a process of perception and personal interpretation. In addition, as this is a retrospective study, their recall and interpretation of past events may be affected by their subsequent experiences. It is likely that many women in the interview were not conscious of potential psychological barriers (shame, fear, etc.) when first confronted with symptoms. Moreover, we believe that the initial interview script did not sufficiently probe gender-related issues. Finally, this study took place in the pre-COVID-19 period, when the healthcare panorama was markedly different. However, since the main constraints on the system—time for each patient and access to diagnostic tests—have only been exacerbated by the pandemic, we believe our findings are more relevant than ever.[46]

CONCLUSION

Women with ovarian cancer reported delays in recognizing bodily symptoms, mainly due to lack of knowledge of symptoms and a failure to interpret them as cancer. Competing demands related to work and family appear to be important barriers to timely help-seeking. Our results support the notion that prediagnostic contact patterns in primary health care may hold missed opportunities to diagnose ovarian cancer. The factors identified in this study can be addressed through individual interventions and community information campaigns, including by providing women with information about the symptoms of ovarian cancer and their individual risk based on their personal or family history, encouraging body literacy and promoting women's confidence in their observations of bodily changes. At the same time, active and empathic listening and respect for women's subjective experiences are essential in healthcare consultations, as is encouraging two-way communication and shared decisions. Further research is needed to investigate the extent to which traditional gender roles and socioeconomic inequalities condition women's ability to manage their own health and to interact with health professionals and the health system.

AUTHOR CONTRIBUTIONS

Conceiving and designing the study: Mercè Marzo-Castillejo, Carmen Vela-Vallespín, Núria Codern-Bové and Josep M. Borras. *Obtaining funding and ethical approval*: Mercè Marzo-Castillejo, Carmen Vela-Vallespín and Josep M. Borras. *Collecting the data*: Núria Codern-Bové. *Analysing the data*: Núria Codern-Bové and Carmen Vela-Vallespín. *Interpreting the data*: Carmen Vela-Vallespín, Mercè Marzo-Castillejo, Laura Medina-Perucha and Constanza Jacques-Aviñó. *Writing the report*: Carmen Vela-Vallespín, Mercè Marzo-Castillejo, Laura Medina-Perucha, Constanza Jacques-Aviñó and Meggan Harris. *Revising the report*: Carmen Vela-Vallespín, Laura Medina-Perucha, Constanza Jacques-Aviñó, Núria Codern-Bové, Meggan Harris, Josep M. Borras and Mercè Marzo-Castillejo.

ACKNOWLEDGEMENTS

We are grateful to the women with ovarian cancer who participated in the study as well as the health professionals who recruited them. We acknowledge the valuable contribution of Àngels Cardona, who assisted in the collection of interview data. We are grateful to Anna Berengera Ossó for comments, editing and suggestions on the manuscript. The project received a competitive research grant from the Generalitat de Catalunya, awarded on the 2016 call under the Health Strategy Action 2016–2020, within the Pla Estratègic de Recerca i Innovació en Salut (PERIS). This was oriented towards planning and coordination, and it defined the general lines of research and innovation in the Department of Health, in line with the priorities of the Health Plan Catalonia 2016-2020 (reference SLT002/16/00200). Carme Vela-Vallespín received a predoctoral grant from the Catalan Health Institute for research training in primary care, in the Northern Territorial Health Management Area, for research on, ‘Evaluation of the care process in women suspected of having ovarian cancer in Catalan primary care’.

CONFLICT OF INTEREST

The authors declare that they have no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of the study are available from the corresponding authors upon reasonable request.

ETHICS STATEMENT

This study was conducted in accordance with the Declaration of Helsinki and received ethics approval from the Ethics Committee of the Primary Health Care Research Jordi Gol i Gurina (IDIAPJGol), number P17/088. The confidentiality of the participants is guaranteed under the Organic Law on the Protection of Data of a Personal Nature (03/2018, December 5) and in accordance with the provisions of Regulation (EU) 2016/679 of the European Parliament and Council of 27 April on Data Protection (GDPR) and relevant national legislation.

DETAILS

Subject:	Reassurance; Ovarian carcinoma; Health care policy; Medical diagnosis; Cysts; Sex roles; Health care; Diagnosis; Inequality; Human papillomavirus; Ovarian cancer; Delayed; Alternative medicine; Decision making; Emergency medical care; Health literacy; Womens health; Brain cancer; Mutation; Content analysis; Women; Gender; Private medical care; Health professional-Patient interactions; Breast cancer; Access; Symptoms; Clinical outcomes; Drugs; Age; Medical personnel; Clinical decision making; Gynecology; Interviews; Cancer; Primary care; Patient participation; Qualitative research; Public participation; Health services
Identifier / keyword:	early detection; healthcare seeking; ovarian cancer; prehospital care; primary health care; qualitative research; women
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	476-487
Publication year:	2023
Publication date:	Feb 2023
Section:	ORIGINAL ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513

e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-29
Milestone dates:	2022-11-16 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-10-07 (manuscriptReceived); 2022-11-29 (publishedOnlineEarlyUnpaginated); 2022-11-17 (manuscriptAccepted)
Publication history :	
First posting date:	29 Nov 2022
DOI:	https://doi.org/10.1111/hex.13681
ProQuest document ID:	2767101840
Document URL:	https://www.proquest.com/scholarly-journals/womens-experiences-along-ovarian-cancer/docview/2767101840/se-2?accountid=211160
Copyright:	© 2023. This work is published under http://creativecommons.org/licenses/by/4.0/ (the “License”). Notwithstanding the ProQuest Terms and Conditions, you may use this content in accordance with the terms of the License.
Last updated:	2023-04-24
Database:	Public Health Database,Publicly Available Content Database

Conceptual and practical challenges associated with understanding patient safety within community-based mental health services

Averill, Phoebe [1]
; Vincent, Charles [2]
; Reen, Gurpreet [2]
; Henderson, Claire [1]
; Sevdalis, Nick [1]

[1] Centre for Implementation Science, Health Service and Population Research Department, Institute of Psychiatry, Psychology and Neuroscience, King's College London, London, UK [2] Department of

Experimental Psychology, University of Oxford, Oxford, UK

ABSTRACT (ENGLISH)

Introduction

Patient safety problems stemming from healthcare delivery constitute a global public health concern and represent a pervasive barrier to improving care quality and clinical outcomes. However, evidence generation into safety in mental health care, particularly regarding community-based mental health services, has long fallen behind that of physical health care, forming the focus of fewer research publications and developed largely in isolation from the wider improvement science discipline. We aimed to investigate the state of the field, along with key conceptual and empirical challenges to understanding patient safety in community-based mental health care.

Methods

A narrative review surveyed the literature to appraise the conceptual obstacles to advancing the science of patient safety in community-based mental health services. Sources were identified through a combination of a systematic search strategy and targeted searches of theoretical and empirical evidence from the fields of mental health care, patient safety and improvement science.

Results

Amongst available evidence, challenges in defining safety in the context of community mental health care, evaluating safety in long-term care journeys and establishing what constitutes a 'preventable' safety problem, were identified. A dominant risk management approach to safety in mental health care, positioning service users as the origin of risk, has seemingly prevented a focus on proactive safety promotion, considering iatrogenic harm and latent system hazards.

Conclusion

We propose a wider conceptualization of safety and discuss the next steps for the integration and mobilization of disparate sources of 'safety intelligence', to advance how safety is conceived and addressed within community mental health care.

Patient and Public Contribution

This paper was part of a larger research project aimed at understanding and improving patient safety in community-based mental health care. Although service users, carers and healthcare professionals were not involved as part of this narrative review, the views of these stakeholder groups were central to shaping the wider research project. For a qualitative interview and focus group study conducted alongside this review, interview topic guides were informed by this narrative analysis, designed jointly and piloted with a consultation group of service users and carers with experience of community-based mental health services for working-age adults, who advised on key questioning priorities.

FULL TEXT

INTRODUCTION

Physical healthcare services have benefited from over two decades of patient safety research, a discipline concerned with 'the avoidance, prevention and amelioration of adverse outcomes or injuries stemming from the process of healthcare'.[1] Despite achievements within other clinical specialities, safety in mental health care is largely uncharted. However, interest in the safety and quality of mental health care is gathering momentum. In the United Kingdom, recent publications have advanced learning about areas such as care transitions, service user, carer, and provider perspectives on safety, and factors affecting incident reporting.[2-8] However, further efforts are required to unify research on safety in mental health care with research from the physical healthcare-focused patient safety science tradition.[9]

Existing research into safety in mental health care has almost exclusively prioritized inpatient services. This reflects

comparable challenges to those documented in physical healthcare literature, where measuring safety and implementing interventions is more complex in community settings due to a lack of robust safety indicators, lower frequency of care encounters and limited ability to influence treatment adherence or the safety of patient home environments.[10,11] Despite these commonalities, there are likely safety concerns unique to community mental healthcare settings. For example, issues surrounding risky behaviour, decision-making capacity and compulsory treatment pose specific challenges.[12,13]

Aims and overview

In this paper, we aim to provide a brief narrative review of the present state of patient safety with a focus on community-based mental health services. We seek to illuminate key conceptual and empirical challenges associated with understanding safety in the aforementioned settings. Finally, considering research gaps and the limitations of existing safety knowledge, we propose an approach to consolidating and furthering the evidence base for safety in community mental health care.

As issues concerning general services for working-age adults are better documented, we focus primarily on this population (rather than services for children, older adults or specialist community care pathways, e.g., mental health learning disabilities care). Herein, community-based mental health services are defined broadly as noninstitutional mental health services which deliver care to people living in community settings. This includes provision within primary care (e.g., care from a general practitioner [GP] or Improving Access to Psychological Therapies services), and secondary care mental health services (e.g., Community Mental Health Teams). Multidisciplinary care in these settings may involve a range of clinical professionals, including mental health nurses, psychiatrists, social workers, clinical psychologists and occupational therapists. Likewise, given the paucity of research exploring patient safety issues in community-based mental health care, in places, we draw upon literature from psychiatric inpatient settings to illustrate the issues discussed. In these cases, we reflect upon further challenges introduced when applying safety concepts to community mental health settings.

METHODSStudy design

We conducted a narrative review to critically assess extant research, appraise knowledge gaps and examine conceptual issues within the field, seeking to contribute to the conceptual advancement of patient safety science as applied to community-based mental health care. The present research was devised in response to difficulties encountered in our efforts to formulate appropriate search and screening criteria for a separate systematic scoping review by the same authorship team, which focused on the nature of patient safety problems in these services. Issues comprised problems in identifying relevant articles, lack of consensus over safety-relevant outcomes in community-based mental health care and difficulties in establishing the boundaries of safety and harm preventability within these settings.

Given these unresolved challenges, a narrative review was deemed a necessary initial step to take stock of this research area. The benefits of narrative review approaches lie in their permitting of a broader exploration of topics,[14] and contribution to the conceptual development of a given area. Historical narrative reviews have been described as ‘irreplaceable to track the development of a scientific principle or clinical concept’, where the ‘narrative thread could be lost in the restrictive rules of a systematic review’ (p. 231).[14]

Literature search and selection

This review drew upon literature identified through several means. A subset of articles was retrieved from a systematic search developed for the separate systematic scoping review discussed above.[15] This search, executed in June 2020, focused around three key elements: ‘mental health’, ‘patient safety’ and ‘community-based mental healthcare’. Once the need for the present narrative exploration was ascertained, further targeted searches were performed, centring on investigating identified challenges detailed within this paper. These searches focused on mental health care and the wider fields of patient safety and improvement science, also involving patient safety literature developed in other comparatively better-evidenced care settings, such as general hospital services. Articles were purposively selected for inclusion according to their conceptual contribution to the debates addressed within this review,[16] and were discussed and agreed upon amongst the review team. This review sought to provide

an overview of a broad range of issues, rather than comprehensive coverage of all relevant papers. A diversity of literature was surveyed, with no restrictions applied on the basis of study design or publication status (e.g., 'grey' literature). Whilst the study limitations, suitability of methods and quality of obtained findings were considered,[14] no formal quality assessment was undertaken. Findings were synthesized narratively and organized around key conceptual themes. The review was devised according to guidelines for the quality assessment of narrative review articles.[17]

RESULTS AND DISCUSSION

This narrative analysis was informed by 71 sources, including empirical research and contextual material such as policy documents and healthcare news announcements. A combination of published (70.4%) and unpublished (29.6%) literature was purposively selected for inclusion, focusing on a range of different care settings, including community-based mental health services (17.0%), inpatient mental health services (19.7%) or mixed mental healthcare settings (47.9%). Other included sources related to physical healthcare services (2.8%), or mixed healthcare settings (12.7%). Of the literature consisting of research or reviews of research, 30.0% reported on quantitative data, 21.7% on qualitative data and 31.7% had mixed study designs, with a further proportion constituting editorials or position pieces (16.7%).

