Review

Views, Use, and Experiences of Web-Based Access to Pediatric Electronic Health Records for Children, Adolescents, and Parents: Scoping Review

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Abstract

Background: Ongoing efforts worldwide to provide patients with patient-accessible electronic health records (PAEHRs) have led to variability in adolescent and parental access across providers, regions, and countries. There is no compilation of evidence to guide policy decisions in matters such as access age and the extent of parent proxy access. In this paper, we outline our scoping review of different stakeholders' (including but not limited to end users) views, use, and experiences pertaining to web-based access to electronic health records (EHRs) by children, adolescents, and parents.

Objective: The aim of this study was to identify, categorize, and summarize knowledge about different stakeholders' (eg, children and adolescents, parents, health care professionals [HCPs], policy makers, and designers of patient portals or PAEHRs) views, use, and experiences of EHR access for children, adolescents, and parents.

Methods: A scoping review was conducted according to the Arksey and O'Malley framework. A literature search identified eligible papers that focused on EHR access for children, adolescents, and parents that were published between 2007 and 2021. A number of databases were used to search for literature (PubMed, CINAHL, and PsycINFO).

Results: The approach resulted in 4817 identified articles and 74 (1.54%) included articles. The papers were predominantly viewpoints based in the United States, and the number of studies on parents was larger than that on adolescents and HCPs combined. First, adolescents and parents without access anticipated low literacy and confidentiality issues; however, adolescents and parents who had accessed their records did not report such concerns. Second, the main issue for HCPs was maintaining adolescent confidentiality. This remained an issue after using PAEHRs for parents, HCPs, and other stakeholders but was not an experienced issue for adolescents. Third, the viewpoints of other stakeholders provided a number of suggestions to mitigate issues. Finally, education is needed for adolescents, parents, and HCPs.

Conclusions: There is limited research on pediatric PAEHRs, particularly outside the United States, and on adolescents' experiences with web-based access to their records. These findings could inform the design and implementation of future regulations regarding access to PAEHRs. Further examination is warranted on the experiences of adolescents, parents, and HCPs to improve usability and utility, inform universal principles reducing the current arbitrariness in the child's age for own and parental access to EHRs among providers worldwide, and ensure that portals are equipped to safely and appropriately manage a wide variety of patient circumstances.

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KEYWORDS

electronic health record; patient-accessible electronic health record; adolescents; parents; children; patient experience; patient portal; electronic portal; review; scoping review; youth; patient perspective; user experience; patient access; mobile phone

Introduction

Background

Patients being enabled to read their health records on the web is a growing phenomenon. Patient-accessible electronic health records (PAEHRs) commonly include clinical information (eg, physician visit notes, laboratory test results, medications, diagnoses, and referrals), and enabling patients to access their electronic health records (EHRs) is thought to promote patient empowerment by involving patients in their own care [1]. The term *open notes* is often used to describe the specific practice of giving patients access to the free-text entries written by clinicians [2] and is considered an important part of any PAEHR. The websites that host PAEHRs, commonly developed by so-called EHR vendors, are often referred to as patient portals and, for the purposes of this study, *patient portals* will refer to tethered, secure websites that hold any type of health information recorded by a health care provider that users have access to. Today, health institutions in >15 countries are developing patient portals [3], and there is continuous adaptation of legal frameworks at a national level to improve use and ensure patients' privacy [3,4].

An often-cited challenge to PAEHR implementation concerns how to manage access for parents, children, and adolescents [3]. The transfer of proxy access being managed by the parent or guardian (hereafter referred to as *parents*) into own access for the child is often conducted during adolescence, with the aim of protecting the adolescent's privacy as well as to support the transition to adulthood. The need for protection arises as the individual begins seeking care for sensitive medical conditions such as mental health or reproductivity. The child's need for autonomy in their relationship with their health care professional (HCP) is compromised during shared access with parents. So far, providers and countries have approached this dilemma in different ways. For example, the access age of the child varies, as well as when parents lose access and the age when patients gain self-access to their records. In some countries (eg, Finland and Estonia), parents are provided access (unless actively restricted by the child), whereas in other countries (eg, Sweden and Norway), parents are blocked from accessing records by law when their children reach a certain age threshold [5]. In France, adolescents receive access at the age of 18 years, when, in turn, the parents lose access. Decisions regarding earlier access in France can also depend on the perceived maturity of the minor. In many countries and regions, a lack of continuity in access to care is apparent [3]. In the United States, policies regarding age and privacy exceptions are dependent on state laws, which vary throughout the country. In 2021, the 21st Century Cures Act made it mandatory for health care providers to provide every patient with free electronic access to their clinical notes [6]. There is a possibility for withholding confidential information; however, questions still remain [7]. Evidently, the current lack of an international consensus on regulations for EHR access for parents, children, and adolescents has led to great variability.

The research of views, use, and experiences of PAEHRs to date has focused on HCPs and patients of the general adult population. The effects of PAEHRs are not conclusive, yet they indicate benefits including improved medication adherence and self-care as well as improved relationships between patients and their physicians [8-10]. However, a growing yet scarce body of literature is exploring access to EHRs for parents, children, and adolescents in particular. Although parents appreciate having access to their child's records into adolescence [11], shared access to PAEHRs for parents and adolescents runs the risk of causing ethical dilemmas for HCPs. For example, some health information may be considered sensitive by adolescents, such as health care data pertaining to the disclosure of alcohol or drug abuse, sexual activity, or stigmatized illnesses such as anxiety or depression. Adolescents have also been observed to withhold information from HCPs if they are uncertain about who may access it [12,13]. With regard to adolescents' self-access, it is thought that EHR access offers information transparency that might contribute to patient empowerment and enhanced health care; however, evidence suggests that the adolescent population requires targeted analysis. To date, one systematic review [14] has examined patient portals among pediatric patients. The review included only parents and adolescents and focused on empirical studies, and 10 of the 11 studies were based in the United States. Mostly positive feedback was found; however, there was some concern about medical literacy and its effects on the communication between adolescents, parents, and HCPs.

Study Objectives

The objective was to identify, categorize, and summarize knowledge about different stakeholders' (eg, children and adolescents, parents, HCPs, policy makers, and patient portal designers) views, use, and experiences of PAEHR access for children, adolescents, and parents. The findings will aid policy makers in designing future regulations regarding EHR access for parents and adolescents and potentially improve the design and implementation of PAEHRs to meet the needs of the end users. The concept "view" refers to attitudes, expectations, and thoughts; "use" refers to portal feature use and use rates; and "experience" includes experiences pertaining to, for example, satisfaction, concerns, and literacy. We use the definition of Davis et al [15] for a scoping review—"a synthesis and analysis of a wide range of research and nonresearch material to provide greater conceptual clarity about a specific topic or field of evidence"—with the adjustment of not including nonresearch material because of restrictions of the study search strategy. We defined *policy maker* as an agent with capacity or responsibility for deciding policies on PAEHRs (either national, regional, institutional, or as an HCP). The following research question was examined in detail: how do different stakeholders experience children's, adolescents', and parents' web-based access to the EHRs of children and adolescents? With regard to experiences of HCPs and HCP experts (among other stakeholders) who document in the records or manage the records within their professions, we focused on how these individuals perceive or are affected by the situation where children, adolescents, and parents have access to the EHRs of children and adolescents.

Methods

Scoping Review Approach

The full protocol for this review has been published previously [16]. To summarize, a literature search on PAEHRs for children, adolescents, and parents was conducted using the Arksey and O'Malley [17] framework. The framework includes six stages: (1) identifying the research question; (2) identifying relevant studies; (3) study selection; (4) charting the data; (5) collating, summarizing, and reporting the results; and (6) consulting with relevant stakeholders. To ensure reproducibility and traceability, the scoping review was conducted according to the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) extension for Scoping Reviews (PRISMA-ScR) checklist to report our results (Multimedia Appendix 1) [18].

Stage 1: Identifying the Research Question

Our research question was as follows: how do different stakeholders experience children's, adolescents', and parents' web-based access to the EHRs of children and adolescents?

Stage 2: Identifying Relevant Studies

A comprehensive literature search was conducted on June 23, 2021, by an experienced research librarian at Uppsala University, who provided the research team with the results immediately after conducting the search. The search included the following electronic literature databases: PubMed, CINAHL, and PsycINFO. The search included peer-reviewed literature published between 2005 and September 2021, where the year 2005 was chosen as a cutoff as we expected to not identify any relevant publications on pediatric PAEHRs before this. Search terms were identified using input from the research team and the literature. The references of the identified articles were scanned backward to identify prior work to consider for the research topic. The search query with Boolean operators was presented in the published protocol [16].

Stage 3: Selecting Eligible Studies

The inclusion and exclusion criteria were informed by the review process and were applied at the study selection stage.

Inclusion Criteria

Studies were included if they met the following criteria: (1) the patient user population was children, adolescents, and parents; (2) the population studied was children, adolescents, parents, HCPs, and other stakeholders; (3) outcomes were views, use, or experiences of access or proxy access to PAEHRs; and (4) the study design was all study types.

We defined patients aged ≤12 years as children, patients aged 13 to 18 years as adolescents, and those aged ≥18 years as adults. However, to increase the number of eligible studies for the adolescent population, the age of 19 to 20 years was included if a study participant group included a majority of adolescents (eg, aged 15-19 years).

Exclusion Criteria

Studies were excluded if they (1) were not written in English, (2) were published outside the study period, or (3) did not focus on pediatric PAEHRs.

Search Strategy

The search results were imported into the software program Rayyan (Rayyan Systems Inc) [19] according to the following headings: publication type, publication year, country, sample characteristics, setting, study aim, research question, and conclusions. Duplicates were removed. Titles and abstracts were screened by the authors with consideration of the eligibility criteria. The articles were divided between the investigators (excluding IS) so that each article was screened by at least 2 people. Any disagreements were resolved through group discussion and, if needed, with the addition of a third reader.

Stage 4: Charting the Data

The first author set up a Microsoft Excel spreadsheet to which all researchers added information independently, including the following study characteristics: reference ID, type of identification, title, authors, year, journal, type of publication, study design, participant description, country, treatment setting, clinical field, research question, and main conclusions. The first author held the main responsibility for verifying the accuracy of the data (Multimedia Appendix 2 [11,20-92]). If the abstract and title were insufficient for assessment, the full text was screened. Multiple authors could provide an assessment of the same paper, and instances of disagreement were resolved through discussion. In the second stage, full-text papers were evenly assigned to 2 authors. Instances of disagreement were resolved through discussion and sometimes by bringing in a third reader. The ideas that emerged during the process were discussed among the authors in regular meetings set up by the main author.

Stage 5: Collating, Summarizing, and Reporting the Results

The results reported in the included studies were compiled and read multiple times. In the Microsoft Excel spreadsheet, papers were categorized according to the stakeholder group studied: children and adolescents, parents, HCPs, or expert viewpoints. In total, 2 students categorized the viewpoints into three groups: (1) experts, such as HCPs, IT experts, or researchers; (2) policy makers; and (3) public opinion. In a meeting, 2 authors were assigned to each stakeholder group through discussion, where the first of the following authors listed was mainly responsible: children and adolescents were assigned to JH and BH, parents were assigned to MH and SH, HCPs were assigned to CB and IS, and viewpoints were assigned to JH and MH (as first and senior author, respectively). The results from the included studies were then independently analyzed and jointly drafted in a shared Google Docs. For organization of the results, key themes were adapted from a previous scoping review of the literature on PAEHRs in mental health [93]. These were refined by the main author using thematic analysis [94]. During this process, the material was gathered according to themes, and themes were reviewed and defined. This synthetization of results was conducted primarily by the main author but was discussed in research team meetings.

Stage 6: Consultation

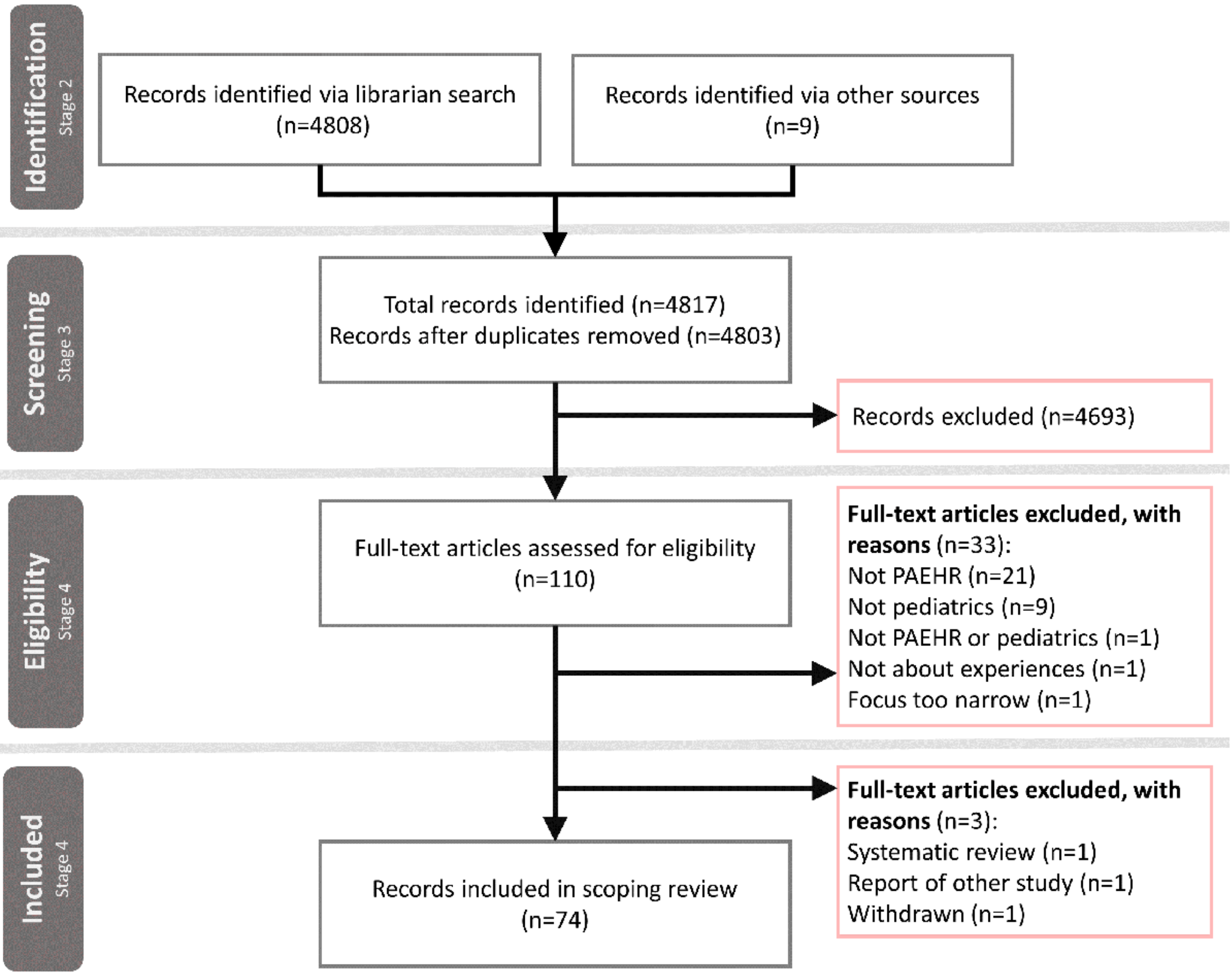
To gain further insights on the topic, the results were shared with stakeholder representatives, including a pediatric oncologist, members of a young patient council at a public hospital in Sweden, and the Ombudsman for Children in Sweden. The representatives were provided with material via email and invited to choose to provide their thoughts in text via email or verbally during a web-based meeting.

Results

Study Selection

Figure 1 shows the study selection process in a PRISMA diagram [95]. In total, 4817 records were identified, of which 4808 (99.81%) were identified via a database search and 9 (0.19%) were identified via other sources. After removing duplicates, 99.71% (4803/4817) of the records remained for screening of abstracts, titles, and keywords. In this process, 97.71% (4693/4803) of the records were excluded, resulting in 110 full-text articles to be assessed for eligibility. As a result of this, 1.6% (77/4817) of the total records identified met the inclusion criteria. During the analysis, 0.06% (3/4817) of the records were excluded, leaving 1.54% (74/4817) of articles included in the review.

Figure 1. PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flow diagram adapted from Moher et al [22]. PAEHR: patient-accessible electronic health record.



Basic Characteristics of the Body of Evidence

The included studies were mainly viewpoint papers or used quantitative methods (Table 1), and 92% (68/74) were based in the United States (Figure 2). The number of articles published in the area of PAEHRs for parents, children, and adolescents was fairly stable over time (Figure 3), ranging from 3% (2/74) of the articles in 2007 to 16% (12/74) in 2021. An increase can be observed for 2018 and 2021, and none of the articles during these years belonged to a special issue.

Table 1. Basic characteristics of the included studies (N=74).[a]

Parameter	Total, n (%)
Study design	
Viewpoint or comment	27 (36.5)
Quantitative	27 (36.5)
Qualitative	13 (17.6)
Mixed methods	7 (9.5)
Publication year	
2007-2009	7 (9.5)
2010-2012	7 (9.5)
2013-2015	13 (17.6)
2016-2018	23 (31.1)
2019-2021	24 (32.4)
Country	
Australia	3 (4.1)
Canada	1 (1.4)
New Zealand	1 (1.4)
United Kingdom	1 (1.4)
United States	68 (91.9)
Study participants[b]	
Children and adolescents	6454 (5.5)
Parents	110,184 (94.1)
Health care professionals	496 (0.4)
N/A[c] (no participants or not specified; studies)	34 (45.9)
Treatment setting	
Pediatric	34 (45.9)
Adolescent	15 (20.3)
Adult	2 (2.7)
Inpatient	15 (20.3)
Outpatient	20 (27)
Academic	1 (1.4)
N/A	7 (9.5)
Clinical field	
Chronic illnesses (cystic fibrosis, juvenile idiopathic arthritis, or diabetes mellitus)	6 (8.1)
Psychiatry	4 (5.4)
Intensive care	4 (5.4)
Gastroenterology	2 (2.7)
Hematology	2 (2.7)
Obstetrics and gynecology	2 (2.7)
Neonatal care	2 (2.7)
Cancer	1 (1.4)
Cardiology	1 (1.4)
Pulmonology	1 (1.4)

Parameter	Total, n (%)
Emergency	1 (1.4)
Hepatology	1 (1.4)
Subspeciality	1 (1.4)
Radiology	1 (1.4)
N/A	7 (9.5)

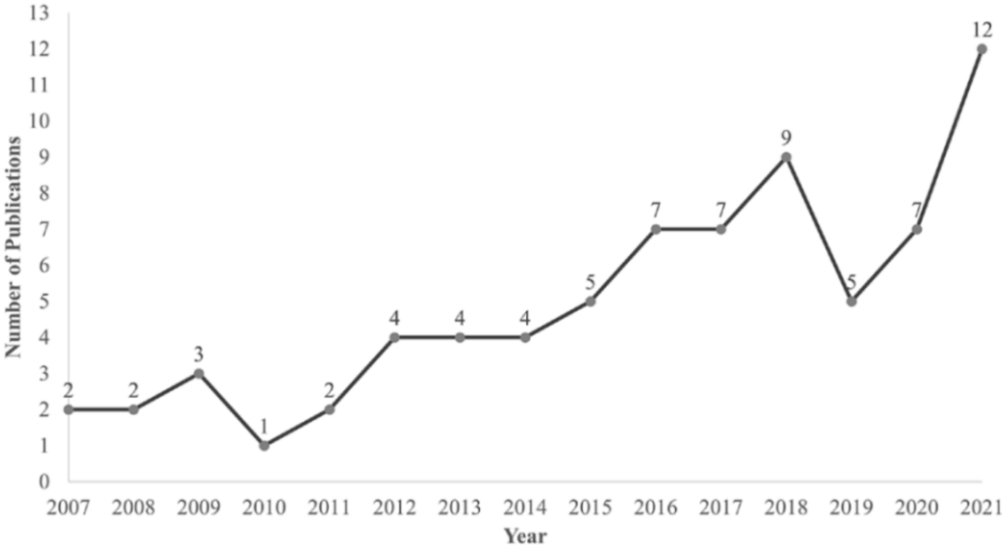
[a] Individual papers can be assigned to various subparameters at the same time, which means that percentage totals of >100% can be achieved.

[b] The number of study participants was accumulated based on empirical and observational studies that included a reported number of study participants.

[c] N/A: not applicable.

Figure 2. Included publications by country and studied stakeholder group. HCP: health care professional; UK: United Kingdom; USA: United States of America.

Figure 3. Number of publications by year.



Search Results

The results were divided into four groups of stakeholders: (1) children and adolescents, (2) parents, (3) HCPs, and (4) other stakeholders. For children, adolescents, and parents, the identified categories were adoption and use, positive views and experiences, and concerns and negative experiences. For HCPs, the identified categories were positive views and experiences and concerns and negative experiences. For expert analysis or viewpoints, the identified categories were positive views and experiences and concerns.

Children and Adolescents

Overview

Views, use, or experiences of PAEHRs among children and adolescents comprised a relatively small part (16/74, 22%) of the included studies. Of these 16 studies, 14 (88%) were conducted in the United States, 1 (6%) was conducted in Australia [20], and 1 (6%) was conducted in Canada [21]. Most of these studies were observational (6/16, 38%), followed by surveys (4/16, 25%), qualitative studies (focus groups or interviews; 3/16, 19%), and mixed methods studies (2/16, 12%). Only 1 opinion paper was included, authored by a male patient aged 15 years [22]. Survey studies ranged from 20 [23] to 1006 [21] participants. Qualitative studies used focus group interviews (2/16, 12% of the studies) [20,24] and individual interviews (1/16, 6% of the studies) [25]. The most frequent care settings were pediatric inpatient care, primary care, psychiatry, and nonclinical care. In total, 12% (2/16) of the studies focused on the general population [20,21]. Observational studies focused on adoption and use over time, demographic data, and frequently used functions of patient portals [26-31], whereas survey studies explored satisfaction with reading the records [23,32,33], literacy [23,32], intervention effects [33], attitudes toward web-based patient portals [21,34], and barriers to adoption [34]. The studies included adolescents aged between 12 and 20 years, and 12% (2/16) of the studies included patients aged ≥18 years [21,32]. A few studies (2/16, 12%) included adolescents and proxy users and did not distinguish adolescent patients from proxy users in their analyses [26,31].

Adoption and Use

A number of studies (4/16, 25%) reported low adoption and use of patient portals among adolescents compared with other age groups [27-29,31]. In a US study, 11% of patients aged 10 to 17 years had activated an account at a patient portal implemented 3 years before [27]. Similarly, a study that allowed for surrogate access and individual accounts for patients aged >13 years with parental consent found that adolescent patients composed 16.5% of all log-ins, although use increased during late adolescence [29]. A study based in Canada identified an age-related difference where younger adolescents (aged 12-15 years) were more open than older adolescents (aged 16-19 years) to sharing their notes with parents [21]; however, a US study with a smaller sample size observed a similar tendency but no significant difference [33]. For adolescent patients with cancer, the perceived value of record access decreased during recovery [35]. Knowledge of PAEHRs was reported as low not only among adolescents without access to a patient portal in US studies [21,24,34] but also in a focus group study based in Australia where adolescents had access to their EHR from the age of 14 years [20]. The studies were inconclusive on gender differences in adolescents' PAEHR adoption, finding either no differences [30,33] or a greater inclination among female patients [27]. In 6% (1/16) of the studies, male patients aged between 12 and 17 years had the lowest percentage of viewing their results in the patient portal (<1%) [28]. A study of 96 urban, low-income African American late adolescents in outpatient care found that male patients were more likely than female patients (P=.001) to consider allowing proxy access [33]. Regarding mode of access, adolescents in 12% (2/16) of the studies reported a preference for smartphones or tablet devices over computers [34,35].

Positive Views and Experiences

Studies that explored views on PAEHRs among adolescents who had not previously accessed their records identified a strong interest in access [20,21,24,33,34]. For example, among 1006 adolescents, 84% supported the idea that adolescents should be able to read their records on the web [21]. Adolescents wished to receive information about EHRs from HCPs according to their future needs [20,24]. Notably, an intervention study in which adolescents in primary care were informed about a patient portal observed an increase in portal account activation but not in use [33].

Positive expectations were confirmed by adolescents reading their records, with high satisfaction reported by studies in gastroenterology (9.2/10) [32], psychiatry (8.8/10) [23], and primary care (79%) [33]. In the study by Hong et al [35], adolescents with cancer and blood disorders read their records to ensure accuracy and check for updates. For these adolescents, reading their records led to reduced anxiety, enhanced knowledge about their illness, an ability to ask informed questions, and more reflection on their health. If needed, they consulted the internet or asked their parents. A US study conducted in a psychiatric ward found that having record access led patients' trust in their health provider to either increase or remain the same [23]. In total, 12% (2/16) of the studies observed adequate literacy, with almost no exceptions among patients in psychiatry [23,32]. Both adolescents with and without experience of having access to their records foresaw empowerment [22,24,25,35]; a male patient aged 17 years stated in an opinion paper [22] that access "could help my generation learn about our health care system" and "encourage [adolescents] to take more responsibility for our health." Patients with cancer anticipated that PAEHRs could support the transition from pediatric to adult care [35]. A high school senior in hematology who had used a patient portal suggested that the records could be jointly managed by themselves and their parents during the transition to adult care [25].

Better recall was an anticipated benefit among adolescents who did not access their records [20,22]. Furthermore, adolescents who did not have access to their records foresaw the utility of checking test results [21,24,34], messaging [20,24], viewing medications [20,21,34], reading visit notes [20], reviewing appointments [21,24], and viewing allergies [24]. In primary care contexts, adolescents valued being able to ask questions via email rather than in person, particularly concerning sensitive information [24]. Similarly, the most accessed information in observational studies was commonly test results [27,30,35], messaging [27,31,35], appointments [27,30,35], and medications [30,35]. Reminders were considered useful for planning around daily life [25,33,35], and a frequently asked questions section was suggested for ease of use [24].

Adolescents with cancer or a blood disorder who had accessed their records reported no concerns about what their parents would see in the EHR [35]. In an institute providing primary and mental health care that used a patient portal where a minor's consent was required when aged >10 years to release information to parents, HCPs had received no complaints about confidentiality from adolescents since the implementation [27]. In an Australian focus group study, a participant noted that, in spite of valuing privacy, timely access to medical data in a critical situation was more meaningful [20].

Concerns and Negative Experiences

Although relatively few, the leading concern was health literacy among adolescents. Adolescents without access to their EHRs expressed worry about not being able to understand and appropriately interpret the information in the EHRs [21,24]. Among patients in psychiatry who read their records, half reported not understanding the discharge criteria [23]. In studies where adolescent patients had the option to suggest note changes, edits concerned personal history and anthropometrics [23] as well as allergies and medication reconciliation [32].

Concerns about internet security and confidentiality whereby parents might access their EHR were expressed by adolescents with no access to their records and who were patients in an outpatient or nonclinical setting [21,24]. A teenager in another study suggested that the relationship with the parent may affect the teenager's feelings toward parental access, and in case of shared access, a private email option would be useful [25]. Adolescents without EHR access reported feeling uncomfortable with sharing their health information on social media [21].

Parents

Overview

Parents' and guardians' experiences with web-based access to health records comprised more than a third of the studies (33/74, 45%). Of these 33 studies, 31 (94%) were conducted in the United States, 1 (3%) was conducted in Australia [36], and 1 (3%) was conducted in the United Kingdom [37]. The most common studies were surveys (10/33, 30%), followed by qualitative (9/33, 27%), observational (9/33, 27%), and mixed methods (5/33, 15%) studies. Among these were an opinion piece coauthored by a parent [25] and a usability test where 16 parents evaluated the usability of a patient portal prototype [38]. The most common settings were pediatric inpatient care, outpatient care, in-hospital care, primary care, congenital cardiac surgery, and hematology. The observational studies focused on adoption and use over time, demographic data, and frequently used functions of patient portals. The qualitative studies included both individual interviews (5/33, 15% of the studies, whereof 1/5, 20% also included observations) and focus group interviews (3/33, 9% of the studies). The survey studies ranged from 25 [39] to 3672 [40] participants. A total of 12% (4/33) of the studies had <100 participants, and only 6% (2/33) of the studies had >500 participants. Of the survey studies, 12% (4/33) explored parents' thoughts about using a web-based patient portal [41-43] or their teenagers using such a portal [44] in the future. Of the remaining 8 survey studies, 3 (38%) focused specifically on errors in the record and patient safety issues [26,40,45]. In total, 6% (2/33) of the studies did not distinguish between parents and patients in their analyses [40,45]**.**

Adoption and Use

The studies reported high rates of patient portal adoption and use among parents during the first years of the child's life [29,30]. In both Australia and the United States, studies identified the highest rates of patient portal activation for the youngest children of both sexes aged 0 to 11 [28] and 0 to 14 years [36]. In studies that required the assent of older adolescents for parental access, parents' use of patient portals decreased. A study of a patient portal that required such assent received no applications for unrestricted access, and 80.4% of parents or guardians who enrolled had children aged <10 years [27]. In a longitudinal study where there were no restrictions, 93.62%

(16,036/17,128) of all pediatric patients during the study period had a surrogate (parents or legal guardians), and surrogate users accounted for 83.2% of all log-ins for adolescent patients [29]. There was higher use among parents of children with chronic diseases [46]. Another study observed a 100% adoption rate among parents as proxy users for children aged 0 to 11 years, whereas merely 5.9% of parents of adolescents enrolled [30].

In an inpatient setting, a study [47] found that 27.89% (530/1900) of families created a patient portal account, 47.8% (238/498) used the portal within 3 months of registration, and 15.9% (79/498) continued using the portal 3 to 6 months after creating the account. A US study identified disparities in social demographics; parents who identified as Hispanic, Asian, or "other races" than White were less likely to use a patient portal, which was hypothesized to be related to language barriers and device accessibility [48]. The same and another study identified that privately insured parents were more likely to enroll in portal activation than those with public insurance [46,48]. In a study in which 12 children died during the study period, most families continued accessing their children's records after their death [49]. A study of parents of children with attention-deficit/hyperactivity disorder found that, although half of the participants used their home computer to read the records, one-third accessed the portal on their smartphone and that barriers to use included lack of awareness, lack of internet access, lack of time, and password problems [50]. Schneider et al [37] identified four different use styles families at a children's hospital in the United Kingdom applied to access the children's records: controlling (approach-oriented and highly motivated to use PAEHRs), collaborating (approach-oriented and motivated to use PAEHRs), co-operating (avoidance-oriented and less motivated to use PAEHRs), and avoiding (very avoidance-oriented and not motivated to use PAEHRs).

Positive Views and Experiences

Several studies (4/33, 12%) focused on parents' expectations or thoughts about PAEHR use before actually having experienced access to their child's EHR [24,42,51,52]. In a 2013 US study, parents were approached in the waiting room and given a demonstration of the patient portal. A total of 72% (46/64) of the participants had not heard of the patient portal before, and only 28% (5/18) of those who had heard of the portal had used it. Nearly 70% (44/64) of the parents intended to use the patient portal after the demonstration [42]. Expectations were mostly positive and confirmed by studies with parents who had experience of record access, yet concerns were also discussed, which will be presented in the following section.

Better recall or reinforcement of information was reported as a benefit in many studies (7/33, 21%) [24,38,45,51-54], as was improved parental knowledge and understanding of their child's health [39,51,53,55-59,96] and a sense of control [39]. In addition to access to information, parents reported enhanced communication and partnership with providers [11,39,45,51,53,55-58]. In a study on parents of hospitalized children, the addition to the PAEHR of pictures of staff taking care of the child was highly appreciated [58]. Another reported benefit was not having to bother clinicians [56,57,96]. As anticipated by parents of hospitalized children [51], having access to the child's record also improved parental empowerment and the parents' ability to advocate for their child [11,43,53,55,56]. Furthermore, parents of children with cancer or chronic illnesses described reduced anxiety as a positive result of having access to their children's records [11,96]. The benefit of error detection was both reported by parents who had experiences of accessing their child's records [35,55,56,58] and anticipated by those who did not [51].

Records were used to prepare for discussions with clinicians [35,39,56], formulate questions, and ask for explanations [35,56]. Another study observed that parents used the portal to ask questions about their children's minor illnesses and request medication refills [27]. In studies in which parents were asked for suggestions for portal improvement, they often cited more information, such as a portal use tutorial [56], more educational links and resources [57,58], medical explanations or interpretations [38], and clarification of medical jargon [38]. However, in a survey study of 25 parents with real-time access to their children's EHR, none considered notes more confusing than helpful [39].

Studies varied in the available portal features and details of reporting use. In total, 6% (2/33) of the studies provided a similar broad functionality, consequently seeing a similar use where one study [30] found high use of appointment reviews (85%), messaging (84%), test results (79%), and immunizations (79%) and the other [46] found parents to frequently access immunizations (80%), messaging (72%), appointment reviews (55%), and test results (50%).

Parents of children who were seriously ill consistently reported positive experiences, for example, parents who had immediate access to laboratory test results in an inpatient portal during a child's hospitalization [55] and parents of children diagnosed with cystic fibrosis, juvenile idiopathic arthritis, or diabetes mellitus [11]. Chung et al [43] reported that 92% (78/85) of parents of hospitalized neonatal children wished to receive information even if it was "bad news." A study among parents of patients in pediatric radiology found that, although only 12.1% (18/104) accessed a web-based portal to check their children's test results, 65% prioritized minimal waiting time as the most important aspect for receiving results [60].

Some studies explored parents' views on their teenagers accessing their own records, and parents saw it as a way for the teenagers to take better control of their own health care [24,96]. When parents of adolescents in juvenile detention were asked for their opinion on giving their teenagers access to their health records, 70% were positive, and 100% felt that the adolescent should be able to share this information with their parents through the web-based system [61]. Parents also felt that the PAEHR would be useful when transitioning to adult care or another care provider [35,51].

Concerns and Negative Experiences

Before having access to the record, parents worried about information being released without face-to-face communication [51,53]. When it came to adolescents having access to their own records, parents had privacy concerns that the portal might be hacked [61], that the teenager would be pressured to share information [61], or that billing of confidential services would cause privacy breaches [24]. Some requested that parents be required to consent to teenagers having access to portals [24,61] and were worried that teenagers would make appointments without parents knowing and wanted to be informed about email conversations [24]. Moreover, parents worried that adolescents might not reveal sensitive information if they knew it would be visible to their parents [51]. In a US survey study of 93 parents where half were parents of adolescents, 68% were negative about their children receiving information from their HCP through a secure web portal [44]. In a study in which parents of children in an intensive care unit were provided with real-time EHR access on a large monitor, parents expressed concern about visibility to bypassers [56]. Issues around parents' loss of access to the record as the child enters adolescence were highlighted by Carlson et al [25], suggesting that record access needs to be an integrated part of the transition from childhood through adolescence and into adult health care. In the study by Hong et al [35], it was found that parents of teenagers with cancer would act as intermediaries in communication with HCPs as teenagers preferred to discuss their health with their parents rather than with clinicians. Thus, proxy access was considered essential. Parents in this study also expressed concerns about negative results being immediately available to teenagers, worrying that they might cause anxiety [35].

Some felt that teenagers may not understand all medical information, including test results [24], and that they might use the portal inappropriately and would need education [24,25]. Medical jargon was reported as an expected challenge in several studies (4/33, 12%) [38,43,51,96] as well as not being able to interpret complex results without context or explanation [56]. Parents of teenagers with cancer reported searching on the web to help make sense of the medical record and seeking additional information not readily available on MyChart [35]. Among 270 parents in a pediatric outpatient setting, 52.5% expected to read the medical records if they had access to them, with one-third indicating that they "sometimes" needed help reading health materials [41]. In another US study, 5% of surveyed parents of children with cancer reported understanding the notes to be "somewhat" or "very difficult" [59]. However, a study found that, among patients and families finding a serious mistake in visit notes, only approximately half reported the mistake, barriers including lack of knowledge of how to report but also fear or retribution [40].

Among concerns about PAEHRs, increased confusion, distress, or anxiety were anticipated by parents with no access [53]. Both parents with and without experience of PAEHRs worried about record access impairing the parents' relationship with the provider [11,53] and, in turn, negatively affecting collaboration [53]. Another concern stemmed from empathy with HCPs, worrying that parental record access could increase the workload and lead to complications [51,53] or restrict communication between HCPs through the record [51].

HCP Stakeholders

Overview

Comparatively fewer studies (11/74, 15%) explored HCPs' experiences of or opinions on web-based access to EHRs for children, adolescents, and parents. Of these 11 studies, 8 (73%) were conducted in the United States, 2 (18%) were conducted in Australia [20,62], and 1 (9%) was conducted in the United Kingdom [37]. Most of these studies (6/11, 55%) were qualitative (focus groups or interviews), although the sample sizes were small; 18% (2/11) of the studies had a sample size of 1 [23,25]. In total, 9% (1/11) of the studies used a web-based survey [63], and 18% (2/11) used paper-based surveys [43,52]. Many studies (6/11, 55%) recruited representatives from a wide variety of clinicians, including, for example, specialist physicians, general practitioners, hospitalists, nurse practitioners, nurses, mental health clinicians, physician assistants, dietitians, physiotherapists, and pharmacists [20,37,43,53,62,63]. Survey studies ranged from 1 [23] to 212 [63] participants. Notably, only 18% (2/11) of the studies exclusively solicited the views of pediatric health professionals [62,64]. Several studies explored HCPs' broad experiences with sharing PAEHRs with patients and parents [37,52,62]; a few focused on HCPs' anticipation of the practice among children or adolescents and parents [25,43,53]. In total, 18% (2/11) of survey studies exclusively focused on providers' perspectives on adolescent confidentiality with PAEHRs [63,64]. Only 12% (1/8) of the US studies reported on both accessibility and age of access: of 212 clinicians, 87.6% reported that their institution offered PAEHRs to both the adolescent and their parent or guardian, and most (69.1%) reported a minimum age requirement, with most (42.2%) citing between 12 and 14 years [63].

Positive Views and Experiences

Studies that explored HCPs' experiences with PAEHRs among children or adolescent patients and parents reported positive experiences. For example, among 96 providers with experience sharing access at a children's hospital, Kelly et al [52] found that 92% wanted patients and parents to continue to use the portal. They reported that patients and parents asked questions about the information they read, including laboratory test results (45%), medications (13%), and errors or mistakes in their care (3%). Exploring the views of HCPs in pediatric settings, Janssen et al [62] found that staff appreciated enhanced communication with patients, especially regarding coordinating appointments with parents and the potential for families or patients to ask questions. A study soliciting the views of 1 provider working in an adolescent inpatient psychiatric setting reported that clinical note sharing helped inpatient counseling sessions and compliance [23]. A study including 25 physicians identified experiences of increased transparency, improved documentation, reassurance or validation of concerns, and enhanced care plan clarity [39].

Among the anticipated benefits of sharing PAEHRs with child or adolescent patients and parents among HCPs with no experience of the practice, Kelly et al [53] reported that clinicians (including 8 nurses, 5 residents, and 7 hospitalists) predicted reinforced information, improved parental knowledge and empowerment, enhanced parent communication and partnership with providers, and increased provider accountability and documentation quality. Among 133 surveyed medical professionals, Chung et al [43] reported that 63.2% (84/133) believed that parental access may help identify incorrect information, and 61.7% (82/133) believed that access may reassure parents of the care provided to their child. In a qualitative study based in Australia by Beaton et al [20], school-based clinicians anticipated that adolescent patients with multiple providers would benefit from reduced duplication of investigations, ineffective treatment strategies, and more timely access to information.

Concerns and Negative Experiences

In several studies (4/11, 36%), patient confidentiality breaches and managing private patient information among children and adolescents was the leading concern [20,25,63,64]; as 1 surveyed clinician noted, "Privacy is just the biggest thing" [20]. In 18% (2/11) of the studies, HCPs reported that, despite sharing PAEHRs with other patients, they had precluded sharing information with adolescents because of privacy concerns, such as that savvy parents would be able to access it [20,25]; attesting to this, lack of clinician familiarity with PAEHR utility and technical implementation among minors was another expressed concern in both studies. Among clinicians with experience of PAEHRs, in a US study of 212 clinicians, nearly 4 in 10 (39.6%) were not at all confident that their EHR maintained privacy for minors, with 81.7% expressing concerns about maintaining confidentiality [63]. In another US study of 26 pediatric health care providers with experience of sharing PAEHRs with adolescents, Stablein et al [64] reported that confidentiality concerns affected documentation practices, such as worries that all HCPs involved in the child's care will not be aware of what information in the record is private from parents versus what the parent needs to know, in addition to the fact that the record has a multifold purpose (eg, billing and communication with families). As a result, providers reported selectively omitting or concealing information and using codes on the EHR designed to alert other providers to confidential information.

Kelly et al [53] reported that HCPs with no experience of the practice (including 8 nurses, 5 residents, and 7 hospitalists) foresaw increased provider workload, heightened parental confusion, distress or anxiety, impaired parental relationship with providers, and compromised note quality and purpose. In a US study, 34% (17/50) of attending and intern physicians were concerned that parents would be confused by reading their child's notes [39]. Among 133 surveyed medical professionals, Chung et al [43] reported that 114 (85.7%) believed that parental access may make medical professionals apprehensive about charting certain information, and 75 (56.4%) believed that parental access may increase the time spent updating parents, with approximately half (64/133, 48.1%) believing that parental access may increase the probability of a lawsuit. A study of inpatient pediatric physicians with experience of access found that 11% reported increased workload and 4% reported not being satisfied with portal use by patients or families [52].

Other Stakeholders

Overview

The viewpoints of other stakeholders on pediatric PAEHRs constituted most of the included studies (30/74, 41%). These studies comprised three types of stakeholders: (1) experts (27/30, 90%) such as HCPs, IT experts, or researchers; (2) policy makers (4/30, 13%); and (3) the public (1/30, 3%). Of these 30 studies, 28 (93%) were conducted in the United States, 1 (3%) was conducted in Canada [21], and 1 (3%) was conducted in New Zealand [65]. The aim of many studies (15/30, 50%) was focused on ethical issues related to adolescent PAEHRs, and a few (2/30, 7%) described the development of a portal solution [66,67].

Positive Views and Experiences

Viewpoints focused mainly on concerns (which we describe in the following section) but included a number of positive views of PAEHRs for a pediatric population. Among informants from 25 medical organizations, it was stated that adolescent patients with chronic diseases benefited the most from parents having access [68]. In fact, pediatricians claimed that parents of children with chronic diseases should be offered full access to their children's EHRs [69]. Jasik [70] advocated that PAEHRs could be useful in health education, in support of care transition for adolescents with chronic illnesses, and in risk behavior screening. Several viewpoints (3/30, 10%) argued that adding educational materials to the PAEHRs may facilitate literacy and comprehension for families [67,71,72]. Some noted unfulfilled potential for pediatric PAEHRs, for example, in the areas of patient data contribution [66], developmental screening [73], and research trial participation [74].

Green-Shook [75] anticipated that HCPs' control of their schedule may increase with PAEHRs because of communication with patients via messaging rather than telephone, an anticipation that was subsequently observed in a primary care setting [48]. Several papers (4/11, 36%) reported a need for availability on mobile devices to increase accessibility and practicality for users [67,69,70,74], and a medical director developing a mobile PAEHR app advocated for the integration of various functions in one app [67].