The current state of the field

In their review of the UK National Health Service (NHS) mental healthcare provision, the Care Quality Commission cited safety as a key priority.[18] Some 43% of community-based mental health services for working-age adults were rated 'requires improvement' for safety at the most recent inspection.[19] Likewise, an independent investigation into the safety of risk assessment in community mental health teams was recently announced.[20]

The developing evidence base for mental health patient safety

Mental health patient safety research has focused predominantly on hospital settings,[21] which constitute a minority of mental healthcare encounters. Service users' care journeys are increasingly comprised of contact with community-based mental health services, with UK psychiatric inpatient bed numbers falling by over 55% since 2000,[22] and efforts to reduce the length of inpatient stays and avoid admissions through community-based alternatives.[23] Consequently, community services are treating larger numbers of sicker patients.[24]

Nevertheless, a handful of studies have included data about patient safety concerns across several types of mental healthcare settings, with participants reporting on experiences of inpatient or community services.[4,5,25,26] Some study findings are of evident relevance to community care, in highlighting safety risks relating to lengthy community treatment waiting times, care discontinuity, inadequate crisis services and challenges in managing acute risk in the community.[4,5,25] Different perceptions of safety in the community versus hospital settings were also directly discussed in one paper.[4] Other insights are likely of relevance across all mental healthcare settings, though may not have been explicitly explored or evidenced in terms of community services within the articles. These include risks stemming from workforce issues such as inadequate staffing levels, training and staff burnout.[4,5,25] It is plausible that data have not always been disaggregated to analyse by mental healthcare setting type, which could form a pertinent point of analysis in future work. Indeed, a study of staff-reported risk assessment and safety management processes revealed important differences in risk assessment practices between mental healthcare settings, including a greater focus on patients' family and social context in the community than in hospital-based care.[26] Further research centring explicitly on issues of safety in community-based mental health services is however warranted.

Aside from gaps in the evidence base, the research field faces further challenges. There is a lack of integration between the field of safety in mental health care and the wider safety and quality evidence base.[9,27] Much of the research about safety in mental health care has neglected to acknowledge and build upon established patient safety science literature, developed primarily in physical healthcare contexts. Indeed, a substantial body of literature discusses topics widely regarded as safety-relevant (e.g., self-harm, violence and aggression), though does not necessarily situate itself within existing patient safety theory, such as human factors or systems approaches.

This divide is mirrored within the quality and safety academic discipline. In searches of three journals containing traditional patient safety literature, few titles corresponding to safety in mental health settings were identified.[9] The

authors determined that research into safety in mental health care tended instead to be published within mental health-specific journals.[9] This separation impedes system-wide learning on safety events and principles which apply across clinical settings.

A further body of evidence discusses issues likely to affect safety across the care journey (e.g., care team communication problems), though neither embeds relevant patient safety science literature, nor is it conceived of as safety-relevant research. Such articles instead identify themselves within the spectrum of care quality,[28] or focus on specific issues, such as inadequate care planning,[29] without linking these findings to safety implications. A bibliometric study of research activity on patient safety in community mental health services exemplifies this problem.[30] Searches of 'patient safety' and 'community mental health services' across two bibliographic databases retrieved only two articles covering safety in community mental health care, neither of which centred specifically on this setting.[31,32] The apparent lack of relevant research is likely in part due to the failure of researchers to contextualize their findings within the parameters of patient safety.

Dominant approaches to safety in mental health care

Service users' interests have not been placed at the heart of a nascent focus on improving safety in mental health care, where 'safety' has typically been approached as the inverse of risk in these services.[33–35] Since widespread moves away from institutionalization towards community care, incidents of violence and aggression have received significant attention, due to high-profile public inquiries and media coverage of a small number of homicides by service users.[36,37] This focus is further sustained by rates of patient assault on mental health staff, which are the highest amongst the healthcare sector.[12,38]

The importance of staff safety cannot be overstated, and this issue may have interrelated consequences for patient safety. Workplace violence is associated with poor well-being and burnout amongst healthcare staff, which in turn link to factors affecting patient safety and care quality.[39,40] For example, beyond potential safety consequences immediately following an assault, such as a risk of patient injury during physical restraint,[41] repeated exposure to staff-directed violence may contribute to workforce issues including absenteeism and staff turnover, and overuse of restrictive practices.[42,43] Such issues provoke further safety risks. Nevertheless, efforts to improve safety in mental health care must not be limited to violence reduction.

In prioritizing risks that service users may present to themselves or others when acutely unwell, the term 'patient safety' has been somewhat misappropriated. This interpretation situates patients as the origin of risk, prohibiting discussion about iatrogenic harm or hazards elicited by the process of health care itself.[27,44] The corresponding risk management culture has resulted in a narrow safety research agenda, centred around topics such as suicide, self-harm and violence. Indeed, a recent review exploring patient involvement in the development of safety improvement interventions in acute inpatient mental healthcare settings identified that almost two thirds of included studies were concentrated on the reduction of restrictive practices, rather than on interventions aimed at advancing the therapeutic culture of these settings.[45] Where resource allocation, team culture and training are concentrated primarily around maintaining personal and public safety,[35,46] there may be overreliance on coercive or restrictive interventions, curtailing a broader focus on proactive promotion of safe care.[33] Possible sources of harm in community-based mental health care are summarized in Box 1.

1. **BoxPotential areas for patient harm in community-based mental health services**

Harm from ineffective risk management	Harm from inadequate or unsuccessful prevention and management of risk, such as self-harm, suicide or risks of violence and aggression. Capacity for prevention of these events by services may not always be clear.

Harm due to failure to provide appropriate treatment	Service users routinely do not receive optimal or evidence-based standards of care, which may contribute to harm. For example, staffing shortages may result in service users not being assigned a care coordinator when one is required, or not receiving care within safe timeframes.
Medication-related harm	Medications prescribed for mental health problems may result in adverse drug reactions, unpleasant or harmful side-effects, or contribute to the development of comorbid physical health conditions. Medication errors, on part of care teams or service users and their carers, may also result in harm. This is increasingly relevant in community care, where service users play a larger role in their own medication management.
Harm from restrictive or coercive care	Harm may stem from the use of restrictive practices in mental health care, including scenarios where there is contact with other services which are less equipped to address a mental health crisis (e.g., the police). Service users may feel that they have little control over their own lives.
Harm due to undertreatment	Avoidable harm may result from under-detection and undertreatment of risks associated with prescribed medications, such as failure to prescribe metformin for antipsychotic-induced dyslipidaemia. Similarly, access to interventions, such as psychological therapies, amongst service users who would benefit from such treatment may be inequitable and more readily offered to those perceived to be assertive or articulate.
Harm relating to diagnosis	Misdiagnosis, missed diagnosis or delayed diagnosis can cause harm by delaying access to the appropriate course of treatment. Delays may contribute to deterioration and loss of confidence in mental health services. Service users may also experience harm associated with the specifics of the diagnosis they receive. For example, those with a personality disorder diagnosis may be faced with lack of adequate treatment pathways or stigma from care teams.
Psychological harm	Unhelpful or distressing encounters with community-based mental health services may cause service users to feel unsafe when using these services. Similarly, prior experiences of compulsory treatment under Mental Health Act legislation may erode trust in care systems, potentially leading service users to conceal important risk information from care teams.

Key challenges for understanding patient safety in community mental health care

In what follows, we discuss theoretical and empirical issues in understanding and conceptualizing safety in community mental health care.

Defining patient safety in mental health care

To date, patient safety definitions have been derived largely from physical healthcare contexts.[6] Concepts of

adverse events, errors and near misses are also shaped by terminology originating outside of mental healthcare settings.[21] The United States Agency for Healthcare Research and Quality offer a well-cited definition of patient safety as: 'freedom from accidental or preventable injuries produced by medical care'.[47]

The focus on 'medical care' offers little scope to apply safety principles to the wealth of nonpharmacological treatments provided in mental health care, where the workforce itself has been described as the main therapeutic intervention.[48] Additionally, positioning 'injury' as the safety outcome of interest obscures key types of iatrogenic harm. For instance, assessment and detention under Mental Health Act legislation can impact patients' psychological safety, potentially invoking trauma or replicating prior traumatic experiences.[49] This example exposes further tensions for the concept of 'patient safety' in this care context. Community clinicians routinely face competing potentially harmful scenarios, whereby delaying or choosing not to pursue a Mental Health Act assessment on the grounds of providing the least restrictive level of care may increase risks of adverse safety outcomes associated with relapse in the community (e.g., suicide, harm to others).

The constraint of safety concerns to injuries 'produced by' healthcare services marks a further problem with this definition when applied to mental health care. As discussed, service users may be at risk from their own actions when acutely unwell (e.g., self-harming behaviours). This definition may be poorly aligned with certain hazards prevalent in mental health care, but also exemplifies key disparities between mental health services compared to other care specialities. Indeed, mental health teams tend to focus on risks generated by service users, rather than by the care itself.[26]

Research also suggests that the poorly defined nature of patient safety in mental health care may create challenges for clinical care. Qualitative interviews of NHS psychiatrists revealed limited agreement on definitions of 'patient safety' and 'quality'.[50] Clinicians lacked awareness of the wider safety context in the NHS, including relevant high-profile publications such as *An Organisation with a Memory*.[51] Critically, participants failed to recognize certain types of potential safety incidents, culminating in shortfalls in both incident reporting and opportunities for learning.[50] This is problematic, as whether a given factor is conceived of as a safety issue may impact awareness of these risks as they arise and motivation to take preventative measures.

Conceptual complexities associated with safety in community settingsLocating the boundaries of the phenomenon of interest

Mental healthcare providers assume wider responsibility to care for the 'whole person', than do their physical healthcare counterparts. This is most pronounced within community services, where alongside mental healthcare provision, care teams have oversight of service users' needs in relation to housing, risk of victimization, financial problems and physical health, including comorbidities exacerbated by psychotropic medications.[52,53] When a service user comes to harm at home, it is difficult to disentangle the potential for prevention, if any, by mental health services. Equally, where a comprehensive harm definition is used, including subjective experiences of psychological harm associated with unfavourable care experiences, the line at which such events constitute a patient safety problem is ambiguous.

Mental health teams may also undertake clinical activities outside of their core expertise.[54] Where a service user is reluctant to engage with their GP, psychiatrists and nursing staff have reported performing and interpreting physical healthcare investigations, such as phlebotomy and electrocardiograms, despite acknowledging their lack of confidence in carrying out these tasks.[53] In these circumstances, one must consider whether imperfect test result interpretation is preferable to the absence of such investigations. These nuances must be reflected in our understanding of safety in mental healthcare contexts.

Measuring safety in long-term care

Compared to acute psychiatric inpatient care, the longer-term nature of patient journeys through community-based services presents challenges for operationalizing safety. Efforts to understand safety in clinical practice have centred on so-called active failures, corresponding incidents and their analysis.[1] Severe incidents resulting in immediate, observable harm are most likely to be reported and selected for in-depth clinical incident analysis. However, this may be at odds with the context of care delivery in community settings.