Concerns

Most viewpoint papers included concerns about adolescent confidentiality [72,73,76-84]. HCPs in gynecology and psychiatry reported that adolescents may be less willing to seek health care if they are uncertain about confidentiality [82,83], and 83% of respondents in a public opinion survey [85] deemed adolescents less likely to discuss sensitive issues with HCPs when parents had access to their EHRs. An American organization advocating for adolescents' health warned that adolescent aversion toward PAEHRs caused by confidentiality concerns and an uneven internet access could increase health disparities [86].

The studies described concerns in terms of portal functionality. Many insisted on an option for HCPs to label information as confidential [68,69,76,83,87] and enable adolescents to restrict parental access [80,86], some pointing to the variable definition of "sensitive" [68,81], which portal features contain such information [83], and division into "portions" of notes [84]. Psychiatric PAEHRs have been noted as unique in need of confidentiality, and Kendrick and Benson [83] listed portal functions that may hold information pertaining to sensitive topics in mental health, noting that sexual activity, gender identity, and substance abuse may be accessed in all portal areas. Bayer et al [80] posited that the release of sensitive information to the parent should require the adolescent's consent, whereas Bourgeois et al [88] urged HCPs to carefully review notes to prevent leakage of sensitive information. Not only concerning children, 7% (2/30) of the studies noted the need for protecting caregiver privacy [73,76]. In fact, medical professionals favored customizable controls of information display for both parents and adolescents [69], and several studies prompted considering family circumstances [65,89]. A group of pediatricians suggested that structured data content could improve efficiency and consistency [73].

Jasik [70] asserted a lack of stakeholder investment in PAEHR development for adolescents and that current portals are usually not designed to deal with privacy issues. Attesting to this, pediatricians noted that adolescent access to patient portals is hindered by time-consuming decision-making and lacking technology and manpower and that implementation variability is a result of absent guidelines and vague laws [68]. Anoshiravani et al [69] proposed that portal access for adolescents should be limited until the privacy functionality is more robust.

Set age limits for patient and parental access to mitigate confidentiality issues has raised concerns and been the topic of much debate. Taylor et al [89] suggested different content access for three subgroups in the pediatric population (aged <13 years, 13-18 years, and >18 years) based on information sensitivity. Various studies (2/11, 18%) held that default ages may enable long-term consistency [65,68], allow for automated notifications, and facilitate policy making [65]. Conversely, viewpoint papers cautioned that age-based loss of access could seriously affect families reliant on EHR access in the care for a child [68]. With regard to the transition from child to adult, Sittig and Singh [78] discussed the transfer of EHRs created when the patient was a child, whereas Bourgeois et al [88] reported that their institution provided access "prospectively," keeping confidential information suppressed also when the individual became an adult.

Several viewpoints (3/11, 27%) advocated for education on PAEHRs for various stakeholders [75,86,87], for example, that early HCP-initiated conversations with parents and adolescents may reduce parental concerns, increase acceptance [65], and set clear expectations [88]. Obstetrician-gynecologists have argued that adolescents should be informed if parents will have access to the EHRs [84], and Sherek and Gray [90] stated that, when possible, parents need to be informed of how to extend access to the child's record. In a short paper, the American Academy of Child and Adolescent Psychiatry [91] provided advice for parents on questions for their child's psychologist. In total, 13% (4/30) of the studies noted that insurance claims can lead to confidentiality issues [75,80,84,87], especially with uninformed use of the PAEHRs. The importance of guidance for staff has also been stated [69,88,92] as well as communication between staff and EHR vendors [68]. In pediatric psychiatry, Nielsen [81] advocated for training graduate students in penning PAEHRs. On the topic, a group of pediatric gastroenterologists recommended removing irrelevant details, not labeling emotions, and spell-checking [71].

Among other concerns, Gracy et al [73] described the divergent needs of pediatric portals compared with those of adult populations. Spooner [72] listed the critical areas for pediatric PAEHRs as immunizations, growth tracking, medication dosing, patient identification, norms for pediatric data, and privacy.

Visual Summary of Stakeholders' Expectations and Experiences

Figure 4 presents a visualization of the findings on adoption and use among adolescents and parents. Furthermore, Figure 5 provides a visualization of the findings based on expectations and experiences. Here, "expectations" is, as mentioned previously, a type of view in which the stakeholder has no previous experience of web-based record access.

Figure 4. Summary of results of adoption and use of patient-accessible electronic health records among parents and adolescents. FAQ: frequently asked questions.

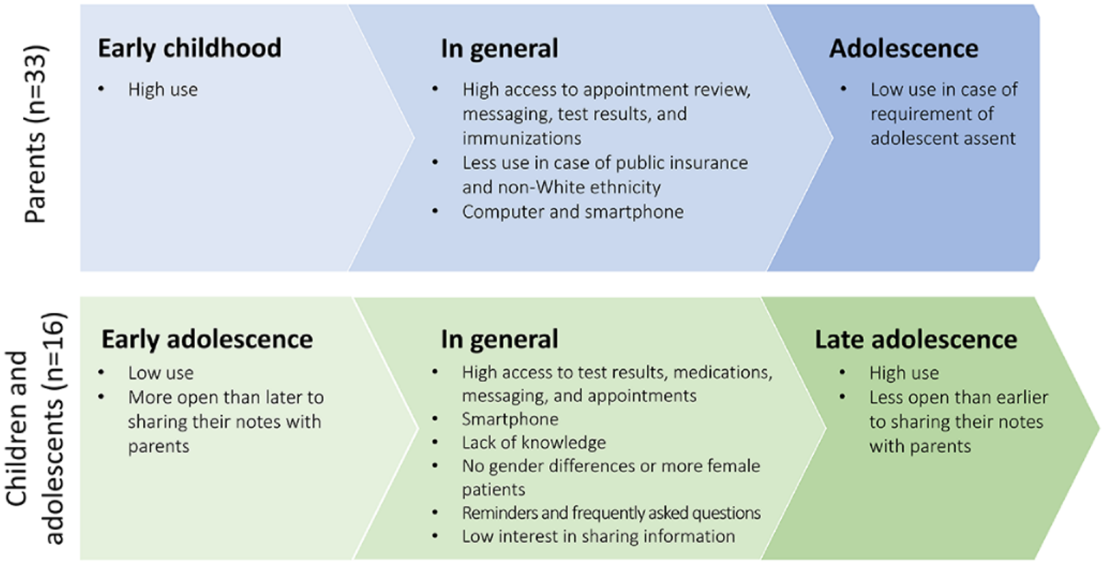
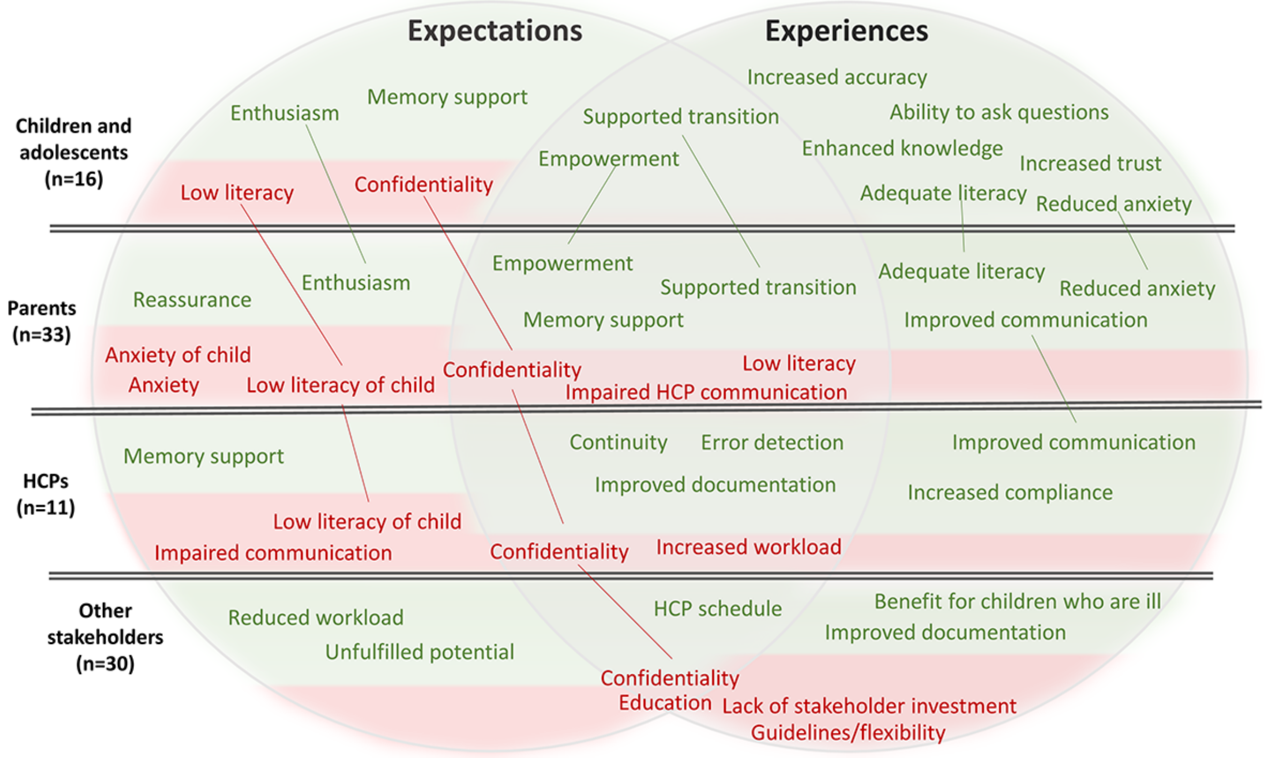


Figure 5. Summary of results of expectations and experiences of patient-accessible electronic health records among children/ and adolescents, parents, health care professionals (HCPs), and other stakeholders. Green text depicts positive views and experiences, and red text depicts negative views and experiences. Color in the various boxes illustrates the distribution of positive and negative views and experiences for the stakeholder group.



Discussion

Principal Findings

The results of the 74 studies included in this scoping review contribute to the understanding of factors associated with stakeholders' views, use, and experiences of children's, adolescents', and parents' web-based access to the EHRs of children and adolescents. The reviewed studies consistently observed positive views and experiences on the part of parents and particularly of adolescents, whereas HCPs and other stakeholders held many concerns. In this section, we will (1) compare stakeholders' views on and experiences with PAEHRs, (2) discuss some of the challenges that are unique to the PAEHRs of children and adolescents, (3) comment on the implications for design and implementation, and (4) suggest future research.

Limitations

Although it followed the scoping review methodology, the review was limited by not assessing the quality of the included studies. By only including studies written in English, we may have missed important papers written in other languages. Considering that 92% (68/74) of the included studies were based in the United States, we do not know whether an information bias affected the findings. Among the identified studies, some merged adolescents with young adults or parent proxies, which complicated the analysis of specific groups. Furthermore, the studies' definitions of adolescents varied, with the upper age limit ranging from 17 to 20 years. The studies did not always distinguish between positive and negative views or experiences. For example, the provision of education and guidance could be deemed as both a benefit and a concern. Furthermore, several expert viewpoints provided recommendations for the future based on concerns about PAEHRs, omitting to mention benefits. For the purpose of this study, we referred to the effects of PAEHRs that appeared beneficial to the patient as "benefits." Finally, conducting stakeholder consultations after completing the review prevented any integration of their results into the study. Future scoping reviews may wish to invite stakeholders to a more active participation earlier or to provide input throughout the process.

Expectations Versus Experiences Among Adolescents and HCPs

The findings suggest a similar pattern for adolescents to that previously observed in adult populations [9,10,93,97-99], where adolescents' positive experiences contrast with HCPs' concerns. For example, HCPs and parents imagined that adolescents would not understand the information in their notes and experience negative emotions as a result. However, adolescents reported high satisfaction and literacy even in the much-debated field of psychiatry. Another interesting aspect was that, although adolescents who had not previously accessed their EHR notes did have concerns about not understanding the notes and what parents may have access to, those with experience of accessing their records reported no such concerns.

A possible explanation for this might be a different perspective as most nonobservational studies exploring adolescents' experiences with PAEHRs (4/5, 80%) included patients with serious illness or in inpatient settings. It might be that children and teenagers with serious illnesses may have a better understanding of medical jargon. In addition, they may be familiar with being dependent on parental insight into their care and involving parents in their health care issues. Thus, the adolescent's desire for privacy is likely to depend on many factors, and there is still a need to provide confidentiality for those who require it, which was mentioned in many viewpoint papers.

The one existing review in the field [14] did not include expectations of PAEHRs; however, its findings in terms of experiences were aligned with our included evidence. For example, there was enthusiasm among adolescents and interest among parents in using patient portals, whereas medical literacy and confidentiality were the main concerns. Similarities are not surprising as, of the 11 included papers in the aforementioned review, only 3 were not included in this review (because they did not have a focus on pediatric PAEHRs). Except for not focusing on expectations, among the differences were that the previous review included use barriers and clinical outcomes and did not include the perspectives of HCPs and other stakeholders.

Interestingly, all but one of the parents' concerns about adolescents' confidentiality referred to external parties rather than the self as a parent as a threat to their adolescent's privacy. It is difficult to explain this result, but it might be related to the fact that parents have been found to value the importance of their involvement highly out of concerns about not being apprised of important information and uncertainty of the child's ability to manage their own care [100]. Instructing HCPs to engage parents and adolescents in a dialogue on confidentiality has been mentioned previously as a strategy to mitigate parents' worries, although current extensive pressure on HCPs may necessitate new approaches to such education.

Special Challenges for Pediatrics

A key challenge for PAEHRs is balancing confidentiality and information privacy for adolescent patients with the need for parental involvement in the adolescent's care. Several viewpoint papers focused on guidelines regarding when and how to grant access to parents and adolescents. The results are inconclusive and reflect the complexity of this issue. A health institute argued that allowing for manual changes to parental access can signal that the child has received some type of sensitive care [27]. Set age limits for automatic gain and loss of access could be beneficial, yet an extensive variety of potential circumstances do call for customizability according to the situation. A lack of investment and priority of portal development for adolescents and parents was indicated, which one could argue causes a waste of potential of PAEHRs and a loss for the health care system in the long term. One such function advocated by numerous viewpoints was the possibility of designating information as confidential. Still, efforts to hide sensitive information from parental view could be counteracted by parents evading the system to access their adolescents' accounts directly. If a parent perceives their adolescent incapable or unwilling to manage their own health care, they may consider it necessary and part of their parental responsibility to find a work-around. A recent UK article published outside the search period indicated that more than half of the messages to adolescents' accounts were accessed by guardians [101]. In addition to protecting the adolescent, a few papers stressed the importance of considering caregiver privacy in cases where parents disclose confidential information with regard to the child's care. Furthermore, modern family constellations vary, which may require the consideration of access provision based on the type of parental or legal guardianship. In a case study, health data coordinators at a US medical center described using different rules of access for a "natural or adoptive parent," legal guardian, or stepparent [90]. The same institution denied parents aged <18 years access to their child's EHR before becoming an adult, highlighting another potential issue. Differences between countries further complicate the issue of PAEHRs in pediatrics; for example, the definition of policy maker in the PAEHR context varies considerably by country, whereby HCPs in some countries are required to decide on policies themselves.

Consultation With Stakeholder Representatives in Sweden

We consulted on the findings with a pediatric oncologist, a young patient council at a public hospital in Sweden, and the Ombudsman for Children in Sweden. All reported their feedback via email. First, the pediatric oncologist reported not missing any aspect in the results. She reported that she considered the findings highly interesting and the biggest takeaway to be the positive effects on adolescents and parents of reading the PAEHRs and that security seemed to be the main cause of worry. Second, the young patient council reported to the first author, after discussion in a meeting, that the findings "looked very good" and dovetailed with their own experiences of having access to the EHRs. They reported that they had nothing to add to our findings. Third, the Ombudsman for Children in Sweden expressed positivity toward this overview as none has so far been done. He had questions about the findings, such as about results that confirmed his suspicions (eg, that male patients were more likely than female patients to consider allowing proxy access), as well as whether there was a complete lack of Swedish studies. He also asked for clarification of one case of unclear wording. An area that he saw as missing was the perceptions of shared access among adolescents and parents. As a result, we clarified some wordings and included the perspective of shared access in the Results and Discussion sections.

Implications of the Findings

Consistent findings can be summarized into four implications for PAEHR implementation: (1) adolescents and parents should be educated on PAEHR use and confidentiality (eg, information visibility for children, adolescents, and parents; possibility to restrict information; reasons for age limits; children's and adolescents' need for privacy; the moment when parents will lose access; and procedures for parents to stay involved in the child's care); (2) HCPs should communicate with EHR vendors and be educated on PAEHRs (eg, use; updates; privacy functionality; and information visibility for children, adolescents, and parents); (3) PAEHRs should be available on mobile devices, and functions need to be integrated; and (4) there should be options on a portal for HCPs and patients to label information as confidential.

Future Research

There is a lack of studies examining the effects of PAEHRs among children and adolescents. Although the Nordic countries are often considered to be at the forefront of PAEHR implementation [1] and access has been available longer at the national level in Sweden than in most other countries, no survey studies targeting a pediatric population in Sweden have been published to date. However, there is ongoing research within the NORDeHEALTH project [1] (with some of the authors' involvement) that aims to rectify this situation. One way is to explore approaches that have already been implemented and conduct comparative studies on the benefits and risks of access or exclusions among children and adolescents. Owing to the current scarcity, investigations with focus on literacy and confidentiality in adolescent outpatient or nonclinical populations are suggested. In addition, there is a need to explore the anticipation of parents and adolescents that shared access may support the transition to adulthood. Furthermore, there is little evidence on the efficiency of PAEHRs in the pediatric population, and work should be undertaken to better understand the effects on documentation time for HCPs and the potential cost-effectiveness of PAEHRs for families and adolescents in the long term. Finally, questions remain with respect to how PAEHRs affect the quality of documentation [102]. In this area, the approach of natural language processing has been increasingly used to quantitatively examine note changes, for example, according to ethnicity and disease chronicity [103].

Conclusions

This study consisted of a scoping review of 74 studies on PAEHRs for parents, children, and adolescents. Most studies (27/74, 36%) were comment papers as, despite the urgency of the matter, there is limited research, particularly regarding adolescents' experiences with web-based access to their records and outside the United States. Existing literature on pediatric PAEHRs indicates a pattern similar to that observed in adult populations, whereby adolescents' and parents' strong interest and positive experiences of accessing the records are juxtaposed with and obstructed by concerns among HCPs and other stakeholders, confidentiality being the key issue. Our findings could inform the design and implementation of future regulations regarding access to PAEHRs. Further examination of the experiences of adolescents, parents, and HCPs is warranted to improve usability and utility, inform universal principles reducing the current arbitrariness in the child's age for own and parental access to EHRs among providers worldwide, and ensure that portals are equipped to safely and appropriately manage a wide variety of patient circumstances.

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Authors' Contributions

JH wrote the manuscript, created the tables, and designed the figures. MH, CB, BH, and SH contributed to study design, results analysis, and writing of the Results section. MH was responsible for the conception of the study. All authors read, provided feedback, and approved the paper for submission.

Conflicts of Interest

CB is employed by OpenNotes, a research and advocacy unit that investigates and promotes patients' access to their clinical records.

Multimedia Appendix 1

PRISMA-ScR (Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for Scoping Reviews) checklist.
[PDF File (Adobe PDF File), 168 KB-Multimedia Appendix 1]

Multimedia Appendix 2

Summary of the included studies.
[PDF File (Adobe PDF File), 281 KB-Multimedia Appendix 2]

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Abbreviations

EHR: electronic health record
HCP: health care professional
PAEHR: patient-accessible electronic health record
PRISMA: Preferred Reporting Items for Systematic Reviews and Meta-Analyses
PRISMA-ScR: Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for Scoping Reviews

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Original Paper

The Generation of a Lung Cancer Health Factor Distribution Using Patient Graphs Constructed From Electronic Medical Records: Retrospective Study

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Abstract

Background: Electronic medical records (EMRs) of patients with lung cancer (LC) capture a variety of health factors. Understanding the distribution of these factors will help identify key factors for risk prediction in preventive screening for LC.

Objective: We aimed to generate an integrated biomedical graph from EMR data and Unified Medical Language System (UMLS) ontology for LC, and to generate an LC health factor distribution from a hospital EMR of approximately 1 million patients.

Methods: The data were collected from 2 sets of 1397 patients with and those without LC. A patient-centered health factor graph was plotted with 108,000 standardized data, and a graph database was generated to integrate the graphs of patient health factors and the UMLS ontology. With the patient graph, we calculated the connection delta ratio (CDR) for each of the health factors to measure the relative strength of the factor's relationship to LC.

Results: The patient graph had 93,000 relations between the 2794 patient nodes and 650 factor nodes. An LC graph with 187 related biomedical concepts and 188 horizontal biomedical relations was plotted and linked to the patient graph. Searching the integrated biomedical graph with any number or category of health factors resulted in graphical representations of relationships between patients and factors, while searches using any patient presented the patient's health factors from the EMR and the LC knowledge graph (KG) from the UMLS in the same graph. Sorting the health factors by CDR in descending order generated a distribution of health factors for LC. The top 70 CDR-ranked factors of disease, symptom, medical history, observation, and laboratory test categories were verified to be concordant with those found in the literature.

Conclusions: By collecting standardized data of thousands of patients with and those without LC from the EMR, it was possible to generate a hospital-wide patient-centered health factor graph for graph search and presentation. The patient graph could be integrated with the UMLS KG for LC and thus enable hospitals to bring continuously updated international standard biomedical KGs from the UMLS for clinical use in hospitals. CDR analysis of the graph of patients with LC generated a CDR-sorted distribution of health factors, in which the top CDR-ranked health factors were concordant with the literature. The resulting distribution of LC health factors can be used to help personalize risk evaluation and preventive screening recommendations.

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KEYWORDS

lung cancer; risk factor; patient graph; UMLS knowledge graph; Unified Medical Language System; connection delta ratio; EMR; electronic health record; EHR; electronic health record; cancer

Introduction

Early lung cancer (LC) detection is a key strategy to combat this deadly disease worldwide [1]. The National Lung Screening Trial in the United States and similar clinical trials around the world have shown an approximately 20% reduction in mortality from LC as a result of screening with low-dose computed tomography [2]. Based on these studies, LC screening medical guidelines as well as statistical risk prediction models including $PLCO_{M2012}$ have been implemented to recommend screening for smokers [3]. However, screening is not commonly recommended for nonsmokers even though they represent a significant percentage of patients with LC worldwide, 15%-20% among male patients and over 50% among female patients [4]. In addition, adoption of LC screening is still very low. For example, only approximately 5% of the at-risk population received their annual screening in the United States [5].

Risk-based or personalized screening approaches are being studied to overcome these challenges [6]. We believe that a deeper understanding of the spectrum of risk factors for LC and applying technologies such as machine learning and knowledge graphs (KGs) will generate more cost-effective screening solutions.

KGs have been widely applied in biomedical research. For interpreting proteomics data, a large-scale clinical KG has been plotted from biomedical data using the Neo4j tool [7]. Open-source graph databases and tools including Neo4j have made it easier to build and analyze KGs [8]. Studies have also demonstrated that construction of high-quality patient KGs from electronic medical records (EMRs) using rudimentary concept extraction is feasible and that the KGs can be used to predict diagnosis on the basis of symptoms [9]. Even though graphical representation of patient data holds the promise to illuminate insights in health care and to transform such insights gleaned from EMR data into actionable knowledge, the application of EMR-wide graphs for studying individual disease diagnosis journeys or treatment processes is still limited [10]. A graphical data model has been constructed, integrating clinical and molecular data of patients with non–small cell LC in the Cancer Genome Atlas LC data sets [11]. Another recent study of synthetic patients proposed a new graphical method to identify any particular disease's potential risk factor distribution from EMR (personal communication by A Chen, March 1, 2022).

The Unified Medical Language System (UMLS) ontology, freely available from the National Library of Medicine, is a KG consisting of millions of nodes and relationships [12]. It forms the foundation of interoperable biomedical information systems and services, including electronic health records. Connecting the UMLS KG to patient graphs may enable semantic search of patient data and support clinical decision-making [13].

This study aimed to construct a patient health factor graph for LC from a hospital EMR and integrate it with the UMLS KG for graph search and risk factor analysis. Through graph search, the study also aimed to generate a distribution of LC health factors, which was expected to help implement personalized LC risk evaluation for preventive screening.

Methods

EMR Health Factor Data Collection

We deidentified the patient records from January 2018 to June 2021 and saved them on a secured data server controlled by the hospital's informatics department. The data set had approximately 1 million patients and 7 million encounters including both outpatients and inpatients, in which patient names, dates of birth, contacts, and addresses were removed. The original identifiers of patients and encounters were replaced by irrelevant random numbers. Before using the data, our research team members were trained in the hospital's patient data security and privacy policy.

Because the EMR data had no usable codes associated with the diagnoses, synonyms of LC in Chinese were used to search for patients with LC. A total of 1397 patients with LC aged ≥30 years were included in the target data set. The same number (n=1397) of patients without LC and aged ≥30 years were randomly selected as control (or background) patients for comparison purposes.

Deidentified records of outpatient and inpatient visits, diagnoses, laboratory tests, and procedures were imported into a custom data collection tool on the secured data server. The data tool automatically extracted laboratory test data and saved them in the database. Researchers manually selected data from text records and entered them into the database. Because the records were not coded, practical rules were developed to improve consistency in the data collection process. Synonyms were automatically converted to "local standard terms" and the resulting data were called "local standard data." For each patient, only data from before the final diagnosis of LC were collected for studying disease risk factors, and a patient diagnosis journey (PDJ) object was created in the data tool to contain 1 or multiple encounters leading to the final diagnosis. When exporting PDJ data to a CSV file for analysis, only the latest data for each health factor in PDJ were selected. The final raw data set contained near 50,000 data from patients with LC and over 58,000 data from background patients. There were over 3000 different health factors identified in these data.

Patient Graph Construction

To simplify the patient graph, continuous numerical data were converted to categorical data. For example, values of age were converted to categories (ranges), including 30-50, 50-70, and >70 years; the value of drinking was "true" if the patient consumed >1 drink per day; the value of smoking was "true" if the patient smoked >1 cigarette per day. Laboratory findings from the EMR were already recorded as categorical variables: normal or abnormal; true or false; positive or negative; high, medium, or low; and up, down, or normal. After value conversion, approximately 93,000 standard data for about 550 factors (ie, codes) that appeared in at least 10 patients with LC were selected and saved into a factor import CSV file. The format of the factor import file was as follows: virtual-id, category, code, term, value, unit, converted-value, and date. Patients with LC and background patients (N=2794) were both saved in a patient import file, one patient per line, with the following format: virtual-id, LC-label (1 for LC, 0 for background), and factor-count.

We used the Neo4j Desktop tool (version 4.4) available freely from Neo4j Inc, which is a graph database with a graphical user interface (Neo4j Browser) to query with Cypher language and view graphs. It provides an application programming interface through a Python driver. It can load data from CSV files to construct graphs. In our patient-centered graph model, each patient was represented by a “Patient” node (total of 2794 patient nodes), while health factor and value pairs were represented by 650 factor nodes. Because all values were categorical and some health factors had more than 1 piece of categorical data, the number of factor-value pair nodes increased from 550 to 650. The health factors were further subdivided into the following categories: Condition, Symptom, Observation, History, RiskFactor, Labtest, Procedure, Medication, and Treatment. The graph drew over 93,000 connections from patients to factors. Constraints were created on each label to ensure uniqueness. Patient nodes required virtual-id while all factor nodes required category, code, and converted-value as node key.

UMLS Disease Subgraph Construction

The UMLS 2020AB release was downloaded from the National Library of Medicine’s UMLS website and installed locally by following the provided instructions. The local UMLS ontology had 2.8 million concepts, 8.3 million terms, and 39.1 million relationships. For generating an LC UMLS subgraph, we directly used the concept file MRCONSO.RRF and relation file MRREL.RRF in rich release format to generate Neo4j graph import files. The LC codes were first expanded to a more complete set of LC codes using the UMLS hierarchy (Table 1). We then used the expanded concept unique identifiers to find all horizontal relations (approximately 1100) between these LC target concepts and other biomedical concepts from over 39 million relations in UMLS ontology. The relations discovered were filtered by a selected set of UMLS relationship attributes for biological or medical concepts (Textbox 1); these were categorized into either biological concept relationships (called “biorel”) or medical concept relationships (called “medrel”). To visualize this simple categorization of biomedical knowledge, we added RelCat nodes between TargetConcept nodes and related Concept nodes in the UMLS subgraph as shown in Figure 1. We then introduced a single AbstractPatient node to connect with all LC TargetConcept nodes. Connecting the patient nodes in EMR graph to the single AbstractPatient node resulted in an integrated biomedical graph that can present any patient’s health factors together with biomedical knowledge from UMLS ontology for LC.

Table 1. Expanded lung cancer concepts in the Unified Medical Language System (UMLS) hierarchy.

UMLS concept unique identifiers	Term	SNOMEDCT code
C0581834	Suspected lung cancer	162573006
C0242379	Malignant neoplasm of lung	363358000
C0149925	Small cell carcinoma of lung	254632001
C0007131	Non-Small Cell Lung Carcinoma	254637007
C0152013	Adenocarcinoma of lung (disorder)	254626006
C0149782	Squamous cell carcinoma of lung	254634000
C1306460	Primary malignant neoplasm of lung	93880001
C0153676	Secondary malignant neoplasm of lung	94391008

Textbox 1. List of Unified Medical Language System (UMLS) relationship attributes and categories.

Biological concept relationships:

- gene_associated_with_disease
- gene_involved_in_pathogenesis_of_disease
- gene_mapped_to_disease
- gene_product_malfunction_associated_with_disease
- gene_product_is_biomarker_of
- may_be_cytogenetic_abnormality_of_disease
- may_be_molecular_abnormality_of_disease

Medical concept relationships:

- may_treat
- regimen_has_accepted_use_for_disease
- has_associated_finding
- associated_finding_of
- associated_disease
- is_finding_of_disease
- related_to
- clinically_associated_with
- co-occurs_with
- may_be_associated_disease_of_disease
- may_be_finding_of_disease

Figure 1. Biomedical graph model for the integration of the electronic medical record patient graph with the Unified Medical Language System knowledge graph of lung cancer. Numbered relationship labels are listed in Table 2.

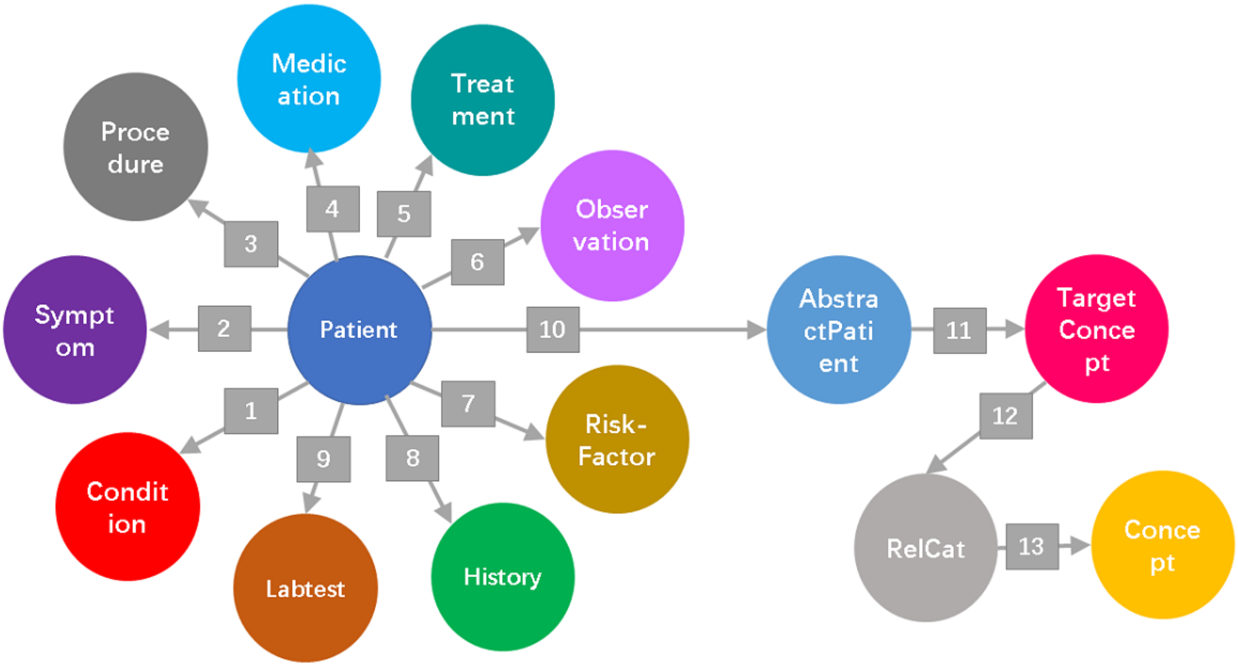


Table 2. Node and relationship labels in the integrated biomedical graph model (shown in Figure 1).

Number	From node label	Relationship labels	To node label
1	Patient	HAS_CONDITION	Condition
2	Patient	HAS_SYMPTOM	Symptom
3	Patient	HAS_PROCEDURE	Procedure
4	Patient	HAS_MEDICATION	Medication
5	Patient	HAS_TREATMENT	Treatment
6	Patient	HAS_OBSERVATION	Observation
7	Patient	HAS_RISKFACTOR	RiskFactor
8	Patient	HAS_HISTORY	History
9	Patient	HAS_LABTEST	Labtest
10	Patient	INSTANCE_OF	AbstractPatient
11	AbstractPatient	MAY_HAVE_TARGET	TargetConcept
12	TargetConcept	HAS_RELCAT	RelCat
13	RelCat	HAS_RELA	Concept

Patient Health Factor Distribution

We developed a Python script to automatically query the patient graph with each of the health factors. The number of connections from each factor to LC target patients (depicted as "TPC" in equation 1) and background patients (depicted as "BPC" in equation 1) in the search results were counted separately. For each factor, the delta of patient connection counts was calculated by subtracting the number of background patient connections from that of the target patient connections. Division of the delta by the total number of patient connections yielded the "connection delta ratio" (CDR), a relative measure of the strength of connections from a factor to the target patient. Sorting factors by CDR and plotting a graph of CDR versus the sorted factors yielded a distribution of LC health factors from high to low strength.

$$CDR = (TPC - BPC) / (TPC + BPC) \quad \textbf{(1)}$$

A CDR between 1 and 0 implied that the factor was more related to the target patient, 1 being most related. A CDR below 0 implied that the factor was more related to the background patient.

In this study, factors with a CDR of >0.5 and having connections with at least 10 patients with LC were selected for literature verification. The local standard terms were first translated to English and the corresponding UMLS concepts as well as standard codes from SNOMEDCT_US, LOINC, or RxNORM if possible. We then searched the research literature on Google, Google Scholar, PubMed for each health factor and reviewed the published studies to verify whether the health factors were confirmed risk factors, correlated with LC, were unrelated to LC, or had an unsure relationship with LC. If a factor's relationship with LC was inconclusive in existing research reports, the factor was tagged as "unsure." For example, to look up the factor "Hypocalcemia," search terms included "Lung cancer risk factor Hypocalcemia" and other variations if necessary.

Ethical Considerations

This retrospective study of EMR patient data has been approved by the institutional review board of Guilin Medical University Associated Hospital in China (QTLL202139).

Results

Integrated Graph Model of the EMR Patient Graph and the UMLS KG

To study the spectrum of health factors related to LC in the hospital EMR, we applied a new graph method that we recently developed using synthetic patient data. Figure 1 shows the graph model integrating the EMR patient graph and UMLS knowledge subgraph for LC. The patient graph is patient-centered with patient nodes connecting to different categories of health factor nodes. Table 2 lists the relationships between nodes, as generated in the graph database. The UMLS subgraph in this model is focused on the horizontal biomedical relationships between LC nodes and related concept nodes. Such an integration model enables the presentation of a patient's actual health factors together with the UMLS KG's related biomedical factors in the same graph.

Patient Health Factor Graph Based on EMR Data

From the hospital EMR, 1397 patients with LC were selected along with the same number of background patients without LC. After deidentified data of laboratory tests and procedures were integrated into the corresponding encounters, a total of 108,000 standard data for various categories of health factors were extracted from patient encounters. Although over 3000 different factors were collected, only approximately 550 factors shared by at least 10 patients with LC were used for building the patient health factor graph.

The patient health factor graph was constructed by importing patient properties for the patient nodes and factor properties for the corresponding health factor nodes. The resulting patient graph had 93,000 relations between the 2794 patient nodes and

650 factor-value pair nodes. Table 3 lists several examples of Cypher queries for searching patients with various factors. For example, clinicians can easily search for patients with LC with 1 or more co-occurring diseases (Figure 2), with 1 or more nonlaboratory factors (symptoms, medical histories, and observations; Figure 3), or laboratory tests (Figure 4). One can also easily search for any number of health factors shared by patients among patients with LC.

Table 3. Examples of graph search tasks and queries using Cypher language.

Number	Graph search task	Cypher query[a,b]
1	• Search for patients with LC with 1-6 co-occurring diseases and present the topology. • C-389764: Hypocalcemia • C-172569: Bacterial Infection • C-765209: Obstructive pneumonia • C-305976: Pneumothorax • C-352894: Leukopenia • C-654730: Pneumonia	match (p:Patient {label:'1'})-->(f {cat: 'dac'}) where f.code = 'C-389764' or f.code = 'C-172569' or f.code = 'C-765209' or f.code = 'C-305976' or f.code = 'C-352894' or f.code = 'C-654730' return p, f;
2	• Search for patients with LC with 1-5 nonlaboratory factors and present the topology • C-549780: Pain • C-289547: Bloodstained sputum • C-127089: Hoarseness • C-029761: Productive Cough • C-294680: Swollen Lymph Node in head and neck	match (p:Patient {label:'1'})-->(f) where (f.code = 'C-549780' and f.valcvt = 'true') or (f.code = 'C-289547' and f.valcvt='true') or (f.code = 'C-127089' and f.valcvt='true') or (f.code = 'C-029761' and f.valcvt='true') or (f.code = 'C-294680' and f.valcvt='true') return p, f;
3	• Search for patients with LC with 1-5 laboratory test values and present the topology. • C-659218: Hepatitis B virus • C-493765: Squamous cell carcinoma antigen • C-573086: Neuron-specific enolase measurement • C-120948: Gastrin-releasing peptide precursor increased • C-814793: Mycoplasma pneumoniae antibody	match (p:Patient {label:'1'})-->(f {cat: 'lab'}) where (f.code = 'C-659218' and f.valcvt = 'true') or (f.code = 'C-493765' and f.valcvt = 'up') or (f.code = 'C-573086' and f.valcvt = 'up') or (f.code = 'C-120948' and f.valcvt = 'abnormal') or (f.code = 'C-814793' and f.valcvt = 'abnormal') return p, f;
4	• Search for 1 patient, show the electronic medical record health factor graph and the Unified Medical Language System knowledge graph together	match (p:Patient {label:'1', vpid:'_8908085766'})-->(f) match (p)-->(ap:AbstractPatient)-->(tc:TargetConcept)-->(cr:RelCat)-->(c:Concept) return p, f, ap, tc, cr, c;

[a] Using Neo4j Cypher query language.

[b] Patient with LC: label=1; background patient: label=0. Factor property f.code: unique local code. Factor property f.valcvt: converted value.

Figure 2. Topology of an example patient graph searched with 6 disease factors. Search query 1 in Table 3 was used. Patient nodes are shown in blue and factor nodes are shown in red. Lines represent relationships between a patient and factors.

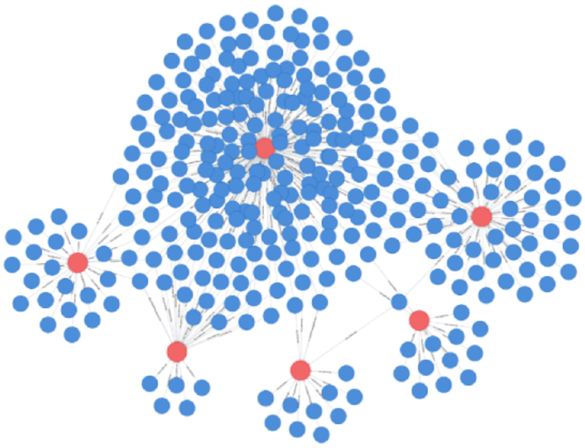


Figure 3. Topology of an example patient graph searched with 5 nonlaboratory factors. Search query 2 in Table 3 was used. Patient nodes are shown in blue and factor nodes are shown in pink. Lines represent relationships between a patient and factors.

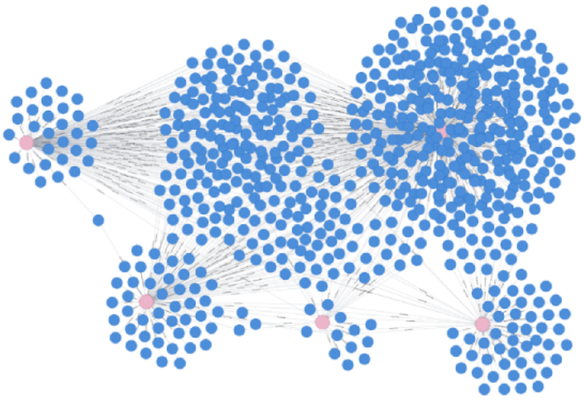
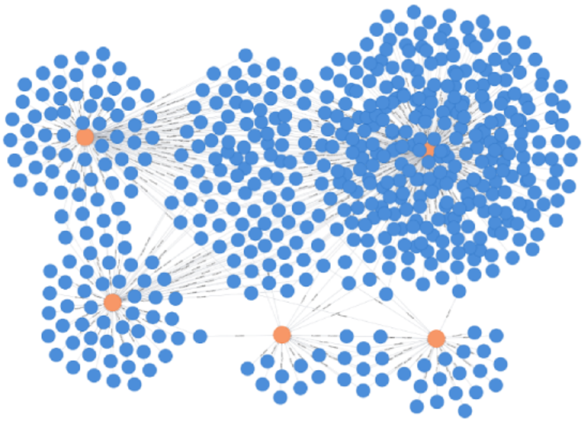


Figure 4. Topology of an example patient graph searched with 5 laboratory factors. Search query 3 in Table 3 was used. Patient nodes are shown in blue and factor nodes are shown in orange. Lines represent relationships between a patient and factors.