Safety problems which manifest in long-term community care may be better understood by examining the dynamic accumulation of risk from unsafe care processes and care delivery problems over time. Unlike in hospital admissions, community-based clinicians perform in the context of long-term management of myriad complexities. For example, monitoring of stable or deteriorating chronic illnesses over time, rising multimorbidity and communication challenges from care delivery across multiple, fragmented settings are some of the difficulties teams face.[55–57] Harm from such risks may be less evident, and less immediate, and their role in safety event causation may be less easily established. These complications thus align poorly with the traditional focus on specific errors, lapses and other 'sharp end' performance failures which are temporally or physically proximal to an incident and thus more easily measurable.

Community care is upstream and preventative

An overlapping challenge for observing and measuring safety concerns the upstream nature of much of community health care. Detection, prevention and maintenance of conditions are the mainstay of community-based care, seeking to avert adverse outcomes such as deterioration or hospitalization. Consequently, the safety impacts of care in primary and community mental health care may not be quantifiable until further up the succession of health care, in terms of either causing or preventing adverse outcomes (e.g., psychiatric inpatient admissions, or long-term psychotropic medication-related physical health complications). Indeed, the consequences of misdiagnosis in primary care may not reach clinical attention for several years.[58]

Likewise, as clinical guidance is typically disorder-specific,[58] providers are sometimes obliged to depart from these decision-making supports, to weigh up the relative benefits and potential harms of treatment. For example, clinicians may consider the cardiometabolic burden associated with long-term antipsychotic medication, versus the potential reduction in suicide risk.[59,60] These examples reinforce the importance of attending to system-wide care processes over time, rather than incidents alone.

Conceptualization of patient safety in a community mental health setting: A case example

When seeking to understand safety in the community, several factors warrant consideration. Care journeys may span months to decades, with a much lower intensity of care offered than in inpatient settings. Accordingly, the pace of care may be much slower, often with little to no community team involvement between appointments. Access issues are of greater significance to safety in community care, where waiting times often extend over several months for specialist psychological therapy, or to receive any care upon referral to secondary care community services.[61–63] Rather than a direct relationship between care delivery failures and immediate safety consequences, safety problems in community settings may less resemble an 'incident'. Risks may build over time where care is delivered across multiple, dispersed community settings, with patients and their carers playing a bigger role in patient safety, alongside involvement from several providers (e.g., GP practice, community mental health team, social care, community pharmacy).

A worked example (see Box 2) illustrates the operation of risks and their influence on organizational safety across a 1-year period in a patient journey. It is informed by a systems perspective, according to the Yorkshire Contributory Factors Framework mental healthcare adaptation (YCFF-MH).[25] This hypothetical scenario is based on a recent announcement by a UK pharmaceutical company of intentions to discontinue the production of Priadel® brand lithium carbonate modified-release tablets.[64]

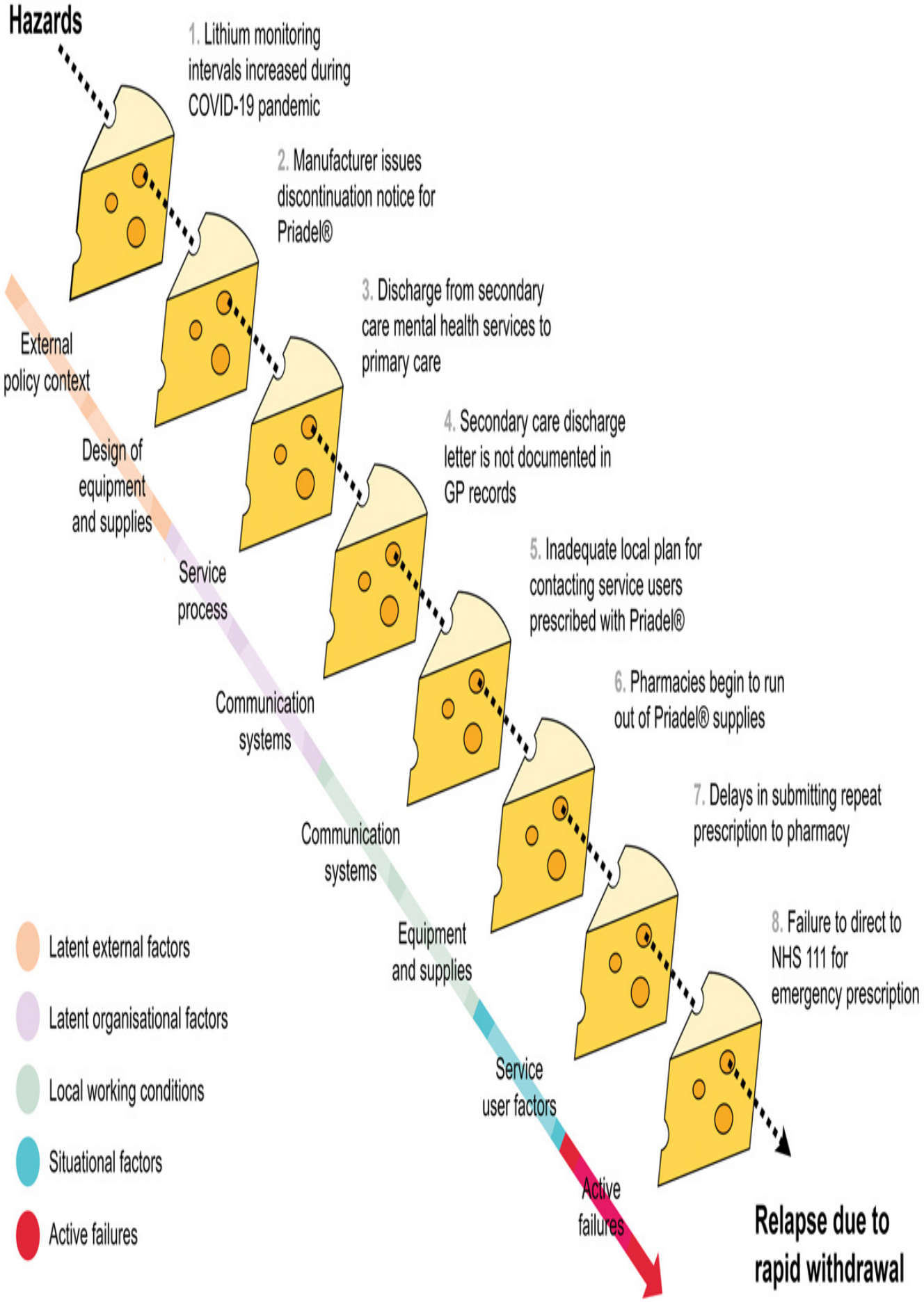
2.**BoxA hypothetical case example of a safety event in community-based mental health services**

Safety event	Relapse of service user with bipolar disorder after a long period of stability, caused by sudden discontinuation of lithium carbonate medication.
Outcome(s)	Psychiatric hospitalization, severe depressive episode and minor accidental physical injuries due to risky behaviours in manic episode.

Description of contributory factors	Due to the COVID-19 pandemic, guidance was issued recommending that lithium monitoring intervals were to be increased from 6 up to 9 months for stable patients (April 2020). Service user had been responding well to lithium carbonate (Priadel® brand) for 2 years whilst under the care of Community Mental Health Team. Six-monthly routine physical monitoring indicated no abnormalities (blood levels within therapeutic range, no problems with renal or thyroid function). Due to stability on medication regimen, service user was discharged from secondary care for continued monitoring in primary care. Service user informed by care coordinator that their GP practice would contact them directly when required to arrange a follow-up consultation in primary care (July 2020). No documentation of secondary care discharge letter in GP records. Therefore, Community Mental Health Team still presumed to be responsible for medication and physical health monitoring. No attempt made to contact service user (July 2020). Essential Pharma Ltd. announced a discontinuation of Priadel® from April 2021 onwards (September 2020). GP practice began to contact service users prescribed with Priadel® to arrange medication reviews and to plan transition to a different brand of lithium. Service users only contacted if records indicate that there is no current Community Mental Health Team care package in place. No check made on whether secondary care services were managing these transitions for their caseloads. No attempt made to contact service user (September 2020). Service user running low on their medication supply and was delayed in submitting repeat prescription to pharmacy (February 2021). Pharmacy supplies of Priadel® had already run out when service user submitted repeat prescription form (March 2021). Pharmacist explained to service user about discontinuation of Priadel® brand, recommended alerting their GP and requesting prescription review. Pharmacist failed to advise service user to contact NHS 111 for advice or emergency prescription. No check of how many tablets service user had left (March 2021). Service user abruptly ran out of medication before contact could be made with GP. Sudden discontinuation led to acute episode of mania resulting in hospitalization (March 2021).

Lithium carbonate is prescribed for mood stabilization in individuals with bipolar disorder and treatment-resistant depression. Due to its narrow therapeutic index, regular monitoring must be undertaken, owing to the risks of toxicity. The Priadel® brand of lithium carbonate is widely used in the United Kingdom, with over 750,000 prescriptions of either the 200 or 400 mg strength tablets documented by GP practices across England in the year preceding November 2020.[65] Due to pressure from prescribers,[66] the Department for Health and Social Care reached an agreement with the manufacturers for continued supply of the medication to the United Kingdom.[67] This scenario, based on its hypothetical discontinuation, exemplifies the importance of investigating safety over a

longer period of patients' community care journeys. Event reviews confined to so-called ‘sharp end’ factors immediately preceding the service user's hospitalization would likely fail to capture broader systemic problems, obscuring key sources of learning. The roles of multiple contributory factors across the wider system are exhibited, including changes to national care guidance, communication problems and active failures. These accumulating hazards, which successfully surmount defences built into the system, are depicted across the trajectory of a patient's care journey in Figure 1.



Enlarge this image.

Implications and proposed directions for future research and practice

Seeking a resolution to the problems described, in what follows, we propose several directions for safety science

applied to community mental health care (Box 3).

3.**BoxLooking forward: Next steps to advance mental health patient safety in the community**

Agreed definitions of safety	Shared definitions of what constitutes a patient safety problem in this context must be developed, to agree on an agenda for improving safety.
A wider remit for safety	Evaluation of 'safe' community services must not be centred around a limited number of recognized adverse incidents only (e.g., incidents of violence and aggression). Attention must be paid to what makes service users feel safe or unsafe, along with broader, upstream determinants of safety (e.g., safe waiting times).
Measure safety over time	Efforts to investigate and measure safety in community-based mental health care must be designed with long-term care journeys in mind.
Make greater use of theory and evidence	Opportunities should be identified to learn from existing patient safety theory and evidence, which may have its origins within other care specialties or settings.
A nuanced approach to intervention	A wider range of safety interventions are needed to target systems factors which impact the safety of care, moving beyond a focus on direct service user and staff factors alone.
Draw on wider sources of 'safety intelligence'	Beyond traditional academic evidence, we must also seek to understand what can be learned from existing unpublished literature and local quality improvement work. Likewise, our approach to safety improvement in community services must be shaped by and with mental health service users and carers.

A need for shared definitions of patient safety in mental health care

Without shared definitions, nomenclature and agreed indicators of safety as applied to mental health care, research has observed safety through an overly narrow lens. A significant proportion of published research in this field concerns self-harm and suicides among mental health patients, due to the severity of harm caused by these incidents.[68–70] Although this research is vital to suicide reduction, other key risks must be identified and targeted in future research.