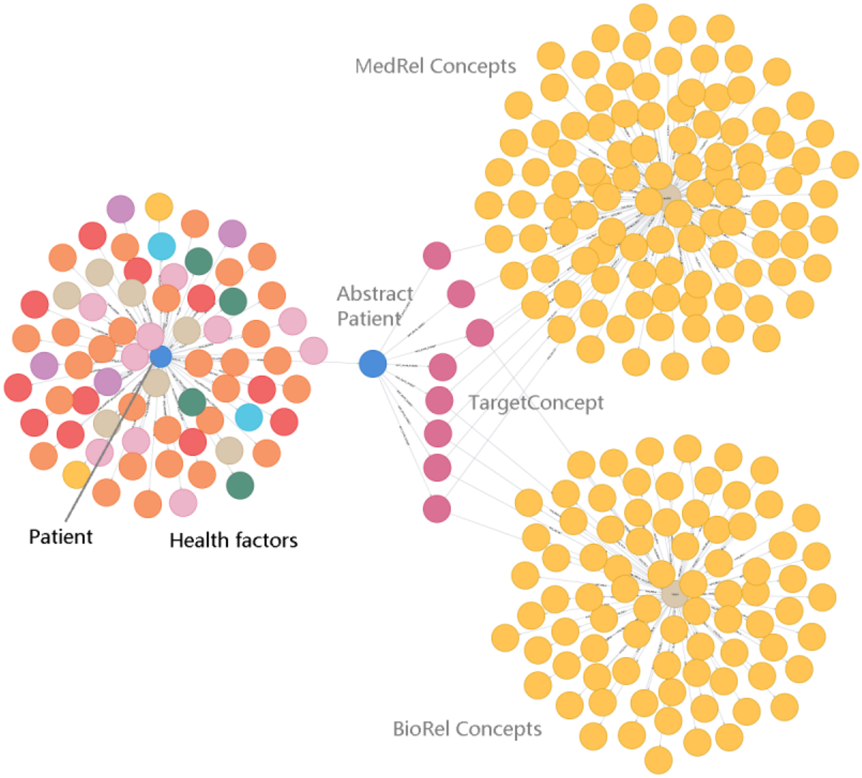


Integration of the EMR Patient Graph With the UMLS Subgraph

As the largest integrated biomedical ontology, the UMLS graph contains hierarchies of diseases and horizontal relationships with other entities. Within a disease family such as LC, the various types of LCs are horizontally connected to a myriad of related biomedical concepts including genes, proteins, symptoms, observations, medication, and treatments. This study is focused on the UMLS knowledge subgraph containing horizontal relationships for LC. Using the UMLS LC hierarchy, the target LC codes found in EMRs were expanded to 8 main LC concepts (Table 1). From these concepts, approximately 1100 relations were identified in the UMLS ontology. Most of the relations were hierarchical—for example, a parent-child relationship—and thus the relations were further filtered by the biomedical relationships that we were interested in (Textbox 1). The resulting UMLS LC biomedical subgraph had 8 LC concept nodes, 187 related biomedical concepts, and 188 horizontal biomedical relations (Figure 5).

Through a single AbstractPatient node, the EMR patient graph was connected to the UMLS subgraph for LC. Search query 4 in Table 3 and its search result in Figure 5 show an example presentation of both actual patient's health factors in the EMR and relevant biomedical knowledge in the UMLS in the same graph.

Figure 5. Example search result of the integrated biomedical graph. Search query 4 in Table 3 was used to search 1 specific ID of a patient with lung cancer. Left side: health factors from the electronic medical record of one patient with lung cancer. Right side: lung cancer biomedical knowledge from the Unified Medical Language System. Middle: single AbstractPatient as the connection. BioRel: biological concept relationship; MedRel: medical concept relationship.



Generation of the Distribution of LC Health Factors From the EMR

With the patient health factor graph, we searched for patients with LC and background patients with each of the health factors and its value. The connection delta ratios were calculated for each factor from the number of connections to patients with LC and the number of connections to background patients. Sorting factors by CDR in descending order generated a distribution of health factors for LC found in the EMR. The complete distribution of top-ranked factors over a CDR cutoff of 0.5 are shown in Table A1 in Multimedia Appendix 1 and plotted in Figure 6. As examples, up to 5 top health factors in each category are shown in Table 4. For understanding LC risk factors, this distribution excluded the various cancers, all procedures and medications related to cancers, and treatments.

Figure 6. Distribution curve of lung cancer health factors sorted by the connection delta ratio (CDR; cutoff=0.5). Only partial codes are visible on the x-axis. The full spectrum of lung cancer health factors can be found in Table A1 in Multimedia Appendix 1.

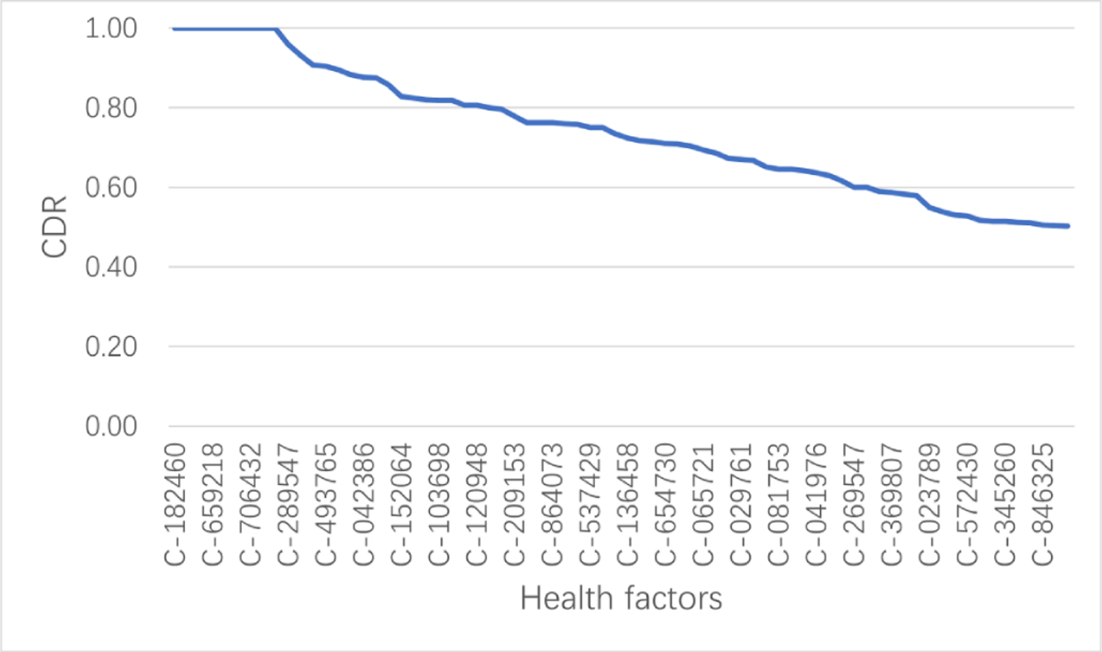


Table 4. Partial distribution of lung cancer health factors sorted by category and connection delta ratio (cutoff=0.5) as examples. The full distribution of lung cancer health factors is provided in Table A1 in Multimedia Appendix 1.

Category[a]	Local code	Term	Value	Connection delta ratio	Tag
dac	C-182460	Left lung pulmonary obstructive pneumonia	TRUE	1.00	confirmed
dac	C-248056	Right lung pulmonary obstructive pneumonia	TRUE	1.00	confirmed
dac	C-765209	Obstructive pneumonia	TRUE	1.00	confirmed
dac	C-305976	Pneumothorax	TRUE	0.93	correlated
dac	C-172569	Bacterial Infection	TRUE	0.88	correlated
lab	C-659218	Hepatitis B virus	TRUE	1.00	correlated
lab	C-493765	Squamous cell carcinoma antigen	up	0.90	confirmed
lab	C-573086	Neuron-specific enolase measurement	up	0.82	correlated
lab	C-952408	Non-small cell lung cancer associated-antigen	up	0.82	confirmed
lab	C-103698	Superoxide dismutase measurement	down	0.82	correlated
obs	C-039824	Mediastinal mass	TRUE	1.00	confirmed
obs	C-706432	Lung mass	TRUE	1.00	confirmed
obs	C-748932	Lung mass found in checkup	TRUE	1.00	confirmed
obs	C-134276	Lung shadow	TRUE	0.91	confirmed
obs	C-706281	Bronchial stenosis	TRUE	0.89	correlated
rf	C-902187	Smoking	TRUE	0.50	confirmed
smp	C-549780	Pain	TRUE	1.00	confirmed
smp	C-289547	Bloodstained sputum	TRUE	0.96	confirmed
smp	C-152064	Hemoptysis (cough up blood)	TRUE	0.83	correlated
smp	C-243071	Shoulder Pain	TRUE	0.82	confirmed
smp	C-127089	Hoarseness	TRUE	0.80	correlated

[a] Categories include condition (dac), laboratory test (lab), observation (obs), risk factor (rf), and symptom (smp).

We checked the medical literature for any associations between these top CDR-ranked health factors and LC [14-26]. This literature review confirmed that 70 out of the 71 factors (Table A1 in Multimedia Appendix 1) were LC risk factors or were correlated with LC. The relationship between 1 factor, laboratory test for immunoglobulin E levels, and LC was unsure according to the literature [27]. This high degree of concordance between the results of our CDR analysis and the literature suggests that the patient graph CDR method was effective in generating a reliable distribution of LC health factors from EMR patient data.

Discussion

Using hospital EMR patient data and applying the new patient graph CDR method recently developed from synthetic patient data, this study was able to construct an integrated biomedical graph for LC. From searching the graph, the study created a distribution of health factors for LC, which were verified through literature review. Our results show that the new strategy of first using synthetic patients for method development and then applying the methods with real patient data is valid and effective.

This study has implications for hospitals with regard to harnessing KG databases and technologies. First, generating an integrated biomedical graph with hospital EMR data may enable medical professionals to view individual patient’s health factor graphs along with the related UMLS KGs for comprehensive comparisons. Current medical concept nodes horizontally related to the LC nodes are mostly genes and gene-related biological information, as well as drugs and treatment-related information from the UMLS ontology (see Figure 5). Since the UMLS is updated quarterly, the LC integrated biomedical graph will grow as the UMLS grows. Thus, this KG integration offers a new way for hospitals to bring continuously updated international standard biomedical knowledge to patient care. The current graph model is designed specifically for searching risk factors; however, it can be modified for other clinical information tasks. It may also be integrated with cancer-associated lifestyle KGs for disease management information [28].

The second implication of this study may be applying the CDR-ranked distribution of health factors to build more effective or practical machine learning models for LC risk prediction. Because the distribution ranks factors from higher to lower relative strength, they may be used to help select more health factors to build prediction models; that is, feature engineering. For example, we have an ongoing project experimenting with the factor distribution in building LC risk prediction machine learning models. Knowing the risk factors actually found in the

EMR data, we could focus on these risk factors and reduce the variables from over 100 to less than 30 in the machine learning models that were generated from EMR-wide data. To increase the LC screening rate in larger populations, machine learning models with a small number of variables for which data can be readily available in community and rural clinics are necessary.

In addition, the patient health factor graphs generated from EMR data may enable hospitals to study the effect of various types of factors in diagnosis, medication, treatment, and disease management. Such graph analysis complements existing statistical analysis. Traditionally, studies on individual risk factors are hypothesis driven and use a clinical trial or case-control study design [29]. The literature found in this study for verification of the health factor distribution collectively indicate the use of this approach [14-27]. Because this study's patient graph method is EMR data driven, it can reveal potential new risk factors or inconclusive risk factors that deserve additional research. For example, the factor "laboratory test for immunoglobulin E levels" was tagged as "unsure" in the distribution because prior studies were inclusive. Our CDR analysis suggests that this immunoglobulin E factor requires further clinical validation [30].

Because EMR data sometimes have biases and missing data, the EMR data–driven patient graph CDR method has limitations. CDR is a simple measurement of a factor's relative strength, but caution should be taken when considering factors with a high CDR but a small number of connections. The higher the number of connections, the more reliable the CDR. Hence, studies should set a cutoff for the CDR as well as the minimal number of connections to ensure that the study uses enough data. It is also important to recognize factors that might be affected by data biases and to exclude them from CDR analysis [31]. For EMRs lacking standardized and structured data, collecting standardized data is crucial but challenging. If a data collection pipeline is not fully automated, collecting enough unbiased standardized patient profile data will be a very time-consuming process.

In conclusion, by collecting standardized data of thousands of patients with and those without LC from EMRs, it was possible to generate a hospital-wide patient-centered health factor graph for graph search and presentation. It was also practical to integrate the patient graph with the UMLS KG for LC, enabling hospitals to bring continuously updated international standard biomedical KGs from the UMLS to clinical care. Applying CDR analysis to the graph of patients with LC yielded a CDR-sorted distribution of health factors, where top CDR-ranked health factors showed a high degree of concordance with the literature. The resulting distribution of LC health factors can be used to help personalize risk evaluation and preventive screening recommendations.

Acknowledgments

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Authors' Contributions

AC designed the study and drafted the manuscript. R Huang wrote programs and analyzed data. EW and R Han collected the data. JW supervised the study. ZZ, QL, and BS proposed the study, obtained funding, and directed the study.

Conflicts of Interest

None declared.

Multimedia Appendix 1

Complete lung cancer health factor distribution sorted by category (Cat) and connection delta ratio (CDR).
[PDF File (Adobe PDF File), 159 KB-Multimedia Appendix 1]

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Abbreviations

CDR: connection delta ratio
EMR: electronic medical record
KG: knowledge graph
LC: lung cancer
PDJ: patient diagnosis journey
UMLS: Unified Medical Language System

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Corrigenda and Addenda

Correction: Understanding the Social Mechanism of Cancer Misinformation Spread on YouTube and Lessons Learned: Infodemiological Study

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In "Understanding the Social Mechanism of Cancer Misinformation Spread on YouTube and Lessons Learned: Infodemiological Study" (J Med Internet Res 2022;24(11):e39571), the authors made the following changes.

1. The *Acknowledgments* section was inadvertently excluded in the originally published article.

In the corrected version, the following statement has been added under the new *Acknowledgments* section.

> *This study was supported by the National R&D Program for Cancer Control through the National Cancer Center (NCC) funded by the Ministry of Health & Welfare, Republic of Korea (HA21C0048).*

2. In the originally published article, Affiliations 7 and 8 were incorrectly presented as two separate affiliations.

In the corrected version, Affiliation 7 is revised as follows to present the original two affiliations:

> *Yonsei Cancer Center, Yonsei University Health System, Seoul, Republic of Korea*

The updated list of affiliations and its attribution to the authors are as follows:

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The correction will appear in the online version of the paper on the JMIR Publications website on November 25, 2022, together with the publication of this correction notice. Because this was made after submission to PubMed, PubMed Central, and other full-text repositories, the corrected article has also been resubmitted to those repositories.

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Original Paper

Economic and Environmental Impact of Digital Health App Video Consultations in Follow-up Care for Patients in Orthopedic and Trauma Surgery in Germany: Randomized Controlled Trial

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Abstract

Background: Following the Riyadh Declaration, digital health technologies were prioritized in many countries to address the challenges of the COVID-19 pandemic. Digital health apps for telemedicine and video consultations help reduce potential disease spread in routine health care, including follow-up care in orthopedic and trauma surgery. In addition to the satisfaction, efficiency, and safety of telemedicine, its economic and environmental effects are highly relevant to decision makers, particularly for the goal of reaching carbon neutrality of health care systems.

Objective: This study aims to provide the first comprehensive health economic and environmental analysis of video consultations in follow-up care after knee and shoulder interventions in an orthopedic and trauma surgery department of a German university hospital. The analysis is conducted from a societal perspective. We analyze both economic and environmental impacts of video consultations, taking into account the goal of carbon neutrality for the German health care system by 2030.

Methods: We conducted a prospective randomized controlled trial comparing follow-up care with digital health app video consultations (intervention group) to conventional face-to-face consultations in the clinic (control group). Economic impact included the analysis of travel and time costs and production losses. Examination of the environmental impact comprised the emissions of greenhouse gases, carbon monoxide, volatile hydrocarbons, nitrogen oxides, and particulates, and the calculation of environmental costs. Sensitivity analysis included calculations with a higher cost per ton of carbon dioxide equivalent, which gives equal weight to the welfare of present and future generations.

Results: Data from 52 patients indicated that, from the patients’ point of view, telemedicine helped reduce travel costs, time costs, and production losses, resulting in mean cost savings of €76.52 per video consultation. In addition, emissions of 11.248 kg of greenhouse gases, 0.070 kg of carbon monoxide, 0.011 kg of volatile hydrocarbons, 0.028 kg of nitrogen oxides, and 0.0004 kg of particulates could be saved per patient through avoided travel. This resulted in savings of environmental costs between €3.73 and €9.53 per patient.

Conclusions: We presented the first comprehensive analysis of economic and environmental effects of telemedicine in the follow-up care of patients in orthopedic and trauma surgery in Germany. Video consultations were found to reduce the environmental footprint of follow-up care; saved travel costs, travel time, and time costs for patients; and helped to lower production losses. Our findings can support the decision-making on the use of digital health during and beyond the COVID-19 pandemic, providing decision makers with data for both economic and environmental effects. Thanks to the pragmatic design of our study, our findings can be applied to a wide range of clinical contexts and potential digital health applications that substitute outpatient hospital visits with video consultations.

Trial Registration: German Clinical Trials Register DRKS00023445; https://tinyurl.com/4pcvhz4n

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KEYWORDS

carbon neutrality; digital health; environmental impact; health economics; net-zero; orthopedic; sustainability; telemedicine; trauma surgery; video consultations

Introduction

Medical care does not always require patients' attendance in the hospital [1], mainly because digital health affords physicians and patients the opportunity to have synchronous video consultations online [2]. When used for outpatient follow-up care in orthopedic and trauma surgery, for example, video consultations can relieve patients of any restrictions on their mobility or of the need to travel long distances [3-5]. Patient satisfaction, physician satisfaction, and clinical outcomes often show comparable results between telemedicine and conventional face-to-face (F2F) examinations in the hospital, demonstrating that video consultations can be a safe and efficient alternative for patient care in orthopedic and trauma surgery [6-13].

After the outbreak of the COVID-19 pandemic, the role of digital health has been highlighted by the Riyadh Declaration [14]. Following the global pandemic response, there has been an increasing interest in telemedicine in clinical practice to reduce potential disease spread as well as in science, which is reflected in a growing number of literature reviews [2,15-20]. The number of clinical trials, however, remains limited. In particular, there are only a few health economic analyses of the use of telemedicine in orthopedic and trauma surgery follow-up care [15,21].

In addition to patient satisfaction and quality of care, the societal perspective needs to consider both economic and environmental effects in order to support stakeholders in deciding whether to implement telemedicine in orthopedic and trauma surgery. Following the United Nations Sustainable Development Goals, the 125th German Medical Assembly declared in 2021 that the German health care system should become carbon-neutral by 2030 [22]. One way of meeting this requirement might be the implementation of video consultations to supplement or substitute clinic consultations. Whether this is possible, however, must first be determined by investigations. A positive environmental impact of telemedicine has already been demonstrated in certain cases: for example, in the reduction of carbon monoxide, carbon dioxide, and nitric oxides [23-25]. However, analyses of the environmental impact of video consultations in the field of orthopedic and trauma surgery are limited, and no studies based on German data exist to date.

The aim of this study is to provide the first health economic analysis comparing telemedicine in the follow-up of patients in orthopedic and trauma surgery with knee and shoulder disorders with conventional F2F examinations in the clinic in Germany. The analysis focuses on the societal perspective, considering, on the one hand, the patients' point of view in terms of potential time and cost savings and, on the other hand, the environmental impact regarding potential savings of emissions and environmental costs.

Methods

Study Design

The data used for the health economic analysis were obtained by a prospective randomized controlled trial (RCT) conducted at a single German university hospital—University Hospital Giessen, Department of Trauma, Hand and Reconstructive Surgery, Level-1 trauma center—between September 2020 and April 2021. The RCT was reported according to the Consolidated Standards of Reporting Trials (CONSORT) [26]. Patients in orthopedic and trauma surgery were randomly assigned 1 to 1 to an intervention group or a control group for a single follow-up appointment. The intervention group did not attend a standard outpatient follow-up appointment in the clinic but had a real-time online video consultation with the treating physician instead. The control group, on the other hand, was treated conventionally and received a F2F examination in the clinic. In both the intervention group and the control group, the examinations were performed by the same physicians. The study population had already received conservative or surgical treatment for various knee and shoulder conditions in the clinic.

Ethical Considerations

Patients who were eligible for the study based on the inclusion and exclusion criteria in Textbox 1 were asked either at the clinic or by telephone if they wished to participate in the RCT. After a detailed verbal explanation of the study, including the conduct of a health economic analysis as part of the study, all study participants provided written informed consent. To protect the privacy of participating patients, pseudonymization of the study data took place. Study participants were not compensated for their participation. The local ethics committee of the University of Giessen approved the RCT (AZ 73/20), and the study was registered in the German Clinical Trials Register (DRKS00023445).

Textbox 1. Inclusion and exclusion criteria of the randomized controlled trial.

Inclusion criteria:

- 18 years or older
- Previous outpatient or inpatient stay at the clinic, with an operation or conservative therapy
- Need of a follow-up that does not require more than a visual examination
- Ownership of a computer, laptop, tablet, or smartphone with microphone and camera
- Stable internet connection
- Mental and physical ability to consent and to participate
- Sufficient knowledge of German in order to understand the declaration of consent
- Shoulder International Classification of Diseases, Tenth Revision (ICD-10) codes: M75.1, M75.6, M75.0, Z96.60, M75.4, M19.91, S43.1, S42.20, S42.00, M75.2, M75.3, and S43.0
- Knee ICD-10 codes: S83.53, S83.54, S83.2, S83.0, M22.0, M23.32, M23.35, M17.1, M17.5, M21.16, M21.06, S83.3, S83.44, S83.43, S82.18, S82.0, S72.3, S72.43, M25.56, M76.5, S83.6, S76.1, and S86.8

Exclusion criteria:

- Neurological diseases that preclude the use of digital devices
- Diagnosis of dementia, blindness, or deafness
- Need for presence in the clinic and on-site treatment and diagnostics (ie, imaging, laboratory, stitches, and drainage)
- Appointments where the patient has to be touched and moved by the treating physicians
- Lack of willingness to participate
- Failure to consent

Sample Size and Randomization

The sample size calculation of the underlying RCT was based on an a priori power analysis. As a conservative estimate, we used half of the effect size of 2.19 that was observed for the findings of patient satisfaction with telemedicine in a study by Sharareh and Schwarzkopf [8]. The effect size of 1.095 yielded 19 patients per study arm for a power of 90% in a 2-sided *t* test with a 5% significance level. To increase statistical power and to compensate for potential withdrawals and dropouts, missing responses, and a skewed distribution of results, the number of participants was expanded to 30 patients for each group. In total, 60 eligible patients were recruited for the study.

Using block randomization with randomly varying block sizes (ie, 4, 6, and 8), 30 patients were assigned to a follow-up with telemedicine (intervention group), and 30 patients were assigned to a conventional F2F follow-up in the clinic (control group). The parallel-design randomization and assignment process was performed independently of the treating physicians by study staff using sealed envelopes.

Course of the Study

The video consultations in the intervention group were browser based for physicians and multiplatform for patients, including a digital health app or browser-based software from a German telemedicine provider. The software complies with the legal requirements in Germany and is recognized by the National Association of Statutory Health Insurance Physicians. The university hospital paid a monthly fee for each physician to use the software. Video consultation procedures were deliberately kept as simple and as functional as possible to ensure that they would be viable in regular clinical practice: all video consultations were performed directly between the physicians in the clinic and the patients, regardless of their location. No other medical providers, such as local caregivers or others, were involved. Patients received written instructions on how to conduct the video consultation, and no additional clinical staff were required to assist the patients. This pragmatic study design appeared to be the most promising one for a health economic evaluation seeking to produce valid, generalizable results [27]. Patients in the intervention group did not have to bear any additional costs or out-of-pocket payments for using telemedicine, as the digital health app or browser-based software was free for them to use. They were only required to have a smartphone, tablet, laptop, or computer with a microphone and camera, and an adequate internet connection. The examination itself was paid for by their respective health insurance. Patients in the control group did not have to pay any additional costs either; their costs for an in-clinic follow-up appointment (eg, travel costs) were the same as those they would have paid outside of study participation.

After the follow-up appointments, patients in both the intervention and control groups completed questionnaires. These questionnaires included questions about the distance between the patients' homes and the clinic, the amount of time spent for the appointment (eg, travel and waiting time), and the potential need to be absent from work to attend the appointment. Further information on the study can be found in a previous publication by Muschol et al [13].

Statistical Analysis and Health Economic Evaluation

The RCT data are presented as mean and SD, median and IQR, or percentage. To compare the intervention and control groups, the Mann-Whitney *U* test was used for continuous variables and the Fisher exact test was used for categorical ones. Statistical significance was assumed at $P \leq .05$.

The health economic analysis was based on data collected from the questionnaires and other official, external data. The study design was guided by recommendations for health economic analyses in the context of eHealth interventions, and the study examined non–health care costs associated with the use of telemedicine from a societal perspective [27,28]. The analysis proceeded in two steps. In the first step, economic effects of the societal perspective were examined from the patients' point of view. This involved, firstly, calculating and comparing three types of non–health care costs associated with medical appointments:

1. Travel costs were calculated following recommendations for empirical standard costs for health economic evaluations in Germany [29].
2. Time costs were assessed by assigning monetary values to patients' travel time, waiting time, and total time spent on appointments based on Verbooy et al's [30] valuation approach to unpaid work and leisure time.
3. Production losses due to patients' absence from work while attending their appointments were computed using Germany's average gross hourly wage in 2021 and average working hours for all German full-time and part-time employees in 2019 [31,32].

When tallying total costs from a societal perspective, it was felt to be appropriate to differentiate between patients who were employed and patients who were not employed, given that production losses are only relevant for patients who are employed.

In the second step, the effects of the societal perspective were evaluated in the form of the environmental impact of telemedicine. The analysis of the environmental impact was conducted using data from the German Federal Environment Agency. It comprised three different aspects. First, the environmental impact in terms of greenhouse gases, carbon monoxide, volatile hydrocarbons, nitrogen oxides, and particulates was calculated by multiplying the average emissions per passenger-kilometer (pkm) by the kilometers patients traveled by car to and from the clinic. This calculation was based on an average car occupancy of 1.4 passengers, as the average emissions are specified by the Federal Environment Agency on the basis of this value [33]. A separate calculation of emissions from public transportation was not performed within the study because only 1 patient in the control group and 1 patient in the telemedicine group used or would have used public transportation. Second, the average environmental costs incurred per pkm by the patients' trips per car were calculated. For this purpose, the cost rate of the Federal Environment Agency of €195 per ton of carbon dioxide equivalent was applied (a currency exchange rate of €1=US $0.97 is applicable) [34,35]. This value is based on a higher weighting of the welfare of current versus future generations [35].

In a third step, the potential savings in emissions and environmental costs were estimated in a model calculation if 8 patients per week would conduct a video consultation instead of a clinic consultation, as was the case in our study [33-35].

Sensitivity Analysis

Finally, a sensitivity analysis was performed to evaluate the robustness of the findings. For the patients' point of view in the societal perspective, this analysis studied the effect of differentiating between full-time and part-time employment when calculating production losses [32]. For the environmental impact of the societal perspective, the sensitivity analysis considered the following:

1. A cost rate from the Federal Environment Agency for the calculation of the environmental costs of €680 per ton of carbon dioxide equivalent, which gives equal weight to the welfare of present and future generations [34,35].
2. A total of 16 patients with a video consultation per week for the analysis of potential savings in emissions and environmental costs [33-35].

For the calculation of the environmental costs, both €195 and €680 per ton of carbon dioxide equivalent were considered [34,35]. As the Federal Environment Agency reports both cost rates, the aim of the sensitivity analysis was to show how the equal weighting of the welfare of present and future generations (€680) compared to the higher weighting of the welfare of present versus future generations (€195) affects the environmental costs.

Results

General Findings

Of the 60 patients recruited—intervention group (n=30) and control group (n=30)—4 patients in each of the groups withdrew from the study. Thus, data from a total of 52 patients could be considered for the health economic evaluation, with several variables displaying a lower n value due to missing items on some patient questionnaires. The progress of the recruited patients through the trial is shown in a CONSORT flow diagram in Multimedia Appendix 1.

Demographic patient characteristics are shown in Table 1. No significant differences were observed between the telemedicine group and the control group.

Regarding the variables used for calculating costs, however, the differences between the groups were partially significant, as shown in Table 2. Treatment duration in the intervention group, at 8.23 minutes on average, was significantly shorter than that in the control group, at 10.92 minutes on average (P=.02). The average waiting time in the online waiting room for the telemedicine software was also significantly shorter than that experienced in the clinic (6.73 minutes vs 36.88 minutes, respectively; P<.001). The largest intergroup difference, however, was observed in total patient time spent per follow-up appointment. An appointment in the telemedicine group took an average of 21.92 minutes out of the patients' days, whereas an appointment in the control group required patients to spend 154.80 minutes on average (P<.001). There was no significant difference between the potential travel distance and travel time the telemedicine group would have faced if required to travel to an in-clinic appointment and the actual travel distance and travel time faced by the control group. The groups also did not differ significantly in patients' absence from work due to their appointments. Nevertheless, of the employed patients, only 5% (1/20) were absent from work so they could attend the appointment in the telemedicine group, compared with 16% (3/19) in the control group, as shown with the Fisher exact test (*P*=.34). In the telemedicine group, 1 patient had to visit the clinic again for further treatment. As this would also have been required after an F2F consultation and, therefore, occurred independently of the video consultation, this additional visit was not included in the cost calculation.

Table 1. Demographic characteristics of patients.

Characteristics	Telemedicine group (n=26), n (%)	Control group (n=26), n (%)	*P* value[a]
Medical indication			.99
Knee	10 (38)	9 (35)	
Shoulder	16 (62)	17 (65)	
Age (years)			.36
18-40	7 (27)	5 (19)	
41-60	17 (65)	15 (58)	
>60	2 (8)	6 (23)	
Female	11 (42)	10 (38)	.99
Employed	20 (77)	19 (76)[b]	.99

[a] *P* values were based on the Fisher exact test.

[b] Percentage of n=25 due to missing item on questionnaire.

Table 2. Variables included for cost calculation.

Variables	Telemedicine group (n=26)			Control group (n=26)			*P* value[a]
	Participants, n (%)	Mean (SD)	Median (IQR)	Participants, n (%)	Mean (SD)	Median (IQR)	
Treatment duration (minutes)	26 (100)	8.23 (4.45)	6.00 (5-10)	25 (96)	10.92 (5.58)	10.00 (8-14.5)	.02
Travel distance (kilometers)	26 (100)	37.00 (32.06)	30.00 (10-46.25)	25 (96)	31.58 (22.62)	28.00 (15.5-45)	.65
Actual and potential travel time (minutes)	26 (100)	38.46 (21.72)	40.00 (18.75-46.25)	25 (96)	34.80 (20.89)	30.00 (20-40)	.42
Waiting time (minutes)	26 (100)	6.73 (6.84)	5.00 (1.75-10)	24 (92)	36.88 (27.54)	30.00 (15-48.75)	<.001
Total time spent on appointment (minutes)	26 (100)	21.92 (10.40)	22.50 (13.75-30)	25 (96)	154.80 (79.75)	150.00 (105-197.5)	<.001

[a] *P* values were based on the Mann-Whitney *U* test.

Patients' Perspectives

The cost calculation from the patients' point of view in the societal perspective showed that patients in the control group had to pay an average of €18.95 in travel costs, based on a cost of €0.30 for each kilometer travelled to and from the clinic, as shown in Table 3. There were no travel costs for patients in the telemedicine group because they did not have to attend the clinic. If they had had an in-clinic follow-up, however, their average travel costs would have been €22.20.

The time costs resulting from follow-up appointments in both groups were estimated at €16.00 per hour to account for both unpaid work time and leisure time that patients lost. The average cost of patients' travel time was €18.56 in the control group. Again, patients in the telemedicine group faced no travel time costs due to the trip they avoided. Yet, the potential cost of their travel time would have been €20.51. The increased waiting time in the clinic was reflected in time costs of €9.83 in the control group, compared with €1.79 in the intervention group.

The difference in time costs between the groups became even more pronounced when the total time patients spent on their follow-up appointments was valued. Whereas patients with a telemedical appointment had average total time costs of €5.85, those with an in-clinic appointment had total time costs of €41.28. In other words, a telemedical rather than an in-clinic follow-up appointment would have saved patients €35.43 in average time costs.

Finally, the production loss due to patients' absence from work while they were attending their appointments was calculated. This was based on an average hourly wage of €29.48 in Germany and an overall average of 6.96 working hours per day per full-time or part-time German employee. With 1 patient absent in the telemedicine group and 3 patients absent in the control group, total production losses were €205.18 and €615.54, respectively. With 20 employed patients in the telemedicine group and 19 employed patients in the control group, the costs due to lost production averaged €10.26 for a telemedical follow-up and €32.40 for an in-clinic one.

Taking employment status into account, the total costs of a follow-up appointment were €16.11 for an employed patient in the telemedicine group and €92.63 for an employed patient in the control group. For an unemployed patient, the total costs decreased to €5.85 in the telemedicine group and to €60.23 in the control group due to the irrelevant production loss. Multimedia Appendix 2 presents the cost calculations in detail.

Table 3. Cost calculation from the patients' perspective.

Costs	Telemedicine group	Control group	Difference
Travel costs (€[a]), mean (SD)	0 (0)	18.95 (13.57)	18.95
Travel time costs (€), mean (SD)	0 (0)	18.56 (11.14)	18.56
Waiting time costs (€), mean (SD)	1.79 (1.82)	9.83 (7.34)	8.04
Total time costs (€), mean (SD)	5.85 (2.77)	41.28 (21.27)	35.43
Production loss (€)	205.18	615.54	410.36

[a] A currency exchange rate of €1=US $0.97 is applicable.

Environmental Impact

To calculate the emissions saved in the telemedicine group due to the avoided trips to and from the clinic, 152 g/pkm for greenhouse gases, 0.94 g/pkm for carbon monoxide, 0.15 g/pkm for volatile hydrocarbons, 0.38 g/pkm for nitrogen oxides, and 0.006 g/pkm for particulates were applied based on an average car occupancy of 1.4 passengers. This led to the result that around 11.248 kg of greenhouse gases, 0.070 kg of carbon monoxide, 0.011 kg of volatile hydrocarbons, 0.028 kg of nitrogen oxides, and 0.0004 kg of particulates were saved per patient with the help of video consultations. Table 4 also shows the total emissions saved for the 26 patients in the telemedicine group. For example, as a result of the video consultations, emissions of 292.448 kg of greenhouse gases could be avoided in our study. The calculation of environmental costs saved in the telemedicine group is based on environmental costs of €0.05045 per pkm. This value represents the average environmental costs of gasoline and diesel powered cars. The use of telemedicine saved approximately €3.73 in environmental costs per patient, resulting in a total of €97.07 for all patients in our study. Finally, the potential savings can also be seen in the model calculation for 1 year if 8 patients per week had a video consultation instead of a clinic consultation, as was the case in our study. For this calculation, the average distance between the home of the patients in the telemedicine group and control group and the clinic was used. With a total of 384 patients who would not have to travel to the clinic each year due to video consultations, a total of 4009.88 kg of greenhouse gases, 24.80 kg of carbon monoxide, 3.96 kg of volatile hydrocarbons, 10.02 kg of nitrogen oxides, and 0.16 kg of particulates could be avoided. In addition, at €195 per ton of carbon dioxide equivalent, €1330.91 in environmental costs could be saved.

Table 4. Saved emissions and environmental costs in the telemedicine group.

Emissions and costs	Per patient	Total
Greenhouse gases (kg)	11.248	292.448
Carbon monoxide (kg)	0.070	1.809
Volatile hydrocarbons (kg)	0.011	0.289
Nitrogen oxides (kg)	0.028	0.731
Particulates (kg)	0.0004	0.012
Environmental costs (€[a])	3.73	97.07

[a] A currency exchange rate of €1=US $0.97 is applicable.

Sensitivity Analysis

In the subsequent sensitivity analysis, several adjustments were made. First, the cost calculation from the patients' point of view was modified to test the effect of alternative assumptions on the valuation of production losses. Assuming that all patients who were absent from work were employed full time (ie, 8.2 hours per day), the societal cost of lost production would have increased to €241.74 (mean €12.09, SD 54.05) in the telemedicine group and to €725.21 (mean €38.17, SD 90.56) in the control group. In contrast, assuming only part-time employment of 3.9 hours per day for all patients who were absent from work, the costs of lost production would have decreased to €114.97 (mean €5.75, SD 25.71) in the telemedicine group and to €344.92 (mean €18.15, SD 43.07) in the control group. These assumptions would have changed the total costs for employed patients to €17.94 for full-time employees and €11.60 for part-time employees in the telemedicine group, as well as to €98.40 for full-time employees and €78.38 for part-time employees in the control group.

Second, the calculation of environmental costs was adjusted to the cost rate of €680 per ton of carbon dioxide equivalent, which increased the average environmental costs of gasoline and diesel cars to €0.12885 per pkm. Due to this adjustment, the environmental costs saved in the telemedicine group would have been €9.53 per patient and €247.91 in total.

In addition, if a total of 16 patients per week had a video consultation instead of a clinic consultation, approximate emissions of 8019.76 kg of greenhouse gases, 49.60 kg of carbon monoxide, 7.91 kg of volatile hydrocarbons, 20.05 kg of nitrogen oxides, and 0.32 kg of particulates could be saved. Environmental costs could furthermore be reduced by €2661.82, at €195 per ton of carbon dioxide equivalent, or by €6798.33, at €680 per ton of carbon dioxide equivalent.

Discussion

Principal Findings

This analysis of the economics of using telemedicine in follow-up care for patients in orthopedic and trauma surgery in a German university hospital showed that implementing video consultations enabled time and cost savings for patients, savings in environmental costs, and reductions in emissions.

Implications for Patients

Seen from the patients' point of view in the societal perspective of the health economic analysis, the use of telemedicine was not associated with additional costs (eg, out-of-pocket payments) for the patients in our study. On the contrary, compared with the control group, telemedical appointments resulted in cost savings due to the avoidance of travel and the reduction in time costs.

Previous economic evaluations by Buvik et al [36] and Ohinmaa et al [37] also showed that telemedicine saved travel time and travel distance—and, thus, travel costs—in sparsely populated Scandinavian countries even though patients had to travel to a local caregiver for their appointment [36,37]. Similarly, RCTs by Sathiyakumar et al [9] and Kane et al [12] found savings in travel distances and time spent as well, but these studies did not feature economic analyses [9,12]. Reducing travel burdens is an important societal benefit of telemedicine, as it can ensure better access to medical care. In particular, patients in rural regions and hospitals that seek to offer their medical services beyond their own region stand to benefit. At the same time, however, all patients must still be able to reach their local clinic when video consultations are not sufficient.

Since our trial ended in 2021, our analysis did not consider the energy pricing dynamics following the 2022 European energy crisis. Actual savings in travel costs could be far higher in future digital health deployments.

In addition, the results of the analysis showed that the average costs of lost production were lower for a video consultation compared to a clinical consultation, indicating that telemedicine may have a positive impact in this regard as well. The potential of telemedicine to reduce lost work time—and, thus, production losses—reported here is consistent with the findings of other RCTs [9,12,36,37].

From a societal point of view, the use of telemedicine saved average total costs for employed patients of €76.52 per follow-up appointment, ranging from €66.78 to €80.46 in the sensitivity analysis. Most likely, the real savings would be even higher, as patients often wish or require an accompanying person for a clinic consultation, and the cost and time savings of companions were not considered in the study. The finding that video consultations save overall costs compared with conventional F2F examinations in follow-up care is also confirmed by Buvik et al's [36] analysis. It should be noted, however, that in our calculation patient time lost due to a follow-up appointment was assigned a monetary value independently of any production losses, because including such time costs is strongly recommended in health economic methodology [28,30].

Implications for the Environment

In addition, from the environmental point of view in the societal perspective, our analysis showed that for each patient who received a video consultation instead of a clinic consultation, emissions of 11.248 kg of greenhouse gases, 0.070 kg of carbon monoxide, 0.011 kg of volatile hydrocarbons, 0.028 kg of nitrogen oxides, and 0.0004 kg of particulates could be saved due to avoiding traveling by car. International studies have also demonstrated the reduction of emissions through the use of telemedicine, although the level of individual emissions differs in the respective studies [38,39]. For example, in a study by Udayaraj et al [23], telemedicine led to a reduction of 3527 miles and saved 1035 kg of carbon dioxide for kidney transplant patients in the United Kingdom. A retrospective analysis of patients in vascular surgery in the United States by Paquette and Lin [24] found a reduction of 1632 kg of carbon dioxide; 42,867 g of carbon monoxide; and 3160 g of nitric oxides by performing a total of 146 telemedicine encounters. In addition, based on Spanish data, a study by Vidal-Alaball et al [25] showed an average reduction of 3248.3 g of carbon dioxide, 4.05 g of carbon monoxide, and 4.86 g of nitric oxides per patient in a telemedicine program that included different specialties.

In our study, up to 8 patients could be treated weekly via telemedicine, which can lead to an annual improvement in the environmental footprint for a single German university orthopedic and trauma surgery department alone. Although the performance of telemedicine is not suitable for all patients in orthopedic and trauma surgery, the reduction in emissions could be improved by increasing the number of patients treated by video consultations each week. If the number of patients were expanded to the 1903 hospitals in Germany and included specialties suitable for telemedicine, such as general and visceral surgeries or dermatology, the call of the 125th German Medical Assembly in 2021 for a net-zero German health care system could be substantially supported [22].

In addition to the emission savings themselves, our study also showed that the introduction of telemedicine can also contribute to a reduction in environmental costs from the societal perspective.

Implications for Practice

This health economic analysis provides clinical evidence that can improve stakeholders' decision-making on implementing telemedicine both in and beyond the current COVID-19 pandemic. It was shown that the use of telemedicine in the follow-up care of orthopedic and trauma surgery benefits both patients and the environment from an economic perspective. Given the pragmatic design of this study, it can be expected that its main findings can be applied by decision makers in other clinical contexts as well.

When deciding whether to implement telemedicine, however, health care providers should consider other aspects besides the economic and environmental benefits. First, the quality of care provided by telemedicine must be ensured. Patient and physician satisfaction, efficiency, and the safety of the video consultations in terms of the same clinical outcomes achieved in F2F consultations play an important role. Various studies show that these goals can be achieved by introducing telemedicine in orthopedic and trauma surgery [6-12]. In addition, we have extensively analyzed patient and physician satisfaction, as well as quality of care for the study cohort in a previous publication [13]. Second, the costs of the technological infrastructure for telemedicine (eg, for electricity, internet connection, and hardware, such as computers and laptops with cameras and microphones) have to be considered. This infrastructure, however, is expected to be part of the standard equipment in most hospitals, as was the case in our study.

Limitations

This study also has some limitations that should be noted. First, although the results were primarily based on actual data collected in the course of an RCT, some assumptions had to be made to be able to calculate costs. Travel costs saved, for example, were calculated based on the assumption that patients have their video consultations at home. In fact, they could have them anywhere, meaning that patients' actual travel costs from that place to the hospital may well be higher or lower. The distance from home and the time spent on the appointments (eg, travel and waiting times) were furthermore queried via a questionnaire, and the actual distances and times could potentially differ slightly from the information provided by the patients. In addition, the original calculation of production loss lacked information on whether patients were employed full time or part time. For this reason, a sensitivity analysis sought to identify possible deviations and to evaluate the robustness of the findings.

Furthermore, given that data on time costs for German patients were missing in the literature, Verbooy et al's [30] valuation approach was used, which was based on Dutch data. However, assuming that the Dutch population is reasonably similar to the German one, this minor inconsistency appears unlikely to have distorted overall results.

Finally, one of the inclusion criteria of the study was patients' ownership of a technical device (smartphone, computer, etc) that allowed them to make video calls. This requirement could lead to socioeconomic inequalities being exacerbated, because only patients with adequate financial means might be able to benefit from cost savings due to telemedicine [40]. This inequity could not be avoided within the study, but it is an important issue with practical relevance and should be taken into account by policy makers.

Conclusions

The use of telemedicine was found to reduce the environmental footprint and to save travel costs, travel time, and time costs for patients, and it helped to lower production losses from a societal perspective compared to F2F consultations in Germany. Thus, telemedicine helps to reduce costs in multiple dimensions. These results were demonstrated in the first health economic analysis of the use of telemedicine in follow-up care for patients with knee and shoulder disorders in orthopedic and trauma surgery, based on data from Germany. Simultaneously, this study provided economic and environmental evidence supporting stakeholders, such as hospitals, patients, and policy makers, who may consider extending the use of telemedicine in and beyond the COVID-19 pandemic. In addition, these findings might be relevant beyond the medical specialty of orthopedic and trauma surgery; they could be applied to other clinical contexts and to a wide range of potential digital health applications that substitute outpatient hospital visits with video consultations.

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Data Availability

The data sets generated and analyzed during this study are available from the corresponding author upon reasonable request.

Conflicts of Interest

None declared.

Multimedia Appendix 1

Consolidated Standards of Reporting Trials (CONSORT) flow diagram.
[DOCX File , 87 KB-Multimedia Appendix 1]

Multimedia Appendix 2

Detailed presentation of cost calculations.
[DOCX File , 30 KB-Multimedia Appendix 2]

Multimedia Appendix 3

CONSORT-eHEALTH checklist (V 1.6.1).
[PDF File (Adobe PDF File), 345 KB-Multimedia Appendix 3]

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Abbreviations

CONSORT: Consolidated Standards of Reporting Trials
F2F: face-to-face
pkm: passenger-kilometer
RCT: randomized controlled trial

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Original Paper

Online Health Information Seeking Among Patients With Chronic Conditions: Integrating the Health Belief Model and Social Support Theory

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Abstract

Background: Chronic diseases are the leading causes of death and disability. With the growing patient population and climbing health care expenditures, researchers and policy makers are seeking new approaches to improve the accessibility of health information on chronic diseases while lowering costs. Online health information sources can play a substantial role in effective patient education and health communication. However, some contradictory evidence suggests that patients with chronic conditions may not necessarily seek online health information.

Objective: This study aims to integrate 2 theories (ie, the health belief model and social support theory) and a critical health literacy perspective to understand online health information seeking (OHIS) among patients with chronic conditions.

Methods: We used the survey method to collect data from online chronic disease communities and groups on social media platforms. Eligible participants were consumers with at least 1 chronic condition and those who have experience with OHIS. A total of 390 valid questionnaires were collected. The partial least squares approach to structural equation modeling was employed to analyze the data.

Results: The results suggested that perceived risk (t=3.989, P<.001) and perceived benefits (t=3.632, P<.001) significantly affected patients' OHIS. Perceived susceptibility (t=7.743, P<.001) and perceived severity (t=8.852, P<.001) were found to influence the perceived risk of chronic diseases significantly. Informational support (t=5.761, P<.001) and emotional support (t=5.748, P<.001) also impacted the perceived benefits of online sources for patients. In addition, moderation analysis showed that critical health literacy significantly moderated the link between perceived risk and OHIS (t=3.097, P=.002) but not the relationship between perceived benefits and OHIS (t=0.288, P=.774).

Conclusions: This study shows that the health belief model, when combined with social support theory, can predict patients' OHIS. The perceived susceptibility and severity can effectively explain perceived risk, further predicting patients' OHIS. Informational support and emotional support can contribute to perceived benefits, thereby positively affecting patients' OHIS. This study also demonstrated the important negative moderating effects of critical health literacy on the association between perceived risk and OHIS.

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KEYWORDS

health information seeking; patients with chronic conditions; health belief model, social support; critical health literacy

Introduction

Background

Chronic diseases are the leading global causes of death and disability. In the United States, 6 in 10 adults have 1 chronic disease, and 4 in 10 adults live with 2 or more chronic conditions [1]. According to the US Centers for Disease Control and Prevention (CDC), chronic diseases account for 3.8 trillion dollars in annual health care expenses in the United States [1]. In China, 3 chronic diseases (ie, cardiovascular diseases, cancer, and chronic respiratory diseases) were responsible for 80.7% of total deaths in 2019 [2]. Despite causing huge burdens, chronic diseases are influenced by several risk factors (eg, poor diet, physical inactivity, hyperlipidemia, and uncontrolled high blood pressure) that are generally preventable and manageable [3]. However, people living with chronic diseases often reported limited knowledge of the causes and consequences of their conditions [4]. Studies revealed that better informed patients are more likely to manage their chronic conditions, prevent exacerbations, and lower costs [5]. Due to the growing patient population and climbing health care expenditures, researchers and policy makers are seeking new approaches to improve the accessibility of health information on chronic diseases while lowering costs. Online health information sources can play a substantial role in facilitating effective patient education and health communication.

It is widely assumed that online health information seeking (OHIS) plays a significant role in the health management of patients with chronic diseases. Some evidence accords with this notion. For example, Madrigal and Escoffery [6] found that patients with chronic diseases are more likely to perform OHIS than those who are healthy and that patients with chronic diseases are more knowledgeable in OHIS. The phenomenon may be explained by the fact that health information needs trigger the OHIS process. Patients with chronic conditions have more explicit information needs than general consumers, including information on disease causes, lab testing results, and coping strategies [7-9]. Online sources are more convenient and accessible than formal health care services, so patients are assumed to perform OHIS frequently.

However, some contradictory evidence suggests that patients with chronic conditions may not necessarily seek health information. For example, McCloud and colleagues [10] conducted a mail-based survey in the United States and found that 1 in 3 cancer survivors intentionally avoided cancer-related information. Li et al [11] carried out a randomized field experiment in China and revealed that people avoid information on cancer and diabetes tests even when there is no monetary or transaction cost. A recent metareview concluded that health status is not a strong predictor of health information seeking [12]. Therefore, aside from health information needs, research questions of whether and why patients with chronic conditions seek health information online remain unresolved.

The existing research has applied many well-established theories to the portrayal of health behaviors among general consumers, such as the health belief model (HBM), social support theory, and health literacy. However, few attempts have been made to integrate these theories to understand health information behaviors comprehensively. Therefore, this paper aims to integrate 2 long-standing theories (ie, the health belief model and social support theory) and a critical health literacy perspective to understand online health information seeking among patients with chronic conditions.

Research Model and Hypotheses

OHIS Among Patients With Chronic Conditions

Patients with chronic conditions have long-term health management demands; thus, many health experts call for patient activation, an ideal state wherein patients know how to manage their conditions, keep functioning, and prevent health declines [13]. The extrinsic needs related to health management (eg, to get better informed and to manage chronic conditions) and intrinsic motivations (eg, to seek social support) motivate patients to perform OHIS [14].

Moreover, the internet provides patients with a supportive environment for OHIS. Conventional online health information sources include general search engines [15], medical databases [16], online forums [17], and so forth. Recently, social media has become one of the most popular online health information sources among users [18]. Song et al [19,20] suggest that although many social media platforms were not intentionally designed for OHIS, the rich sets of technological affordances embedded in these platforms allow users to search for health-related content and facilitate user engagement. For example, YouTube empowers patients in chronic condition management [21], and TikTok has also been a critical channel for delivering chronic disease information [22].

HBM As an Explanatory Framework in Health Behavior Research

Historically, the HBM has been widely used to understand why patients engage in proactive health behaviors. Social psychologists developed the HBM in the 1950s to explain preventive health behaviors [23]. The model assumes that the intentions of taking proactive health actions rely more on individual beliefs about a particular condition than the objective facts of the condition [24]. According to the HBM, people's proactive health behaviors are primarily determined by their *perceived susceptibility* to disease-related conditions, *perceived severity* of the consequences of disease-related conditions, *perceived benefits* of the behaviors in reducing the threats, and *perceived barriers* to the negative aspects of the health behaviors [25].

Numerous studies have investigated various health behaviors through the lens of HBM to contextualize health behaviors including a healthy diet [26], cancer screening [27], vaccination [28], medical help seeking [29], and preventive behaviors during epidemics [30]. For example, Hochbaum [31] applied the HBM when examining X-ray screening for tuberculosis and found that perceived susceptibility to tuberculosis and perceived benefits of screening varied across participants who had and had not received chest X-rays. More recently, Wong et al [28] employed the HBM to assess the acceptance of the COVID-19 vaccine and revealed that perceived severity of contracting COVID-19 and perceived benefits of receiving the vaccine positively predicted vaccine acceptance. Overall, these studies produced internally consistent results that provided fairly strong support for HBM and informed the subsequent use of HBM to understand health behaviors. Despite the intensive use of HBM in health and medical contexts, the model is less adopted to investigate health information behaviors. Given the considerable explanatory power of HBM in health sciences, this study will employ the HBM to investigate OHIS intentions among patients with chronic conditions.

Although the HBM does not specify the variable ordering, it implicitly purports the idea that perceived susceptibility and severity jointly lead to a perception of the risk of disease, and perceived benefits influence an individual's assessment of the outcome of the proactive health behaviors [32]. As such, the risk-benefit consideration motivates the individual to take action. Noteworthily, the HBM does not provide rules of combinations of the constructs. For example, Harrison et al [33] did not include the cues to action and health motivation components in their analyses. Ahadzadeh et al [34] only included risk perceptions (ie, perceived susceptibility and perceived severity) when using the HBM. According to a recent systematic review [27], the risk-benefit aspect is the most frequently explored component in prior studies. Therefore, this study will also focus on the risk-benefit perspective.

The risk-benefit relationship posited by HBM has been partially examined in prior studies. For example, Ahadzadeh et al [34] found that risk perceptions had an indirect positive effect on Malaysian women's online health-related internet use. Mou et al [35] observed that perceived benefits of online health websites, perceived susceptibility, and perceived severity of one's health conditions were significant predictors of online health information seeking. Accordingly, our study proposes 2 hypotheses based on the parsimonious form of the HBM: (1) The OHIS of patients with chronic diseases is positively influenced by the perceived risk of chronic diseases (H1a) and the perceived benefits of performing OHIS (H1b); and (2) the perceived risk of chronic diseases of patients with chronic conditions is positively influenced by perceived susceptibility (H2a) and severity (H2b).

However, explicating the relationship between the HBM constructs cannot resolve all the theoretical limitations of the HMB. To overcome these constraints, researchers have often treated the HBM as an overarching framework [36] and combined its constructs with other theories [37]. For instance, Ahadzadeh et al [34] incorporated the HBM and the technology acceptance model to understand users' online health-related internet behaviors. Mou et al [35] integrated the HBM, the extended valence framework, and the perspective of self-efficacy to explain users' OHIS. Since prior work suggested that OHIS is associated with social support and health literacy [38], we will integrate the perspectives of social support and health literacy in this study.

Social Support in OHIS

Social support is often described as the comfort, help, or information that an individual obtains from others [39]. In offline settings, social support is often provided by friends and relatives [40]. In online environments, social media serves as an important source of social support for patients. For example, Zhang and He [41] found that people living with diabetes exchange medical and lifestyle information and provide and seek social support in Facebook groups. These Facebook diabetes groups share a broad variety of topics, such as nutrition, medications, blood glucose screening, and physical activity [42].

Social support has been extensively examined in health-related fields, with many studies finding positive associations between social support and people's physical and mental health [43,44]. The benefits of social support are especially evident in patients' self-management of chronic conditions [45]. However, despite its promising positive impacts, the mechanisms of how social support influences health behaviors remain underexplored. A couple of studies examined the direct associations between informational and emotional support and health behaviors or conditions. For example, Wang and Parameswaran [46] suggested that adequate online social support is correlated with better self-care behaviors of HIV patients. However, other studies revealed that the impacts of social support on health behaviors are mediated by different factors, such as health self-efficacy and health information seeking [47,48].

Although social support is a multifaceted concept with different subdimensions, informational and emotional supports are the most frequently studied aspects in the existing health literature [49]. Savolainen [50] found that dietary information seekers solicited emotional support in health blogs by describing their dieting problems, and readers responded by offering considerable informational and emotional support. Stellefson and Paige [42] surveyed the 34 largest diabetes support groups on Facebook and revealed that informational and emotional support exchanges were the 2 most common purposes for creating those groups. Therefore, this study will focus on these 2 main types of social support.

Regarding patients' motivations for seeking online sources for social support, some researchers suggest a compensation view and posit that online sources can fulfill patients' social support deficits from offline settings [51,52]. However, Guillory and Niederdeppe [53] found that patients who already had sufficient social support from families and friends were also likely to seek online health information. McKinley and Wright [47] assert that although their inconsistent findings cannot fully support the compensation view, they demonstrate that online social supports are helpful for the end users. Accordingly, we propose our third hypothesis (H3): The perceived benefits of online sources for patients with chronic conditions are positively influenced by online emotional support (H3a) and informational support (H3b).

Critical Health Literacy in OHIS

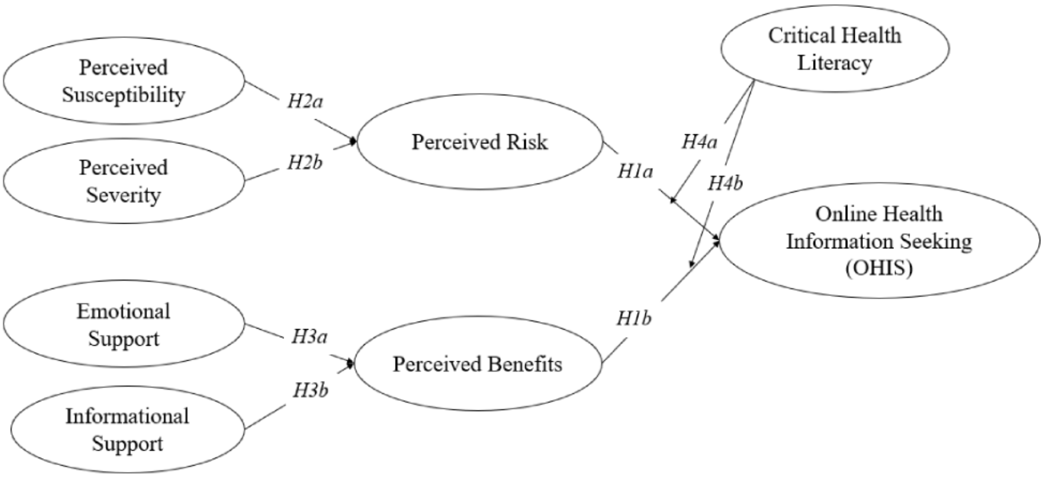
Health literacy refers to "the degree to which individuals can obtain, process, understand, and communicate about health-related information needed to make informed health decisions" [54]. According to Nutbeam [54], health literacy is a hierarchical concept consisting of multiple layers, depending on different levels of advancement of the literacy. While functional literacy refers to basic skills in reading and writing regarding health information, critical literacy refers to the advanced cognitive skills in analyzing health information critically.

Early studies treated health literacy as a holistic concept and found varied associations between health literacy and patients' health behaviors [55]. However, many recent studies revealed that the different components of health literacy have different power in explaining health behaviors. For example, Heijmans and Waverijn [56] found that critical health literacy is related to self-management, but functional health literacy is not. Matsuoka and Tsuchihashi-Makaya [57] revealed similar findings that critical health literacy influences self-care and consulting behaviors but functional health literacy does not. Based on these findings, we argue that critical health literacy may influence patients' information behaviors.

Moreover, prior studies suggested that patients with chronic conditions were concerned about the information quality, although they mostly agreed that online health information was easy to find [58]. These findings indicated that some patients might be knowledgeable about their health conditions [9] and thus are more critical when it comes to health information assessment. Therefore, we posit that the effects of the perceived risk and benefits of OHIS are moderated by critical health literacy. When patients have higher critical health literacy, they are more cautious when choosing online health information sources and may turn to authoritative sources such as offline health care providers. Thus, we propose the following hypotheses (H4): Critical health literacy negatively moderates the associations between perceived risk (H4a) and perceived benefits (H4b) and patients' OHIS.

The research model and hypotheses are shown in Figure 1.

Figure 1. Proposed research model.



Methods

Measurement Instrument

Most of the construct items in this study were adapted from validated existing scales. Each item was measured following a 5-point Likert scale ranging from 1 (strongly disagree) to 5 (strongly agree). The 3 OHIS items were adapted from studies by Deng and Liu [48] and Li and Wang [59]. The 3 items measuring perceived risk were developed from Kahlor [60]. The perceived benefits scales were adjusted from McKinley and Wright [47]. The perceived susceptibility and severity were gauged based on studies by Ahadzadeh et al [34] and Shang and Zhou [61]. Measurements of emotional and informational support were derived from studies by Deng and Liu [48] and Li and Wang [59]. Three items for critical health literacy drew on the measurement developed by Ishikawa and Takeuchi [62] and converted into an index. The constructs and measures are shown in Table 1.

The questionnaire was formed in 2 stages. First, we used translation (from English to Chinese) and back-translation (from Chinese to English) techniques to design the questionnaire to ensure its reliability. Second, we invited 20 patients living with chronic disease to participate in a pilot survey. We gathered their feedback and suggestions during the completion of the initial questionnaire to further modify the questionnaire, which resulted in the final version of the questionnaire.

Table 1. Constructs and measures.

Constructs	Measures	References
Online health information seeking	- OHIS[a]1: I want to seek health information often on the internet. - OHIS2: I am willing to search the internet for relevant health information when I need it. - OHIS3: I will seek health information on the internet before making health decisions.	Deng and Liu [48]; Li and Wang [59]
Perceived risk	- PCR[b]1: I am constantly worried about my health condition. - PCR2: I fear that my chronic condition would probably attack or worsen. - PCR3: If my chronic disease attacks or worsens, it would have a serious impact on my work or life.	Kahlor [60]
Perceived benefits	- PBF[c]1: Health information on the internet could be useful for me. - PBF2: Health information on the internet could be helpful to me. - PBF3: Health information on the internet could help me become familiar with health knowledge.	McKinley and Wright [47]
Perceived susceptibility	- PSU[d]1: The health-related issues mentioned in the internet health information are likely to happen on me. - PSU2: There is a good possibility that I will experience the health-related issues mentioned in the internet health information. - PSU3: I am likely to contract the health-related issues mentioned in the internet health information.	Ahadzadeh et al [34]; Shang and Zhou [61]
Perceived severity	- PSE[e]1: The consequences of the health-related issues mentioned in the internet health information may be serious for me. - PSE2: Contracting the health-related issues mentioned in the internet health information would be likely to cause me major problems. - PSE3: Suffering from the health-related issues mentioned in the internet health information is a serious problem for me.	Shang and Zhou [61]
Emotional support	- ES[f]1: When faced with difficulties, some individuals on the internet comforted and encouraged me. - ES2: When faced with difficulties, some individuals on the internet expressed interest in and concern for my well-being. - ES3: When faced with difficulties, some individuals on the internet are on side with me.	Deng and Liu [48]; Li and Wang [59]
Informational support	- IS[g]1: When faced with difficulties, some individuals on the internet would offer suggestions when I needed help. - IS2: When faced with difficulties, some individuals on the internet would give me information to help me overcome the problem. - IS3: When faced with difficulties, some individuals on the internet would help me discover the cause and provide me with suggestions.	Li and Wang [59]
Critical health literacy	- CHL[h]1: Since being diagnosed with chronic diseases, I have considered whether the information was applicable to my situation. - CHL2: Since being diagnosed with chronic diseases, I have considered the credibility of the information. - CHL3: Since being diagnosed with chronic disease, I have checked whether the information was valid and reliable.	Ishikawa and Takeuchi [62]

[a] OHIS: online health information seeking.

[b] PCR: perceived risk.

[c] PBF: perceived benefit.

[d] PSU: perceived susceptibility.

[e] PSE: perceived severity.

[f] ES: emotional support.

[g] IS: informational support.

[h] CHL: critical health literacy.

Ethics Approval

This study was approved by the Institutional Review Boards of the School of Economics and Management of the Nanjing University of Science and Technology (20201101).

Data Collection

The questionnaire was distributed from 2 main channels. First, we recruited participants through online chronic disease health communities. Five typical online health forums (ie, diabetes, hypertension, chronic gastritis, hyperlipoidemia, and rhinitis) were chosen in each of the leading Chinese communities (ie, Baidu Tieba and Douban groups). We also distributed the questionnaire through chronic disease health groups on general social media platforms (eg, WeChat). Eligible participants were consumers with at least 1 chronic condition who sought health information online during the past 12 months. The questionnaire contained a consent form that included the details of the study. Participants who agreed to the consent continued to the questionnaire. Each participant received a cash incentive of 5 renminbi (RMB) (about US $0.8) after completing the questionnaire. We received 426 questionnaires from October 18 to 29, 2021. After eliminating incomplete and invalid questionnaires by applying the eligibility criteria, we finally obtained a sample consisting of 390 valid responses.

Statistical Analysis

The respondents' characteristics are illustrated in Table 2. Of the participants, 64.1% (n=250) were male, and 35.9% (n=140) were female. The age coverage was relatively broad, comprising young people under the age of 20 and older adults above the age of 60 years. Respondents' places of residence were relatively balanced, with 46.7% (n=182) of participants living in urban areas and 53.3% (n=208) living in rural areas. Approximately half (n=192, 49.2%) of the participants had college degrees. In terms of health status, 38.5% (n=150) of the participants reported feeling normal, 25.6% (n=100) felt bad, and 35.9% (n=140) felt good or very good. Participants reported various types of chronic conditions. Chronic gastritis (n=146, 37.4%) was the most frequently mentioned condition, followed by diabetes (n=114, 29.2%) and hyperlipidemia (n=98, 25%). About half (n=193, 49.5%) of the participants had 1 chronic condition, 31.79% (n=124) had 2, and 4% (n=17) had 4 or more conditions.

We also measured the types of health information that participants sought using a typology from Zhao and Zhao [38]. Participants most frequently sought health information about disease symptoms (n=209, 53.6%), medical resources (n=201, 51.5%), and health prevention (n=199, 51%). Additionally, we counted the online health information sources that the participants used. Medical and health apps (n=187, 48%) were the most frequently reported online health information source, followed by social question-and-answer platforms (n=179, 46%) and short video platforms (n=174, 44.6%). Regarding OHIS frequency, all the participants reported they had sought online health information at least once during the past 6 months, and 39.5% (n=154) participants reported that they had sought online health information relatively often or very frequently.

Table 2. Characteristics of respondents.

Measure and item	Value, n (%)
Sex	
Male	250 (64.1)
Female	140 (35.9)
Age	
＜20	13 (3.33)
20-29	131 (33.6)
30-39	137 (35.1)
40-49	58 (14.9)
50＜59	35 (9)
≥60	16 (4.1)
Place of residence	
Urban	182 (46.7)
Rural	208 (53.3)
Education level	
Junior high school or below	58 (14.9)
Senior high school	98 (25.1)
Technical secondary school	42 (10.8)
Associate degree	72 (18.5)
Bachelor’s degree	103 (26.4)
Master’s degree	17 (4.4)
Monthly income (RMB[a])	
＜1500	17 (4.4)
1500-2999	55 (14.1)
3000-3999	112 (28.7)
4000-4999	68 (17.4)
5000-5999	71 (18.2)
6000-6999	29 (7.4)
≥7000	38 (9.7)
Profession	
Currently in health care profession	46 (11.8)
Past worked in health care profession	208 (53.3)
Never worked in health care profession	136 (34.9)
Health status	
Very bad	17 (4.4)
Relatively bad	83 (21.3)
Normal	150 (38.5)
Relatively good	100 (25.6)
Very good	40 (10.3)
Type of chronic disease	
Chronic gastritis	146 (37.4)
Diabetes	114 (29.2)
Hyperlipoidemia	98 (25.1)

Measure and item	Value, n (%)
Hypertension	76 (19.5)
Rhinitis	72 (18.5)
Rheumatism	62 (15.9)
Lumbar disc bulging	37 (9.5)
Asthma	33 (8.5)
Chronic conjunctivitis	33 (8.5)
Other	10 (2.6)
Number of chronic diseases	
1	193 (49.5)
2	124 (31.8)
3	56 (14.4)
4	11 (2.8)
>4	6 (1.5)
Type of health information	
Disease symptoms	209 (53.6)
Medical resource	201 (51.5)
Health prevention	199 (51)
Medication/treatment	111 (28.5)
Health promotion	94 (24.1)
Other	4 (1)
Source of health information	
Medical and health apps	187 (48)
Social question-and-answer platforms	179 (45.9)
Short video platforms	174 (44.6)
Social platforms	122 (31.3)
Search engines	111 (28.5)
News clients	56 (14.4)
Other	8 (2.1)
Frequency of searching	
Occasionally	83 (21.3)
Sometimes	153 (39.2)
Relatively often	127 (32.6)
Very frequently	27 (6.9)

[a] RMB: renminbi.

Results

Approach

We employed a partial least squares (PLS) approach to structural equation modeling (SEM) on testing the proposed model. Previous studies have shown that the PLS-SEM method is suitable for testing theoretically constructed models [63] and validating relatively complex models [64]. In addition, PLS-SEM can deal with nonnormally distributed samples, which is advantageous when processing relatively small sample sizes [65]. We used SmartPLS 3 software (SmartPLS GmbH) to analyze the data and test the structural model.

Measurement Model

Drawing on Shang and Zhou [61], we adopted reliability, convergent, and discriminant validity to evaluate the measurement model. Table 3 reports the reliability and convergence validity results. The reliability was judged based on the Cronbach alpha and composite reliability values. The results show that all Cronbach alpha and composite reliability values were greater than the proposed threshold of 0.7 [66], indicating qualified reliability. The convergence validity was examined by the values of average variance extracted (AVE). The results show that AVEs were higher than the recommended value of 0.5 [67], and all indicator loadings exceeded the threshold of 0.7, suggesting satisfactory convergence validity.

The discriminant validity was checked by testing both the Fornell-Larcker criteria [68] and the heterotrait-monotrait ratio (HTMT) [69]. Table 4 suggested that the square root of AVE values for each construct exceeded all its correlation coefficients with other constructs, indicating promising discriminant validity [68]. Moreover, all HTMT values were below the recommended value of 0.85 (Table 5), suggesting good discriminant validity [69]. The foregoing results verify the discriminant validity of all the constructs in our study.

Table 3. Reliability and convergence validity.

Constructs and items	Indicator loading	Cronbach alpha	Composite reliability	AVE[a]
Perceived susceptibility		.814	.890	.729
PSU[b]1	.881			
PSU2	.795			
PSU3	.883			
Perceived severity		.852	.910	.772
PSE[c]1	.888			
PSE2	.861			
PSE3	.886			
Informational support		.831	.898	.747
IS[d]1	.883			
IS2	.832			
IS3	.878			
Emotional support		.856	.913	.777
ES[e]1	.896			
ES2	.861			
ES3	.888			
Perceived risk		.835	.901	.752
PCR[f]1	.882			
PCR2	.834			
PCR3	.885			
Perceived benefits		.821	.894	.737
PBF[g]1	.867			
PBF2	.834			
PBF3	.874			
Online health information seeking		.824	.895	.740
OHIS[h]1	.881			
OHIS2	.823			
OHIS3	.874			

[a] AVE: average variance extracted.

[b] PSU: perceived susceptibility.

[c] PSE: perceived severity.

[d] IS: informational support.

[e] ES: emotional support.

[f] PCR: perceived risk.

[g] PBF: perceived benefit.

[h] OHIS: online health information seeking.

Table 4. Discriminant validity (Fornell-Larcker criterion)[a].

Constructs	1	2	3	4	5	6	7
1. Emotional support	.881	—	—	—	—	—	—
2. Online health information seeking	.571	.860	—	—	—	—	—
3. Informational support	.621	.526	.864	—	—	—	—
4. Perceived benefits	.684	.660	.676	.858	—	—	—
5. Perceived risk	.526	.578	.461	.585	.867	—	—
6. Perceived severity	.522	.576	.460	.582	.717	.879	—
7. Perceived susceptibility	.513	.529	.442	.488	.698	.629	.854

[a] Values on the diagonal represent the square root of average variance extracted (AVE) for each construct.

Table 5. Discriminant validity (heterotrait-monotrait ratio).

Items	1	2	3	4	5	6	7
1. Emotional support							
2.Online health information seeking	.677						
3. Informational support	.735	.633					
4. Perceived benefits	.816	.799	.815				
5. Perceived risk	.624	.692	.550	.707			
6. Perceived severity	.610	.687	.544	.694	.848		
7. Perceived susceptibility	.615	.643	.537	.598	.844	.754	

Structural Model

We adopted standard bootstrap in SmartPLS 3 on 5000 bootstrapping samples to examine the structural model's path coefficients and corresponding significance levels. Figure 2 shows the results of the PLS-SEM analysis, where perceived risk, perceived benefits, and online health seeking behavior are explained by the independent variables with variance values of 62.2%, 57%, and 61.5%, respectively, indicating a good explanation of the structural model.

The hypotheses testing results (Table 6) show that perceived risk (β=.188, *P*<.001) and perceived benefits (β=.222, *P*<.001) have significant positive effects on OHIS, supporting both H1a and H1b. As for health beliefs, perceived susceptibility (β=.408, *P*<.001) and perceived severity (β=.461, *P*<.001) significantly influence perceived risk, indicating that both H2a and H2b are supported. Concerning social support, both emotional support (β=.431, *P*<.001) and informational support (β=.408, *P*<.001) have positive effects on perceived risk, supporting H3a and H3b. Moreover, we tested the moderating effects of critical health literacy. The results show that critical health literacy (β=−.133, *P*=.002) has negative moderating effects on the relationship between perceived risk and OHIS, which supports H4a. However, critical health literacy cannot significantly moderate the relationship between perceived benefits and OHIS (β=−.012, *P*=.774). Therefore, H4b is not supported.

Figure 2. Structural model results. ns: nonsignificant. ****P*<.001, ***P*<.01, and **P*<.05.

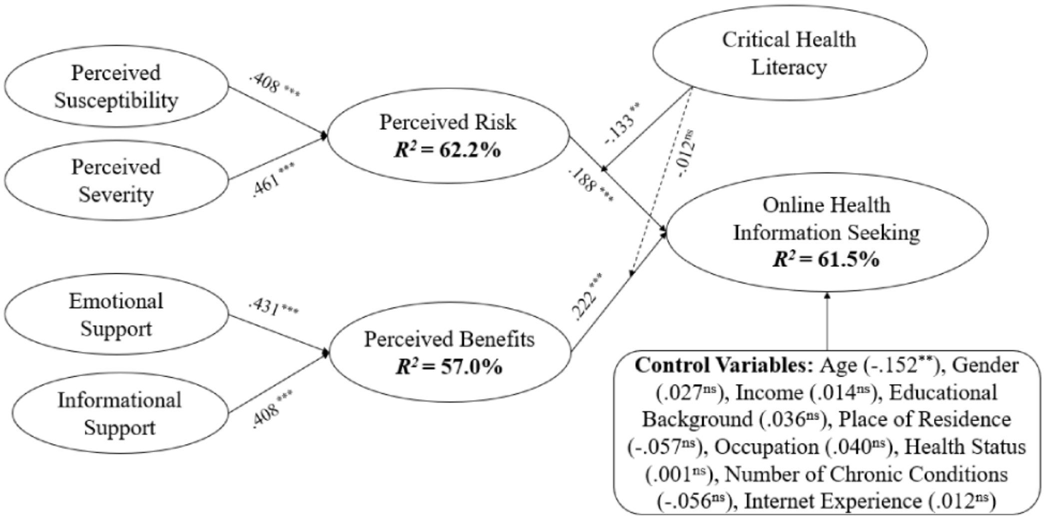


Table 6. Hypotheses testing results.

Hypotheses	Paths	Path coefficients	*t*-statistic	*P* value	Hypothesis validation
H1a	PCR -> OHIS	.188	3.989	<.001	Supported
H1b	PBF -> OHIS	.222	3.632	<.001	Supported
H2a	PSU -> PCR	.408	7.743	<.001	Supported
H2b	PSE -> PCR	.461	8.852	<.001	Supported
H3a	ES -> PBF	.431	5.748	<.001	Supported
H3b	IS -> PBF	.408	5.761	<.001	Supported
H4a	PCR×CHL -> OHIS	−.133	3.097	.002	Supported
H4b	PBF×CHL -> OHIS	−.012	0.288	.774	Not supported

Discussion

Principle Findings

In this study, we investigated the effects of perceived risk and perceived benefits on OHIS among patients with chronic conditions. Based on HBM, we examined the influencing factors of perceived risk using 2 antecedents: perceived susceptibility and perceived severity. Additionally, drawing on social support theory, we explored the impact of informational and emotional support on perceived benefits of patients' OHIS. This study also focused on critical health literacy and how it moderates the effects of perceived risk and perceived benefits on OHIS. We proposed a research model by integrating the aforementioned theories and developed corresponding measurement instruments. Data were collected from online chronic disease communities and social media groups using the survey method and analyzed using the PLS-SEM method.

The results suggested that perceived risk (t=3.989, P<.001) and perceived benefits (t=3.632, P<.001) significantly affected patients' OHIS. Perceived susceptibility (t=7.743, P<.001) and perceived severity (t=8.852, P<.001) were found to significantly influence the perceived risk of chronic diseases. Informational support (t=5.761, P<.001) and emotional support (t=5.748, P<.001) also impacted the perceived benefits of online sources for patients. In addition, moderation analysis showed that critical health literacy significantly moderates the relationship between perceived risk and OHIS (t=3.097, P=.002) but not the relationship between perceived benefits and OHIS (t=0.288, P=.774).

Implications

This study makes contributions to both theory and practice. From a theoretical perspective, we extend the HBM into information behavior research by integrating it with the social support theory. The HBM suggests that belief in health risk predicts the likelihood of engaging in health-related behaviors [37]. Prior work shows that individuals with higher perceived risk have a stronger motivation to perform health-related behaviors and change their health conditions [34,70]. Among them, patient-initiated OHIS can undoubtedly meet patients' health information needs and promote positive health information behaviors to a certain extent. In addition to patients' spontaneous health beliefs, this paper argues that social determinants of health can largely contribute to patients' health information behaviors—social support as an intermediary social determinant predicts patients' OHIS. We believe this assertion can simultaneously enrich the HBM and literature on health information behaviors. Our empirical study confirms the validity of this extension. Wilson [71] suggested that the disciplines of health and medical sciences and information sciences share a prominent common interest in information behavior research, and the flows of ideas and theories from the community of interest would also benefit information behavior research.

Additionally, we contextualize health literacy in chronic diseases by proposing and testing how critical health literacy moderates the relationship between health beliefs and social support to patients' OHIS. Prior work has explored the measurement of critical health literacy for patients with chronic diseases and the impact on self-management of health [56,72]. However, few studies have analyzed the impact of critical health literacy on OHIS. Our analysis contributes to the literature by uncovering a negative moderating effect between perceived risk and OHIS. We speculated that patients with higher critical health literacy may also be more capable in health information seeking and source selections. When patients with higher critical health literacy perceive a greater health risk, they may not necessarily search for health information on the internet and social media, given the general information quality concerns with online sources; instead, they are likely to seek more professional medical advice and visit doctors directly. This finding allows us to reexamine the compound influences of OHIS and seek more theoretical support from a psychological perspective.

From a practical perspective, this study suggests that online health communities should provide sufficient social support to patients and create a reciprocal virtual community. This social support can come from high-quality content created by professionals or emotional support generated by the mutual help between patient-patient and doctor-patient interactions. Meanwhile, online health communities should encourage surrogate health information seeking among patients and enhance the sense of belonging to the virtual community through gamification incentives and participatory design methods.

Finally, online health platforms need to better segment their users by providing targeted professional services to differentiated patients according to their varied health literacy levels instead of the traditional demographic profiles. Patients can become well informed about their health conditions and evolve into "expert patients." Expert patients with high health literacy usually have higher health information quality standards and prefer to go to offline professional medical institutions for consultation. Therefore, online health communities could consider inviting health care experts to carry out freemium consultations with more specialized, personalized, and accurate services to retain patients with higher critical health literacy and enhance their stickiness and loyalty to online health platforms.

Limitations and Future Work

This study has several limitations. First, the underlying influence mechanism between the 2 theories (ie, the health belief model and the social support theory) needs to be further empirically demonstrated. Future research could consider health beliefs as mediating constructs to unravel the effects of social determinants of health on individuals' perceived risks and benefits and further draw on social cognitive theory to empirically explore this mediating effect.

Second, we identified the moderating effect of critical health literacy in OHIS; however, the moderation analysis indicates that more contextualized measures are needed to validate the working mechanisms of critical health literacy. Future research needs to uncover how critical health literacy moderates the patients' OHIS intentions. Additionally, future research could further empirically analyze the constituent domains of critical health literacy [72] in terms of the dimensions of the constructs and how they are measured. Furthermore, researchers may also consider a randomized controlled trial to explore the effects of improved critical health literacy on OHIS.

Third, the generalizability may be limited as our sample is restricted to chronic disease patients in China. Our findings may not be applicable to other countries, regions, and contexts. Future work may conduct cross-cultural and cross-national comparisons to better generalize this study's results. Moreover, this is a cross-sectional study; due to the diversity of chronic diseases and the dynamic nature of chronic conditions, more longitudinal studies are needed in the future to reveal the dynamic effects of changes in health beliefs and social support on OHIS among patients with chronic diseases. Experience sampling methods and action research approaches are recommended to improve the validity of the research through multiwave data collection.

Conclusions

This paper contributes to the literature on OHIS by integrating the HBM and the social support theory. The integrated model suggested that health beliefs and social support positively impact OHIS among patients with chronic diseases. In particular, perceived susceptibility and severity can positively impact perceived risk, further influencing patients' OHIS. Informational support and emotional support can contribute to perceived benefits, further positively affecting patients' OHIS. This study also demonstrated critical health literacy's important negative moderating effects on the association between perceived risk and OHIS. Theoretical and practical implications for leveraging OHIS for patients with chronic diseases were also provided.

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Data Availability

The data sets generated and analyzed during this study are available from the corresponding author on reasonable request.

Conflicts of Interest

None declared.

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Abbreviations

CDC: Centers for Disease Control and Prevention
HBM: health belief model
HTMT: heterotrait-monotrait ratio
OHIS: online health information seeking
PLS: partial least square
RMB: renminbi
SEM: structural equation modeling

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Review

Ethical and Methodological Considerations of Twitter Data for Public Health Research: Systematic Review

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Abstract

Background: Much research is being carried out using publicly available Twitter data in the field of public health, but the types of research questions that these data are being used to answer and the extent to which these projects require ethical oversight are not clear.

Objective: This review describes the current state of public health research using Twitter data in terms of methods and research questions, geographic focus, and ethical considerations including obtaining informed consent from Twitter handlers.

Methods: We implemented a systematic review, following PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines, of articles published between January 2006 and October 31, 2019, using Twitter data in secondary analyses for public health research, which were found using standardized search criteria on SocINDEX, PsycINFO, and PubMed. Studies were excluded when using Twitter for primary data collection, such as for study recruitment or as part of a dissemination intervention.

Results: We identified 367 articles that met eligibility criteria. Infectious disease (n=80, 22%) and substance use (n=66, 18%) were the most common topics for these studies, and sentiment mining (n=227, 62%), surveillance (n=224, 61%), and thematic exploration (n=217, 59%) were the most common methodologies employed. Approximately one-third of articles had a global or worldwide geographic focus; another one-third focused on the United States. The majority (n=222, 60%) of articles used a native Twitter application programming interface, and a significant amount of the remainder (n=102, 28%) used a third-party application programming interface. Only one-third (n=119, 32%) of studies sought ethical approval from an institutional review board, while 17% of them (n=62) included identifying information on Twitter users or tweets and 36% of them (n=131) attempted to anonymize identifiers. Most studies (n=272, 79%) included a discussion on the validity of the measures and reliability of coding (70% for interreliability of human coding and 70% for computer algorithm checks), but less attention was paid to the sampling frame, and what underlying population the sample represented.

Conclusions: Twitter data may be useful in public health research, given its access to publicly available information. However, studies should exercise greater caution in considering the data sources, accession method, and external validity of the sampling frame. Further, an ethical framework is necessary to help guide future research in this area, especially when individual, identifiable Twitter users and tweets are shared and discussed.

Trial Registration: PROSPERO CRD42020148170; https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=148170

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KEYWORDS

systematic review; Twitter; social media; public health ethics; public health; ethics; ethical considerations; public health research; research topics; Twitter data; ethical framework; research ethics

Introduction

Since its launch in 2006, Twitter has become one of the most popular social media sites as a platform that allows users to post and interact with short messages known as tweets. According to a 2019 survey by Pew Research Center [1], 1 in 5 (23%) adults in the United States report using Twitter. While Twitter users are not representative of the general population (users tend to be younger, more educated, and located in urban or suburban areas) [2], the volume of publicly available tweets allows for research to be conducted on large data sets, eschewing a common perceived limitation of small samples.

Public health researchers have identified “big data” from Twitter as a new wellspring from which research can be conducted [3]. However, the utility of these data depends on the appropriateness of the research questions and the methodological approaches used in sampling and analyzing the data. Previous systematic reviews have explored how Twitter data have been used. A systematic review by Sinnenberg et al [4] of 137 articles using Twitter in health research between 2010 and 2015 found that the main research questions explored with Twitter data involved content analysis, surveillance, engagement, recruitment, intervention, and network analysis. Similarly, a scoping review from 2020 [5] found 92 articles that fell within 6 domains: surveillance, event detection, pharmacovigilance, forecasting, disease tracking, and geographic identification. Additional systematic reviews of social media, beyond Twitter alone, have examined specific domains, for instance, exploring how these data, including Twitter, are being used for public health surveillance [6-8] or pharmacovigilance [9-11].

While social media provides new opportunities for data sources in research, some unique obstacles are also present. For instance, the presence of spam and noisy data can make it difficult for researchers to identify a legitimate signal for the research topic in question [12]. To navigate this issue, researchers sometimes opt to employ traditional manual coding of content; however, this can be a nonideal solution given the size of the data sets and the time and effort required for these analyses [13]. Other teams have used natural language processing (NLP) or machine learning approaches, which present their own problems; one study [14] found that among the algorithms built to classify emotions, the highest performing model had an accuracy of 65%. The landscape of social media necessitates understanding of the mechanisms and limitations of the platforms, as well as adaptations to the requirements of this landscape.

In addition to the research questions and methodological approaches used with Twitter data, the extent to which social media data are in general considered public, and what this means for ethical research oversight are unclear. There is substantial literature discussing the ethics of using social media data for public health research, but clear ethical guidelines have not been established [15-24].

The need for these guidelines is increasingly pressing, as leveraging social media for public health research raises questions about privacy and anonymity; properly deidentifying user data requires the researchers to understand an “increasingly networked, pervasive, and ultimately searchable dataverse” [18]. Information shared on social media can often be intensely personal; hence, anonymity would be even more important for research involving sensitive data such as health conditions and disease [23]. This is particularly relevant for the field of public health, since the data collected and analyzed for public health research will often fall into these more sensitive categories.

Beyond the questions of user anonymity, when conducting research on more sensitive health information, traditional research protocols center the importance of informed consent among participants. However, there are currently no established guidelines for the expectation of consent when leveraging publicly available social media data. Some theorists in the realm of internet research ethics have proposed an assessment model that determines the need for consent based on possibility of pain or discomfort. They further suggest that this assessment should consider the vulnerability of the population being studied and the sensitivity of the topics [22].