Efforts are required to define and determine the remit of patient safety in mental health care, which influences the aims of corresponding interventional work. It has been argued that safety in mental health care can be defined narrowly or broadly, with the former comprising a series of adverse events such as suicide and medication errors and the latter encompassing wider matters pertaining to the quality of care, service access and stigma.[21] An ambitious, broader mandate risks weakening the impact of efforts to improve safety, yet the authors anticipate that an overly narrow conceptualization could undermine its effectiveness, by failing to target underlying systemic factors which obstruct safe care.[21] Standardization of language, definitions and development of practice standards in mental health patient safety are important prerequisites to effective safety measurement and improvement.[12]

A broader conceptualization of safety

Given the unmapped nature of research into safety in community-based mental health services, a robust approach to conceptualizing safety problems and wider system-level risk factors is essential. The following safety definition provides a starting point by beginning to accommodate the specifics of mental health care: 'the avoidance of

unintended unsafe or iatrogenic harm associated with mental healthcare—either an error in inappropriate treatment or an omission to detect unsafe behaviour'.[71] However, terminology directly associated with 'sharp end' failures or incidents (e.g., 'error', 'omission'), may not adequately represent latent, systemic contributors to safety that better reflect community care processes. This definition must be expanded to encompass the full range of safety issues associated with community care.

The appeal for a broader safety agenda in mental health care is echoed widely. Creative research methods are thought necessary to overcome dominant views of patient safety in mental health care,[72] which may have 'filtered out' certain safety considerations, such as the use of practices which cause patients to feel distress and powerlessness. Other researchers note that the boundaries of safety are blurred in community mental health care, aligning more closely with what has been conceptualized as 'quality' issues in other specialities.[73] It is plausible that accumulated poor-quality care experiences may culminate in less safe care.

Evaluate safety across the whole care journey

Recognizing the complexity of measuring safety in long-term and community care scenarios, a departure is needed from a simplistic incident-focused perspective, to suitably capture key determinants of safety which impact care over time (Figure 1). As such, a wider frame of safety event analysis is required to incorporate the whole patient journey.[74] Regarding community care, it has been asserted that: 'The concept of a patient safety incident, or even of adverse events, breaks down in these settings or is at least stretched to its limit' (p. 6).[57] These considerations are pertinent to understanding safety in any long-term, community-based care scenario.

Making greater use of safety theory and evidence

Without employing established patient safety theories and models, mental health services are unlikely to observe safety improvements beyond those already achieved by the immediate, service user and staff-directed interventions. Incidents of violence or self-harm on inpatient units have often been attributed to proximal factors,[75] including attentional lapses by staff members, or patients labelled as 'challenging'. However, the contributory role of wider organizational characteristics has seldom been explored.

A systems approach to safety views incidents in the context of dynamic, complex healthcare system factors which precipitate safety events.[76] The role of latent conditions (e.g., organizational culture, service resourcing), alongside localized workplace factors (e.g., team characteristics, staffing levels), have been identified in multiple models delineating systems safety.[25,76–78] To advance the field, system-wide conditions must be acknowledged.

Developing a wider range of safety interventions

Interventions to improve safety in mental health care have primarily targeted patient and staff-level factors only, with limited consideration of wider systems influences. For example, service users might receive directly therapeutic psychological interventions in response to self-harming incidents. Likewise, care teams may receive training aimed at preventing errors or other performance-related factors deemed relevant to the incident causation.

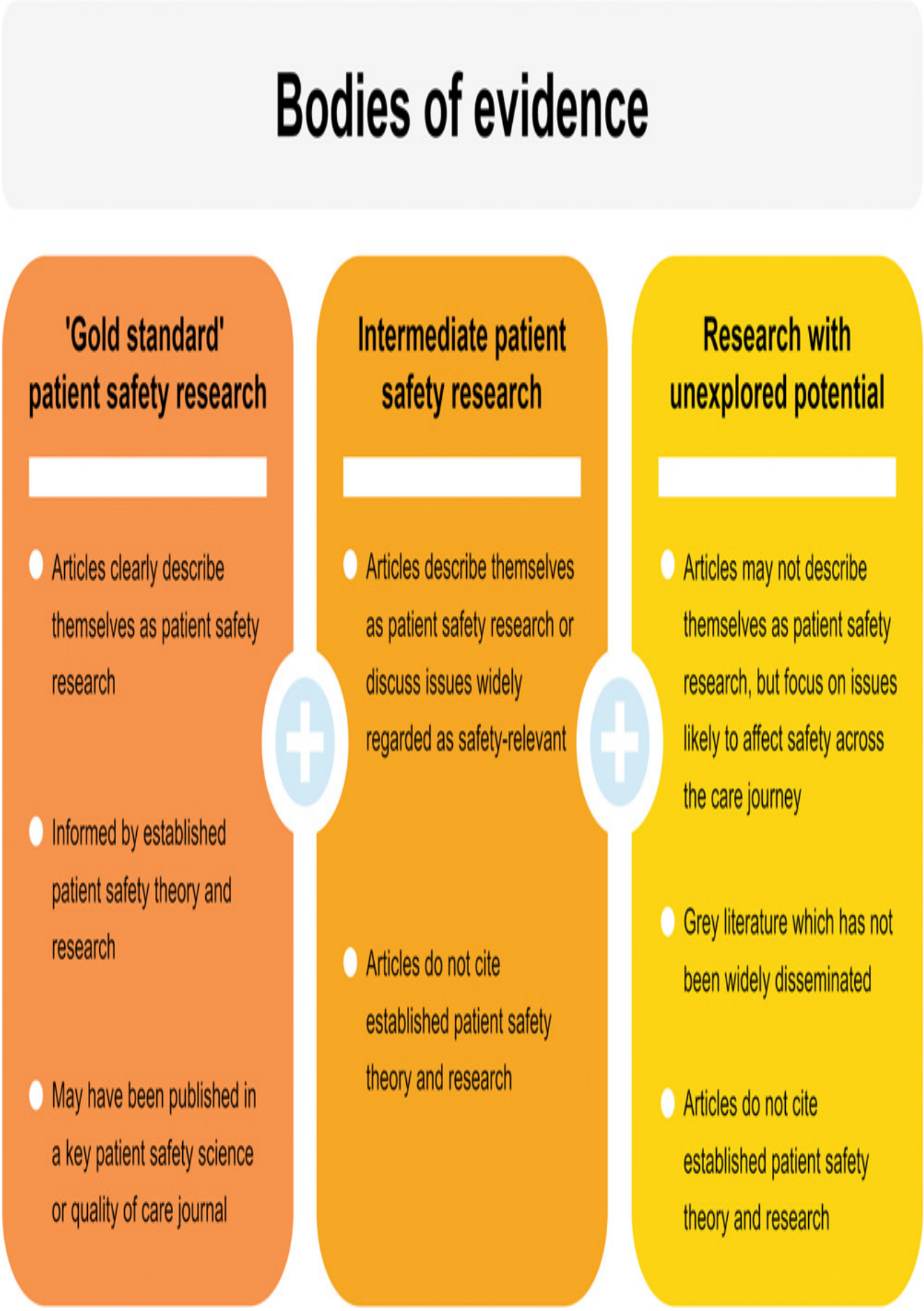
Whilst direct interventions are undoubtedly important, a broader, more nuanced approach is likely required to drive additive safety improvements. For instance, boredom due to inadequate activity provision in inpatient environments has been linked to aggression and self-harming.[79] Provision of structured evening activities (e.g., drama, animal therapy), was associated with reduced proportions of adolescent patients self-harming during evenings.[80] Similarly, the Safewards model,[81] evidenced reductions in safety outcomes such as self-harm, violence and restrictive practices within the inpatient environment, using an approach which addressed both local and systemic factors. Moreover, before-and-after analyses indicated that organizational and indirect factors (e.g., low staff turnover and family involvement in learning from suicides) were associated with reductions in suicide rates amongst mental health service users.[82] These factors comprise part of a toolkit for specialist mental health services and primary care, aimed at improving safety.[83] Together, these works suggest the causal interplay of both local and distal systemic conditions in safety events. For research to be best positioned to improve care, such factors must inform our understanding of organizational safety.

A place at the table for a wider range of informants on safety

Going forward, echoing views expressed by other researchers, we call for closer collaboration between mental

health care and the wider field of patient safety.[9] Future mental health safety-relevant research must position itself within the patient safety science discipline so that these services are acknowledged as a priority in global safety improvement agendas. Where synergies exist, safety research across different clinical specialities must aspire to build upon each other, rather than progressing in isolation. Moreover, for greater unification of the evidence base, barriers faced by quality improvement professionals to publishing their findings in traditional patient safety journals must be surmounted.[84] Where there is mission alignment to improve safety, 'messy' real-world improvement work of relevance to long-term conditions must be represented alongside traditional research methodologies within our growing understanding of safety.

However, to make the best use of existing evidence, our perspective of safety must not be limited only to research which fits well within both mental health and patient safety science research areas. This may obscure learning from other evidence bodies. Patient safety researchers must accommodate broader bodies of research, which may not yet have harnessed theories and principles from patient safety science, or have contextualized itself within the patient safety evidence base, alongside local service improvement reports or other 'grey' literature (see Figure 2).



Enlarge this image.

Likewise, we support calls for a bottom-up approach to safety in mental health care, both in terms of its conceptualization[85] and improvement initiatives, with the involvement of service users, carers and frontline staff.[8,45]

Historically, safety efforts may have been driven by top-down policy and regulations, to the detriment of aims to produce authentic improvements to service users' care.[86] In integrating diverse sources of safety intelligence, we will be best placed to improve care, by bringing together findings which are embedded in patient safety theory alongside those grounded in the reality of practice. This will provide a meaningful starting point for understanding safety in community mental health services.

CONCLUSION

Community-based mental health services must be at the forefront of future endeavours to define, measure and intervene to improve patient safety in mental health care. Although recent increases in research into safety in mental health care are encouraging, future research programmes must seek to expand the evidence base beyond psychiatric inpatient settings. It is also essential that research and innovation are not constrained to a limited range of safety problems, such as suicide and self-harm. Moreover, there is a lack of shared language and agreement over what constitutes a safety concern in the context of community-based mental health services. Going forward, we argue that the disparate bodies of existing research, with unexplored potential for understanding safety, must be integrated into our developing understanding of patient safety. Likewise, service users and carers must be involved in efforts to improve the safety of services. We hope this exploratory review of theoretical, conceptual and empirical challenges and discussion of potential approaches to their resolution will be useful to those seeking to advance this area of research.

AUTHOR CONTRIBUTIONS

All authors were involved in the conceptualization of this paper. Phoebe Averill developed the first draft of the manuscript. All other authors provided feedback on drafts of this paper, which were used to critically revise the manuscript. All authors have read and agreed to the published version of the manuscript.

ACKNOWLEDGEMENTS

This project is supported by the Health Foundation's grant to the University of Cambridge for The Healthcare Improvement Studies Institute (THIS Institute), grant number PHD-2018-01-026. Open Access publication of this article was made possible by King's College London's ‘Read &Publish’ agreement with Wiley (Wiley—Jisc). Nick Sevdalis' research is supported by the National Institute for Health and Care Research (NIHR) Applied Research Collaboration (ARC) South London at King's College Hospital NHS Foundation Trust. Nick Sevdalis is a member of King's Improvement Science, which offers co-funding to the NIHR ARC South London and comprises a specialist team of improvement scientists and senior researchers based at King's College London. Its work is funded by King's Health Partners (Guy's and St. Thomas' NHS Foundation Trust, King's College Hospital NHS Foundation Trust, King's College London and South London and Maudsley NHS Foundation Trust), and the Guy's and St. Thomas' Foundation. The views expressed in this publication are those of the author(s) and not necessarily those of the Health Foundation, THIS Institute, the NIHR or the Department of Health and Social Care.

CONFLICT OF INTEREST

Nick Sevdalis is the director of London Safety and Training Solutions Ltd., which offers training in patient safety, implementation solutions and human factors to healthcare organizations. The remaining authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no new data were generated or analysed during the current study.