In the systematic review by Sinnenberg et al [4], approximately one-third of the 137 articles included therein mentioned ethical board approval. Given that Twitter usage has changed dramatically in recent years [25], this systematic review is an updated examination of both ethical considerations and research questions or methodologies across all domains of public health research using Twitter.

We sought to investigate the methodological and ethical aspects of using Twitter data for public health research from 2006, when Twitter was launched, to 2019 [26]. Specifically, we describe the measures being used in Twitter research, the extent to which they are validated and reliable, and the extent to which ethical oversight is included in studies using publicly available tweets.

Methods

Design

This review followed the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines [27,28] and was registered with PROSPERO (CRD42020148170).

Eligibility Criteria

The database search was limited to peer-reviewed public health studies originally written in English, which were published between January 2006 and October 31, 2019, and used social media data to explore a public health research question. The social media platforms included in the search were Twitter and Sina Weibo (China’s version of Twitter), Facebook, Instagram, YouTube, Tumblr, or Reddit.

Studies were excluded if they were systematic or literature reviews, marketing or sales research, only investigated organizational-level tweets, investigated tweets from conferences in disciplines other than public health, or included primary data collection asking participants about their social media use. We excluded articles that focused on organizations disseminating information to the public (evaluation of social media dissemination and analysis of organizational- or institutional-level social media data) or testing interventions that used social media as a method (intervention study using social media), as our research question was not related to interventions using social media platforms as a tool but rather explored how existing social media data are being used in secondary analyses in public health research.

Given the volume of studies identified, separate analyses were conducted on Facebook and YouTube; thus, this systematic review focuses solely on Twitter. Studies that included Twitter and other social media platforms were included, but only Twitter findings were extracted.

Information Sources

We searched PubMed, SocINDEX, and PsycINFO for articles about social media and public health after consulting with our institutional librarian on the best approaches to the search.

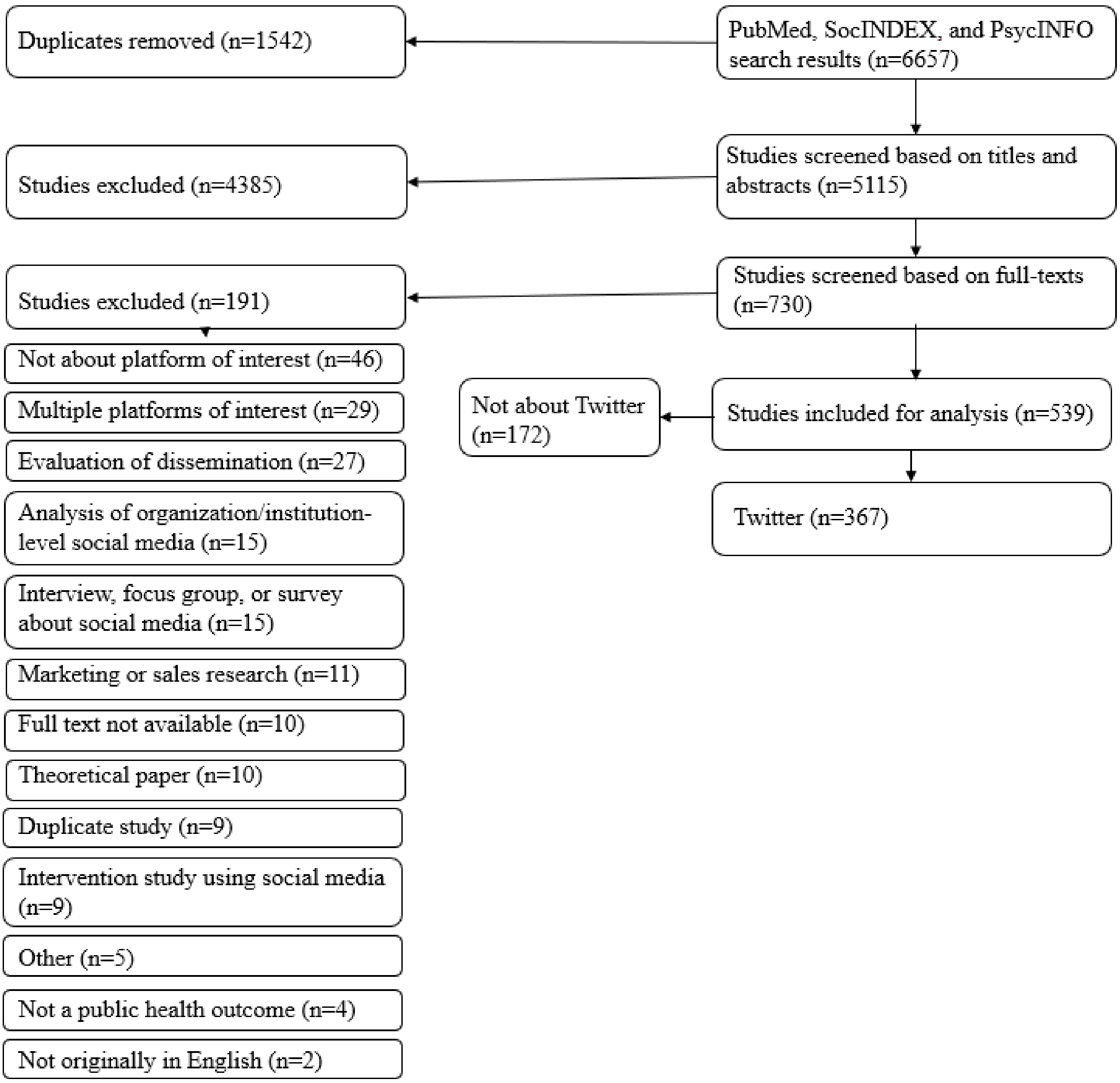
Search

The search strategy consisted of the Boolean search term: ((“Social media” OR twitter OR tweet* OR facebook OR instagram OR youtube OR tumblr OR reddit OR “web 2.0” OR “public comments” OR hashtag*) AND (“public health” OR “health research” OR “community health” OR “population health”)).

Study Selection

Three authors reviewed abstracts for eligibility in a 2-step process, with each abstract reviewed by 2 authors independently. A first screen was performed on the basis of the title and abstract; if deemed ineligible, the study was excluded from further screening. Disagreements were resolved through discussion and consensus. Full texts of the remaining articles were retrieved for the second screen and reasons for exclusion were coded and ranked by the priority of exclusion criteria for cases in which more than one exclusion criterion was applied (Figure 1). Disagreements about inclusion and exclusion criteria were resolved through discussion and consensus.

Figure 1. PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flowchart for systematic review of methodological approaches and ethical considerations for public health research using Twitter data, 2006-2019.



Data Collection Process

Data were extracted using a standardized data extraction spreadsheet, which was developed a priori and refined during the data extraction process. This refinement resulted in the removal of data elements; new data elements were not added. To establish consistency in extractions, 2 reviewers independently extracted data from the same 5 articles and compared the results. This process continued during weekly meetings, in which papers of varying complexity were discussed until consensus was reached. No studies were excluded on the basis of their quality.

Data Items

The data items in this review categorized information about the study within 4 domains: (1) study characteristics: public health topic, year, and country of publication; (2) study design and results: sample size, Twitter data extraction method, operationalization (ie, which data points were collected from social media posts and how researchers quantified these data), methodologic and analytic approaches, primary results, and descriptions of linking or account data; (3) ethical considerations: ethical approval, discussion of informed consent, and general discussion of ethical issues; and (4) risk of bias or methodological checks: quality assessment, validity, reliability, and accuracy checks implemented. We defined methodological approach as the overall objective of a research project coupled with the operationalization of methods to fulfill this objective.

Quality assessment metrics were adapted from existing quality assessment tools used for systematic reviews [29-31]. The specific quality assessment metrics were the following: whether the stated research question matches the data-defined research question, the presence of a clearly defined objective or hypothesis, validity of measures, reliability of measures, validation of computer algorithms, whether the data analysis is sufficiently grounded, whether findings logically flow from the analysis and address the research questions, and the presence of a clear description of limitations. A study was considered to have addressed validity if the measures used were based on validated measures, previous studies, or existing frameworks. A study addressed reliability if manual coding efforts incorporated checks or assessed intercoder reliability, descriptions of reliability were not expected for studies that only used machine learning. Accuracy checks were described if manual checks were performed by researchers or validation of computer algorithms used for studies using machine learning algorithms and NLP.

Summary Measures

The summary measures related to methods and study design include the following: the frequency of studies by topic, geographic focus, year of publication, analytic approach, sampling approach, and overall methodological approach or objective of the study (ie, surveillance, content exploration, sentiment mining, network science, and model development and testing). The summary measures related to ethical considerations include the frequency of studies that sought institutional review board (IRB) review or approval, included informed consent from Twitter handlers, discussed ethical considerations within the paper, and reported identifying results (ie, verbatim tweets). For quality assessment, we present information on the validity and reliability of measures used; a full summary of quality assessments is provided in Multimedia Appendix 1.

Results

Our search resulted in 6657 unique studies for review, of which 730 required full-text review (Figure 1). We identified 539 studies across all social media platforms; 367 used Twitter data forming the analytic sample for this review (Multimedia Appendix 2 for the full list of included articles with all data extraction fields; for readability of text, references are only included when details of specific articles are provided as contextual examples).

Study Characteristics

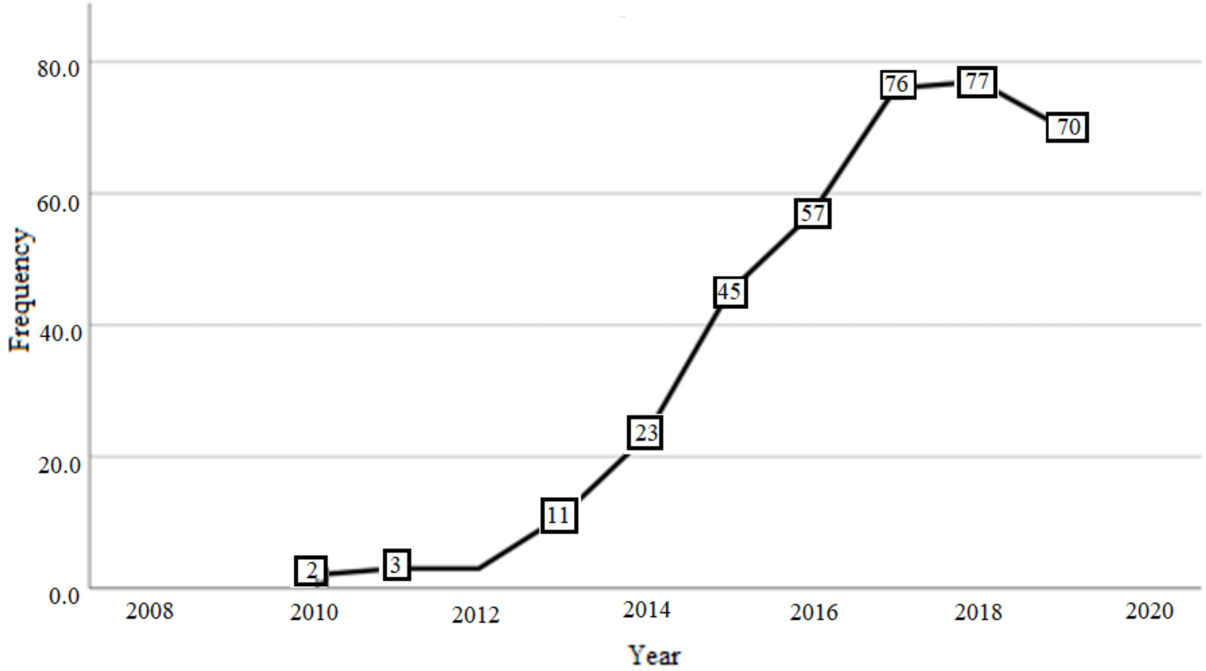
Public Health Research Topics

The most common public health topics among the articles reviewed were communicable diseases (eg, influenza, Ebola, and Zika; n=80, 22%), substance use (n=66, 18%), health promotion (n=63, 17%), chronic disease (eg, cancer; n=48, 13%), and environmental health (n=48, 13%; Multimedia Appendix 1).

Year of Publication

The year of publication for the articles in this review ranged from 2010 to 2019. A sharp increase in the number of Twitter articles was observed from 2012 to 2017 (Figure 2). Two preprint articles on October 31, 2019, were included in the count for 2019 [32,33].

Figure 2. Number of articles published by year for systematic review of methodological approaches and ethical considerations for public health research using Twitter data, 2006-2019.



Geographic Focus

Most studies analyzed tweets originating from the United States (n=158, 43%) or worldwide (n=134, 36%); only 75 (20%) of them focused on non-US regions or countries. Of the articles that had a global geographic focus, 23 (17%) of them collected geotags and reported on geospatial metrics within the body of the article. Despite having a worldwide focus, these 23 articles demonstrated a bias toward the United States, western Europe (namely the United Kingdom), Canada, and Australia; the majority of the data collected in these studies were posts originating in these countries, with a distinct minority representing other regions or countries.

Study Design and Results

Sample Size and Unit of Analysis

Of the 367 articles reviewed here, 355 (97%) used individual tweets as the unit of analysis and 11 (3%) used Twitter accounts (or "handles") as the unit of analysis. One article (0.3%) used keywords as the unit of analysis, as the study sought to identify keywords that would help researchers detect influenza epidemics via Twitter [34].

There was a wide range of sample sizes. For studies with tweets as the unit of analysis (n=353), the number of analyzed tweets ranged from 82 [35] to 2.77 billion [36] (median=74,000), with 90 papers having a sample size larger than 1 million. Similarly, for studies using Twitter handles as the unit of analysis (n=11), the sample size ranged from 18 [37] to 217,623 [32].

Methods for Accessing Data

To pull data from Twitter, most studies used application programming interfaces (APIs) that were developed by Twitter (eg, Gardenhose and Firehose) and could be integrated into statistical software packages. Third-party APIs (eg, Twitonomy and Radian6) were also used frequently, either through contracting with a commercial vendor, purchasing tweets that match specified criteria, or using software developed by an entity outside of Twitter. Most studies either mentioned that they used an API without indicating the specific type (37%) or did not mention their method of tweet accession (13%; Table 1). Of papers that identified the API used, purposive and random sampling were equally employed. However, only 22 (7%) articles explicitly mentioned whether the API used was purposive or random in its sampling technique; when the API was named (eg, decahose, search API, and Gardenhose) but the sampling type was not noted in the article, we looked up the sampling technique in use by the API.

We also found that the description of the sampling method was often not described. For instance, some Twitter APIs are purposive in nature (eg, Twitter Search API) and some are random (Twitter Firehose API) or systematic (some REST APIs). Many studies did not specify what type of sampling was used to extract tweets from Twitter or did not fully explain retrieval limitations (eg, how it might affect the sample population if only a certain number of tweets could be retrieved daily through an API).

Table 1. Frequency of studies by access method and data source from a systematic review of methodological approaches and ethical considerations for public health research using Twitter data, 2006-2019.

Method or source for Twitter data	Frequency (N=367), n (%)
Access method	
Unspecified application programming interface (API)	136 (37)
Purposive sampling[a]	88 (24)
Random sampling[a]	84 (23)
Existing database	10 (3)
Unspecified method of accession	49 (13)
Data source	
Native Twitter API/functionality	222 (60)
Third-party API	102 (28)
Unknown	34 (9)
In-house program	9 (3)

[a] Accession methods and sampling type are differentiated as random or purposive in accordance with reports from the articles' authors or Twitter.

Methodological Approach

As seen in Table 2, the most common methodological approaches were as follows: thematic exploration (eg, describing the themes of conversations about e-cigarettes on Twitter) [38], sentiment mining (eg, assessing if tweets about vaccines are positive, negative, or neutral) [39], and surveillance (eg, tracking the patterns of information spread about an Ebola outbreak) [40]. Less common methodological approaches were tool evaluation (eg, using Twitter data to predict population health indices) [41] and network science (eg, examining health information flows) [42]. Different methodological approaches tended to be pursued for different topics. For example, most infectious disease research was in the domain of surveillance, whereas research about mental health and experiences with the health care system was more conducive to thematic exploration and sentiment mining.

Across the 3 most common study methodological approaches (thematic exploration, sentiment mining, and surveillance), approximately one-third of the papers (36%) used machine learning (Table 2). Machine learning here is defined as an application of algorithms and statistical modeling to reveal patterns and relationships in data without explicit instruction (eg, to identify the patterns of dissemination related to Zika virus–related information on Twitter) [43]. This can be contrasted to NLP, which necessitates explicit instruction; often, NLP is used to identify and classify words or phrases from a predefined list in large data sets (eg, to identify the most common key topics used by Twitter users regarding the opioid epidemic) [44]. Of the articles reviewed, NLP was more prevalent in sentiment mining than in other types of methodological approaches.

Table 2. Frequency of studies by methodological approach and analytical technique from a systematic review of methodological approaches and ethical considerations for public health research using Twitter data, 2006-2019.

Methodological approach and analytical technique[a]	Frequency (N=367), n (%)
Sentiment mining	227 (62)
Natural language processing	145 (64)
Machine learning	66 (29)
Spatial analysis	12 (5)
Descriptive analyses or frequencies	4 (2)
Surveillance	224 (61)
Natural language processing	104 (46)
Machine learning	85 (38)
Spatial analysis	17 (8)
Descriptive analyses or frequencies	18 (8)
Thematic exploration	217 (59)
Natural language processing	114 (52)
Machine learning	81 (37)
Spatial analysis	13 (6)
Descriptive analyses or frequencies	9 (4)
Tool evaluation	61 (16)
Network science	36 (10)

[a] Multiple responses were allowed.

Ethical Considerations

Presence of Identifying Information

Just under half (n=174, 47%) of the articles reviewed did not contain any identifying information of Twitter accounts or tweets, 36% (n=131) of them contained anonymized account information or paraphrased tweets, and 17% (n=62) of them contained direct quotes of tweets or identifiable information such as Twitter handles or account names (Table 3). Of the 62 articles that included verbatim tweets or identifying information about the user, one-third (n=21, 34%) of them included a discussion of ethics in the paper (eg, Berry et al [45]).

Less than half of the articles (n=173, 47%) indicated that they did not use any of the metadata (eg, username, demographics, and geolocation) associated with the tweet (Multimedia Appendix 1). Approximately one-third of the articles (n=110, 30%) used geographic information associated with the tweet, and a much smaller number of articles (n=15, 4%) included photos associated with the account or health information (such as illness disclosure or mentions of medications taken). Of the articles analyzing tweets from either the United States or another specific region or country (n=233), 37% (n=86) of them used geotags of Twitter accounts to identify the location of the tweets; of the articles that did not specify a geographic region (n=134), 17% (n=23) of them used geotagging.

Though research on infectious disease and health promotion were most likely to include user metadata in their data analyses, linked health information was most often used in papers about infectious disease and mental health, often in the form of medical self-disclosures.

IRB Approval and Informed Consent

Just under one-third of the articles reviewed (n=119; 32%) explicitly stated that those studies sought and received IRB review or approval (Table 3). The majority (n=226, 61%) of them did not mention IRB approval, although many of these articles included statements about the nature of Twitter posts being publicly available. Only a small subset (n=23, 6%) of studies explicitly stated that IRB approval was not necessary.

Among those that sought IRB approval (n=119), over half (n=68, 57%) of them were granted exemptions; just under half (n=49, 41%) of them did not specify the type of approval received. Two studies [46,47] received full IRB approval. One of them [46] retrospectively examined existing public data about health beliefs regarding the human papillomavirus and was approved with a waiver of consent owing to its retrospective design. The other study [47] had 2 parts: study 1 consisted of a survey of self-reported stress following a school lockdown, and study 2 consisted of data mining of community-level rumor generation during the lockdown on Twitter. The survey necessitated informed consent as it involved human participants; hence, the full scope of the study (parts 1 and 2) had to undergo IRB review. None of the studies using only Twitter data sought informed consent, even when including identifying information from Twitter handlers or tweets. Over two-thirds of the articles (n=258, 70%) did not include a discussion of ethics or privacy concerns.

Additionally, 53 (49%) articles discussed the anonymization of data used in their study either by omitting usernames and Twitter handles [48] or by providing only paraphrased tweets to prevent exact-match searching [49]. Only 5 studies included specific and extensive discussions around the ethical implications of social media research and went beyond disclaimer statements about the publicly available nature of tweets. One study [50] described consulting guidelines for internet research from various organizations and researchers, while another [51] included a long "ethical considerations" section that described needing to "weigh threats to safety and privacy against benefits gained by using novel approaches to study suicide," and acknowledged vulnerable populations and risks of stigma and discrimination. Another study [52] raised the challenge of social media research given the lack of relevant ethical frameworks.

Table 3. Frequency of studies by ethics-related factors from a systematic review of methodological approaches and ethical considerations for public health research using Twitter data, 2006-2019.

Ethics-related factors	Frequency (N=367), n (%)
Level of identification	
No identifying information	174 (47)
Anonymized data and paraphrased tweets	131 (36)
Identifiable information and direct quotes	62 (17)
Institutional review board (IRB) approval obtained	
Yes	119 (32)
No	23 (6)
Not mentioned/unclear	225 (61)
Among those with IRB approval (n=119)	
Exempt	68 (57)
Nonexempt	2 (2)
Not specified (eg, "approved")	49 (41)
Informed consent of Twitter handler attempted	
Yes	0 (0)
No	119 (100)
Any discussion of ethical considerations, including disclaimers	
Yes[a]	109 (30)
Discussion of anonymization process	53 (49)
Extensive discussion[b]	5 (5)
Other discussion, including disclaimers	54 (49)
No	258 (70)

[a] Note that 3 articles included both an extensive discussion of ethics as well as details regarding their anonymization process.

[b] The denominator for the articles that discussed ethics is 109.

Risk of Bias in Individual Studies

We found that 270 (74%) articles included a clear description of the validity of measures; 21 (6%) articles were purely exploratory in nature and collected only counts of tweets, so we deemed them exempt from an assessment of validity of measures; 76 (21%) articles did not include efforts at establishing measurement validity. Further, of the 264 articles involving human coding, 184 (70%) included a description of intercoder reliability and quality assurance checks, while 80 (30%) did not. Similarly, 235 articles involved computer algorithms or automated coding, of which 165 (70%) explicitly described accuracy checks or validation of the algorithms, while 70 (39%) did not.

In addition to concerns about validity and reliability of measures, one of the main sources of bias was the sampling frame. The self-selection of Twitter users was discussed in most of the studies, with 85% (n=314) of them describing this as a potential limitation.

Discussion

Principal Findings

Summary Measures

We saw evidence of a steep increase in publications using Twitter data after 2012, which may be due to Twitter releasing its native standard (version 1.1) API in 2012, which made mining of its data much more accessible to the general public without the need for complex coding capabilities [53]. The prevalence of research using "big data" from Twitter is increasing and will likely continue to do so in the coming years [50].

Infectious disease was the most common topic of the research papers, which may indicate a burgeoning interest in using social media to detect disease outbreaks. It is likely that a review of studies using Twitter data that picks up from where this study left off (ie, after October 31, 2019) would support this finding given the onset of the COVID-19 pandemic in late 2019.

There are some major considerations that this review highlights for the future of public health research using Twitter data. Most of the research focused on Twitter users in the United States; this includes the articles with a global focus that demonstrated a bias toward the anglophone world. Three articles appeared to genuinely have a representative global scope; interestingly, two of these were about the Zika virus. This indicates the data scraped from Twitter tends to be heavily focused on the United States and English-speaking settings.

Another major consideration is that of the accession method used to build a data set. Most of the studies examined in this review used APIs or variations thereof; only 10 studies used alternative accession methods. Those 10 studies used data either extracted from Twitter for previous studies or hosted in pre-existing databases. Of the remaining studies that used an API, only 22 studies explained whether the API used was purposive or random in nature. This is of interest because the sampling technique of APIs has been called into question in previous papers [54,55]. In particular, the Twitter Streaming API is considered to produce less representative samples and should be approached with caution; this API is susceptible to intentional or accidental bias based on inclusion and exclusion criteria selected for a particular study [56]. Owing to the "black box" nature (ie, lack of documentation of the sampling approach) of native Twitter APIs, it cannot be determined that data retrieved using Twitter APIs are truly random [57,58].

In addition to the aforementioned obstacles, there are questions about the accuracy of algorithms using machine learning and NLP. A little less than half of the papers reviewed for this systematic review involved surveillance and prediction, and approximately one-sixth of them evaluated new tools or frameworks in the realm of Twitter data. Machine learning was commonly used for these methodological approaches. However, a previous evaluation of the efficacy of using various machine learning algorithms to automatically identify emotions expressed on Twitter found that the highest performing algorithm achieved an accuracy rate of 65% [14]. Another recent article found that machine learning was not effective in making meaningful predictions about users' mental health from language use on social media; further, Twitter metadata and language use was not specific to any one mental health condition [59].

This raises concerns about the overall use of social media data for research, as data science in general and public health research in particular use data to make insights; these data "then get acted upon and the decisions impact people's lives" [20]. Hence, conscientious planning is advised when using publicly available social media data for the purpose of public health research.

Discussion of Ethics

Given that slightly over one-third of studies anonymized Tweets or Twitter users, many researchers seem to think that there are ethical considerations when using these data, even if they are publicly available. Nevertheless, the majority of projects did not seek IRB review or approval. This contradiction suggests an implicit understanding that while there are no international or place-specific ethical guidelines around research using social media data, there is something unique about the nature of this research that distinguishes it from truly public data.

International ethical standards for biomedical and public health research already exist, and these standards often continue to influence the national guidelines that develop within a given country [60-62]. Given the global scope of social media, it may be most prudent for guidelines to be established on an international scale and then adapted to place-specific committees and ethics boards. However, this is complicated by the ever-evolving landscape of social media use and data agreements. The field of research ethics has yet to fully address the introduction of new media as sources of data; even before a comprehensive international framework is introduced, it may be advisable for institutions and regions to enact their own interim frameworks to mitigate possible harm and preserve user privacy and anonymity to the extent possible.

Limitations

This systematic review has a number of limitations. Owing to the iterative nature of data extraction for a large number of articles included, it is possible that there were differences in how data were coded as we refined our process. However, we attempted to minimize this concern through weekly research team meetings during the extraction process. Another limitation is that because we only examined articles originally published in English, we may be underestimating the number of articles that were conducting research in a specific geographic area other than the United States. The influence of this underestimation should be minimal; however, as most leading journals for health research are published in English [63]. One final limitation is that the literature review spanned from 2010 to 2019, so we are not capturing changes since then, which may have taken place in the approach to ethics or methodology in research using social media data since then. This is an evolving field of research; hence, we anticipate that standards and norms may have also evolved.

Comparison With Prior Work

Similar to Sinnenberg et al's [4] review, this study examined whether ethics board approvals were sought when using social media data for public health research, finding equivalent proportions of articles that obtained IRB approval. Our study further explored whether there were other types of ethical considerations (eg, ethical discussion) present in the body of the articles. We also assessed the presence and use of identifiable information such as personal health information, verbatim Tweets, and user account metadata. In both this review and in that of Sinnenberg et al [4], many articles noted that the public nature of tweets allows researchers to observe the content. This presents a clear need for an ethical guideline framework for researchers using Twitter, especially when including identifying information.

Conclusions

Twitter data appear to be an increasingly important source of data in public health research. However, attention needs to be paid to sampling constraints, ethical considerations involved in using these data, and the specific methodologies to be used to ensure the rigorous conduct of this research.

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Supplementary tables.
[DOCX File , 16 KB-Multimedia Appendix 1]

Multimedia Appendix 2

Full data extraction sheet.
[XLSX File (Microsoft Excel File), 165 KB-Multimedia Appendix 2]

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Abbreviations

API: application programming interface
IRB: institutional review board
NLP: natural language processing
PRISMA: Preferred Reporting Items for Systematic Reviews and Meta-Analyses

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Original Paper

Factors Associated With Telemedicine Use Among German General Practitioners and Rheumatologists: Secondary Analysis of Data From a Nationwide Survey

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Abstract

Background: Previous studies have demonstrated telemedicine (TM) to be an effective tool to complement rheumatology care and address workforce shortage. With the outbreak of the SARS-CoV-2 pandemic, TM experienced a massive upswing. However, in rheumatology care, the use of TM stagnated again shortly thereafter. Consequently, the factors associated with physicians' willingness to use TM (TM willingness) and actual use of TM (TM use) need to be thoroughly investigated.

Objective: This study aimed to identify the factors that determine TM use and TM willingness among German general practitioners and rheumatologists.

Methods: We conducted a secondary analysis of data from a German nationwide cross-sectional survey with general practitioners and rheumatologists. Bayesian univariate and multivariate logistic regression analyses were applied to the data to determine which factors were associated with TM use and TM willingness. The predictor variables (covariates) that were studied individually included sociodemographic factors (eg, age and sex), work characteristics (eg, practice location and medical specialty), and self-assessed knowledge of TM. All the variables positively and negatively associated with TM use and TM willingness in the univariate analysis were then considered for Bayesian model averaging analysis after a selection based on the variance inflation factor (≤2.5). All analyses were stratified by sex.

Results: Univariate analysis revealed that out of 83 variables, 36 (43%) and 34 (41%) variables were positively or negatively associated (region of practical equivalence≤5%) with TM use and TM willingness, respectively. The Bayesian model averaging analysis allowed us to identify 13 and 17 factors of TM use and TM willingness, respectively. Among these factors, being female, having very poor knowledge of TM, treating <500 patients per quarter, and not being willing to use TM were negatively associated with TM use, whereas having good knowledge of TM and treating >1000 patients per quarter were positively associated with TM use. In addition, being aged 51 to 60 years, thinking that TM is not important for current and future work, and not currently using TM were negatively associated with TM willingness, whereas owning a smart device and working in an urban area were positively associated with TM willingness.

Conclusions: The results point to the close connection between health care professionals’ knowledge of TM and actual TM use. These results lend support to the integration of digital competencies into medical education as well as hands-on training for health care professionals. Incentive programs for physicians aged >50 years and practicing in rural areas could further encourage TM willingness.

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KEYWORDS

telemedicine; rheumatology; primary care; secondary analysis; health services research

Introduction

Telemedicine (TM) offers the opportunity to overcome spatial distances in health care delivery [1]. Thus, TM represents a promising way to support rheumatology care [2,3] in light of the rising worldwide burden of musculoskeletal diseases [4] and growing workforce shortage [5,6]. However, the effective implementation of TM in standard care is only possible if the end users are willing and able to use TM [7,8].

With the outbreak of the SARS-CoV-2 pandemic, physicians’ face-to-face consultations declined considerably [9,10]. The possibility of contactless medical care is now more important. Advantageously, through TM, medical care could be provided, avoiding contacts and thus infections [11,12]. Hence, TM has received a tremendous upswing worldwide [13] and regionally [9,14]. Although the pandemic situation, involving social distancing and multiple lockdowns, provided an ideal environment for the implementation of TM, this momentum soon stagnated again [10,15]. Particularly in rheumatology, health care professionals’ use and acceptance of TM fell short of expectations [10]. Apparently, other factors may play a role in the willingness to use TM (TM willingness) and actual use of TM (TM use) among general practitioners (GPs) and rheumatologists. Identifying these factors is a rather challenging task but could have implications for the development of TM strategies aiming to improve health outcomes and access to care and make health care delivery systems more efficient and cost-effective.

To gain a better understanding of these factors, we performed a secondary analysis using data from a nationwide cross-sectional survey conducted earlier in Germany [7]. Our objective was to identify the underlying factors associated with TM use and the TM willingness among German GPs and rheumatologists.

Methods

Overview

This work reports on findings from a secondary analysis of data collected as part of a cross-sectional, self-completed, and paper-based survey of German GPs and outpatient rheumatologists. The initial study was conducted from September to November 2018 and investigated the acceptance, opportunities, and obstacles to the implementation of TM. Of the 2395 questionnaires that were sent out, 497 (20.75%) were returned. Of the 497 responses, 12 (2.4%) were excluded from the data set because fewer than half of the questions were answered. The final response rates were 18.94% (437/2307) and 55% (48/88) for GPs and rheumatologists, respectively. The exact methodology applied for the nationwide survey has been described previously [7].

Regression Analysis

Both Bayesian univariate and multivariate logistic regression analyses were applied to the data to determine which factors were associated with TM use (question [Q]3) and TM willingness (Q4A), respectively. In total, 22 independent variables were considered for each univariate regression analysis (Multimedia Appendix 1). The individuals who missed providing information on age or gender or answers to Q3 (467/492, 5.1%) and Q4A (454/492, 7.7%) were excluded. Otherwise, missing values (no answer) were considered as a new category for the univariate regression analysis. For instance, Q28, “assigning physician or rheumatologist,” previously had 2 categories and was revised to have 3 categories, “assigning physician,” “rheumatologist,” or “not answered”. For statistical analysis, all the categorical variables having >2 modalities, for example, “yes,” “no,” or “do not know,” were transformed into dummy or binary variables. For instance, Q21 was transformed into 3 dummy variables.

For each model, odds ratios (ORs) with 95% credible interval (CI) are presented. All the individual variables associated (positively or negatively) with TM use and TM willingness in the Bayesian univariate analysis were considered for analysis in the later Bayesian multivariate analysis (model selection) after variable selection. This variable selection was based on the region of practical equivalence (ROPE) percentage (ROPE%≤5) [16] and a subsequent selection based first on the variance inflation factor (VIF) [17]. Collinear covariates, with a VIF>2.5, were excluded in the multivariate models [18]. Finally, the determinants of TM use and TM willingness were identified through Bayesian model averaging (BMA) [19]. The “best” model (ie, model with the highest posterior probability) from BMA was detailed. All models were stratified by sex. In addition, determinant factors (question answers), defined as variables with a posterior probability of ≥10% with BMA, were identified and used to establish the profile of the individuals using or willing to use TM and the profile of the individuals not using or not willing to use TM using spider charts. For each determinant factor, the percentage of individuals who chose a specific answer was displayed on the spider chart. This percentage could range from 0 (the inner circular line, the closest to the center) if no individuals chose the specified answer for the considered question to 100 (the outer circular line, the farthest from the radar center) if all individuals answered the question with the specified answer. Green points and lines on the spider charts refer to the individuals who use or want to use

TM, whereas red points and lines correspond to the individuals not willing to use or not using TM. For each question, there were 3 possible situations. When the green and red points overlapped (were similar), it meant that there was no difference between the individuals whether they were using TM or not or willing or not to use TM, that is, the proportion of similar answers was high. When the green point was higher (higher percentage) than the red point, it indicated that the individuals using or willing to use TM chose the specified answer more often than those not willing to use or not using TM, which meant that this factor (question) had a positive impact on TM use or TM willingness. Finally, when the green point was lower (lower percentage) than the red point, it indicated that the individuals willing to use or using TM chose the specified answer less often than the individuals not willing to use or not using TM, which meant that this factor (question) had a negative impact on TM use or TM willingness.

All statistical analyses were performed using R software (version 4.1.2, R Foundation for Statistical Computing) for Windows 10. The *tidyverse* package (version 1.3.2) was used [20]. VIFs were calculated using the *car* package (version 3.1-0) [21]. Bayesian estimation was performed using the *rstanarm* package (version 2.21.1) [22,23]. Weakly informative priors (default priors in *rstarnarm*) were used. The default priors in *rstanarm* 2.21.1 are designed to be weakly informative. The Bayesian model adds priors (independent by default) to the coefficients of the generalized linear model. The Bayesian estimation was performed via the Markov chain Monte Carlo Bernoulli model, with 4 randomly initialized Markov chains, each for 2000 iterations (including a warm-up period of 1000 iterations that is discarded). Posterior distributions were described using the *bayestestR* package (version 0.12.1) [24]. The selection of the "best" model through BMA was undertaken using the *BMA* package (version 3.18.15) [25]. Regarding priors for BMA, we assumed that all candidate models were equally likely a priori (same prior weight). The spider charts were created using the *fmsb* package (version 0.7.3) [26].

Ethics Approval

Primary data collection was conducted in compliance with the current data protection regulations of the General Data Protection Regulation [27] and the Helsinki Declaration. All study participants were informed about the research project and provided written informed consent. Data were anonymized before analysis. The ethics committee of the Theodor Fontane Medical School in Brandenburg stated that no written consent was necessary owing to the noninterventional study design, which also applies to the secondary analysis.

Results

Population Characteristics

A total of 94.9% (467/492) and 92.3% (454/492) of individuals were selected for the analysis of TM use and TM willingness, respectively. Most participants (247/454, 54.4%) were female. Most individuals were GPs (408/454, 89.9%) and were aged between 51 and 60 years (215/454, 47.4%). Although most individuals were not using TM (344/454, 75.8%), two-thirds (282/454, 62.1%) were willing to use it in the future.

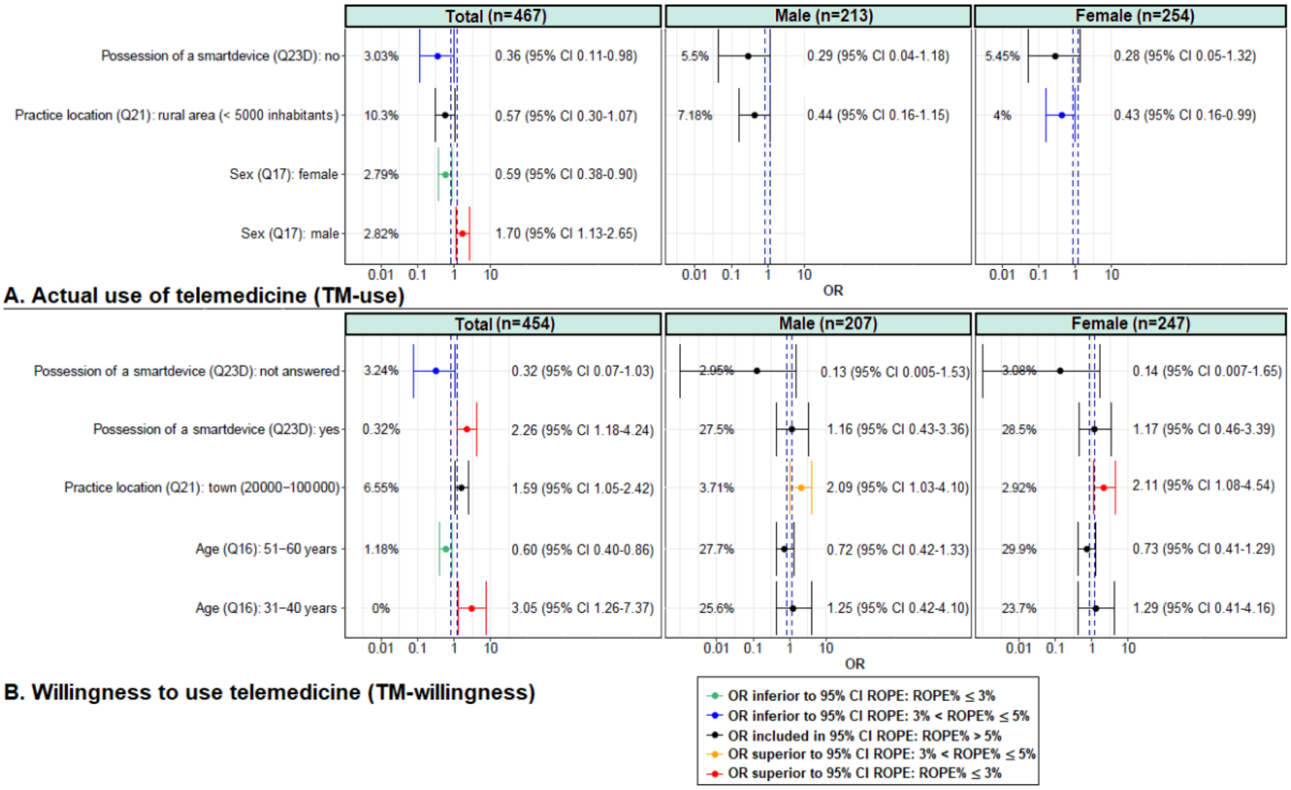
Bayesian Univariate Logistic Regression Analysis

Only significant results are presented in the main text, but all the results can be found in the Multimedia Appendices 1-5 and Figures S1-S4 in Multimedia Appendix 6. A total of 26 questions were answered (83 answers) and analyzed using the univariate logistic regression analysis. Out of 83 variables, 36 (43%) and 34 (41%) variables were found to be positively or negatively associated (ROPE%≤5%) with TM use and TM willingness, respectively (Multimedia Appendix 2). Regarding sociodemographic factors (Figure 1), not owning a smart device (OR 0.36, 95% CI 0.11-0.99; ROPE%=3.0); being female (OR 0.59, 95% CI 0.38-0.90; ROPE%=2.8); and being female with a practice located in rural area (<5000 inhabitants; OR 0.43, 95% CI 0.16-0.99; ROPE%=4.0) were negatively associated with TM use, whereas being aged between 51 and 60 years (OR 0.60, 95% CI 0.40-0.86; ROPE%=1.2) was negatively associated with TM willingness. By contrast, being male (OR 1.70, 95% CI 1.13-2.65; ROPE%=2.8) was positively associated with TM use, whereas owning a smart device (OR 2.26, 95% CI 1.18-4.24; ROPE%=0.3); being aged 31 to 40 years (OR 3.05 95% CI 1.26-7.37; ROPE%=0); and having a practice located in town (20,000-100,000 inhabitants) were positively associated with TM willingness. For more details, please refer to Figures S1 and S2 in Multimedia Appendix 6.

Regarding work characteristics, being a rheumatologist, working in a medical care center, and treating >1000 patients per quarter were positively associated with TM use, whereas treating <500 patients per quarter and being an assigning physician were negatively associated (Multimedia Appendix 2 and Figures S3 and S4 in Multimedia Appendix 6).

Regarding the opinion and knowledge about TM, having at least good TM knowledge, thinking that TM is suitable for exchange in rheumatology, wanting to exchange information with specialists via TM, and thinking that TM is at least rather important for current and future work were positively associated with both TM use and TM willingness (Multimedia Appendix 2 and Figures S3-S6 in Multimedia Appendix 6). By contrast, having poor or very poor TM knowledge and thinking that TM is not important at all for current and future work were both negatively associated with both TM use and TM willingness. Individuals willing to use TM were strongly and positively associated with TM use.

Figure 1. Bayesian univariate logistic regression—Relationship between the actual use of telemedicine (TM use) or willingness to use telemedicine (TM willingness) and sociodemographic factors. The percentage indicates the region of practical equivalence (ROPE) percentage, that is, the probability that the considered credible factor values are not negligible. The dashed lines indicate the 95% credible interval (CI) of the ROPE. OR: odds ratio; Q: question.



BMA and Bayesian Multivariate Logistic Regression Analysis

A total of 6 BMA analyses were conducted, with 3 (both sexes, male, and female) for TM use and 3 for TM willingness. Figure 2 presents the determinants identified through BMA for the 6 analyses. Only variables with a posterior probability of ≥10% were considered determinant factors. A total of 16 answers were selected using BMA. Variables above the dashed horizontal line refer to factors positively associated with TM use or TM willingness (cells with color from light yellow to red). By contrast, variables under the dashed horizontal line refer to factors negatively associated with TM use or TM willingness (cells with colors from light green to dark blue). The value in each cell corresponds to the posterior probability that the considered variable is nonzero (in percentage). Darker the color, the higher the posterior probability percentage.

Regarding TM use, a total of 13 determinant factors (13 answers from 8 questions) were identified. Being female, having very poor knowledge of TM, treating <500 patients per quarter, thinking that TM is not important at all for current work, and not being willing to use TM were negatively associated with TM use. By contrast, having good or very good knowledge of TM, thinking that TM is important or very important for current work and at least rather not important for future work, treating >1000 patients per quarter, and thinking that TM is suitable for exchange in rheumatology were positively associated with TM use.

Regarding TM willingness, a total of 17 determinant factors (17 answers from 11 questions) were identified. Not wanting to exchange information with specialists using TM, thinking TM services have no place in the care process, being aged 51 to 60 years, thinking that TM is not important for current and future work, and not currently using TM were negatively associated with TM willingness. By contrast, owning a smart device, thinking that TM is at least rather not important for future work, thinking that TM is relevant in subareas in rheumatology, and thinking that there should be exchange with TM were positively associated with TM willingness.

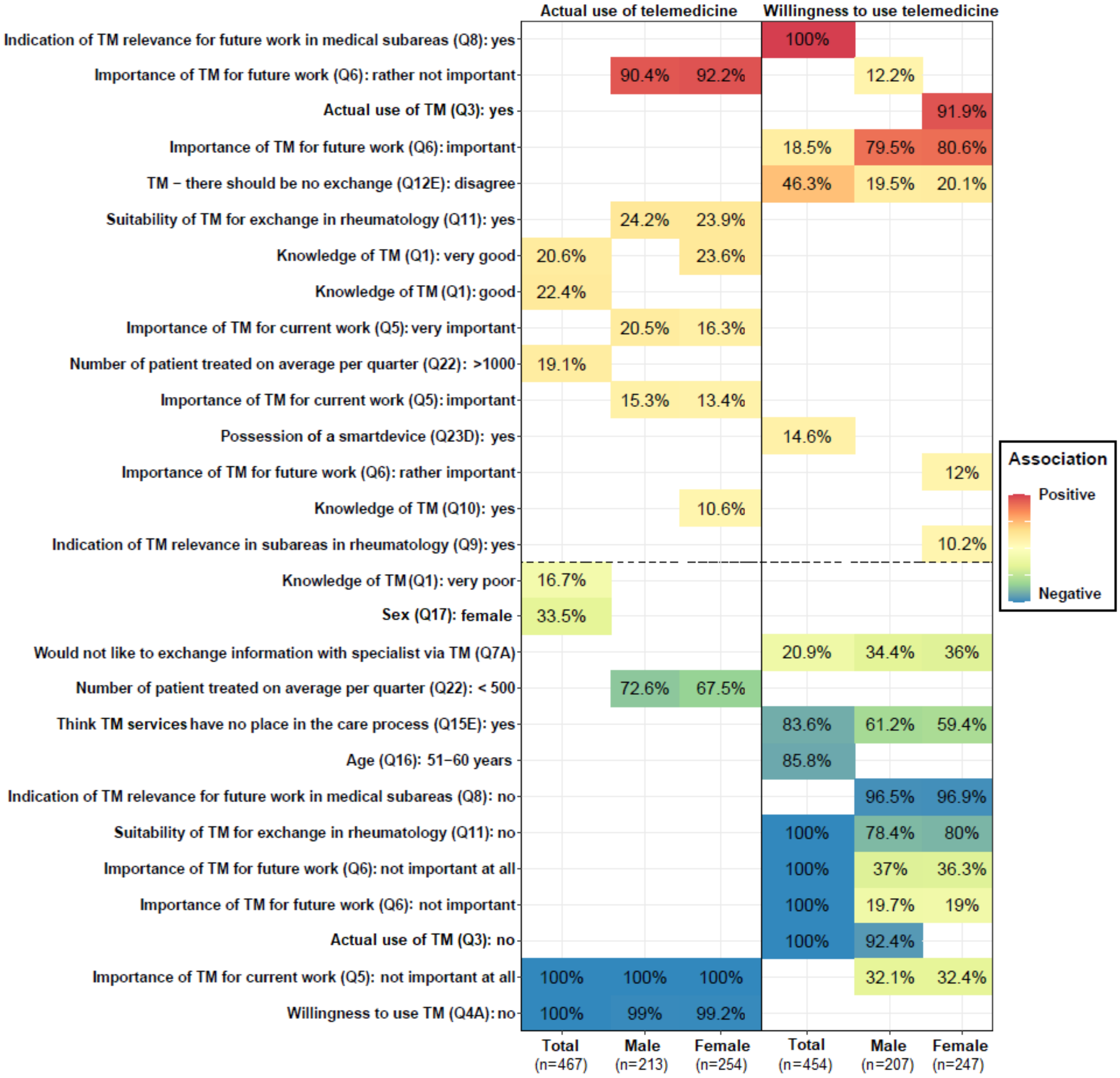
For more details about the BMA analysis, please refer to Multimedia Appendix 4, which synthesizes BMA results for the top 5 models, as well as to Figures S7-S11 in Multimedia Appendix 6 for TM use and for TM willingness, which represent all the variables considered (in the y-axis) for the full list of models selected (in the x-axis). Blue color indicates variables negatively associated with TM use or TM willingness, whereas red color indicates variables that are positively associated.

Results for the "best" model identified through BMA indicated that being female (OR 0.57, 95% CI 0.35-0.90; ROPE%=3.2); thinking that TM is not important at all for current work (OR 0.15, 95% CI 0.08-0.29; ROPE%=0); and not being willing to use TM (OR 0.22, 95% CI 0.10-0.38; ROPE%=0) were negatively associated with TM use for both sexes. When stratified by sex, it was found that treating <500 patients per quarter was negatively associated with TM use. Regarding TM willingness, being aged 51 to 60 years (OR 0.43, 95% CI 0.26-0.74; ROPE%=0); not using TM (OR 0.14, 95% CI

0.06-0.31; ROPE%=0); thinking that TM is not suitable for exchange in rheumatology (OR 0.13, 95% CI 0.05-0.35; ROPE%=0); and thinking that it is not important for future work (OR 0.13, 95% CI 0.05-0.35; ROPE%=0) were factors negatively associated with TM willingness for both sexes.

More details about the “best” models are available in Multimedia Appendix 5.

Figure 2. Determinants of the actual use of telemedicine (TM use) or willingness to use telemedicine (TM willingness) identified through the Bayesian model averaging analysis. A total of 28 answers from 16 questions were selected with Bayesian model averaging. The value in each cell corresponds to the posterior probability that the considered variable is nonzero (in percentage). Q: question.



	Actual use of telemedicine			Willingness to use telemedicine		
	Total (n=467)	Male (n=213)	Female (n=254)	Total (n=454)	Male (n=207)	Female (n=247)
Indication of TM relevance for future work in medical subareas (Q8): yes				100%		
Importance of TM for future work (Q6): rather not important		90.4%	92.2%		12.2%	
Actual use of TM (Q3): yes						91.9%
Importance of TM for future work (Q6): important				18.5%	79.5%	80.6%
TM – there should be no exchange (Q12E): disagree				46.3%	19.5%	20.1%
Suitability of TM for exchange in rheumatology (Q11): yes		24.2%	23.9%			
Knowledge of TM (Q1): very good	20.6%		23.6%			
Knowledge of TM (Q1): good	22.4%					
Importance of TM for current work (Q5): very important		20.5%	16.3%			
Number of patient treated on average per quarter (Q22): >1000	19.1%					
Importance of TM for current work (Q5): important		15.3%	13.4%			
Possession of a smartdevice (Q23D): yes				14.6%		
Importance of TM for future work (Q6): rather important						12%
Knowledge of TM (Q10): yes			10.6%			
Indication of TM relevance in subareas in rheumatology (Q9): yes						10.2%
Knowledge of TM (Q1): very poor	16.7%					
Sex (Q17): female	33.5%					
Would not like to exchange information with specialist via TM (Q7A)				20.9%	34.4%	36%
Number of patient treated on average per quarter (Q22): < 500		72.6%	67.5%			
Think TM services have no place in the care process (Q15E): yes				83.6%	61.2%	59.4%
Age (Q16): 51−60 years				85.8%		
Indication of TM relevance for future work in medical subareas (Q8): no					96.5%	96.9%
Suitability of TM for exchange in rheumatology (Q11): no				100%	78.4%	80%
Importance of TM for future work (Q6): not important at all				100%	37%	36.3%
Importance of TM for future work (Q6): not important				100%	19.7%	19%
Actual use of TM (Q3): no				100%	92.4%	
Importance of TM for current work (Q5): not important at all	100%	100%	100%		32.1%	32.4%
Willingness to use TM (Q4A): no	100%	99%	99.2%			

Profile of TM Users or Individuals Willing to Use TM

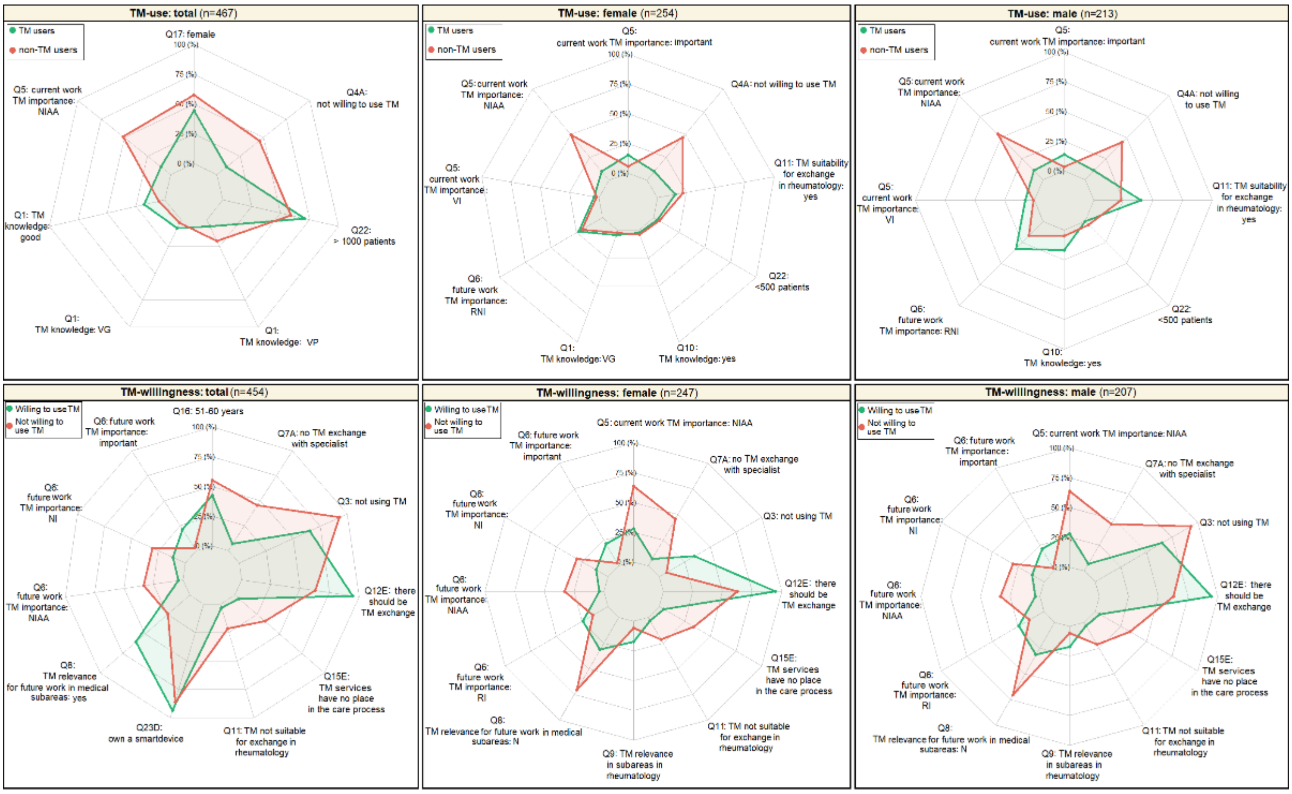
Determinant factors, defined as variables with a posterior probability of ≥10% with BMA, were identified and used to establish the profile of individuals using or willing to use TM and the profile of individuals not using or not willing to use TM. Figure 3 presents the profiles identified based on gender.

Regarding TM use, TM users more frequently had TM knowledge and treated, on average, more patients (>1000 patients per quarter) than non-TM users.

TM users were more often women, more often thought that TM is not important at all for current work, more frequently had very poor TM knowledge, and were less inclined to use TM compared with TM users.

Regarding TM willingness, the individuals who were willing to use TM owned a smart device and thought that there should be TM exchange more often than the individuals who were not willing to use TM. By contrast, the individuals not willing to use TM were more often aged 51 to 60 years and more frequently thought that TM is not suitable for exchange in rheumatology, is not important at all for current and future work, is not relevant for future work in medical subareas, and has no place in the care process. In addition, they used TM less often than the individuals who were willing to use TM.

Figure 3. Profile of telemedicine (TM) users versus nonusers and individuals willing to use TM versus those not willing to use TM using Bayesian model averaging (BMA). Variables displayed on the spider or radar chart correspond to factors selected with BMA that had a posterior probability of ≥10%. Percentages refer to the percentage of individuals with the answer specified for each question. NI: not important; NIAA: not important at all; RI: rather important; RNI: rather not important; VG: very good; VI: very important; VP: very poor; TM willingness: willingness to use telemedicine; Q: question.



Discussion

Overview

We performed a secondary analysis to identify factors associated with TM use and TM willingness on data collected as part of a cross-sectional, self-completed, and paper-based survey of German GPs and outpatient rheumatologists. The initial study [7] was conducted from September to November 2018, with the goal of exploring general acceptance, opportunities, and obstacles for the implementation of TM. The current secondary analysis was conducted to identify the most relevant factors affecting TM use and TM willingness to enable more effective TM strategies.

Principal Findings

Regarding the factors associated with TM use, our results revealed that having good or very good knowledge of TM and treating >1000 patients per quarter were positively associated with TM use. By contrast, being female, having very poor knowledge of TM, treating <500 patients per quarter, not owning a smart device, working in a rural area, thinking that TM is not important at all for current work, and not being willing to use TM were negatively associated with TM use.

Regarding the factors associated with TM willingness, owning a smart device, thinking that TM is relevant in subareas in rheumatology, working in urban areas, and thinking that there should be exchange with TM were positively associated with TM willingness. By contrast, not wanting to exchange information with specialists using TM, thinking that TM services have no place in the care process, being aged 51 to 60 years, thinking that TM is not important for current and future work, and not currently using TM were negatively associated with TM willingness.

Comparison With Prior Work

To the best of our knowledge, this is the first work analyzing specific factors influencing TM use and TM willingness among German GPs and rheumatologists. A major strength of this study lies in its ability to guide TM implementation strategies.

Our results underline the close connection between knowledge and technology use, as described by Paul Attewell [28]. According to his theory on technology diffusion and organizational learning, knowledge barriers—that is, the lack of knowledge about the technology and how this technology can be applied in an organizational setting—are in fact the reasons why technology diffusion remains low. Consistently, we found that having good or very good self-perceived knowledge of TM is positively associated with TM use, whereas having very poor knowledge is negatively associated with TM use. Similarly, a previous survey study identified the unawareness of suitable software solutions as the main factor that prevented rheumatologists from using electronic instead of paper-based questionnaires [29]. Concurringly, German rheumatologists were only aware of a fraction of the available rheumatology apps, limiting their use in clinical routine [30].

Tanriverdi and Iacono [31] extended Attewell's theory to a multidimensional concept including the economic, organizational, and behavioral knowledge barriers that hamper the diffusion of TM. Our results support this multidimensionality. For instance, larger medical practices providing for more patients are more likely to use TM than smaller organizational units. Furthermore, in line with the results of Knörr et al [32], physicians in rural areas appear to use TM less frequently than physicians in urban areas, which seems counterintuitive and might also be due to the limited technical infrastructure in rural areas in Germany [8]. However, Vossen et al [33] reported a positive correlation between the traveling time to the treating rheumatologist and the willingness of German patients with rheumatoid arthritis to use video consultations.

In addition, the purchase of technology equipment, administration effort, and inadequate reimbursement (system) of TM services in Germany were identified as the main barriers to TM use in the primary analysis [7]. These barriers were later confirmed in a multiprofessional survey to impact TM use in other medical domains as well [34].

In line with the previous results reported by Alkureishi [35], our analysis results indicated a negative association between being female and TM use. We were surprised by this finding, as eHealth literacy was recently reported to be higher among women, both among health care professionals [36] and the overall German population [37]. Apparently, higher eHealth literacy does not translate directly into higher TM use. The reasons for the gender difference need to be specifically explored in further research, particularly as the proportion of women among physicians continues to increase in Germany [38]. Furthermore, the negative association between being aged 51 to 60 years and TM willingness is striking, as the average age of physicians in Germany is currently 54.2 years with an increasing trend [38]. This is linked to substantial concerns about increasing workforce shortage [5], which TM is actually intended to address [6,39]. However, a previous study on mobile health found no gender differences in patients with rheumatoid arthritis yet revealed a negative correlation with age [40]. Thus, the differences between the study findings may also be explained by specific TM approaches queried and terminology, which should be further researched.

Implications

Because TM use is closely intertwined with physicians' knowledge in this domain, we strongly support the integration of digital competencies into medical education and offering of dedicated training courses for physicians [41-43]. Continuous education in this area seems to be particularly important, as telemedical options continuously increase, including not only medical apps but also completely new procedures such as patient self-sampling. Health care professionals also seem concerned with an increasing workload due to increasing communication and transmitted information via TM [8]; education could help to implement the most successful TM strategies. As Tanriverdi and Iacono [31] discussed earlier, these training courses should also reflect on the multidimensionality of knowledge barriers by addressing the economic, organizational, and behavioral framework conditions of digital health implementation. Administrative, technical, and reimbursement requirements should be addressed first, as these have been reported as key barriers to the use of TM [7], just as they have recently been to the use of prescribed and regulated digital therapies in Germany [44].

Concomitantly, our data point to the importance of the organizational determinants of TM use. Although there are already numerous studies that point to the effectiveness of TM use [3], it remains unclear how TM needs to be integrated into organizational structures to ensure its effective and sustainable use in routine health care. Therefore, we recommend investigating the organizational and social factors of the implementation of TM and digital health in health care delivery.

Furthermore, our findings will inform private and public stakeholders on TM implementation. Public stakeholders, such as health policy makers, might use our findings to promote TM and upgrade infrastructure in rural areas. Specific target groups for incentive schemes could be female physicians aged 51 to 60 years in particular. Private stakeholders, such as TM companies or start-ups, might infer from our findings that health care professionals need low-threshold instructions on the use of their products. Finally, we recommend organizational and structural guidance, including setup, staff planning, billing of services, and administration, for the implementation of TM in routine health care delivery.

Limitations

The primary data on which this analysis was based were collected in 2018 before the SARS-CoV-2 outbreak. Owing to the need to reduce physical contact and thus minimize the risk of infection, TM use initially received a major uptake in global health care delivery [13]. Hence, more physicians and likely other subgroups will have tried TM by now [23], which has led to an increased use and awareness of TM in routine practice. Nevertheless, recent studies suggest that even after the SARS-CoV-2 outbreak, the same barriers continue to prevent widespread TM adoption [9,10,35,44,45]. However, a replication of the initial survey is essential to identify whether and how the identified factors have changed in the surveyed target group. Thus, the results from our study represent a baseline to future studies that would investigate the change in TM experience and perceptions due to the SARS-CoV-2 pandemic.

Apart from the aforementioned shortcomings, the limitations of the primary data still apply [7]. Only a relatively small proportion (44/454, 9.7%) of the survey sample are rheumatologists, which accounts for 7% of all of the rheumatologists in outpatient care in Germany [46]. Although the survey was directed at physicians from all over Germany, it was primarily physicians from Brandenburg who participated because of the recruitment strategy. We suspect a high potential of self-selection and nonresponse bias. Health care professionals in inpatient care as well as other professions involved in rheumatology care (eg, nurses) were not included in the survey. Furthermore, our results cover the perspectives of German physicians only. Their acceptance of TM might be strongly influenced by the specifics and policy drivers of the German health system. Previous studies reported [8,45] weak remuneration, high bureaucracy, and a lack of digital infrastructure to hamper TM use in Germany. Owing to these influences, the transferability of our results to other countries and health care systems may be limited. Finally, physician engagement is an important factor in the adoption of telehealth into routine care delivery, but it represents only one side of the coin. The patient perspective and TM willingness represent the other side that needs to be investigated as a priority.

Regarding the statistical analysis, we used a Bayesian approach to conduct the secondary analysis of the aforementioned survey. A practical limitation of the Bayesian approach is that it requires the specification of prior distributions both on the parameters of each model and on the distribution of the models themselves. Because we had no a priori assumption, we used weakly informative priors. Choosing another prior distribution may have had substantial influence on the outcome [47,48]. Regarding the variable and model selections, a 3-step approach was used. First, all the individual variables associated (positively or negatively) with the use of or TM willingness in the Bayesian univariate analysis were selected based on the ROPE percentage (ROPE%≤5). Choosing a different ROPE percentage threshold may have yielded different results. Then, we performed a conservative selection based on the VIF (VIF≤2.5) to deal with potential variable multicollinearity. Finally, we used the remaining variables with BMA for model selection and identification of determinants. BMA was chosen in particular because it reduces overconfidence and is relatively robust against model misspecification [47,49-51]. Markov chain Monte Carlo was used to deal with the intractable computational challenge of BMA that comes from the candidate model enumeration [52].

Conclusions

TM use is intertwined with health care professionals' knowledge of TM. Limited knowledge restricts the implementation of TM in rheumatology care. Dedicated education courses could provide the necessary knowledge and improve TM uptake. These courses need to reflect on the multidimensionality of knowledge barriers by addressing the economic, organizational, and behavioral framework conditions of TM implementation.

TM willingness is associated with age and practice location, and incentive programs for advanced physicians practicing in rural areas have the potential to increase the implementation of TM in standard care.

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Authors' Contributions

All the authors were involved in drafting the article and critically revising it for important intellectual content, and all the authors approved the final version to be submitted for publication. FM had full access to all the data in the study and took responsibility for the integrity of the data and accuracy of the data analysis. FM, JK, NV, and PP conceptualized and designed the study. FM, MW, and NV acquired data. FM, JK, NV, and PP analyzed and interpreted the data.

Conflicts of Interest

None declared.

Multimedia Appendix 1

Regression analysis—variables.
[PDF File (Adobe PDF File), 291 KB-Multimedia Appendix 1]

Multimedia Appendix 2

List of all the variables positively and negatively associated (region of practical equivalence≤5%) with the actual use of telemedicine use and willingness to use telemedicine in the Bayesian univariate logistic regression analysis.
[PDF File (Adobe PDF File), 308 KB-Multimedia Appendix 2]

Multimedia Appendix 3

Bayesian univariate logistic regression analysis results.
[XLSX File (Microsoft Excel File), 49 KB-Multimedia Appendix 3]

Multimedia Appendix 4

Bayesian model averaging results.
[XLSX File (Microsoft Excel File), 28 KB-Multimedia Appendix 4]

Multimedia Appendix 5

Bayesian multivariate logistic regression analysis results for the best model.
[XLSX File (Microsoft Excel File), 14 KB-Multimedia Appendix 5]

Multimedia Appendix 6

Bayesian univariate logistic regression figures.
[DOCX File , 2414 KB-Multimedia Appendix 6]

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Abbreviations

BMA: Bayesian model averaging
CI: credible interval
GP: general practitioner
OR: odds ratio
ROPE: region of practical equivalence
TM use: actual use of telemedicine
TM willingness: willingness to use telemedicine
TM: telemedicine
VIF: variance inflation factor
Q: question

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Original Paper

Perceptions of Quality of Care Among Users of a Web-Based Patient Portal: Cross-sectional Survey Analysis

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Abstract

Background: Web-based patient portals enable patients access to, and interaction with, their personal electronic health records. However, little is known about the impact of patient portals on quality of care. Users of patient portals can contribute important insights toward addressing this knowledge gap.

Objective: We aimed to describe perceived changes in the quality of care among users of a web-based patient portal and to identify the characteristics of patients who perceive the greatest benefit of portal use.

Methods: A cross-sectional web-based survey study was conducted to understand patients' experiences with the Care Information Exchange (CIE) portal. Patient sociodemographic data were collected, including age, sex, ethnicity, educational level, health status, geographic location, motivation to self-manage, and digital health literacy (measured by the eHealth Literacy Scale). Patients with experience using CIE, who specified both age and sex, were included in these analyses. Relevant survey items (closed-ended questions) were mapped to the Institute of Medicine's 6 domains of quality of care. Users' responses were examined to understand their perceptions of how portal use has changed the overall quality of their care, different aspects of care related to the 6 domains of care quality, and patient's satisfaction with care. Multinomial logistic regression analyses were performed to identify patient characteristics associated with perceived improvements in overall care quality and greater satisfaction with care.

Results: Of 445 CIE users, 38.7% (n=172) reported that the overall quality of their care was better; 3.2% (n=14) said their care was worse. In the patient centeredness domain, 61.2% (273/445) of patients felt more in control of their health care, and 53.9% (240/445) felt able to play a greater role in decision-making. Regarding timeliness, 40.2% (179/445) of patients reported they could access appointments, diagnoses, and treatment more quickly. Approximately 30% of CIE users reported better care related to the domains of effectiveness (123/445, 27.6%), safety (138/445, 31%), and efficiency (174/445, 28.6%). Regarding equity, patients self-reporting higher digital health literacy (odds ratio 2.40, 95% CI 1.07-5.42; P=.03) and those belonging to ethnic minority groups (odds ratio 2.27, 95% CI 1.26-3.73; P<.005) were more likely to perceive improvements in care quality. Across ethnic groups, Asian and British Asian patients perceived the greatest benefits. Increased frequency of CIE use also predicted perceived better care quality and greater satisfaction with care.

Conclusions: A large proportion of CIE users perceived better care quality and greater satisfaction with care, although many portal users reported no change. The most favorable perceived improvements related to the domain of patient centeredness. With national policy directed toward addressing health disparities, patient portals could be valuable in improving care quality for ethnic minority groups. Future research should test the causal relationship between patient portal use and care quality.

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KEYWORDS

electronic health records; personal health records; patient participation; patient safety; care quality; digital health literacy

Introduction

Background

Web-based patient portals are thought to contribute to improvements in care quality by providing patients with access to their personal health information, empowering them to self-manage their health and become true partners in their own care [1]. As the trend toward patients being able to access their electronic health records accelerates [2], there is a pressing need to evaluate the impact of patient portals, understand their risks and benefits from both patient and provider perspectives, and generate evidence to inform future health policy [3].

Although care is traditionally delivered through face-to-face clinical consultations, patient-provider communication through patient portals is increasingly common [1]. The Care Information Exchange (CIE) is the largest shared personal health records program in the United Kingdom and provides patients with secure web-based access to their health and social care records. Patients can additionally use CIE in different ways: for example, to self-monitor their health by linking home health care devices (eg, activity tracker and blood pressure monitor) to the portal, to communicate with care providers through messaging and videoconferencing, and to check appointments and test results and be signposted to useful weblinks and resources by health and care professionals.

One of the most influential guides for evaluating health care initiatives is the Institute of Medicine's framework, which includes 6 domains of quality of care: effectiveness, safety, timeliness, efficiency, patient centeredness, and equity [4,5]. Effectiveness is about achieving optimal health outcomes by providing appropriate treatment to patients who could benefit and avoiding the underuse and misuse of health services [4,5]. Patient safety seeks to prevent patients from being harmed by the care that is intended to help them [4,5]. Timeliness is about reducing harmful waits and delays, whereas efficiency is about minimizing resource waste [4,5]. Patient centeredness respects patient preferences and needs and values and ensures these are incorporated into clinical decision-making [4,5]. Equity ensures that care does not vary in quality because of differences in patient characteristics such as ethnicity or geographic location [4,5].

Over the last decade, a considerable body of evidence has uncovered important barriers to portal use, enabling the development of portals in line with patient and health service need [6-8]. In contrast, relatively few studies have investigated the relationship between patient portals and quality of care. Some prior evidence demonstrates the beneficial effects of patient portal use, particularly in supporting preventive behaviors and disease control in people with chronic conditions [3,7,9]. A number of studies have documented positive associations between patient portals and patient safety [3,7,10-13], including improved adherence to medical regimens and reductions in medication discrepancies [3]. However, evidence for the impact of patient portals across other domains of quality is sparse, and where evidence does exist, findings have been mixed [3,7]. Among patients who use web-based portals, little is known about which sociodemographic groups perceive the greatest benefits of access to their personal health records. Furthermore, policy makers agree that more evidence is needed to understand the impact of tools that use digital technologies amidst concerns over a growing *digital divide* [14].

Objectives

The aims of this study were to describe perceptions of quality of care among users of a web-based patient portal and to identify the characteristics of portal users who perceive the greatest benefit of portal use.

Methods

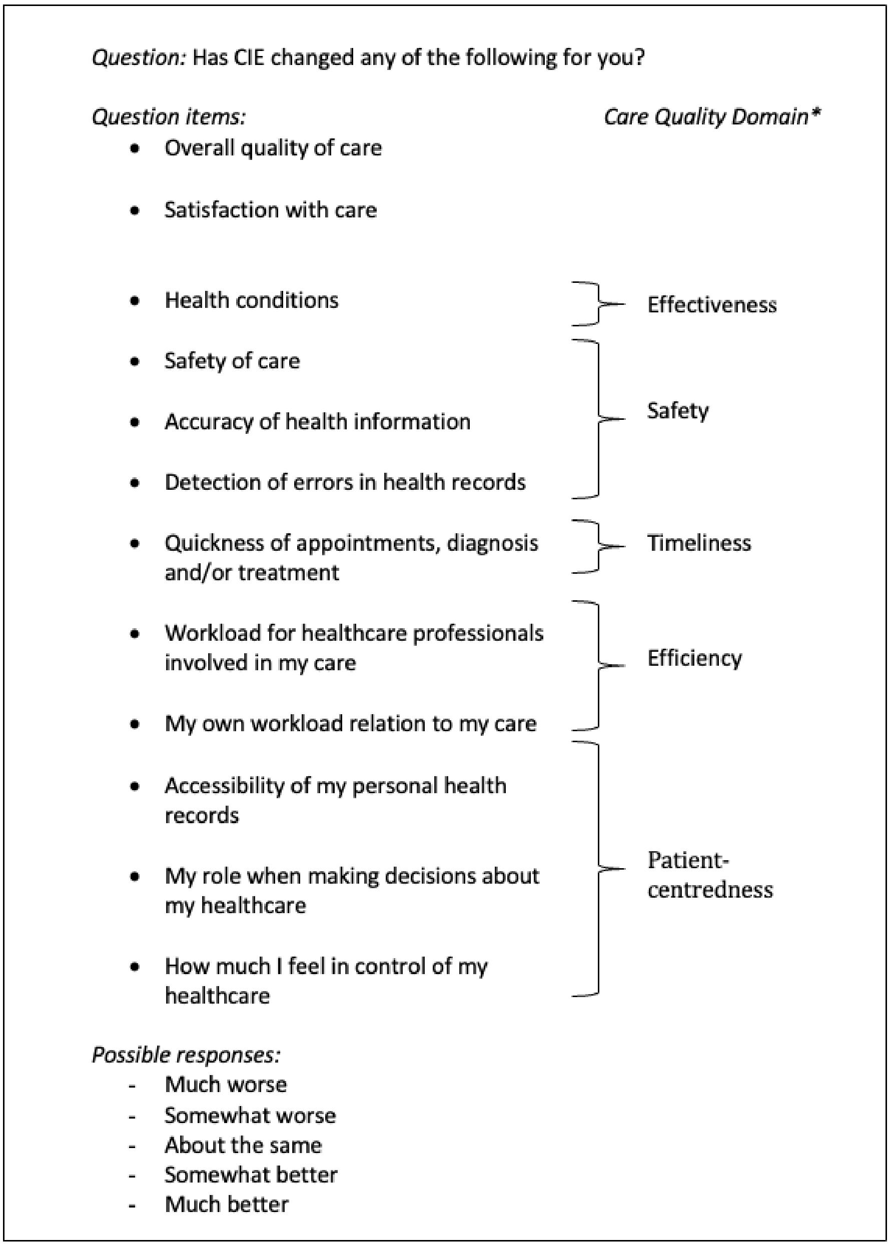
Study Design, Participants, and Data Collection

A cross-sectional survey study was conducted to explore patients' views and experiences of using CIE. The questionnaire was administered via Qualtrics (web-based survey platform) and was open for completion between July 1, 2018, and July 1, 2019. At the time of the survey, CIE was deployed to the diverse 2.3 million patients treated in North West London, including patients residing in London and in other geographic locations across England. CIE held records from hospitals and general practitioners in North West London and from 15 hospitals outside of London, in Birmingham, Bristol, Liverpool, Manchester, Scotland, and Wales. All patients registered with the CIE at the time of the survey were invited via email to complete the questionnaire (n=27,411). The email explained the purpose of the study; informed consent was obtained. Patients accessed the questionnaire via a web link in the portal. Patients had to be aged at least 18 years to be registered with CIE. Not all patients registered with CIE were using the portal. With this data set, we have previously evaluated differences between users and nonusers of CIE with respect to their sociodemographic characteristics and demonstrated the importance of addressing educational aspects and digital literacy to ensure equitable and sustainable portal adoption [15]. Our further work has sought to evaluate the impact of web-based patient portals on safety and quality of care from the patient's perspective. Our recent study found that a large proportion of patients are able and willing to use patient portals to participate in identifying and rectifying errors in their personal health records [16]. This study builds on previous work to understand patients' perceptions of the impacts of CIE across 6 domains of care quality.

For these analyses, we included patients who had previously accessed and used the CIE portal. We excluded patients who did not provide basic demographics regarding age and sex. Considering this population, a CI of 95%, and a margin of error of 5%, the minimum sample size to ensure representativeness was calculated as 379 respondents. We mapped relevant survey items to the Institute of Medicine's domains of quality of care: effectiveness, safety, timeliness, efficiency, and patient centeredness [5,17]. Patients' responses to 12 multiple-choice, closed-ended question items were analyzed. Figure 1 outlines the 12 question items, with mapping to care quality domains.

To evaluate equity, we conducted multivariable regression analyses to determine associations between patients' sociodemographic characteristics and perceptions of the impact of CIE on overall care quality and satisfaction with care. The following information was collected to input into multivariable analyses: age, sex, ethnicity, native language, education level, digital health literacy, motivation to be involved in own care, and health status. Respondents' level of motivation to be involved in their own care was assessed via a multiple-choice question ("In general, how motivated to be involved in your healthcare are you?" Possible responses: "A little," "A moderate amount," "A lot," and "Very much"). Digital health literacy was assessed using the eHealth Literacy Scale (eHEALS), developed and validated by Norman and Skinner [18]. The eHEALS tool is an 8-item measure of patients' combined knowledge, comfort, and perceived skills in finding, evaluating, and using internet health resources for health problems [18]. The 8 items are answered on a 5-point Likert scale (1, strongly disagree to 5, strongly agree); total eHEALS scores range from 8 to 40, with a higher score indicating higher digital literacy.

Figure 1. Questionnaire items mapped to care quality domains. The domain of equity was assessed using the methods described in this section. *As defined by the Institute of Medicine, 2001 [5]. CIE: Care Information Exchange.



Question: Has CIE changed any of the following for you?

Question items:

- Overall quality of care
- Satisfaction with care
- Health conditions
- Safety of care
- Accuracy of health information
- Detection of errors in health records
- Quickness of appointments, diagnosis and/or treatment
- Workload for healthcare professionals involved in my care
- My own workload relation to my care
- Accessibility of my personal health records
- My role when making decisions about my healthcare
- How much I feel in control of my healthcare

*Care Quality Domain**

Effectiveness

Safety

Timeliness

Efficiency

Patient-centredness

Possible responses:

- Much worse
- Somewhat worse
- About the same
- Somewhat better
- Much better

Data Analysis

We used descriptive statistics to summarize respondent characteristics and patients' responses to question items. Counts and proportions were calculated for categorical variables; means and SDs were calculated for continuous variables. Age was categorized into bands (<30, 31-40, 41-50, 51-65, and ≥65 years). Owing to the small numbers of patients self-identifying to individual categories of ethnic minority background, ethnicity was categorized as "ethnic minorities" or "White."

We conducted multinomial regression analyses to identify sociodemographic characteristics that predict patient-perceived improvement in overall care quality and greater satisfaction with care. To overcome the issue of sparse counts in multivariable modeling (Tables S1 and S2 in Multimedia Appendix 1), "age," "motivation to be involved in own care," "digital health literacy," and "frequency of CIE use" were treated as dichotomous variables, and respondents reporting sex as "other" were excluded. Consistent with previous studies, we selected an eHEALS score ≥26 to indicate higher digital health literacy and <26 to indicate lower digital health literacy [19-23]. We also combined categories of the dependent variable (ie, "much worse" and "somewhat worse" were analyzed as a single category; equally, "somewhat better" and "much better" were combined into 1 category). We performed univariate multinomial logistic analyses to identify possible predictors to include in the multivariable model. We adopted the approach by Hosmer et al [24,25] for variable selection: (1) variables that demonstrated significance ($P<.25$) in the univariate analyses were entered into the preliminary multivariable model; (2) variables that were nonsignificant at $P>.05$ according to the likelihood ratio test were removed one at a time according to the variable with the highest P value (backward elimination); (3) to check for suppressor effects, variables excluded during backward selection were re-entered separately into the regression model (forward selection). Only variables that were significant at $P<.05$ (Likelihood Ratio Test) were retained in the final multinomial regression models. Model quality comparisons were conducted using the Akaike Information Criterion [26], and goodness-of-fit was assessed using the Pearson chi-square statistic [25]. Effect estimates are presented as odds ratios (ORs) with their 95% CIs.

To assess the effects of excluding patients with missing data regarding age and sex, we compared the sociodemographic characteristics of the missing data sample (n=78) and the analysis sample (n=445). We ran a Pearson chi-square test of homogeneity (χ^2) to compare the distribution of item responses between the analysis sample and the missing data sample for the perceived impact of CIE on the overall quality of care and satisfaction with care.

Analyses were conducted using Microsoft Excel (version 16.54) and SPSS software (version 27; IBM Corp).

Ethics Approval

The study was approved as a Service Evaluation at Imperial College Health care NHS Trust (registration number: 296/2018).

Reporting

We followed the reporting recommendations in the Strengthening the Reporting of Observational Studies in Epidemiology Statement (Multimedia Appendix 2). [27].

Results

Respondent Characteristics

Of 1083 patients who responded to the survey, 523 (48.29%) patients who were "CIE users" completed the questionnaire. CIE users who provided basic demographic details regarding age and sex were included in the analysis (445/523, 85.1%; +117% of the minimum target sample size); 14.9% (78/523) of respondents with missing data for age and sex were excluded.

Of 445 respondents, most (n=313, 70.3%) were aged >50 years and 276 (62%) were female. Approximately 1 in 5 (97/445, 21.8%) respondents belonged to an ethnic minority group. Most (292/445, 65.6%) respondents were educated to the degree level or higher, and the mean eHEALS score was 33.6 (SD 6.4, range 8-40); a score ≥26 indicates higher digital health literacy. Of 445 patients, 177 (39.8%) patients reported being in good health; 162 (36.4%) of patients reported that the status of their health was poor. Most (278/445, 62.5%) patients reported being very motivated in their own care. Most (284/445, 63.8%) patients said they used CIE at least once a month, and 93.2% (415/445) of patients said they found CIE useful. Patient characteristics are presented in Table 1.

Table 1. Respondent characteristics (N=445).

	Respondents
Sex, n (%)	
Male	167 (37.5)
Female	276 (62)
Other	2 (0.4)
No response	N/A[a]
Age group (years), n (%)	
<30	22 (4.9)
31-40	48 (10.8)
41-50	62 (13.9)
51-64	166 (37.3)
>65	147 (33)
No response	N/A
Ethnicity, n (%)	
Asian or British Asian	44 (9.9)
Black African or Black Caribbean or Black British	20 (4.5)
Mixed or multiple ethnic groups	11 (2.5)
Other	22 (4.9)
White	343 (77.1)
No response	5 (1.1)
Geographic location, n (%)	
London	284 (63.8)
Other locations in England	145 (32.6)
No response	16 (3.6)
Education, n (%)	
Secondary school or below	118 (26.5)
Undergraduate or professional degree	180 (40.4)
Postgraduate or higher	112 (25.2)
No response	35 (7.9)
Language, n (%)	
English	379 (85.2)
Non-English	58 (13.0)
No response	8 (1.8)
eHealth literacy (eHEALS[b] score), mean (SD; range)	33.6 (6.4; 8-40)
Overall health status, n (%)	
Good or very good	177 (39.8)
Neither good nor poor	106 (23.8)
Poor or very poor	162 (36.4)
No response	0 (0)
Motivation to be involved in own care, n (%)	
Not very much	6 (1.3)
A moderate amount	43 (9.7)

	Respondents
A lot	116 (26.1)
Very much	278 (62.5)
No response	2 (0.4)

[a] N/A: not applicable.

[b] eHEALS: eHealth Literacy Scale.

Patients' Perceptions of the Impact of CIE on the Overall Quality of Care

Patients were asked to consider how CIE has changed the overall quality of care they receive. Of 429 patients who answered this question, 172 (38.7%) reported that the quality of their care was better with CIE. A further 54.6% (243/445) said that their care was about the same, and 3.2% (14/445) of patients said their care was worse (Multimedia Appendix 3).

Patients' Perceptions of the Impact of CIE on Satisfaction With Care

When asked to consider how CIE has changed and how satisfied they are with their care, 43.6% (194/445) of patients said their care was better, 47.6% (212/445) said their care was the same, and 4.3% (19/445) said their care was worse. In addition, 4.5% (20/445) of patients did not respond to this question (Multimedia Appendix 3).

Patients' Perceptions of the Impact of CIE Across 6 Domains of Care Quality

Overview

Patients' responses to a further 10 survey items revealed their perceptions of how CIE use has changed the care they receive across the following domains of quality of care: effectiveness, safety, timeliness, efficiency, and patient centeredness (Table 2).

Table 2. Survey items and patients' responses, mapped to the Institute of Medicine's domains of health care quality (N=445).

Health care quality domain[a] and survey item: "Has CIE changed any of the following...?"	Missing data, n (%)	Much worse, n (%)	Somewhat worse, n (%)	About the same, n (%)	Somewhat better, n (%)	Much better, n (%)
Effective						
Health conditions	30 (6.7)	7 (1.6)	9 (2)	276 (62)	61 (13.7)	62 (13.9)
Safe						
Safety of care	33 (7.4)	7 (1.6)	7 (1.6)	260 (58.4)	68 (15.3)	70 (15.7)
Accuracy of health information	25 (5.6)	9 (2)	11 (2.5)	187 (42.0)	117 (26.3)	96 (21.6)
Detection of errors in health records	32 (7.2)	8 (1.8)	13 (2.9)	246 (55.3)	70 (15.7)	76 (17.1)
Timely						
Quickness of appointments, diagnosis, and/or treatment	29 (6.5)	13 (2.9)	12 (2.7)	212 (47.6)	77 (17.3)	102 (22.9)
Efficient						
Workload for health care professionals involved in my care	31 (7.0)	7 (1.6)	15 (3.4)	265 (59.6)	63 (14.2)	64 (14.4)
My own workload relating to my care	28 (6.3)	11 (2.5)	23 (5.2)	209 (47)	92 (20.7)	82 (18.4)
Patient centeredness						
Accessibility of my personal health records	16 (3.6)	6 (1.3)	9 (2.0)	72 (16.2)	112 (25.2)	230 (51.7)
My role when making decisions about my health care	6 (1.3)	11 (2.5)[b]	11 (2.5)[b]	188 (42.2)[c]	240 (53.9)[d]	240 (53.9)[d]
How much I feel in control of my health care	6 (1.3)	19 (4.3)[e]	19 (4.3)[e]	147 (33)[c]	273 (61.2)[f]	273 (61.2)[f]

[a] As defined by the Institute of Medicine [5].