DETAILS

Subject:	Narratives; Mental health; Literature reviews; Public health; Patients; Research; Mental health care; Health care; Avoidable; Health status; Risk management; Community mental health services; Long-term care; Patient safety; Treatment outcomes; Safety; Understanding; Research projects; Health promotion; Caregivers; Empirical analysis; Health services; Mobilization; Quality of care; Intelligence; Risk assessment; Positioning; Clinical outcomes; Teams; Interviews; Safety research; Health professional-Patient communication; Medical personnel; Health care delivery; Long term health care; Safety management
Business indexing term:	Subject: Risk assessment
Location:	United Kingdom--UK
Identifier / keyword:	community mental health services; mental health; patient safety; quality of care
Publication title:	Health Expectations; Oxford
Volume:	26
Issue:	1
Pages:	51-63
Publication year:	2023
Publication date:	Feb 2023
Section:	REVIEW ARTICLES
Publisher:	John Wiley &Sons, Inc.
Place of publication:	Oxford
Country of publication:	United States, Oxford
Publication subject:	Public Health And Safety
ISSN:	13696513
e-ISSN:	13697625
Source type:	Scholarly Journal
Language of publication:	English
Document type:	Journal Article
Publication history :	
Online publication date:	2022-11-12

Milestone dates:	2022-09-27 (manuscriptRevised); 2023-01-20 (publishedOnlineFinalForm); 2022-06-21 (manuscriptReceived); 2022-11-12 (publishedOnlineEarlyUnpaginated); 2022-10-25 (manuscriptAccepted)
Publication history :	
First posting date:	12 Nov 2022
DOI:	https://doi.org/10.1111/hex.13660
ProQuest document ID:	2767101839
Document URL:	https://www.proquest.com/scholarly-journals/conceptual-practical-challenges-associated-with/docview/2767101839/se-2?accountid=211160

| **Last updated:** | 2023-11-08 |
| **Database:** | Public Health Database,Publicly Available Content Database |

Hae-Ra Han, Mendez, K. J. W., Perrin, N., Cudjoe, J., Taylor, G., Baker, D., . . . Sharps, P. (2023). Community-based health literacy focused intervention for cervical cancer control among black women living with human immunodeficiency virus: A randomized pilot trial. Health Expectations, 26(1), 172-182. doi:https://doi.org/10.1111/hex.13644

BackgroundHealth literacy plays an essential role in how individuals process health information to make decisions about health behaviours including cancer screening. Research is scarce to address health literacy as a strategy to improve cancer screening participation among women living with human immunodeficiency virus (HIV), particularly Black women who, despite the heavy burden of cervical cancer, report consistently low screening rates.AimTo assess the feasibility, acceptability and preliminary efficacy of a health literacy-focused intervention called CHECC-uP—Community-based, HEalth literacy focused intervention for Cervical Cancer control—among women living with HIV.MethodsWe conducted a community-based, single-blinded randomized pilot trial. A total of 123 eligible women were enrolled and randomized to one of two conditions, control (i.e., cervical cancer brochure) or intervention (cervical cancer brochure plus 30–60 min health literacy-focused education followed by monthly phone counselling and navigation assistance for 6 months). Study assessments were done at baseline, 3 and 6 months. The final analysis sample included 58 women who completed all data points and whose Papanicolaou (Pap) test status was confirmed by medical records.ResultsAll intervention participants who completed the programme would recommend the CHECC-uP to other women living with HIV. However, adherence in the experimental conditions was low (49.6% attrition rate including 20 women who dropped out before the intervention began) due, in large part, to phone disconnection. Those who had received the intervention had a significantly higher Pap test rate compared to women in the control group at 6 months (50% vs. 21.9%, p = .025). Participation in the intervention programme was associated with improved health literacy and other psychosocial outcomes at 3 months but the trend was attenuated at 6 months.ConclusionsThe CHECC-uP was highly acceptable and led to improved Pap testing rates among Black women living with HIV. Future research should consider addressing social determinants of health such as phone connectivity as part of designing a retention plan targeting low-income Black women living with HIV.ImplicationsThe findings should be incorporated into a future intervention framework to fulfil the unmet needs of Black women living with HIV to facilitate their decision-making about Pap test screening.Patient or Public ContributionNineteen community members including women living with HIV along with HIV advocates and care providers participated in four focus groups to develop cervical cancer screening decision-relevant information and the health literacy intervention. Additionally, a community advisory board was involved to provide guidance in the general design and conduct of the study.

Heckemann, B., Graf, T., Eva, J. U., Jakobsson, S., Ragnarsson, O., Olsson, D. S., & Blomdahl, C. (2023). The importance of personal documentation for patients living with long-term illness symptoms after pituitary surgery: A constructivist grounded Theory study. Health Expectations, 26(1), 226-236. doi:https://doi.org/10.1111/hex.13648

IntroductionDespite surgical treatment, pituitary adenomas often cause long-term illness symptoms, that profoundly impact patients' quality of life physically, psychologically and socially. Healthcare professionals often fail to recognize and discuss the ensuing problems. Personal documentation, such as symptom monitoring, reflective writing or even posts on social media, may help this patient group to manage their daily life and support communication of their care needs. Documentation strategies and the role of documentation for people with long-term symptoms after pituitary adenoma surgery are currently unknown.AimTo examine the effects and strategies of documenting symptoms, activities and physical and emotional well-being among people living with long-term pituitary adenoma.MethodsIn this Constructivist Grounded Theory study, 12 individuals living with long-term illness symptoms after pituitary adenoma surgery described their documentation strategies in in-depth interviews using teleconferencing and photo-elicitation between August and October 2020.ResultsStrategies for documentation included analogue and digital media. One core category (Exercising autonomy) and three categories describing processes (Gaining insight, Striving for control and Sharing) emerged from the analysis. These three interrelated processes become an expression of autonomy to manage life and make sense of chronic illness. Personal documentation is a flexible tool

that is used more extensively in times of ill health and less in times of relative well-being. Sharing documentation with healthcare professionals facilitated care planning and sharing with friends and family fostered emotional well-being.ConclusionPersonal documentation is a valuable resource for managing life after pituitary adenoma surgery. The current findings may be relevant to other chronic illnesses. Further research exploring potential tools for personal documentation is needed.Patient or Public ContributionWe deliberately chose a Constructivist Grounded Theory approach for this interview study. Using Constructivist Grounded Theory, we gave people living with long-term symptoms a voice, allowing them to freely speak about managing their illness in connection with personal documentation. The theoretical sampling approach enabled us to invite participants that could provide a broad overview of the landscape of personal documentation.

De Sola, H., Failde, I., Estalella, I., & Maquibar, A. (2023). Becoming a secondary actor of one's own life: A qualitative study of the experiences of informal caregivers in the care of people with chronic pain. Health Expectations, 26(1), 409-418. doi:https://doi.org/10.1111/hex.13671

IntroductionThe physical limitations experienced by people with chronic pain (CP) produce a greater need for care and assistance, most of which is provided by an informal caregiver (IC). Despite the key role ICs play in the everyday lives of individuals living with CP, knowledge about their experiences and needs is limited. We aimed to address this limitation by exploring the experiences of IC of people with CP.MethodsThis is a qualitative descriptive study using semistructured interviews. Participants were 12 ICs purposively chosen from the Unit of Pain at the University Hospital in Cádiz. Individual interviews were recorded, transcribed verbatim and analysed following thematic analysis.ResultsWe developed one overarching theme ‘Becoming a secondary actor of one's own life’ and three themes: 1. Key elements that shape a caregiver's experiences; 2. It's the hand that life dealt me; 3. The burden of being a caregiver and coping strategies.ConclusionsThis study's findings highlight how the CP impacts IC lives. Being an IC for a relative with CP became the most important role in the IC's life, to the point of casting a shadow over their own needs. Besides, participants felt not having other options but to keep going with that role. Yet, the context was essential in shaping the experiences as caregivers and the burden derived from caregiving. In this line, differences related to gender roles were found in the narratives of participant women and men.Patient or Public ContributionParticipants were purposively chosen from the Unit of Pain at the University Hospital ‘Puerta del Mar’ who attended the consultation accompanying their relatives. All the eligible participants were approached by the clinician. After this initial approach by the clinician, one of the researchers met the potential participant and they went to a quieter place in a clinical setting for the interview, before which the participant was shown a letter with more comprehensive information about the study and its aim. The participants were left alone to read and think carefully before giving their written informed consent. Participation was voluntary and the subjects received no financial contribution for their time.

Babione, J., Panjwani, D., Murphy, S., Kelly, J., Jessica, V. D., Santana, M., . . . Rabi, D. (2023). Alignment of patient-centredness definitions with real-life patient and clinician experiences: A qualitative study. Health Expectations, 26(1), 419-428. doi:https://doi.org/10.1111/hex.13674

IntroductionPatient-centred care (PCC) has come to the forefront for many institutions, funding agencies and clinicians, and is integrated into care. Does a disconnect in understanding still exist between patients, healthcare organizations and clinicians in what PCC means and how outstanding issues might be addressed?MethodsWe conducted interviews and focus groups with self-reported chronic care patients and clinicians providing care to these patients exploring PCC experiences, expectations and practices. These data were initially analysed using inductive thematic analysis. This paper reports on the findings of a secondary analysis examining the alignment between patients and clinicians on five key predetermined dimensions of PCC.ResultsEighteen patients participated, representing a range of chronic conditions. Thirty-eight clinicians participated. One thousand and three hundred patient and 1800 clinician codes were identified and grouped into 5 main topics with 140 unique themes (patients) and 9 main topics with 54 unique themes (clinicians). A total of 166 quotes (patient = 93, clinician = 73) were identified for this PCC definition alignment analysis. Partial or complete alignment of patient and clinician perspectives was seen on most dimensions. Key disconnects were observed in patient involvement, patient empowerment and clinician–patient communication. Only 18% of patients reported experiencing patient-centred communication,

whereas 57% of clinicians reported using patient-focused communication approaches.ConclusionOverall, study patients and clinicians endorse that many PCC elements occur. This study highlights key differences between patients and clinicians, suggesting persistent challenges. Clinician participants relayed their PCC approaches of informing and educating patients; however, patients often perceive these approaches as didactic, role-diminishing and noncollaborative. Collaborative approaches, such as shared decision-making, hold promise to bridge persistent PCC gaps and should be integrated into medical education programmes.Patient or Public ContributionThis project was conceived and executed with a co-design approach wherein patients with chronic conditions who are trained in research (i.e., see descriptions of Patient and Community Engagement Research in the text) were involved in all stages of the research project alongside other researchers on the project team. Healthcare providers were involved as participants and as principal investigators in the project.

Montori, V. M., Ruissen, M. M., Branda, M. E., Hargraves, I. G., & Kunneman, M. (2023). Problem-based shared decision making: The role of canonical SDM steps. Health Expectations, 26(1), 282-289. doi:https://doi.org/10.1111/hex.13654

ObjectiveTo evaluate the extent to which the canonical steps of shared decision making (SDM) take place in clinical encounters in practice and across SDM forms.MethodsWe assessed 100 randomly selected video-recorded primary care encounters, obtained as part of a randomized trial of an SDM intervention in patients with type 2 diabetes. Two coders, working independently, noted each instance of SDM, classified it as one of four problem-based forms to SDM (weighing alternatives, negotiating conflicting issues, solving problems, or developing existential insight), and noted the occurrence and timing of each of the four canonical SDM steps: fostering choice awareness, providing information, stating preferences, and deciding. Descriptive analyses sought to determine the relative frequency of these steps across each of the four SDM forms within each encounter.ResultsThere were 485 SDM steps noted (mean 4.85 steps per encounter), of which providing information and stating preferences were the most common. There were 2.7 (38 steps in 14 encounters) steps per encounter observed in encounters with no discernible SDM form, 3.4 (105 steps in 31 encounters) with one SDM form, 5.2 (129 steps in 25 encounters) with two SDM forms, and 7.1 (213 steps in 30 encounters) when ≥3 SDM forms were observed within the encounter. The prescribed order of the four SDM steps was observed in, at best, 16 of the 100 encounters. Stating preferences was a common step when weighing alternatives (38%) or negotiating conflicts (59.3%) but less common when solving problems (29.2%). The distribution of SDM steps was similar to usual care with or without the SDM intervention.ConclusionThe normative steps of SDM are infrequently observed in their prescribed order regardless of whether an SDM intervention was used. Some steps are more likely in some SDM forms but no pattern of steps appears to distinguish among SDM forms.Clinical Trial RegistrationClinicalTrial.gov: NCT01293578.

Sánchez, T. E., Matvienko-Sikar, K., Meaney, S., & O'Donoghue, K. (2023). Exploring first-time mothers' experiences and knowledge about behavioural risk factors for stillbirth. Health Expectations, 26(1), 329-342. doi:https://doi.org/10.1111/hex.13662

BackgroundModifiable factors such as substance use, lack of attendance at antenatal care, overweight or obesity and sleeping position are associated with a higher risk of stillbirth. This qualitative study aimed to explore women's experiences of modifiable factors during pregnancy and their awareness of stillbirth.MethodsPurposive sampling was implemented by hospital staff in a large tertiary maternity hospital in Ireland between November 2020 and March 2021. Women were approached during their stay in the hospital and were invited to participate in a semistructured interview 3–5 months later. Eligible women were primiparous, >18 years of age and had an uncomplicated pregnancy and delivery. Eighteen women who consented to be followed up were interviewed at 3–5 months postpartum. Thematic analysis was used to analyse the data.ResultsFour themes were identified: attitudes towards behaviour change, awareness regarding stillbirth and risk factors, the silence around stillbirth and risks, and attitudes towards receiving information about stillbirth. Women spoke about behaviour change in terms of outcomes, and most changes (e.g., ceasing alcohol consumption) were perceived as easy to manage. Awareness of stillbirth was limited among the women interviewed, and the association between risk behaviours and stillbirth was not known by any woman. Results suggest that there is a silence around stillbirth, including in antenatal care, which hinders information provision. However, most women highlighted the value of receiving information and extra education

about modifiable risk factors and stillbirth.ConclusionThere is a general lack of understanding of the link between behavioural risk factors and potential pregnancy outcomes such as stillbirth. Providing further information to women about stillbirth and providing additional support with behaviour change might contribute to enhancing preventive efforts.Patient or Public ContributionPatients were involved in this study by providing their experiences of antenatal care which were used as primary data.

Carlisle, E. M., Shinkunas, L. A., Ruba, E., Klipowicz, C. J., Lieberman, M. T., Hoffman, R. M., & Reisinger, H. S. (2023). A valued voice: A qualitative analysis of parental decision-making preferences in emergent paediatric surgery. Health Expectations, 26(1), 531-541. doi:https://doi.org/10.1111/hex.13686

IntroductionShared decision-making, with an emphasis on patient autonomy, is often advised in healthcare decision-making. However, this may be difficult to implement in emergent settings. We have previously demonstrated that when considering emergent operations for their children, parents prefer surgeon guidance as opposed to shared decision-making. Here, we interviewed parents of paediatric patients who had undergone emergent operations to better understand parental decision-making preferences.MethodsParents of paediatric patients who underwent surgery over the past 5 years at a University-based, tertiary children's hospital for cancer, an emergent operation while in the neonatal intensive care unit (NICU) or extracorporeal membrane oxygenation (ECMO) were invited to complete a 60-min semi-structured interview. Interviews were digitally recorded and transcribed verbatim. Thematic content analysis was performed via deductive and inductive analysis. An iterative approach to thematic sampling/data analysis was used.ResultsThematic saturation was achieved after 12 interviews (4 cancer, 5 NICU and 3 ECMO). Five common themes were identified: (1) recommendations from surgeons are valuable; (2) ‘lifesaving mode’: parents felt there were no decisions to be made; (3) effective ways of obtaining information about treatment; (4) shared decision-making as a ‘dialogue’ or ‘discussion’ and (5) parents as a ‘valued voice’ to advocate for their children.ConclusionsWhen engaging in decision-making regarding emergent surgical procedures for their children, parents value a surgeon's recommendation. Parents felt that discussion or dialogue with surgeons defined shared decision-making, and they believed that the opportunity to ask questions gave them a ‘valued voice’, even when they felt there were no decisions to be made.Patient or Public ContributionFor this study, we interviewed parents of paediatric patients who had undergone emergent operations to better understand parental decision-making preferences. Parents thus provided all the data for the study.

Kim, F. S., Sawyer, K., Daryan, S., Allen, J., & Taylor, G. (2023). Service-user experiences of an integrated psychological intervention for depression or anxiety and tobacco smoking in improving access to psychological therapies services: A qualitative investigation into mechanisms of change in quitting smoking. Health Expectations, 26(1), 498-509. doi:https://doi.org/10.1111/hex.13684

IntroductionHigh smoking prevalence leads to increased morbidity and mortality in individuals with depression/anxiety. Integrated interventions targeting both smoking and mood have been found to be more effective than those targeting smoking alone, but the mechanisms of change of these interventions have not been investigated. This qualitative study aimed to understand participants' experiences of the mechanisms underlying change in smoking behaviour following an integrated cognitive behavioural technique-based intervention for smoking cessation and depression/anxiety.MethodsThis study was embedded within an ongoing randomized-controlled acceptability and feasibility trial (http://www.isrctn.com/ISRCTN99531779). Semistructured interviews were conducted with 15 IAPT service users. Data were analysed using thematic analysis. During the interviews, participants were asked open-ended questions about their quitting experience and perception of how the intervention aided their behaviour change.ResultsFive themes were identified. Acquiring an increased awareness of smoking patterns: participants described an increased understanding of how smoking was contributing towards their mental health difficulty. Developing individualized strategies: participants described acquiring ‘a couple of tricks up your sleeve’ that were helpful in making smoking cessation feel more ‘manageable’. Practitioner style as ‘supportive but not lecture-y’: participants expressed how important the therapeutic alliance was in helping change their smoking behaviour. Importance of regular sessions: participants expressed the importance of ‘having someone that's checking in on you’. Having the opportunity to access the intervention at ‘the right time’: participants described the intervention as the ‘push’ that they ‘needed’.ConclusionsParticipants identified key factors towards smoking

behaviour change. Perceived increased awareness of how smoking negatively impacted participants' mental health, and the opportunity to be offered smoking cessation treatment in a ‘non-judgemental’, ‘supportive’ environment, with regular sessions and individualized strategies contributed to successful smoking cessation outcomes. If similar results are found in more diverse samples, these aspects should be embedded within integrated interventions for smoking cessation and depression/anxiety.Patient or Public ContributionPersons with lived experience of depression, anxiety and tobacco addiction contributed towards the design of the interview schedule, participant information sheets and the debriefing process. This was to ensure that interview questions were relevant, nonjudgemental and acceptable for those who did not manage to quit smoking.

Crowther, D., McCulloch, H., Wong, H., Mackay, R., Johnson, C., Chorney, J., . . . Curran, J. (2023). Children, young people and parent engagement in health intervention design and implementation: A scoping review. Health Expectations, 26(1), 1-15. doi:https://doi.org/10.1111/hex.13572

IntroductionEngaging children and young people (CYP) with and without their parents in health research has the potential to improve the development and implementation of health interventions. However, to our knowledge, the scope of engagement activities used with this population and barriers to their engagement is unknown. The objective of this review was to identify and describe CYP engagement with and without their parents in the development and/or implementation of health interventions.MethodsThis scoping review included any primary research studies reporting on engaging CYP, with or without parents, in the design and/or implementation of health interventions. Healthcare professionals had to be involved over the course of the study and the study had to take place in either community, primary or tertiary care settings. The following databases were searched in May 2017, May 2020 and June 2021: Medline (OVID), CINAHL (EBSCO) and Embase (Elsevier). Two independent reviewers screened titles, abstracts and full-text articles and used a previously piloted extraction form to extract and summarize information from the included articles.ResultsTwenty-eight articles discussing twenty-four studies were included. CYP engagement throughout the research cycle was limited. There were no observed differences in the reported presence of engagement, types of interventions or outcomes of engagement between studies engaging CYP or CYP and parents. Studies engaging CYP and parents contained limited information on how these relationships affected outcomes of engagement. Engagement was enabled primarily by the maintenance of resources and relationships among stakeholders.ConclusionsAlthough CYP engagement often influenced health intervention and implementation design, they are inconsistently engaged across the research cycle. It is unclear whether parental involvement enhances CYP engagement. Future research should consider reporting guidelines to clarify the level of CYP and/or parent engagement, and enhance CYP engagement by fostering synergistic and sustainable partnerships with key stakeholders.Patient or Public ContributionA parent partner with codesign experience contributed to the creation of the research questions, screened titles, abstracts and full texts, helped with data extraction and provided feedback on the manuscript.

Talevski, J., Kulnik, S. T., Jessup, R. L., Falls, R., Cvetanovska, N., & Beauchamp, A. (2023). Use of co-design methodology in the development of cardiovascular disease secondary prevention interventions: A scoping review. Health Expectations, 26(1), 16-29. doi:https://doi.org/10.1111/hex.13633

IntroductionThere is growing evidence to support the use of co-design in developing interventions across many disciplines. This scoping review aims to examine how co-design methodology has been used in the development of cardiovascular disease (CVD) secondary prevention interventions within health and community settings.MethodsWe searched four academic databases for studies that used the co-design approach to develop their intervention. Studies were included if consumers (adults with CVD) and key stakeholders (e.g. clinicians, service providers) were involved in the co-design process. The review focused on methodology rather than traditional study outcomes; therefore, co-design processes and activities were extracted and evaluated against a selected co-design framework.ResultsTwenty-two studies were included in this review. Studies were implemented across various settings with consumers and stakeholder groups most frequently consisting of patients and healthcare professionals, respectively. Most studies specifically stated that they used a ‘co-design’ approach (n = 10); others used terms such as participatory action research (n = 3), user-centred design (n = 3) and community-based participatory research (n = 2). Although there was variability in terminology, co-design processes, and participants, all studies adhered to the

key principles of consumer engagement. Predominant co-design activities included semistructured interviews, focus groups, co-design/development workshops and advisory group meetings. Intervention effectiveness was assessed in eight studies showing mixed results.ConclusionsThis review provides an overview of how the co-design approach has previously been used in the development of CVD secondary prevention interventions. These findings provide methodological considerations that can guide researchers and healthcare services when implementing co-design to develop feasible and acceptable interventions that can improve outcomes for CVD populations.Patient or Public ContributionNo patients, service users, caregivers, people with lived experience or members of the public were involved in this scoping review. This review article was written by academics who have undertaken a significant amount of co-design work with consumers and stakeholders.

Hanlon, C. A., McIlroy, D., Poole, H., Chopra, J., & Saini, P. (2023). Evaluating the role and effectiveness of co-produced community-based mental health interventions that aim to reduce suicide among adults: A systematic review. Health Expectations, 26(1), 64-86. doi:https://doi.org/10.1111/hex.13661

BackgroundSuicide is a major public health risk requiring targeted suicide prevention interventions. The principles of co-production are compatible with tailoring suicide prevention interventions to meet an individual's needs.AimsThis review aimed to evaluate the role and effectiveness of co-produced community-based suicide prevention interventions among adults.MethodsFour electronic databases (PsycInfo, CINAHL, MEDLINE and web of science) were systematically searched. A narrative synthesis was conducted.ResultsFrom 590 papers identified through searches, 14 fulfilled the inclusion criteria. Most included studies elicited the views and perspectives of stakeholders in a process of co-design/co-creation of community-based suicide prevention interventions.ConclusionStakeholder involvement in the creation of community-based suicide prevention interventions may improve engagement and give voice to those experiencing suicidal crisis. However, there is limited evaluation extending beyond the design of these interventions. Further research is needed to evaluate the long-term outcomes of co-produced community-based suicide prevention interventions.Patient and Public InvolvementThis paper is a systematic review and did not directly involve patients and/or the public. However, the findings incorporate the views and perspectives of stakeholders as reported within the studies included in this review, and the findings may inform the future involvement of stakeholders in the design, development and delivery of community-based suicide prevention interventions for adults.