[b] I feel I have less of a role.

[c] No change.

[d] I feel I have more of a role.

[e] I feel I have less control.

[f] I feel I have more control.

Effectiveness

Patients were asked whether CIE use had changed their health condition. Most (276/445, 62%) patients responded that their health condition was about the same; however, 27.6% (123/445) patients reported that their health condition had improved with CIE use. Only 3.6% (16/445) said their health condition was worse.

Safety

Although many (260/445, 58.4%) patients reported that the safety of the care was the same with CIE; 31% (138/445) felt that their care was safer. Approximately half (213/445, 47.9%) believed that CIE had led to improvements in the accuracy of their health information, and 32.8% (146/445) of patients felt CIE was associated with better detection of errors in the health record. Only 3.2% (14/445) of patients felt the safety of their care was worse with CIE.

Timeliness

Approximately 40% (179/445) of patients felt that the timeliness of their care (being able to access appointments, diagnoses, and treatment quickly) had improved with CIE. Only 5.6% (25/445) said the timeliness of their care was worse, and 47.6% (212/445) said the timeliness of their care was about the same.

Efficiency

Patients were asked whether CIE had changed the workload relating to their health, including both patients' own workload and the workload of health professionals involved in their care. Many (209/445, 47%) patients reported that their own workload was about the same; however, 28.6% (174/445) felt that their workload was better, and 7.7% (34/445) felt their workload was worse. Regarding the impact of CIE on the workload of health professionals, 39.1% (174/445) of patients perceived that this had improved, 59.6% (265/445) believed it to be about the same, and 5% (22/445) thought that it was worse.

Patient Centeredness

Most (342/445, 76.9%) patients reported that CIE had improved the accessibility of their personal health records. A few (72/445, 16.2%) patients felt that the accessibility of their records was about the same with CIE, whereas only 3.3% (15/445) said their records were less accessible. More than half (240/445, 53.9%) of the survey respondents reported that CIE had led to them having more of a role in decision-making, and 61.3% (273/445) feel they have more control of their health care. Only 2.5% (11/445) of patients reported feeling they have less of a role, and 4.3% (19/445) felt they have less control of their health care with CIE.

Equity

To identify the characteristics of CIE users who perceived better overall quality of care and greater satisfaction with care with portal use, patient characteristics and survey responses were entered into univariate and multivariable multinomial regression models.

For the survey item, "How has CIE changed the overall quality of care you have received?" the final multivariable multinomial regression model with 3 predictor variables (ethnicity, digital health literacy, and frequency of CIE use) predicted significantly better than the null (intercept) model (*P*<.001) and Pearson chi-square statistic indicated satisfactory model fit (χ^2_8=14.4; *P*=.07). The results of the regression are presented in Table 3. Patients with higher digital health literacy (eHEALS score≥26) were more likely to report that the overall quality of their care was better with CIE use (OR 2.40, 95% CI 1.07-5.42; *P*=.03). Compared with their White counterparts, patients self-identifying to an ethnic minority group were also more likely to perceive improvements in care quality based on CIE use (OR 2.27, 95% CI 1.26-3.73; *P*=.005). Across ethnic groups, 68% (30/44) of Asian and British Asian patients reported better overall quality of care with CIE use, compared with 45% (9/20) of Black or African or Caribbean or Black British patients, 36.6% (120/328; missing data, n=15) of White patients, and 36% (4/11) of patients from mixed or multiple ethnic groups (Table S1 in Multimedia Appendix 4).

Table 3. Multinomial regression results of patient characteristics and perceived change in overall quality of care with Care Information Exchange use.

	Univariate[a]				Multivariable[a]			
	Worse care quality vs about the same		Better care quality vs about the same		Worse care quality vs about the same		Better care quality vs about the same	
	Odds ratio (95% CI)	*P* value	Odds ratio (95% CI)	*P* value	Odds ratio (95% CI)	*P* value	Odds ratio (95% CI)	*P* value
Sex								
Female	Reference	Reference	Reference	Reference	Reference	Reference	Reference	Reference
Male	0.47 (0.13-1.74)	.26	1.26 (0.84-1.88)	.26	N/A[b]	N/A	N/A	N/A
Age (years)								
≥65	Reference	Reference	Reference	Reference	Reference	Reference	Reference	Reference
≤64	1.35 (0.41-4.42)	.63	1.28 (0.84-1.94)	.26	N/A	N/A	N/A	N/A
Ethnicity								
White	Reference	Reference	Reference	Reference	Reference	Reference	Reference	Reference
Ethnic minority	1.88 (0.49-7.18)	.36	2.27 (1.37-3.78)	.002	2.44 (0.61-9.80)	.21	2.27 (1.26-3.73)	.005
Native language								
English	Reference	Reference	Reference	Reference	Reference	Reference	Reference	Reference
Non-English	2.56 (0.66-9.91)	.18	1.81 (1.02-3.21)	.04	—[c]	—	—	—
Education								
Secondary or below	Reference	Reference	Reference	Reference	Reference	Reference	Reference	Reference
Undergraduate or professional	4.60 (0.55-38.23)	.16	0.85 (0.53-1.38)	.51	—	—	—	—
Postgraduate or higher	4.00 (0.44-36.76)	.22	0.73 (0.42-1.25)	.25	—	—	—	—
Digital literacy								
Lower digital health literacy	Reference	Reference	Reference	Reference	Reference	Reference	Reference	Reference
Higher digital health literacy	1.57 (0.20-12.63)	.67	2.51 (1.15-5.45)	.02	1.51 (0.18-12.42)	.70	2.40 (1.07-5.42)	.03
Health status								
Neither good nor poor	Reference	Reference	Reference	Reference	Reference	Reference	Reference	Reference
Poor	0.72 (0.20-2.60)	.62	1.29 (0.77-2.16)	.34	N/A	N/A	N/A	N/A
Good	0.52 (0.14-2.02)	.35	1.22 (0.73-2.03)	.45	N/A	N/A	N/A	N/A
Motivation to be involved in own care								
Not very much or a moderate amount	Reference	Reference	Reference	Reference	Reference	Reference	Reference	Reference
A lot or very much	1.92 (0.24-15.19)	.54	1.67 (0.86-3.24)	.13	—	—	—	—
Frequency of Care Information Exchange use								
Once a month or less	Reference	Reference	Reference	Reference	Reference	Reference	Reference	Reference
Once a week or more	1.05 (0.32-3.45)	.94	2.40 (1.59-3.63)	<.001	0.92 (0.24-3.60)	.90	2.31 (1.49-3.58)	<.001

[a] Goodness-of-fit: χ^2_8=14.5; *P*=.07.

[b] N/A: not applicable; variable not entered into the multivariable analyses due to nonsignificance (*P*>.25) in univariate analyses.

[c] Variable excluded from the final multivariable model using a backward elimination approach.

Frequency of CIE Use

Patients using CIE at least once per week were more likely to perceive improved care quality compared with patients using CIE less frequently (OR 2.31, 95% CI 1.49-3.58; *P*<.001). Sensitivity analyses assessing the effects of including or excluding predictor variables that had demonstrated significance in univariate analyses did not alter the results of the multivariable regression.

For the survey item “How has CIE changed how satisfied you are with your care?” the final multivariable model with 3 predictor variables (ethnicity, digital health literacy, and frequency of CIE use) predicted significantly better than the null (intercept) model (*P*<.001) and Pearson chi-square statistic suggested that the model fit the data well (χ^2_8=5.6; *P*=.69). Patients with higher digital health literacy (eHEALS score≥26) were more likely to report greater satisfaction with their care with CIE use, compared with those with lower digital health literacy (OR 2.35, 95% CI 1.09-5.04; *P*=.03; Table 4). CIE use was also associated with greater satisfaction with care among patients belonging to an ethnic minority group compared with White patients (OR 2.12, 95% CI 1.22-3.67; *P*=.007). Cross-tabulation of patients’ ethnicity and perceived change in satisfaction with care revealed that 77% (34/44) of Asian or British Asian patients reported greater satisfaction with care with CIE use, compared with 55% (11/20) of Black or African or Caribbean or Black British patients, 36% (4/11) of patients from mixed or multiple ethnic groups, and 42.1% (137/325; missing data n=18) of White patients (Table S2 in Multimedia Appendix 4).

Patients using CIE at least once per week were more likely to report greater satisfaction with care compared with patients using CIE less frequently (OR 2.03, 95% CI 1.31-3.14; *P*=.002).

Sensitivity analyses assessing the effects of including or excluding predictor variables that had demonstrated significance in univariate analyses did not alter the results of the multivariable analyses.

Table 4. Multinomial regression results of patients' sociodemographic characteristics and impact of Care Information Exchange on patient's satisfaction with care.

	Univariate[a]				Multivariable[a]			
	Worse care quality vs about the same		Better care quality vs about the same		Worse care quality vs about the same		Better care quality vs about the same	
	Odds ratio (95% CI)	*P* value	Odds ratio (95% CI)	*P* value	Odds ratio (95% CI)	*P* value	Odds ratio (95% CI)	*P* value
Sex								
Female	Reference	Reference	Reference	Reference	Reference	Reference	Reference	Reference
Male	0.84 (0.31-2.31)	.74	1.32 (0.88-1.97)	.17	—[b]	—	—	—
Age (years)								
≥65	Reference	Reference	Reference	Reference	Reference	Reference	Reference	Reference
≤64	0.69 (0.27-1.80)	.45	1.215 (0.76-1.75)	.51	N/A[c]	N/A	N/A	N/A
Ethnicity								
White	Reference	Reference	Reference	Reference	Reference	Reference	Reference	Reference
Ethnic minority	1.30 (0.35-4.78)	.70	2.32 (1.38-3.90)	.002	1.68 (0.44-6.41)	.45	2.12 (1.22-3.67)	.007
Native language								
English	Reference	Reference	Reference	Reference	Reference	Reference	Reference	Reference
Non-English	1.74 (0.47-6.52)	.41	1.63 (0.91-2.89)	.10	—	—	—	—
Education								
Secondary or below	Reference	Reference	Reference	Reference	Reference	Reference	Reference	Reference
Undergraduate or Professional	8.15 (1.03-64.80)	.05	1.14 (0.67-1.96)	.63	—	—	—	—
Postgraduate or higher	5.94 (0.67-52.47)	.11	1.11 (0.69-1.80)	.67	—	—	—	—
Digital literacy								
Lower digital health literacy	Reference	Reference	Reference	Reference	Reference	Reference	Reference	Reference
Higher digital health literacy	2.29 (0.29-18.03)	.43	2.47 (1.19-5.13)	.02	2.17 (0.27-17.35)	.46	2.35 (1.09-5.04)	.03
Health status								
Neither good nor poor	Reference	Reference	Reference	Reference	Reference	Reference	Reference	Reference
Poor	0.92 (0.27-3.16)	.89	1.34 (0.80-2.23)	.27	N/A	N/A	N/A	N/A
Good	1.04 (0.32-3.33)	.95	1.07 (0.65-1.78)	.78	N/A	N/A	N/A	N/A
Motivation to be involved in own care								
Not very much or a moderate amount	Reference	Reference	Reference	Reference	Reference	Reference	Reference	Reference
A lot or very much	3.00 (0.39-23.31)	.29	1.99 (1.04-3.82)	.04	X[d]	X	X	X
Frequency of CIE use								
Once a month or less	Reference	Reference	Reference	Reference	Reference	Reference	Reference	Reference
Once a week or more	0.92 (0.32-2.67)	.88	2.13 (1.41-3.23)	<.001	0.90 (0.27-2.95)	.86	2.03 (1.31-3.13)	.002

[a] Goodness-of-fit: χ^2_8=5.6; *P*=.69.

[b] Variable excluded from the final multivariable model using a backward elimination approach.

[c] N/A: not applicable; variable not entered into the multivariable analyses due to nonsignificance (*P*>.25) in univariate analyses.

[d] Variable excluded from the final multivariable model due to 0 cell counts producing unstable estimates.

Missing Data Analysis

Of 523 survey respondents, 78 (14.9%), who had previously used CIE, had missing data regarding age and gender, and these respondents were excluded from our analyses. Meaningful comparisons of sociodemographic characteristics between the missing data sample and the analysis sample were not possible due to considerable additional missing data in the group of 78 respondents excluded from this analysis (Multimedia Appendix 5). There were no differences in the distribution of responses between the analysis sample and the missing data sample for the questionnaire item “How has CIE changed how satisfied you are with your care?” However, patients included in the analysis were more likely to view the impact of CIE on overall quality of care favorably, compared with those in the missing data sample (χ^2_4=10.3; P=.04; Multimedia Appendix 6).

Discussion

Principal Findings

Although many portal users perceived no change with CIE use, a large proportion reported better care quality and greater satisfaction with their care. Around 30% patients perceived their care to be safer, more effective, and more efficient with CIE, and approximately 40% reported that the timeliness of appointments, diagnoses, and treatments had improved. The most positive patient-perceived changes were in the domain of patient centeredness: more than half of patients using CIE felt more in control of their health care and able to play a greater role in decision-making. Patients from ethnic minority groups, those with higher digital health literacy, and those using CIE more frequently were more likely to perceive improvements in overall care quality and greater satisfaction with care. Across ethnic groups, patients of Asian or British Asian ethnicity reported the greatest benefits of portal use in terms of improving care quality and satisfaction with care received.

Comparison With Wider Literature

These reports from users of a web-based patient portal in the United Kingdom are consistent with the findings of other patient experience studies in finding that many patients perceive a range of benefits associated with portal use [28-37]. To our knowledge, this is the first empirical study to map patients’ experiences against the 6 domains of quality of care to provide broad insight into the perceived effects of portal use from the patient perspective.

Regarding the domain of effectiveness, around 1 in 4 patients in our study believed that CIE use contributed to improving their overall health, and this finding echoes the results of other survey studies and meta-analyses of randomized trials [3,38]. We did not collect information about respondents’ medical histories; however, prior studies have shown that portal use may be particularly effective in supporting people with long-term conditions to improve their health, including those with diabetes and hypertension [3,38].

Existing evidence links patient portals to increased medication safety through patients possessing greater knowledge about their medicines, improved medication adherence, and increased reporting of medicine discrepancies [3,39-41]. Our study has shown that patients perceive additional safety impacts of web-based portals including improved accuracy of personal health information and detection of health record errors. Our previous work, together with studies conducted in the United States, has demonstrated that around 1 in 5 patients who access their web-based personal health records can, and do, notice errors in their records, and most patients would like to play an active role in rectifying these discrepancies [16,42]. Moreover, Blease et al [40] have shown that enabling informal carers to access the electronic health records of vulnerable patients (eg, people with serious mental illness) can help to prevent medication errors, delayed diagnoses, and other patient safety risks.

Regarding the efficiency domain, more than one-third of patients in our study perceived their own workload relating to their health had changed for the better. In a previous survey study in Canada, patients reported that web-based portals save time when scheduling appointments, patients needed to repeat themselves less during appointments, and portal use meant that patients could avoid unnecessary clinic visits [43]. Similarly, a review of randomized trials found a reduction in health care use (or no change) when patients have access to their electronic health records [3]. No experimental trials have investigated the impact of web-based portals on the timeliness of care delivery [3]; however, approximately 40% of the patients in our study perceived that CIE enabled them to access appointments, diagnoses, and treatment more quickly.

A growing body of evidence suggests that patients who are engaged in their care are more likely to adhere to medication and treatment plans, take up screening opportunities and prevention practices, participate in the detection of errors and safety risks, and adopt effective management strategies for chronic conditions [28,44-47]. The findings of this and numerous other survey studies have consistently found that patients feel more in control of their health care and better able to play a role in decision-making with access to their personal health records [28,33,34,37,40].

Regarding equity, our findings are consistent with previous research demonstrating that patients experiencing barriers to accessing web-based portals (including low digital literacy), and those with low levels of engagement in technology-enabled care are less likely to report that portals improve their health [38,48]. Previous research has also demonstrated that portal uptake is lower among patients belonging to ethnic minority groups [38]. However, in line with survey studies of portal users in the United States [28,29], we found that CIE users self-identifying to an ethnic minority group were more likely to report better care quality and greater satisfaction with care. Gerard et al [29] found that, compared with White patients, patients of Asian ethnicity in the United States were twice as likely to report the benefits of portal use; our study echoes this finding in the United Kingdom. Sharing electronic health records with patients appears to increase transparency and trust and strengthens the relationship between patients and their providers [44]. These benefits may be particularly important for ethnic minority groups to feel satisfied with their care; however, further qualitative research is needed to understand the mechanisms of portal adoption across different ethnic minority groups.

Of note, we found that patients who use CIE frequently were more likely to perceive improvements in overall care quality (and greater satisfaction with care). However, the direction of this effect is unclear. We suggest that this mechanism is likely to be circular, with initial portal use leading to perceived improvements in care quality, resulting in greater satisfaction with care, prompting increased portal use. In this way, the perception of quality of care could serve as a mechanism of sustained portal adoption. This theory is consistent with the Technology Acceptance Model, which suggests that use behavior (actual use) is partly predicated by the perceived benefits of using the technology [49]. In the study by Portz et al [50], which used the Technology Acceptance Model to explore portal use among older adults with chronic conditions, patient-perceived usefulness (communicating with care provider, saving time and money, addressing concerns without a clinic visit) was linked to frequent use of specific portal features, including the message center, pharmacy center, and viewing laboratory results. Further evaluation of CIE should include developing and testing a "Theory of Change" to determine how and why portal use leads to greater satisfaction with care in some patient groups [51].

Policy Implications and Future Research

This study confirms the importance of addressing "the digital divide" as a policy priority to ensure equitable access to the benefits of patient portals for all patients [14,52]. Crosscutting interventions with system impacts, including user-centric design of portal platforms that adhere to accessibility, legibility and readability standards, and a commitment to "safety net" strategies such as the provision of low-cost, Wi-Fi–enabled devices or patient outreach programs, could all help to ensure that traditionally underserved groups can benefit from portal use [40,53]. More work is required to understand the relative effectiveness of these interventions, such that equity of access and adoption can be achieved for all patients. However, beyond literacy and technology access, our findings suggest that there are other potential avenues for addressing health disparities by expanding patient portal use to underrepresented groups. That ethnic minority groups see greater benefits in accessing their personal health records is worthy of further careful inquiry. Further research using qualitative methods would help to elucidate the mechanisms of patient portal adoption among ethnic minority communities.

Strengths and Limitations

We mapped survey items to the Institute of Medicine's 6 domains to provide a broad overview of perceptions of care quality among CIE users. However, our questionnaire was not designed to evaluate the domains of care quality as multidimensional constructs. There is a need to develop instruments that can measure subjective accounts of care quality as seen through the patient lens; developing and validating such a questionnaire could be the focus of future work.

We recruited a diverse sample, with one-third of respondents residing outside London and 1 in 5 self-identifying to an ethnic minority group. However, the numbers of patients in subgroups of ethnic minority were small. As such, we combined categories of ethnicity for the multivariable regression. Research exploring issues of equity should disaggregate ethnic categories where possible so the experiences of different ethnic groups can be understood [54]. Although we ran cross-tabulations to explore differences between ethnic groups, the numbers were small and may not generalize to larger populations.

Our web-based recruitment strategy may have introduced selection bias because web-based survey studies may favor the inclusion of patients who are digitally literate and more able to fully engage with patient portals. Our sample only included users of a web-based portal, and our findings are based on patient self-reported and perceived changes in care quality based on portal use. As such, and due to the nature of the study design, we cannot make any causal claims about the impact of patient portals on the quality of care. Building on limited existing evidence from controlled trials [2,3], further experimental or quasi-experimental studies should test the relationship between patient portal use and care quality using validated end points.

Conclusions

A large proportion of CIE users perceived better overall quality of care and greater satisfaction with care, although many portal users reported no change. Perceived improvements were reported across all 6 domains of care quality, with the most favorable in the domain of patient centeredness. Patients from ethnic minority backgrounds (particularly Asian or British Asian) and those with higher digital health literacy perceived the greatest benefits of CIE use. With national policy directed toward addressing health disparities, patient portals could be valuable in improving care quality for patients in underrepresented groups, providing the needs of digitally disempowered patients are addressed. Further research should test the relationship between patient portal use and validated measures of the domains of care quality.

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Authors' Contributions

ALN, LF, MK, and EKM designed the study. ALN and LF administered the survey. RL conducted the analyses. RL drafted the manuscript. All authors contributed to the revision, editing, and approval of the final version of the manuscript.

Conflicts of Interest

None declared.

Multimedia Appendix 1

Cross-tabulation of patients' sociodemographic characteristics and their perceptions of the impact of Care Information Exchange on satisfaction with care.
[DOCX File , 42 KB-Multimedia Appendix 1]

Multimedia Appendix 2

The Strengthening the Reporting of Observational Studies in Epidemiology Statement—Checklist of Items That Should Be Addressed in Reports of Observational Studies.
[DOCX File , 328 KB-Multimedia Appendix 2]

Multimedia Appendix 3

Patients' perceptions of the impact of Care Information Exchange on (1) overall quality of care and (2) satisfaction with care.
[DOCX File , 20 KB-Multimedia Appendix 3]

Multimedia Appendix 4

Cross-tabulation of patients' ethnicity and perceived change in overall quality of care with Care Information Exchange use.
[DOCX File , 24 KB-Multimedia Appendix 4]

Multimedia Appendix 5

Sociodemographic characteristics of patients in the missing data sample and in the analysis sample.
[DOCX File , 25 KB-Multimedia Appendix 5]

Multimedia Appendix 6

Missing data analysis for questionnaire items.
[DOCX File , 25 KB-Multimedia Appendix 6]

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Abbreviations

CIE: Care Information Exchange
eHEALS: eHealth Literacy Scale
OR: odds ratio

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Original Paper

Prevalence of Poisoned Google Search Results of Erectile Dysfunction Medications Redirecting to Illegal Internet Pharmacies: Data Analysis Study

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Abstract

Background: Illegal online pharmacies function as affiliate networks, in which search engine results pages (SERPs) are poisoned by several links redirecting site visitors to unlicensed drug distribution pages upon clicking on the link of a legitimate, yet irrelevant domain. This unfair online marketing practice is commonly referred to as search redirection attack, a most frequently used technique in the online illegal pharmaceutical marketplace.

Objective: This study is meant to describe the mechanism of search redirection attacks in Google search results in relation to erectile dysfunction medications in European countries and also to determine the local and global scales of this problem.

Methods: The search engine query results regarding 4 erectile dysfunction medications were documented using Google. The search expressions were “active ingredient” and “buy” in the language of 12 European countries, including Hungary. The final destination website legitimacy was checked at LegitScript, and the estimated number of monthly unique visitors was obtained from SEMrush traffic analytics. Compromised links leading to international illegal medicinal product vendors via redirection were analyzed using Gephi graph visualization software.

Results: Compromised links redirecting to active online pharmacies were present in search query results of all evaluated countries. The prevalence was highest in Spain (62/160, 38.8%), Hungary (52/160, 32.5%), Italy (46/160, 28.8%), and France (37/160, 23.1%), whereas the lowest was in Finland (12/160, 7.5%), Croatia (10/160, 6.3%), and Bulgaria (2/160, 1.3%), as per data recorded in November 2020. A decrease in the number of compromised sites linking visitors to illegitimate medicine sellers was observed in the Hungarian data set between 2019 and 2021, from 41% (33/80) to 5% (4/80), respectively. Out of 1920 search results in the international sample, 380 (19.79%) search query results were compromised, with the majority (n=342, 90%) of links redirecting individuals to 73 international illegal medicinal product vendors. Most of these illegal online pharmacies (41/73, 56%) received only 1 or 2 compromised links, whereas the top 3 domains with the highest in-degree link value received more than one-third of all incoming links. Traffic analysis of 35 pharmacy specific domains, accessible via compromised links in search engine queries, showed a total of 473,118 unique visitors in November 2020.

Conclusions: Although the number of compromised links in SERPs has shown a decreasing tendency in Hungary, an analysis of the European search query data set points to the global significance of search engine poisoning. Our research illustrates that search engine poisoning is a constant threat, as illegitimate affiliate networks continue to flourish while uncoordinated interventions by authorities and individual stakeholders remain insufficient. Ultimately, without a dedicated and comprehensive effort on the part of search engine providers for effectively monitoring and moderating SERPs, they may never be entirely free of compromised links leading to illegal online pharmacy networks.

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KEYWORDS

internet pharmacies; search engine redirection; compromised websites; illegal medicines; patient safety; Europe; erectile dysfunction medications

Introduction

Background

The inherent practicality and convenience of online shopping are proving increasingly influential in consumer's behavior worldwide. Based on the 2020 e-commerce statistics published by Eurostat [1], 89% of all European Union (EU) citizens used the internet within the last 12 months, and 65% of individuals made an online purchase in the same period. Nonprescription medicine or dietary supplements accounted for 28% of these transactions, demonstrating consumers' growing trust in online health- and well-being–related purchases [1]. A large-scale study [2] of changes in information-seeking behavior showed that the most frequently mentioned content is "product information" and "purchase" (30% of all responses in 1997 and 2019), followed by "Health" (18% of all responses in 1997 and 19% in 2019) [2]. Notably, user behavior had been remarkably consistent in the span of 22 years [2].

The use of internet pharmacies and the number of individuals obtaining medications and various health products online are increasing [2]. Several advantages including perceived anonymity, cost savings, and convenience motivate individuals to purchase medications online [3]. Furthermore, the lack of a valid prescription required by legal online and offline vendors is a strong driving force toward illegal online drug purchases [3]. However, several patient safety risks are linked to the procurement of medicines outside the traditional supply chain, including questionable sourcing, poor product quality, substandard and falsified medicines, improper storage, and transportation [4]. Risks are augmented by rogue internet pharmacies considered as a primary source of substandard and falsified medical products in developed countries [5-7].

The widespread availability of search engines and increased public interest in obtaining medicines online imply a major dilemma, whether consumers aiming to purchase medications from the internet are starting their online activity from relevant web pages (eg, a national authority website), or simply searching using their search engine of choice. Most likely the latter is the case. Search engines refer consumers to relevant online resources quickly. Their significance is illustrated by the fact that most trackable website traffic originates from search engines [8], and typically from Google as this platform is handling more than 90% of search queries worldwide. Online distributors choose to use several digital marketing techniques to attract customers via search engines. Website operators apply various search engine optimization (SEO) techniques to improve the visibility of their websites, a practice that is accepted and supported by search engines [9]. SEO is a complex and time-consuming procedure, especially in the international marketplace in which country- and language-specific optimization is required to reach a high-ranking position among organic query results.

For illegal medicine sellers, conventional SEO is neither cost-nor time-effective, as they are constantly threatened with regulatory closure [10]. Furthermore, paid advertisements offering prescription drugs without a prescription by unauthorized pharmacies cannot appear in any of the major paid search advertising services [11,12]. Therefore, alternative dishonest digital marketing methods including web spamming, forum abuse, and additional "black hat" SEO techniques are used by illegal drug distribution websites to promote their links in the unpaid search engine results pages (SERPs) to gain favorable search engine rankings [13,14].

As a result, the user's query on a search engine may contain both "normal" domains (ie, those related to the query) and "compromised/deceptive" domains (ie, ones that are unrelated to the query). The latter domains are promoted in the rank using "black hat" SEO methods, undermining the value proposition of search engines, as search results are presented with deceptive views of a website with inflated relevance to selected search terms. Individuals (search engine users) are referred to low-quality content or malicious websites when clicking on a deceptive search result. Consequently, the deceptive web pages practically "poison" the search result; therefore, this technique is termed as "search engine poisoning" or "search redirection attack" [9,15].

Manipulation of search results for erectile dysfunction medications was published nearly a decade ago by Leontiadis et al [15,16] and Wang et al [17]. Sildenafil was the first commercially available phosphodiesterase type 5 (PDE5) inhibitor available since 1998, followed by vardenafil, tadalafil, and avanafil [18]. Increasing prevalence of erectile dysfunction and widespread use of PDE5 inhibitors as the first-line oral treatment worldwide [19] have resulted in growing demand, which illegal online vendors have been taking advantage of [20].

Objectives

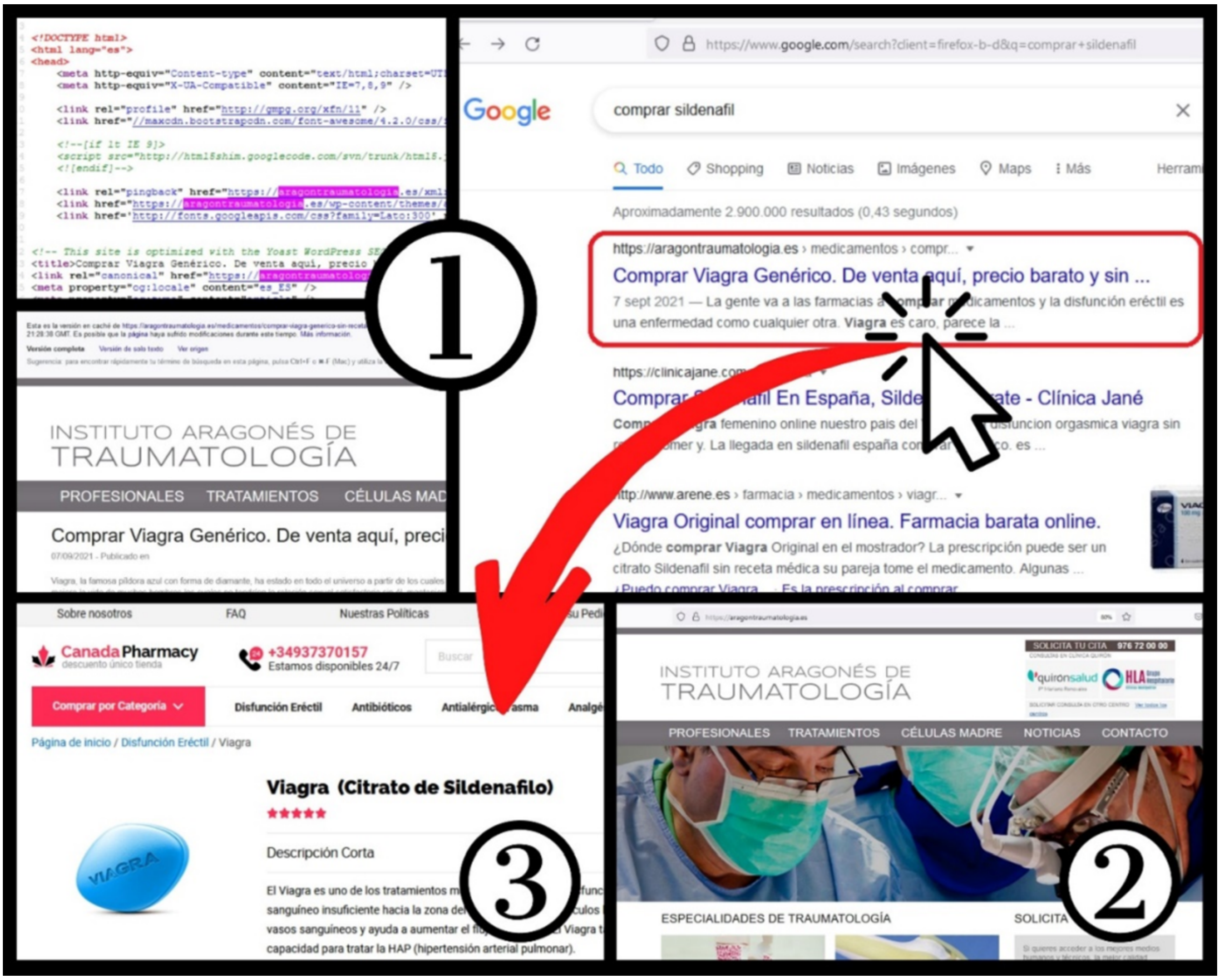
The major aim of our study is to introduce the relatively unknown but significant and persistent issue of poisoning of search engine results (SERs) of erectile dysfunction medications in European countries. Furthermore, the study is meant to measure the scale of the problem and illustrate the redirection networks referring users (patients) to illegal internet pharmacies. Public health significance of the problem is illustrated by the estimation of the likelihood of consumers clicking on poisoned search results and the number of monthly visitors redirected to illicit pharmacy networks. Our utmost aim is to warn the general public and raise the awareness of authorities and law enforcement agencies, thus facilitating long-awaited countermeasures.

Methods

Mechanism of Search Engine Poisoning and Redirection

A search engine poisoning attack begins with an attacker hacking into a vulnerable web page. Common targets are outdated, vulnerable, or complex content management and blogging systems (eg, WordPress; see Figure 1, part 1). Once the attacker has access to the system, a new code is injected, and the hacked website will “interrupt” all incoming HTTP requests to the original web page and respond to these requests differently from the original operation [15]. Typically, users are redirected through a redirection chain, consisting of intermediate pages to a final page. The destination is the illegal pharmacy website most users are unwillingly visiting. However, users do not see the original content of the compromised website after clicking on the search results, because they are presented with the unwanted final page, as hacked websites redirect the web browsers within milliseconds. Redirection attacks—identifiable in various search engines such as Google, Bing, and Yahoo!—disregard term relevance constraints and target search terms of the actual search; however, at the same time, the original content of the hacked website (domain) becomes irrelevant to the search terms used (see Figure 1, part 2).

Figure 1. Illustrative figure of how users pass through a redirection chain from the search result page to the final destination illegal online pharmacy website.



In the case of search engine poisoning attack, it is important that compromised websites look differently, depending on the visitor, due to the so-called cloaking method [13]. The original content stuffed with keywords and links to increase page rank is shown to the automated agent/crawler (eg, Google bot), meanwhile the redirected illegitimate online vendor is displayed to the customer (see Figure 1, part 3) [16]. Currently no efficient technique capable of identifying all spam web pages is available [13]. Because of the cloaking method used by the illegitimate pharmacy operators, the automation of the content evaluation of SERs is difficult and precise detection requires manual assessment or checking.

Obtaining and Evaluating SERs in National and International Data Sets

Search engine query results and links were documented and manually evaluated to simulate and evaluate what consumers see while browsing. Manual data acquisition was necessary as automatic search queries are prohibited by search engine providers and cloaking is difficult to identify automatically. The focus of the research was on erectile dysfunction medications as a popular category affected by illegal online trade and potential source of substandard and falsified medicinal products [20,21]. Consequently, the search queries represent purchase intent (buying prescription medications online), rather than informative types of search (looking for product information). The 4 primary active pharmaceutical ingredients (APIs), sildenafil, tadalafil, vardenafil, and avanafil, were searched for using Google, the most popular search engine. Country-specific data were obtained by individualizing national search using the search terms of the "API" and the "buy" words in the language of the given country (eg, "comprar sildenafil" for Spain). Furthermore, search settings in Google have been adjusted to the preferred region. To track the evolution of the phenomena, the first 20 organic SERs were evaluated during 3 consecutive years: August and October 2019, August 2020, and November 2021 for the national data set. Meanwhile, the first 40 SERs were included in the international data set evaluated in November 2020. Accordingly, we conducted our research on 2 data sets: a long-term evaluation of Hungarian SERs and an international sample in Hungary and an additional 11 other countries (Bulgaria, Croatia, Estonia, Finland, France, Greece, Italy, Romania, Spain, Sweden, and United Kingdom) from different regions of Europe. As most (88%) users click on results appearing in the top 10 SER positions [22], by documenting the top 20 results we consider our findings representative for online queries at the time of evaluation. SER links of websites offering medicinal products for sale were included for evaluation; nonrelevant query results were excluded from our evaluation.

The documented search result data included date, country, search language, API, search phrase, URL and domain name, SER ranking, destination website URL for redirections, and website category. Two figures were used to describe the significance of the phenomena regarding search engine redirection attacks in SERs: (1) prevalence of hacked links in SERPs and (2) cumulative click-through rate (CTR). Both measures correlate with the likelihood of users—intentionally or unintentionally—visiting illegal pharmacies. Prevalence is calculated by dividing the number of infected links by the total number of evaluated links in SERPs. Based on Google's organic search ranking, CTR is a probability value of clicking on a given link assigned to each measured SER position. On the first page of the search (Google) result, 1-10 CTR per ranking values were determined based on the analysis by Sistrix [22], while further CTRs for 11-40 SER positions were computed with the equation of the exponential trend line connecting the first 1-10 SERP datapoints ($y=26{,}76e^{-0.258x}$, where y is the predicted CTR and x is the SER rank; R^2=0.927). Cumulative CTRs express the sum of CTR values regarding all documented positions in SERPs.

Compromised sites redirecting to international illegal medicine retailers have been classified into 3 categories referencing the redirection's life cycle based on Leontiadis et al [16]. First, the compromised site is likely a future redirect (hacked website content with or without links; however, no automatic redirection is yet observed). Second, active redirection to an international illegal medicinal product vendor via a compromised site. Lastly, inactive redirection, that is, sites used to be redirecting, but no longer redirecting, because they are not accessible at the time of evaluation, displaying 404 error code, or similar.

Graph Visualization, Legitimacy, and Traffic Analysis Regarding Destination Websites

Compromised SERP links leading to international illegal medicinal product vendors via redirection (active links) were evaluated and networks have been generated with Gephi [23], an open-source graph visualization and analysis tool. The national and international data sets were visualized as directed graphs illustrating the source and destination website domains. Multiple links from the same domain accounted for increased weight of the edge. The average degree (average number of edges per node in the graph), the in-degree (number of connecting edges), and the page rank (importance score of a node within a directed graph) of nodes were computed.

Destination websites offering products for sale in the national data set were categorized as follows: legitimate online pharmacies, illegal medicine retailers (rogue online pharmacies), or dietary supplement seller (nonpharmacy web shops). Destination website categories were not defined for EU countries, so only links with redirection to illegal online sellers were documented regarding the international data set. Destination website legitimacy was checked at LegitScript [24] and categorized as approved, unlicensed, or rogue (illegitimate). The estimated number of monthly unique visitors of the root domain for all regions at the time of evaluation is provided by SEMrush traffic analytics [25].

Data were analyzed using SPSS Statistics 26 for Windows (IBM Corp.) and MS Excel (Microsoft Inc.).

Ethical Considerations

There were no ethical issues, as only publicly available data obtained from SEs and websites were documented and evaluated. Furthermore, no customer or personal data were measured, recorded, or stored in this study.

Results

Compromised Websites Among SERPs of Medications for Treating Erectile Dysfunction in Hungary Between 2019 and 2021

The results show that during our 3-year observation period, there were no legitimate internet pharmacy websites among the evaluated SERPs. A decrease in the number of compromised sites linking visitors to illegitimate medicine sellers has been observed during our study period, while inaccessible broken links have increased. Similarly, the number of national rogue online pharmacies has increased in SERs up through 2021. All active ingredients have been affected by poisoning, with avanafil showing a somewhat diminished prevalence (Table 1).

Table 1. Top 20 search engine results page link categories for 4 erectile dysfunction medications.

Link category	August 2019, n (%)	October 2019, n (%)	August 2020, n (%)	October 2021, n (%)
Legitimate online pharmacy (n=80)[a]	0 (0)	0 (0)	0 (0)	0 (0)
National illegal medicinal product seller (n=80)	8 (10)	12 (15)	16 (20)	34 (43)
International illegal medicinal product vendor via compromised site and redirection (active; n=80)	43 (54)	33 (41)	25 (31)	4 (5)
Avanafil (n=20)	9 (45)	5 (25)	3 (15)	0 (0)
Sildenafil (n=20)	12 (60)	9 (45)	6 (30)	1 (5)
Tadalafil (n=20)	12 (60)	9 (45)	8 (40)	1 (5)
Vardenafil (n=20)	10 (50)	10 (50)	8 (40)	2 (10)
Compromised site without redirection (n=80)	5 (6)	3 (4)	1 (1)	0 (0)
Not accessible (eg, 404) at the time of evaluation (n=80)	2 (3)	7 (9)	9 (11)	15 (19)
Dietary supplement web shop (n=80)	9 (11)	10 (13)	14 (18)	8 (10)
Other sites not offering products for sale (n=80)	13 (16)	15 (19)	15 (19)	19 (24)

[a] According to national regulations, legitimate online pharmacies in Hungary cannot offer prescription medications—including oral medications for erectile dysfunction—via the internet.

Although most of the compromised websites were “true redirects” transferring individuals to international online sellers, we occasionally came across hacked sites without redirection. For example, in these cases, the rogue online pharmacy was operating under a subpage of the hacked domain, or the medication-related text was filled with keywords and links (so-called keyword stuffing and link building), indicating “black-hat” SEO techniques.

Such pages are likely to rank higher in search engines and develop redirects as time passes. In other instances, the web page we were looking for did not exist on the website’s server. Pages not accessible (eg, 404 error) at the time of evaluation could be related to website administrators identifying the malicious redirect code inserted into a website. According to our observation, hacking is followed by the malicious redirection life cycle, which consists of future (inactive pages ready to become active), active, and finally inactive stages.

The complexity of the graphs decreased (the average degree changed from 1.17 to 0.667), between August 2019 and October 2021 (Figure 2). A majority (11/14, 79%) of the evaluated online pharmacies were categorized as rogue by LegitScript. We identified 5 destination online pharmacy websites in the link network at each evaluation date, except for October 2021. Initially, destination domains (eg, acs-pharmacy.com and evo-pharmacy.com) received numerous incoming links from SERs and played a central role in the network. By the end of the 3-year evaluation period, illegal pharmacy websites in-degree and page rank values underwent substantial reduction (Table 2). Website traffic analytics by SEMrush indicated a high number of monthly visitors (range 370-155,400) for important nodes with high page-rank values within the graph. This value illustrates the destination site’s global visitor count in the given month of evaluation.

Figure 2. Visual graph of SERP links of compromised websites and illegal online medicine vendors accessed via search redirection attack visited in August 2019 (left) and August 2020 (right). SERP: search engine results page.

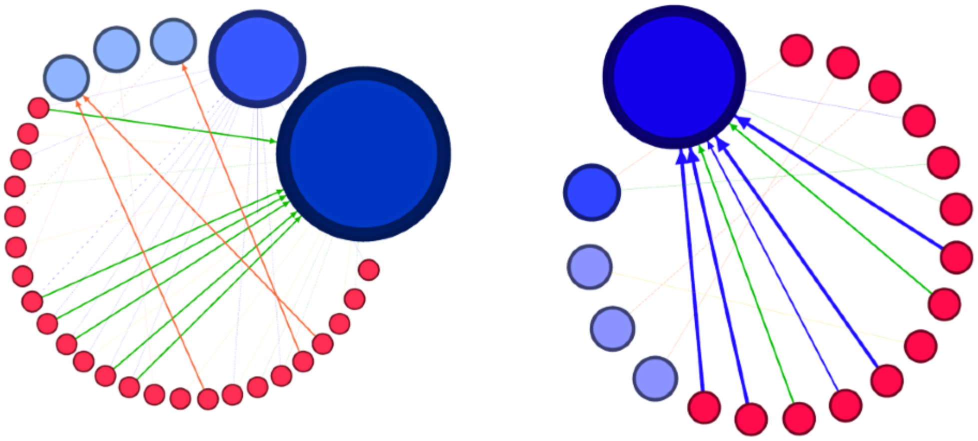


Table 2. Graph statistics, legitimacy rating, and traffic history regarding referred illegal medicine vendors for Hungarian erectile dysfunction medication search queries.