Silcock, J., Marques, I., Olaniyan, J., Raynor, D. K., Baxter, H., Gray, N., . . . Alldred, D. P. (2023). Co-designing an intervention to improve the process of deprescribing for older people living with frailty in the united kingdom. Health Expectations, 26(1), 399-408. doi:https://doi.org/10.1111/hex.13669

BackgroundIn older people living with frailty, polypharmacy can lead to preventable harm like adverse drug reactions and hospitalization. Deprescribing is a strategy to reduce problematic polypharmacy. All stakeholders should be actively involved in developing a person-centred deprescribing process that involves shared decision-making.ObjectiveTo co-design an intervention, supported by a logic model, to increase the engagement of older people living with frailty in the process of deprescribing.DesignExperience-based co-design is an approach to service improvement, which uses service users and providers to identify problems and design solutions. This was used to create a person-centred intervention with the potential to improve the quality and outcomes of the deprescribing process. A ‘trigger film’ showing older people talking about their healthcare experiences was created and facilitated discussions about current problems in the deprescribing process. Problems were then prioritized and appropriate solutions were developed. The review located the solutions in the context of current processes and procedures. An ideal care pathway and a complex intervention to deliver better care were developed.Setting and ParticipantsOlder people living with frailty, their informal carers and professionals living and/or working in West Yorkshire, England, UK. Deprescribing was considered in the context of primary care.ResultsThe current deprescribing process differed from an ideal pathway. A complex intervention containing seven elements was required to move towards the ideal pathway. Three of these elements were prototyped and four still need development. The complex intervention responded to priorities about (a) clarity for older people about what was happening at all stages in the deprescribing process and (b) the quality of one-to-one consultations.ConclusionsPriorities for improving the current deprescribing process were successfully identified.

Solutions were developed and structured as a complex intervention. Further work is underway to (a) complete the prototyping of the intervention and (b) conduct feasibility testing.Patient or Public ContributionOlder people living with frailty (and their informal carers) have made a central contribution, as collaborators, to ensure that a complex intervention has the greatest possible potential to enhance the experience of deprescribing medicines.

Madill, A., Duara, R., Goswami, S., Graber, R., & Hugh-Jones, S. (2023). Pathways to recovery model of youth substance misuse in assam, india. Health Expectations, 26(1), 318-328. doi:https://doi.org/10.1111/hex.13658

IntroductionThere are global calls for better understanding of substance use disorder (SUD) to inform prevention, risk reduction and treatment of this relapse-prone disorder. Our aim in this article is to understand the pathways to recovery of youth in Assam, India who have suffered SUD.MethodsWe recruited 15 participants (11 men and 4 women) via two rehabilitation facilities. All are addicts-in-recovery aged 19–24 years. Material was generated through photo-led interviews, analysed using an inductive variant of thematic analysis and the resulting model refined through expert and participant checks.ResultsWe present a multiroute, multidirectional pathway to recovery model. It has three phases, Recreational Use, Addiction (Relaxed, Chaotic, Strategic) and Supported Recovery, each phase consisting of cycling between, or transitioning through, a series of stages.ConclusionsThe model enhances psycho-socio-cultural insights into the experience of risk and recovery, and informs prevention and treatment for youth substance misuse in Assam. This is the first model of its kind and an important public health resource. We discuss the possible transferability of the model to a wider range of contexts.Patient or Public ContributionThe model presented was generated through analysis of interviews with addicts-in-recovery. Four of these addicts-in-recovery, and two mental health and rehabilitation service providers, conducted participant and expert checks of the model leading to its improvement.

Baz, S. A., Chao, F., Carpentieri, J. D., & Sheard, L. (2023). ‘I don't know what to do or where to go’. experiences of accessing healthcare support from the perspectives of people living with long covid and healthcare professionals: A qualitative study in bradford, UK. Health Expectations, 26(1), 542-554. doi:https://doi.org/10.1111/hex.13687

BackgroundIn October 2022, it was estimated 2.3 million people in the United Kingdom have self-reported Long Covid (LC). Many people have reported not receiving adequate healthcare support. There is a lack of research which provides an in-depth exploration of the barriers faced by people with LC in accessing healthcare support. It is important to understand these barriers to provide better support, care and advice for those experiencing LC.ObjectiveTo understand the barriers faced in accessing primary, secondary and specialist healthcare support for people with LC.Design and Participation40 interviews were conducted with people living with LC in Bradford alongside 12 interviews with healthcare professionals (HCPs) providing LC support in Bradford healthcare settings. Interviews were analysed using reflexive thematic analysis.ResultsPeople living with LC had a large degree of difficulty in accessing healthcare services for LC support. We categorized the healthcare access experiences of participants into five main types: (1) being unable to access primary care, (2) accessing primary care but receiving (perceived) inadequate support, (3) extreme persistence, (4) alternatives to mainstream health care and (5) positive experiences. There was a severe lack of access to specialist LC services. Ethnic minority participants faced a further barrier of mistrust and fear of services deterring them from accessing support. HCPs discussed systemic barriers to delivering services. Experiences were embedded in macrostructural issues further exacerbated by the pandemic.ConclusionTo better support people with LC, the barriers faced in accessing healthcare support must be addressed. Of significance, improvements to general practitioner access are required; especially as GPs are the first line of support for people living with LC.Patient and Public InvolvementA patient and public involvement group is engaged at regular intervals in the project.

McAuliffe, E., Sophie, M. S., Conlon, C., Rogers, L., De Brún, A., Mannion, M., . . . Quinlan, D. (2023). COVID-19 community assessment hubs in ireland: A study of staff and patient perceptions of their value. Health Expectations, 26(1), 119-131. doi:https://doi.org/10.1111/hex.13603

BackgroundCritical care bed capacity per capita in Ireland is among the lowest in Europe. The COVID-19 pandemic has put additional strain on an over-stretched healthcare system. COVID-19 community assessment hubs (CAHs)

were established to prevent unnecessary admission to acute hospitals and to reduce infection spread.ObjectiveThe aim of this study was to assess the effectiveness and acceptability of CAHs and identify how the service might be improved or adapted for possible future use.DesignThis was a mixed methods study, incorporating co-design with clinical stakeholders. Data collection was via an online survey and semistructured telephone interviews with staff and patients conducted between January and May 2021.Setting and participantsThirty-one patients completed the survey and nine were interviewed. Twenty interviews were conducted with staff.ResultsThe findings suggest that the CAH model was successful in providing a dedicated pathway for assessing patients with COVID-19 symptoms, whilst mitigating the risk of infection. Patients were particularly positive about the timely, comprehensive and holistic care they received, as well as the accessibility of the clinics and the friendly attitudes of the staff. Staff welcomed the training and clinical protocols which contributed to their feelings of safety and competency in delivering care to this cohort of patients. They also highlighted the benefits of working in a multidisciplinary environment. Both staff and patients felt that the hubs could be repurposed for alternative use, including the treatment of chronic diseases.DiscussionThis study describes staff and patients' experiences of these hubs. An unexpected outcome of this study is its demonstration of the true value of effective multidisciplinary working, not only for the staff who were deployed to this service but also for the patients in receipt of care in these hubs.ConclusionThis multidisciplinary patient-centred service may provide a useful model for the delivery of other services currently delivered in hospital settings.Patient or Public ContributionAn earlier phase of this study involved interviews with COVID-19-positive patients on a remote monitoring programme. The data informed this phase. Several of the authors had worked in the CAHs and provided valuable input into the design of the staff and patient interviews.

Lian, O. S., Nettleton, S., Grange, H., & Dowrick, C. (2023). ‘It feels like my metabolism has shut down’. negotiating interactional roles and epistemic positions in a primary care consultation. Health Expectations, 26(1), 366-375. doi:https://doi.org/10.1111/hex.13666

IntroductionOur aim is to explore the ways in which a patient and a general practitioner (GP) negotiate knowledge claims stemming from different epistemic domains while dealing with a mismatch between experiential and biomedical knowledge during the clinical consultation. We interpret their interaction in relation to the sociocultural context in which their negotiation is embedded and identify factors facilitating their successful negotiation (a medical error is avoided).MethodsBased on a narrative analysis of a verbatim transcript of a complete naturally occurring primary care consultation, we explore the moment-to-moment unfolding of talk between the patient and the GP (two women).FindingsThe patient experiences symptoms of what she interprets as a thyroid condition, and indirectly asks for medication. She presents her case by drawing on experiential knowledge (‘it feels like my metabolism has shut down’) and biomedical knowledge (while suggesting a diagnosis and a diagnostic test). The GP informs her that her thyroid blood tests are normal and uses biomedical knowledge to explain why she turns down the patient's request. This stages a potential conflict between the patient's embodied experiential knowledge and the doctor's biomedical knowledge. However, during their encounter, the patient and the GP manage to co-construct the patient's illness story and make shared decisions about further actions.ConclusionThe transition from potential conflict to consensus is a result of the mutual efforts of two parties: a patient who persistently claims experiential as well as biomedical knowledge while at the same time deferring to the GP's professional knowledge, and a GP who maintains her epistemic authority while also acknowledging the patient's experiential and biomedical knowledge.Patient and Public ContributionOur empirical data are sourced from a data archive and patients were not involved in the design or conduct of the study, but our study is based on a naturally occurring clinical consultation with a patient.

Helps, Ä., O'Donoghue, K., O'Connell, O., & Leitao, S. (2023). Bereaved parents involvement in maternity hospital perinatal death review processes: ‘Nobody even thought to ask us anything’. Health Expectations, 26(1), 183-198. doi:https://doi.org/10.1111/hex.13645

IntroductionThe death of a baby is devastating for parents, families and staff involved. Involving bereaved parents in their baby's care and in the maternity hospital perinatal death review can help parents manage their bereavement and plan for the future. In Ireland, bereaved parents generally have not been involved in this review process. The aim of our study was to assess parents' perception of how they may be appropriately involved in the maternity hospital perinatal death review in ways that benefit them and the review process itself.MethodsBereaved parents (n

=20) in Ireland were invited to take part in semistructured interviews. Thematic analysis was carried out on the interview transcripts.ResultsFour main themes were identified based on the participants' views and opinions on how they experienced the review process and how they feel this process may be improved. The themes reflect the journey of the parents through the different stages of the review process: Throughout process; On leaving the hospital; Interaction with the hospital ‘waiting in limbo’; Review itself. Identified subthemes highlighted essential aspects of this process and care provided to parents. For the parents, open, honest communication with staff, as well as having a key hospital contact was essential. Parents wished to provide feedback on their experience and wanted to be included in the review of their baby's death, in a way that was sensitive to their needs and the hospital's schedule.ConclusionA respectful, flexible system that allows bereaved parents' involvement in their baby's perinatal death review and is tailored to their needs is essential. A collaborative process between staff and parents can highlight clinical areas in need of change, enhance lessons learned, improve bereavement services and may prevent future perinatal deaths.Public ContributionBereaved parents were interviewed for this study.

Zago, L. F., Correa, J. S., da Silva-Brandão, R.,R., Fracolli, L. A., Padoveze, M. C., de Oliveira, S. M., & Currea, G. C. C. (2023). Experiences of antibiotic use among brazilian healthcare users: An exploratory study. Health Expectations, 26(1), 343-354. doi:https://doi.org/10.1111/hex.13664

IntroductionThis article analyzes experiences of antibiotic use and bacterial infections among Primary Health Care users of the Brazilian Unified Health System (SUS) and the possible implications for antimicrobial resistance (AMR). The aim is to map aspects that shape users' lay knowledge regarding antibiotics use and AMR.MethodsThis is an exploratory study, which consists primarily of individual in-depth interviews with 19 respondents. Recurrent interview topics were coded and analysed according to thematic content analysis.ResultsOur findings show users' lived experiences constitute three dimensions related to users' previous antibiotic use: (1) lay knowledge about medicines; (2) previous bacterial infections and (3) communication during the consultation. Lay knowledge encompasses the users' understanding of how antibiotics work in comparison to other drugs and experimentations they make with medication. Users' narratives about bacterial infections are divided into situations of urinary tract infections and antibiotic treatments for other conditions. Communication during the consultation is mainly characterized by a lack of shared knowledge and trust in the doctor–patient relationship.DiscussionUsers bring together knowledge learned from their own experiences to create the rationale, which shapes how they understand antibiotic use, bacterial infections and medical advice. These experiences are interwoven with information received from healthcare professionals (HPs) on these topics, creating a scenario that goes beyond professional information about antibiotic use. Users have knowledge about medication, antibiotics use and bacterial infection but do not have room to share it with HP, allowing lived experiences to take precedence over professional information.ConclusionUsers ascribe symbolic meanings to antibiotics creating a lay knowledge frame, even if this knowledge is not scientifically correct. The personal experiences of bacterial infections and their treatment are also an important source of knowledge about antibiotic use and AMR among users. Users demand from their HPs both trust and willingness to listen to their health narratives and experiences. By considering lay knowledge as part of the assessment of a user's health condition, rather than dismissing it as erroneous and therefore unworthy of attention, HPs may enhance the compliance of users.Patient or Public ContributionPatients or community members did not participate in the design stage of the study. Primary Care patients were invited to participate as respondents of in-depth interviews, which were carried out by the first author at a Primary Care Unit (PCU) in the suburb of Campo Limpo, Southern region of São Paulo, Brazil. Patients were interviewed after reading and signing a Free and Informed Consent Form, holding with them a copy of the Form. Among the final activities of the project, a feedback session at the same PCU is planned to report on the results of the study. All respondents will have the opportunity to contribute further information regarding their antibiotic use and exchange knowledge and experiences on antimicrobial resistance.

Lockyer, B., Moss, R. H., Endacott, C., Islam, S., & Sheard, L. (2023). Compliant citizens, defiant rebels or neither? exploring change and complexity in COVID-19 vaccine attitudes and decisions in bradford, UK: Findings from a follow-up qualitative study. Health Expectations, 26(1), 376-387. doi:https://doi.org/10.1111/hex.13667

BackgroundCOVID-19 vaccines have been the central pillar of the public health response to the pandemic, intended to enable us to ‘live with Covid’. It is important to understand change and complexity of COVID-19 vaccines attitudes and decisions to maximize uptake through an empathetic lens.ObjectiveTo explore the factors that influenced people's COVID-19 vaccines decisions and how their complex attitudes towards the vaccines had changed in an eventful year.Design and ParticipantsThis is a follow-up study that took place in Bradford, UK between October 2021 and January 2022, 1 year after the original study. In-depth phone interviews were conducted with 12 (of the 20 originally interviewed) people from different ethnic groups and areas of Bradford. Reflexive thematic analysis was conducted.ResultsEleven of the 12 participants interviewed had received both doses of the COVID-19 vaccine and most intended to have a booster dose. Participants described a variety of reasons why they had decided to have the vaccines, including the following: feeling at increased risk at work; protecting family and others in their communities; unrestricted travel and being influenced by the vaccine decisions of family, friends and colleagues. All participants discussed ongoing interaction with COVID-19 misinformation and for some, this meant they were uneasy about their decision to have the vaccine. They described feeling overloaded by and disengaged from COVID-19 information, which they often found contradictory and some felt mistrustful of the UK Government's motives and decisions during the pandemic.ConclusionsThe majority of participants had managed to navigate an overwhelming amount of circulating COVID-19 misinformation and chosen to have two or more COVID-19 vaccines, even if they had been previously said they were unsure. However, these decisions were complicated, demonstrating the continuum of vaccine hesitancy and acceptance. This follow-up study underlines that vaccine attitudes are changeable and contextual.Patient or Public ContributionThe original study was developed through a rapid community and stakeholder engagement process in 2020. Discussion with the Bradford Council Public Health team and the public through the Bradford COVID-19 Community Insights Group was undertaken in 2021 to identify important priorities for this follow-up study.

Lewis, G., Milnes, L., Adams, A., Schwarze, J., & Duff, A. (2023). Influences on indoor environmental trigger remediation uptake for children and young people with asthma: A scoping review. Health Expectations, 26(1), 87-97. doi:https://doi.org/10.1111/hex.13670

IntroductionChildren and young people (CYP) with asthma can benefit from reduced exposure to indoor environmental allergens and triggers but may not consistently have avoidance strategies implemented. To inform future interventions to increase trigger and allergen avoidance and enhance asthma control, a greater understanding of the influences on avoidance behaviours is necessary.MethodsA systematic scoping review was selected to summarize evidence on what influences family uptake of indoor environmental asthma trigger avoidance strategies for CYP with asthma and identify research gaps. Primary studies of any design, including CYP (≤18 years) with asthma, and/or parent-carers, available in English and conducted since 1993, were eligible. Searches included nine databases, hand-searching reference lists and citation searching.FindingsThirty-three articles were included and are summarized narratively due to heterogeneity. Influences appear complex and multifactorial and include barriers to strategy uptake, health beliefs and personal motivation. Research specifically related to family understanding of allergic sensitisation status and exposure risks, and how these may inform avoidance implementation is required. Patient and public involvement (PPI) was not reported in included articles, although two studies used participatory methods.ConclusionThere is limited research on family asthma trigger management, particularly what influences current management behaviours. Variation in families' ability to identify important triggers, understand exposure risk and consistently reduce exposures warrants further exploratory research to explain how families reach avoidance decisions, and what future interventions should aim to address. Further PPI-informed research to address such gaps, could enable theory-based, person-centred interventions to improve the uptake of asthma trigger remediation.Patient or Public ContributionAn asthma-specific PPI group contributed to the decision-making for the funding for the wider project this review sits within. The findings of this scoping review have informed the subsequent phases of the project, and this was discussed with PPI groups (both adult and CYP groups) when proposing the next phases of the project.

Cindy, Y. T., Eliza Lai-Yi Wong, Xu, R. H., Cheung, A. W., Dong, D., & Phoenix K.-H. Mo. (2023). Developing a health literacy scale for adults in hong kong: A modified e-delphi study with healthcare consumers and providers.

Health Expectations, 26(1), 245-255. doi:https://doi.org/10.1111/hex.13651

IntroductionHealth literacy (HL) refers to individuals' abilities to process and use health information to promote health. This study aimed to develop the first HL measurement tool for the Chinese Hong Kong population.MethodsA two-phase methodology was adopted. In Phase I, evidence synthesis with a deductive method was conducted to formulate the item list from the literature. In Phase II, a modified e-Delphi survey was conducted among stakeholders (i.e., healthcare providers and healthcare consumers) to confirm the content validity of the item list. The stakeholders were invited to rate the relevance of each draft item on a 4-point scale and provide suggestions for revisions, removal or adding new items.ResultsIn Phase I, a total of 34 items covering functional, interactive and critical HL were generated. In Phase II, to obtain a balanced view from experts and laypeople, healthcare professionals (n = 12) and consumers (n = 12) were invited to participate in the Delphi panel. The response rates of the three rounds were 100%. After the third round, the consensus was reached for 31 items, and no further comments for adding or revising items were received. All items exhibited excellent content validity (item content validity index: 0.79–1.00; K*: 0.74–1.00).ConclusionsA Health Literacy Scale for Hong Kong was developed. Compared with existing HL scales, the scale fully operationalized the skills involved in functional, interactive and critical HL. The Delphi study shows evidence supporting the high content validity of all items in the scale. In future studies, these items should undergo rigorous testing to examine their psychometric properties in our target population groups. By illuminating the details in the development process, this paper provides a deeper understanding of the scale's scope and limitations for others who are interested in using this tool.Patient or Public ContributionPublic as healthcare consumers, in addition to healthcare providers, were involved in developing a new HL scale for this study. The input from the public contributed to examining the scale's content validity by judging whether all items reflected the skills that they need to find and use health-related information in their daily life.

Olson, R. E., Smith, A., Good, P., Morgan, D., Gurgenci, T., & Hardy, J. (2023). ‘What price do you put on your health?’: Medical cannabis, financial toxicity and patient perspectives on medication access in advanced cancer. Health Expectations, 26(1), 160-171. doi:https://doi.org/10.1111/hex.13642

IntroductionFollowing 2016 legislation permitting limited access to cannabis for research and medicinal purposes, the number of randomized clinical trials (RCTs) investigating the effectiveness of medicinal cannabis (MC) on symptom burden relief in cancer contexts has increased in Australia. This study aimed to understand the perceptions, hopes and concerns of people with advanced cancer regarding the future availability and regulation of MC in Australia.MethodsThis qualitative study draws on semistructured interviews conducted between February 2019 and October 2020 in Brisbane, Australia, as part of an MC RCT substudy. Interviews were undertaken on 48 patients with advanced cancer in palliative care eligible to participate in an MC trial (n = 26 participated in an RCT; n = 2 participated in a pilot study; n = 20 declined). Interviews included a discussion of patients' decision-making regarding trial participation, concerns about MC and perceptions of future availability, including cost. Transcribed interviews were analysed inductively and abductively, informed by constructivist thematic analysis conventions.ResultsOverall, participants supported making MC legally accessible as a prescription-only medication. Fear of financial toxicity, however, compromised this pathway. Steep posttrial costs of accessing MC prompted several people to decline trial participation, and others to predict—if found effective—that many would either access MC through alternative pathways or reduce their prescribed dosage to enable affordable access.ConclusionsThese findings suggest that—despite a relatively robust universal healthcare system—Australians are potentially vulnerable to and fearful of financial toxicity. Prevalent in the United States, financial toxicity occurs when disadvantaged cancer patients access necessary but expensive medications with lasting consequences: bankruptcy, ongoing anxiety and cancer worry. Interview transcripts indicate that financial fears—and the systems sustaining them—may pose a threat to RCT completion and to equitable access to legal MC. Such findings support calls for embedding qualitative substudies and community partnerships within RCTs, while also suggesting the importance of subsidisation to overcoming injustices.Patient or Public ContributionA patient advisory committee informed RCT design. This qualitative substudy foregrounds patients' decision-making, perceptions and experiences.

Dadich, A., Kaplun, S., Kaplun, C., Hopwood, N., & Elliot, C. (2023). ‘it was that … specialist … that finally listened to us … that's probably a weird answer to what you were expecting’: Clinician and carer perspectives on brilliant

feeding care. Health Expectations, 26(1), 488-497. doi:https://doi.org/10.1111/hex.13683

IntroductionTo extend research on positive aspects of health care, this article focusses on health care for children who tube-feed—this is because knowledge about tube-feeding for children is limited and fragmented. This is achieved by consulting with clinicians and carers who supported children who tube-feed to clarify their understandings of and experiences with brilliant feeding care.MethodsNine clinicians and nine carers who supported children who tube-fed were interviewed. The interview transcripts were analysed thematically.ResultsFindings highlighted several features of brilliant feeding care—namely: practices that go above and beyond; attentiveness; empowerment; being ‘on the same page’; hopefulness and normalcy.ConclusionsThese findings show that seemingly trivial or small acts of care can make a significant meaningful difference to carers of children who tube-feed. Such accounts elucidate brilliant care as grounded in feasible, everyday actions, within clinicians' reach. The implications associated with these findings are threefold. First, the findings highlight the need for clinicians to listen, be attuned and committed to the well-being of children who tube-feed and their carers, share decision-making, source resources, and instil hope. Second, the findings suggest that carers should seek out and acknowledge clinicians who listen, involve them in decision-making processes, and continue to source the resources required to optimize child and carer well-being. Third, the findings point to the need for research to clarify the models of care that foster brilliant feeding care, and the conditions required to introduce and sustain these models.Patient or Public ContributionAll of the carers and clinicians who contributed to this study were invited to participate in a workshop to discuss, critique, and sense-check the findings. Three carers and one clinician accepted this invitation. Collectively, they indicated that the findings resonated with them, and they agreed with the themes, which they indicated were well-substantiated by the data.