Domain accessed following search redirection attack	Date	In-degree[a]	Page rank[b]	Legitimacy rating (LegitScript)	Number of unique visitors per month (SEMrush)[c]
acs-pharmacy.com	August 2019	16	0.209	Rogue[d]	155,400
acs-pharmacy.com	October 2019	16	0.332	Rogue	117,000
1-pharm.com	August 2019	12	0.140	Rogue	11,000
specialmedassortment.com	August 2019	2	0.054	Rogue	3600
myworldpharma.com	August 2019	2	0.054	Not in database	4000
pharmpillsonline.com	August 2019	2	0.054	Rogue	800
herbsandmeds.com	October 2019	2	0.061	Rogue	5200
pharmrx-1.com	October 2019	2	0.051	Rogue	6500
cheap-pharma.com	October 2019	1	0.042	Rogue	5100
big-pharmacy.com	October 2019	1	0.032	Rogue	15,600
evo-pharmacy.com	August 2020	9	0.279	Rogue	83,400
evo-pharmacy.com	October 2021	2	0.574	Rogue	30,400
eu-pharm.de	August 2020	2	0.087	Not in database	370
ezshopremedieshere.com	August 2020	1	0.059	Not in database	Not in database
canadarx24h.com	August 2020	1	0.059	Rogue	5200
medsalltheworld.com	August 2020	1	0.059	Rogue	3100

[a] In-degree value shows the number of links adjacent to a domain.

[b] The page rank algorithm measures the importance of each node within the graph.

[c] The estimated number of monthly unique visitors of the root domain for all regions at the time (month) of evaluation provided by SEMrush traffic analytics.

[d] Rogue: online pharmacy website engaged in illegal activity; a rating determined by LegitScript.

International Relevance of Compromised SERPs in Europe 2020

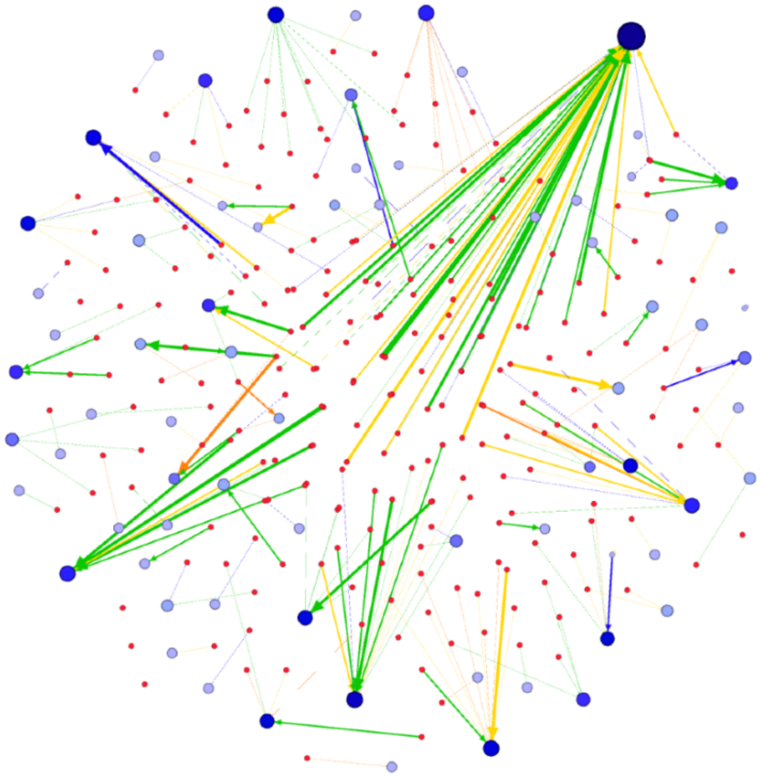
A total of 1920 search results were evaluated in November 2020, in accordance with the results of the aforementioned 4 APIs listed in the top 40 results on the SERP pages throughout 12 European countries. Of those, 380 (19.79%) search query results were compromised, with a majority (n=342, 90%) of the links of the 230 infected source domains redirecting individuals to 73 international illegal medicinal product vendors. The remaining SER links were leading to compromised sites without redirection (6/380, 1.6%) or not accessible web pages/sites (32/380, 8.4%). Descriptive graph statistics of the international data set, website legitimacy category, and traffic history regarding destination online pharmacies with at least five referring links are depicted in Table 3.

The most influential destination domain in the international redirection graph was “ezshopremedieshere.com,” with 79 referring links from search queries in most (8/12, 66%) of the evaluated European countries, and 61,400 unique global visitors in November 2020. Although several destination websites had numerous incoming links, the average in-degree value was 1.11, as most nodes had only 1 (30/79, 38%) or 2 (12/79, 15%) compromised referrals from search engines (Figure 3). The number of monthly global visitors per domain was the highest for “forecastarrays.us,” “cheapshopmed.com,” and “haiyuanpenguan.com,” attaining 566,100, 135,100, and 128,300 visitors, respectively, according to SEMrush traffic analytics. Interestingly, these high-traffic domains had only a small number (1-3) of incoming links from SERs and only 1 European country was affected in each case (Finland, Estonia, and Croatia, respectively). The “cheapshopmed.com” domain is a rogue online pharmacy in the LegitScript database. However, the “forecastarrays.us” and “haiyuanpenguan.com” domains contain compromised pages, including their intended content, and they can be accessed after redirection with an embedded online pharmacy content, so the visitor count of these domains is likely to include nonmedicinal purchase intention also. Website traffic estimation was available for 40 destination domains, with 35 having pharmacy-specific domain names (including terms, such as Rx, pharm, meds, pills). These 35 active online pharmacy domains, accessible from 12 European countries via compromised links in search engine queries, included a total of 473,118 unique visitors during November 2020.

Table 3. Graph statistics, legitimacy rating, and traffic history regarding selected referred illegal medicine vendors for erectile dysfunction medication search queries in 12 European countries (November 2020).

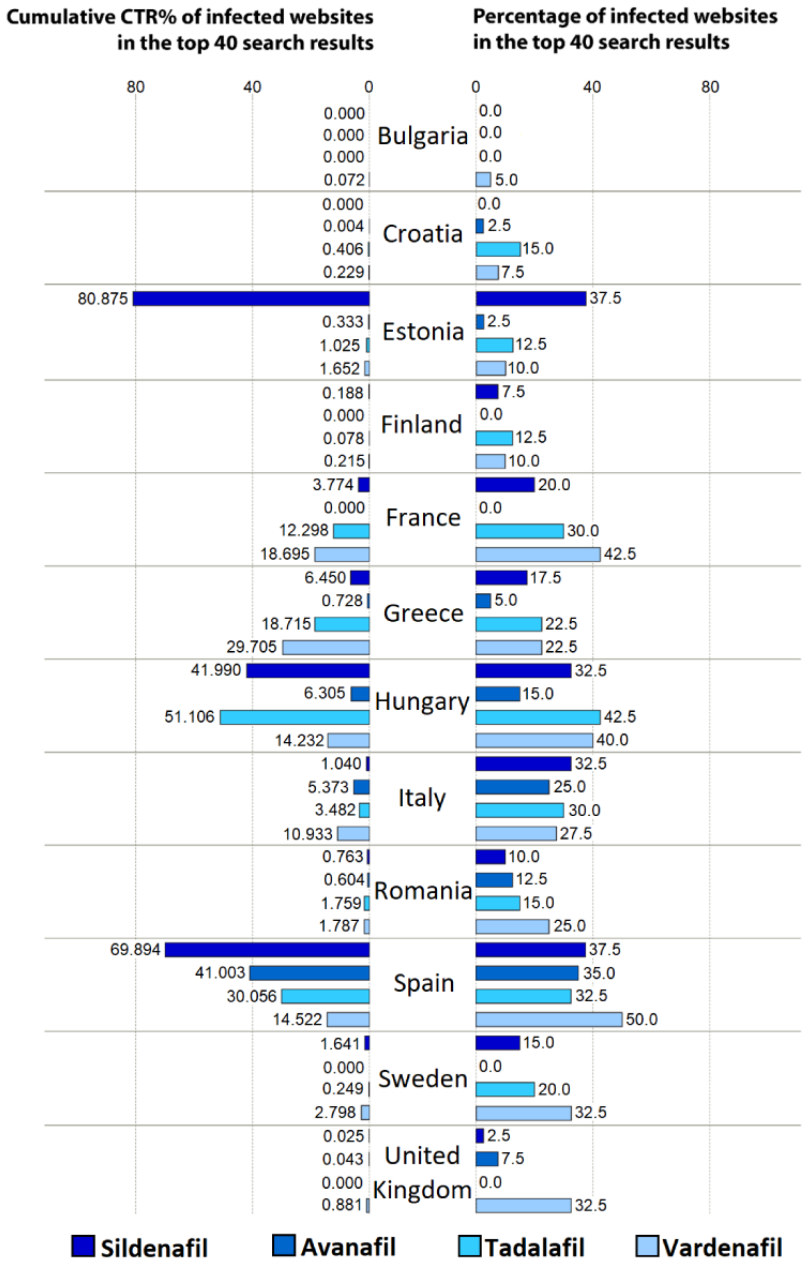
Domain accessed following search redirection attack	In-degree	Page rank	Countries affected	Legitimacy rating (LegitScript)	Number of unique visitors per month (SEMrush)
ezshopremedieshere.com	79	0.080	Croatia, Estonia, France, Greece, Hungary, Italy, Spain, Sweden	Not in database	61,400
evo-pharmacy.com	20	0.017	Hungary	Rogue	Not in database
rx-qualityshop.com	19	0.023	Croatia, Estonia, Finland, Romania, Sweden	Rogue	Not in database
your-meds-store.com	14	0.013	Croatia, Estonia, Finland, Greece, Italy, Romania, Spain	Rogue	4600
onlinepharmacyhub.com	13	0.018	Croatia, UK, Estonia, Romania	Not in database	2300
overnightpharm.com	11	0.015	UK, Estonia, France, Italy, Spain, Sweden	Rogue	321
rx-24-online.com	10	0.018	UK, Sweden	Rogue	Not in database
hot-med.com	9	0.017	Estonia, Spain	Rogue	21,500
usamedicineget.com	8	0.005	Croatia, Estonia, Romania	Rogue	5000
igohealth365.com	8	0.012	UK, France, Italy, Spain	Rogue	Not in database
qualitypillsprovider.com	7	0.007	Hungary, Spain, Sweden	Rogue	519
meds-store-24h.com	7	0.010	Finland, Greece, Italy, Spain	Rogue	7800
pills-group.com	6	0.010	Italy	Not in database	Not in database
vipcanadianstore.com	6	0.008	France, Italy, Sweden	Rogue	Not in database
online-secure-shop24h.com	6	0.009	Bulgaria, Greece, Italy, Spain	Rogue	8400

Figure 3. Graph of compromised websites (n=230) and illegal online medicine vendors (n=73) accessed via search redirection attack in 12 European countries visited in November 2020. Node size—represented by circles—illustrate the in-degree property of a domain in the graph. Small red nodes show compromised website domains in SERs and destination websites are labeled with blue. The edge—representing links—are colored based on the API name used in search queries (blue for sildenafil, green for vardenafil, yellow for tadalafil, and orange for avanafil). API: active pharmaceutical ingredient; SER: search engine result.



The EU countries are affected differently by redirection links within SERPs, leading to illegitimate online pharmacy websites (Figure 4). In the "Methods" section, we proposed 2 metrics to illustrate the magnitude of the problem manifested throughout European countries. The proportion of the hacked pages as a percentage of the total search query results and the cumulative CTR percentages were calculated to illustrate the issue of the compromised websites in a complex manner in each country's SERP. It is important to view cumulative CTR and the number of compromised websites as both unique and complementary factors. To state an example, if a country's SERP has several websites lower down the list, the cumulative CTR will be minimal. However, these websites pose a potential risk of rising surreptitiously quickly through the ranks and gaining higher CTRs.

Figure 4. Cumulative click-through rate (CTR) prevalence of redirection links within search engine result pages leading to illegitimate online pharmacy websites search queries in 12 European countries.



Compromised links redirecting to active online pharmacies were present in search query results of all evaluated countries. The prevalence of compromised links in national SERs was the highest in Spain (62/160, 38.8%), Hungary (52/160, 32.5%), Italy (46/160, 28.8%), and France (37/160, 23.1%), whereas it was the lowest in Finland (12/160, 7.5%), Croatia (10/160, 6.3%), and Bulgaria (2/160, 1.3%). Cumulative CTR values computed for APIs indicated the highest potential impact and danger of search engine redirection attacks for avanafil in Spain (41.0%), sildenafil in Estonia (80.9%), tadalafil in Hungary (51.1%), and vardenafil in Greece (29.7%). Prevalence and cumulative CTR metrics were relatively high for all APIs in Hungary and Spain, indicating a larger number of infected SER links with relatively high-ranking positions in search queries. Accordingly, consumers searching for erectile dysfunction medications online are more likely affected by online medicine purchase opportunities presented by illegal online pharmacies applying search engine redirection attack as a marketing technique in these countries. Although SERs in Romania, Finland, and Greece contain a substantial number of compromised links, because of low rankings, the cumulative CTR vales are low, indicating that consumers are less likely to click on compromised links leading to the destination illegal online pharmacy websites. The complete redirection network is illustrated in Figure 3.

Hacked websites are not specialized in active ingredients and target domains. Of the observed 230 infected source domains, many (n=65, 28.3%) promote various APIs. Although the majority (160/230, 69.6%) of source infections drive traffic to a single destination, many redirect individuals to various online pharmacy websites (range 1-6; mean 1.49 redirection links of independent destination domains).

Discussion

Principal Findings

The evolution of online advertising methods and specialization have led to the development of affiliate networks, an established method for legitimate merchants in which sponsors pay a commission to advertisers delivering traffic to their websites. Unfortunately, illegal online pharmacies are also a typical example of affiliate networks and search engine poisoning is a tool linked to affiliates to convert visitors from search engines. A robust number of independent affiliates, acting as advertisers or traffic brokers, received high (30%-40%) commissions for promoting illegal medication vendors and delivering traffic to the sponsor websites in which medications are sold to customers [14]. This affiliate program business model has numerous advantages for its participants. Sponsors (destination illegal pharmacy websites) do not have to heavily invest in marketing campaigns. Even more advantageous is that they free themselves from direct exposure to the criminal risks associated with large-scale advertising. Affiliates generate sales for sponsors by only focusing on attracting customers without developing web shops, customer service, etc. Online pharmaceutical sales are one of the oldest and largest affiliate program markets, with an estimated turnover of 500,000-600,000 customers, 700,000 billed orders, and US $73,000,000-85,000,000 revenue per 3-year period (2007-2010) analyzed by McCoy et al [14] referencing 2 major affiliate networks (Glavmed and SpamIt). By evaluating the change of new customer acquisitions, the authors concluded that affiliate programs attract new customers at a steady rate (approximately 3300/week). Thus, the market of counterfeit pharmaceuticals was not saturated, suggesting latent customer demand [14]. Furthermore, the same data set provides evidence for customer loyalty and satisfaction regarding online pharmacies, as repeat purchases constitute more than 20% of overall revenue. Our previous findings also indicate that a vast number of online pharmacies operate illegally and offer medicines to buyers in the long run [10].

It has been estimated that the number of men experiencing erectile dysfunction worldwide can reach 332 million by 2025 [19]. Erectile dysfunction medications containing PDE5 inhibitors are highly prone to falsification with proven potential health risk for patients. Analytical investigation of these products often shows the presence of dangerous excipients of nonpharmaceutical origin or quality, more than 1 undeclared PDE5s, and active ingredient amounts higher than declared values often surpassing the maximum therapeutic dose [5]. Previous research [26] regarding patient safety risks assessment of the online market of medicinal products revealed that Google search results include several suspicious links. By clicking on these SERs, the visitor is apparently redirected to an unlicensed drug distribution page by initially clicking on the link of a legitimate, yet irrelevant domain. This unfair online marketing of search redirection attack is thought to play a decisive role in the illegal internet pharmaceutical marketplace. Although search engine redirection attacks leading visitors to illegal online pharmacy networks have been previously published [9,16], we did not find relevant publications in medical informatics journals during the past decade. Admittedly, search engine redirection attacks are not limited to Google, the most popular search engine. The same phenomena could be identified in Microsoft Bing and Yahoo!. Seemingly, this unsolved issue has sunk into oblivion. This study was aimed to describe, map, and highlight its national and international significance.

Nearly half of search results were redirecting individuals to illicit medicine vendor sites during our national results obtained in 2019, with compromised websites being dominant in SERPs. This finding correlates with a previous study by Leontiadis et al [16], highlighting how redirections constitute the most significant proportion of results for the query set implemented in this study. Although the prevalence of compromised links in SERs and the complexity of the graphs have decreased in our national data set between August 2019 and October 2021, the danger has not dissipated. Consumers searching for ivermectin during the COVID-19 pandemic were more likely to find links redirecting to illegal medicine retailers that represent 73.3% of SER links within the first 30 search results in Google in March 2021 [26]. Despite the attempts to prevent this “black hat” SEO technique proposed a decade ago, limited success can be observed [9], and we are facing a constant issue that has not been solved for a relatively lengthy period.

Our international search query data set obtained from a representative sample of SERs among 12 European countries illustrates the international significance of search engine poisoning. All evaluated countries are affected, as at least one of four active ingredients for the treatment of erectile dysfunction was offered for sale via compromised links. The overall prevalence of hacked links in SERs was highest among Spain, Hungary, Italy, and France. Among 1920 manually evaluated links, we documented 380 compromised results from a total of 230 websites (domains) leading to 73 illegal online medicine vendors. The majority of these illegal online pharmacies (41/73, 56%) received only 1 or 2 compromised links. Meanwhile, the top 3 domains with the highest in-degree property received more than one-third of all incoming links. These findings support earlier studies stating that illicit advertising business is dominated by only a handful of big-league players [16].

An important implication regarding our findings is that search-redirection attackers use a complex system with potentially vulnerable elements to convert traffic to their illegitimate destination websites. We conclude that such practices can be disrupted by various stakeholders in a number of ways (Textbox 1).

Most likely, if any 1 or more than 1 of the aforesaid measures are considered, the redirection network collapses, and infected source websites will not appear, nor will they rank high in the search results. Lastly, they will not actively redirect to illegitimate online pharmacy domains.

A common feature of the aforesaid measures is the undisrupted continuity of the system, as it most likely requires time to build up such a complex network among numerous stakeholders. Findings of previously published literature suggest that the median survival time of a source infection is 19 days; however, some claim a lot lengthier time (17% of infections lasted at least six months, while 8% survived for more than 1 year) [16]. Our findings also corroborate this, as 4 compromised pages in our national data set remained in the top 20 results for more than 2 years, between August 2019 and October 2021.

Textbox 1. Possible solutions to overcome search-redirection poisoning redirecting to illegal internet pharmacies.

- Search providers and authorities can identify compromised links by monitoring popular medicinal product–related search terms (eg, brand or active ingredient name of prescription medications), as infected websites contain numerous relevant keywords and links to rank high in search engine results pages (SERPs) for popular queries and to publicize themselves.
- In addition to manual evaluation of SERPs, previously published link-based and content-based algorithms as well as tailor-made automatic detection and classification engines can be used as benchmarks in the effective identification of pharma scam campaigns [27].
- Search engine providers play a decisive role in monitoring and moderating SERPs. Without their dedicated and comprehensive effort, SERPs may never be free of compromised links leading to illegal online pharmacy networks. Automated URL-based classification methods, similar to deSEO [28] proposed in 2011, can only be applied if search engine providers provide search query logs to authority or academic parties.
- If operators fail to identify the infection, compromised websites remain among the top results and maintain the functionality of redirecting. Consequently, the operators of vulnerable legitimate domains should be notified so that they can take action to improve content management system security and remove hacked pages.
- The intermediate redirection chain elements need to remain operational for effective redirection and search engine optimization, so when the webmaster removes the infection triggering the redirection, or any intermediary page, the redirection chain ceases to function.
- The destination illegitimate online pharmacies must stay online to remain operational. Therefore, drug authorities and law enforcement agencies can shut down final destination domains of rogue online pharmacies with a high number of incoming links and unique visitors.

As the number of infected websites appearing in SERPs and all other compromised websites within the redirection chain is considerably high and the number of destination websites are relatively low, it is reasonable to take measures against the latter by shutting down websites and domains. However, the efficacy of this intervention does not seem to be efficient enough, considering the fact that the Operation Pangea coordinated by Interpol has taken down more than 150,000 websites between 2008 and 2020. Despite this large-scale removal, an extremely large number of links (113,020 websites and online marketplaces) were subsequently closed down in 2021 [29,30], demonstrating the substantial scale and recurrence of this issue, which remains unresolved.

Limitations

Admittedly, our study bears several limitations, for instance, the search query results of only 1 search engine have been summarized; however, we believe that the validity of our methodology can be explained by the dominant market share of the search engine. Furthermore, as opposed to brand-name queries, API-based search may offer varied results; however, Google’s complex algorithm is likely to provide results for related searches. API was used because our aim was to find all relevant websites, regardless of their original and generic names, varying from country to country, including unapproved generics and falsified medicines. Legitimacy of all final destination websites cannot be evaluated objectively, as there is no reliable database to evaluate all websites. However, we assumed all online medicine vendors using search engine redirection attack to attract customers and offer prescription medicines for sale most likely bear malicious intent and can be categorized as illegitimate online pharmacies.

In conclusion, our results illustrate that the phenomena of search engine poisoning have been persistent during the past decade and affiliate networks linked to illegitimate online pharmacies are flourishing. This supports the presumption that uncoordinated interventions aiming at ceasing illicit medicinal online purchases by authorities and individual stakeholders are not yet sufficient. It is a problem that has not been solved for more than a decade. Importantly, uncontrolled illegal sale of medications has many unfavorable consequences for the health of consumers and the safety of the pharmaceutical supply chain. Detecting and eliminating malicious links promoting illegal online pharmacies in search engines are of great importance with regard to cybersecurity and patient safety.

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Authors' Contributions

AF was responsible for conceptualization, methodology, writing of original draft, and supervision. PP was responsible for study investigation. ARA performed formal analysis, writing of original draft, and visualization. AP was responsible for study visualization. PI contributed to conceptualization and writing—review and editing.

Conflicts of Interest

None declared.

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Abbreviations

API: active pharmaceutical ingredient
CTR: click-through rate
EU: European Union
PDE5: phosphodiesterase type 5
SEO: search engine optimization
SER: search engine result
SERP: search engine results page

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Review

Digital Devices for Assessing Motor Functions in Mobility-Impaired and Healthy Populations: Systematic Literature Review

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Abstract

Background: With the advent of smart sensing technology, mobile and wearable devices can provide continuous and objective monitoring and assessment of motor function outcomes.

Objective: We aimed to describe the existing scientific literature on wearable and mobile technologies that are being used or tested for assessing motor functions in mobility-impaired and healthy adults and to evaluate the degree to which these devices provide clinically valid measures of motor function in these populations.

Methods: A systematic literature review was conducted by searching Embase, MEDLINE, CENTRAL (January 1, 2015, to June 24, 2020), the United States and European Union clinical trial registries, and the United States Food and Drug Administration website using predefined study selection criteria. Study selection, data extraction, and quality assessment were performed by 2 independent reviewers.

Results: A total of 91 publications representing 87 unique studies were included. The most represented clinical conditions were Parkinson disease (n=51 studies), followed by stroke (n=5), Huntington disease (n=5), and multiple sclerosis (n=2). A total of 42 motion-detecting devices were identified, and the majority (n=27, 64%) were created for the purpose of health care–related data collection, although approximately 25% were personal electronic devices (eg, smartphones and watches) and 11% were entertainment consoles (eg, Microsoft Kinect or Xbox and Nintendo Wii). The primary motion outcomes were related to gait (n=30), gross motor movements (n=25), and fine motor movements (n=23). As a group, sensor-derived motion data showed a mean sensitivity of 0.83 (SD 7.27), a mean specificity of 0.84 (SD 15.40), a mean accuracy of 0.90 (SD 5.87) in discriminating between diseased individuals and healthy controls, and a mean Pearson *r* validity coefficient of 0.52 (SD 0.22) relative to clinical measures. We did not find significant differences in the degree of validity between in-laboratory and at-home sensor-based assessments nor between device class (ie, health care–related device, personal electronic devices, and entertainment consoles).

Conclusions: Sensor-derived motion data can be leveraged to classify and quantify disease status for a variety of neurological conditions. However, most of the recent research on digital clinical measures is derived from proof-of-concept studies with considerable variation in methodological approaches, and much of the reviewed literature has focused on clinical validation, with less than one-quarter of the studies performing analytical validation. Overall, future research is crucially needed to further consolidate that sensor-derived motion data may lead to the development of robust and transformative digital measurements intended to predict, diagnose, and quantify neurological disease state and its longitudinal change.

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KEYWORDS

motor function; medical devices; computers; handheld; smartwatch; smartphone; mobility; wearable electronic devices; Parkinson disease; Parkinsonian disorders; gait; mobile phone

Introduction

Background

Patient care is changing with the dawn of smart sensing technology. Mobile and wearable devices can provide continuous as well as objective monitoring and assessment of many health outcomes [1]. Until recently, outcomes that represent various motor functions (ie, any movement of the entire body or part of the body that is controlled by motor neuron activity) have typically been measured by patient reports (eg, number of falls) or physician assessment (eg, gait abnormalities). Physician assessments are based on very brief observations in an office or clinic [2], whereas self-reported outcomes are subjective and often not as sensitive nor as supervised as in-clinic measures [3]. Finally, measurements may vary between assessors depending on the level of training, familiarity, and experience [4,5].

Wearable technologies have recently emerged as a potential supplemental source of data on motor function. Such technologies could increase the objectivity and ease of assessment for motor functions during clinical trials and care while also allowing for a richer dimension of data to be captured. Real-world and continuous monitoring of patient motor functions through wearable and mobile sensors is increasingly being investigated in areas such as disease progression through motor fluctuations in Parkinson disease [6], detection of amyotrophic lateral sclerosis [7], and tremor activity in essential tremor [8].

Data from digital measurement solutions can enhance the quality of clinical trials, as illustrated by the acceptance of wearable device–measured stride velocity (95th percentile) by the European Medicines Agency (EMA) as an end point in Duchenne muscular dystrophy [9]. Given the implications these new data courses could have on the field, the current regulatory environment for mobile technologies is in flux [10]. US and European regulatory bodies are responding to this emerging opportunity by adapting their regulatory processes to these technological advances [11].

Objectives

Previous reviews have described the characteristics of their patient samples and sensors involved in collecting motor function data [12-20]. However, they do not evaluate the degree of validity produced by such sensors. This review follows the terminology used in previous reviews [21,22] and differentiates between analytical validation (ie, the same motion behavior is measured by an independent source and compared with the sensor-derived motion behavior) and clinical validation (ie, a clinical characteristic or measure of interest is measured and compared with the sensor-derived motion behavior). Gaining insight into the current clinical validity and utility of the data captured by mobile and wearable sensing technologies is of utmost importance. So, the aim of this study was to describe the existing scientific literature on digital measurement solutions that are being used or tested for assessing motor functions in mobility-impaired and healthy adults and to evaluate the degree to which these tools provide clinically valid measures of motor function in these populations. Specifically, we aimed to answer the following research questions: (1) What types of digital devices exist that capture motor function in mobility-impaired and healthy populations? (2) In what types of studies and in what populations have these devices been evaluated? (3) What outcomes do these digital devices measure? (4) What types of technologies and algorithms are used to capture and store the data? (5) To what degree have these technologies and their output been validated using established and recognized criteria?

Methods

Literature Review

This review was conducted in accordance with the Cochrane Handbook for Systematic Reviews of Interventions [23], and reporting is based on the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines [24]. We included clinical trials (randomized and nonrandomized) as well as observational studies (case-control, retrospective cohort, prospective cohort, and cross-sectional) that provided validity estimates from wearable or mobile technologies to assess motor functions in adults (aged ≥18 years). Studies published in English after 2015 were included to focus on the most advanced technologies that are being used to assess motor function.

Study eligibility criteria were defined using an adapted PICO (Population, Intervention, Comparator, Outcomes) framework. We applied criteria based on the technology instead of the intervention or comparator, as the research question focused on the validity of measurement and not treatment efficacy (Table S1 in Multimedia Appendix 1 [25-115]).

A systematic literature search was conducted (January 1, 2015, to June 24, 2020) in the MEDLINE, Embase, and CENTRAL databases. Searches of relevant conferences for the last 3 years (2018-2020) were conducted via Embase. Search strings are available in Tables S2-S6 in Multimedia Appendix 1. Gray literature searches were also conducted to capture studies from sources that were not included in the main literature databases, which included the US Food and Drug Administration website as well as the United States and European clinical trials registry databases for clinical trials which had reported results but were not published in peer-reviewed journals (for the years 2018-2020).

After duplicate removal, all titles and abstracts were screened for potential eligibility according to the prespecified PICO criteria, after which full-text articles were assessed using the same criteria. Study selection was performed by 2 independent reviewers, and disagreements were resolved through discussion.

If no consensus could be achieved, a third researcher was consulted for arbitration.

A total of 2 independent reviewers extracted all relevant data from the final list of included studies. A reconciliation phase was again deployed to resolve any discrepancies between the reviewers, and a third reviewer intervened to resolve any remaining conflicts. The following data were extracted where available: (1) authors, year of publication, country, study setting, and follow-up period; (2) study design; (3) participant characteristics; (4) outcomes; (5) technology characteristics; and (6) validity outcomes. Motor function outcomes were manually sorted into categories by reviewers to facilitate summary where necessary.

Study Quality

A total of 2 independent reviewers assessed the quality of the included studies using the ROBINS-E (Risk Of Bias In Nonrandomized Studies of Exposures) tool [116]. A third investigator intervened to reach consensus if there were any remaining unresolved discrepancies following reconciliation between the decisions of the 2 reviewers.

Statistical Analyses

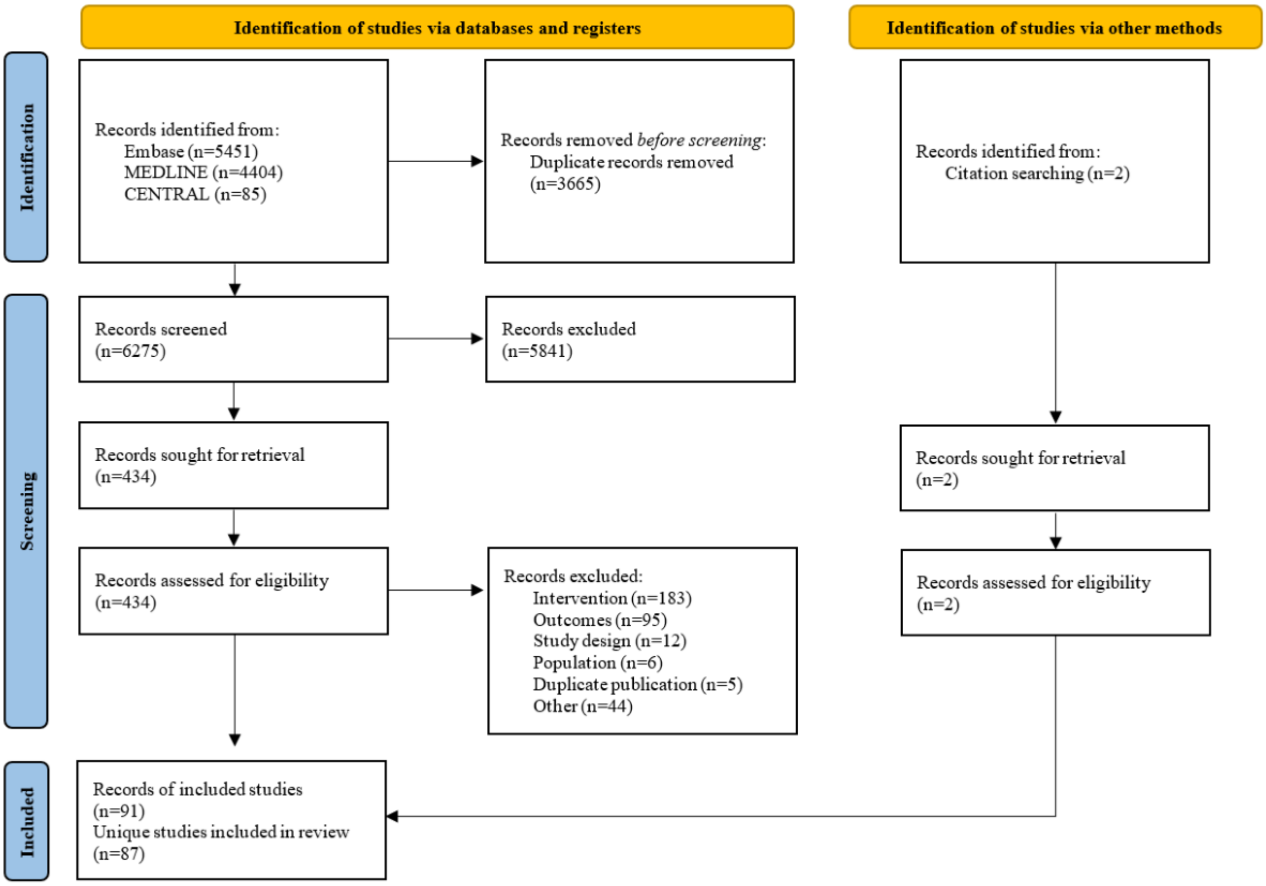
Effect size estimates were extracted from each study where reported, including standardized mean differences (ie, Cohen *d*), correlation coefficients (eg, Pearson *r*), sensitivity, specificity, accuracy, and area under the curve (AUC). In cases where studies provided none of these aforementioned effect size classes, effect sizes were calculated based on the information available in the manuscript using standard formulas [117,118]. To facilitate comparison across the studies, extracted effect sizes were converted to Pearson *r*–based effect size estimates where possible. This extraction and conversion process allows for studies to be directly compared via *r*-based effect sizes, estimates of sensitivity and specificity, and estimates of accuracy. The average effect sizes were calculated across all studies as well as by specific study and sample characteristics of interest. As *r* is bound by −1 and +1, *r*s were transformed into *Zr* using the procedure described by Fisher for analyses [119,120] and then back-transformed for reporting. Differences across groups in the magnitude of obtained effect sizes were tested using restricted information maximum likelihood derived SEs [117] using the inverse variance weight [121]. A random effects approach was taken, which includes in the denominator an extra variance component representing true variation in the population from which the included studies can be considered a random sample. A significance threshold of .05 was used to determine if values significantly differed between groups.

Results

Study Selection

A total of 9940 abstracts were identified from the electronic databases, and 2 articles [25,26] were included from handsearching of a systematic review identified in our searches [122]. After the removal of duplicates and exclusion based on title and abstract screening, 436 records remained for the full-text screening. A list of the records excluded during full-text screening and the reason for exclusion are provided in Table S7 in Multimedia Appendix 1. A total of 91 publications describing 87 primary studies fulfilled all inclusion criteria (Figure 1).

Figure 1. PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flow diagram.



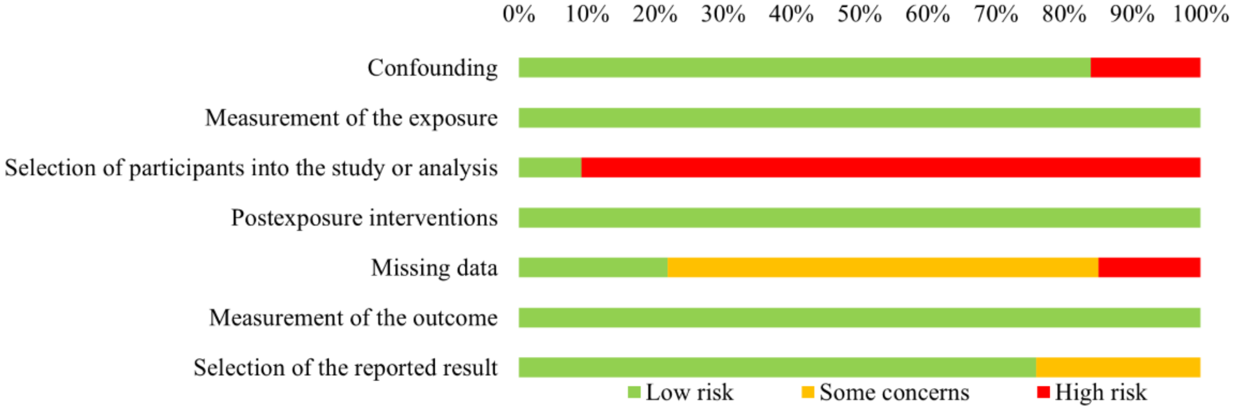
Study Characteristics and Data Collection

Across the 87 studies (n), the most common country settings reported were the United States (n=15) [27-41], United Kingdom (n=10) [42-53], Italy (n=5) [54-58], Spain (n=4) [59-62], South Korea (n=4) [63-66], Germany (n=3) [67-69], and Japan (n=3) [70-72]. At least 1 study was conducted in each of the following countries: Canada (n=2) [73,74], the Netherlands (n=2) [75,76], Portugal (n=2) [77,78], Sweden (n=1) [79,80], Taiwan (n=2) [81,82], Australia (n=1) [83], Brazil (n=1) [84], Demark (n=1) [85], France (n=1) [86], Israel (n=1) [87], Greece (n=1) [88,89], Lithuania (n=1) [90], Norway (n=1) [91], and United Arab Emirates (n=1) [92]. Of the remaining reporting studies, 6 were multinational [93-98]. Sample size ranged from 8 [33] to 1465 [94] (median 40.5 participants). A total of 7995 participants were enrolled in the included studies. Table S8 in Multimedia Appendix 1 presents the list of included publications as well as key study characteristics.

All 87 studies were observational in nature. Most studies (n=50) did not report whether the study was conducted in a single-center or multicenter setting. However, among those that did report, 20 and 17 studies were single center and multicenter, respectively. Approximately half of the included studies were conducted in a laboratory setting (n=42), 11 studies were home based, and 15 were a combination of a laboratory-based and home-based setting. The remaining 19 studies did not specify the study setting. The included studies were categorized into 2 follow-up types: cross-sectional (n=62) with a follow-up period of ≤1 week and longitudinal (n=25) in which participants were followed up for ≥1 week. Follow-up length of longitudinal studies ranged from 7 days [42,45,59,91,99] to 8 years [46]. A total of 30 studies reported the time allocated for data collection; in other words, the time needed to collect data in one session of data collection. In addition, 18 studies were able to capture their data in a session between 20 seconds [52,95] and 24 hours [71]. Moreover, 13 studies required their participant to use the device for multiple days for their collection period, which ranged from 2 [41,62] to 14 consecutive days [40]. This review follows the terminology used in previous reviews [21,22] for analytical validation (ie, the same motion behavior is measured by an independent source and compared with the sensor-derived motion behavior) and clinical validation (ie, a clinical characteristic or measure of interest is measured and compared with the sensor-derived motion behavior). Analytic validation was only performed in 21% (13/62) of cross-sectional studies and 4% (1/25) of longitudinal studies. Most of these studies performed clinical validation of sensor-based motion data. Studies applied a wide variety of technologies to capture motion outcomes. Motion data were captured by ≥30 different devices, including novel wearables (18/42, 43% devices), smartphone or smart watch (13/42, 31%), mass market digital technology (7/42, 17%), other digital technology (eg, PC; 3/42, 7%), and mass market wearables (1/42, 2%). Approximately 1 in 5 studies included a mass market device.

In terms of quality, studies were generally low to moderate risk of bias (Figure 2; Table S9 in Multimedia Appendix 1). Less than 20% (14/42) of studies did not show that groups were balanced in terms of key baseline characteristics and were considered high risk for confounding. The risk of bias arising from measurement of the exposure was most often low because exposures were generally whether the patient had a disease or was healthy, and misclassifications were next to nonexistent. For the domain of selection of participants into the study, studies were often high risk of bias. Disease diagnosis (ie, the exposure) did not generally coincide with the start of follow-up, and the diseases being studied could fluctuate over time. Many of the studies relied on volunteers to participate in the study, and this may have led to participants entering the study if they were in a particularly good or bad disease state (eg, Parkinson disease has *on* and *off* states). Furthermore, no corrections that may have alleviated selection biases in the analysis were conducted. Studies were generally low risk with regard to the domain concerned with the risk of bias owing to postexposure interventions. By design, the included studies did not administer interventions to alleviate the effects of exposures, and therefore, bias was not a concern. Regarding missing data, this was not often accounted for, leading to high risk of bias in that domain. However, studies were generally low risk of bias for measurement of outcomes, as motor function outcomes were assessed objectively and similarly across groups. Finally, over half of the studies were rated low risk for selection of the reported result.

Figure 2. Distribution of study quality across included studies.



Concepts of Interest and Context of Use

Approximately half of the included studies compared the association between sensor-derived motion data and a standardized clinical assessment across diverse disease conditions (n=44). Other studies compared mobility-impaired diseased participants to a healthy control group of participants with no mobility impairment (n=43). The most represented disease condition was, by far, Parkinson disease (n=51); stroke (n=5); Huntington disease (n=5); and depression, cognitive impairment, cerebral palsy, and multiple sclerosis (n=2 for each). All other disease groups were only represented in a single study.

Among the 67 studies that reported the mean age of participants, values ranged from 23.6 years [92] to 77.2 years [95] for mobility-impaired participants and from 19.5 years [29] to 78.9 years [87] for healthy participants. Control groups were generally well-matched by participant age and sex. Among the 71 studies that reported the proportion of males or females in their sample, the average percentage of the sample that were male ranged from 22.8% [62] to 100% [72,84] in mobility-impaired participants and from 11% [41] to 100% [84] in healthy participants. Studies with the largest sex imbalances were those addressing the less frequently studied disease states (ie, represented in only 1 or 2 studies). In contrast, Parkinson disease, Huntington disease, and stroke reflected a more balanced representation of females and males.

The primary motion outcomes were gait (n=30), gross motor movements (n=25), fine motor movements (n=23), motor symptom severity (n=9), bradykinesia (n=7), motor fluctuations (n=6), dyskinesia (n=5), balance control (n=5), postural stability (n=4), voice or speech impairments (n=3), facial expression impairments (n=1), and nocturnal movements (n=1). A summary of commonly reported outcomes by disease that the outcome was measured in is provided in Table 1.

The most common motions that participants were required to enact for sensor data collection across these studies were based on diverse active motor tasks: multimovement tasks (16/87, 18%) including balancing and reaction time during tests such as the Timed Up and Go, the Cognitive Dual Task Timed Up and Go, and the Manual Dual Task Timed Up and Go, unscripted daily activities (17/87, 20%), walking (10/87, 11%), tapping (9/87, 10%), and scripted activities of daily living (7/87, 8%). Less commonly used motions (<5% of studies) included several real-world tasks such as reaching, sit-to-stand motion, seated tremors, wrist pronation-supination tracing or pointing, typing, seated conversation, standing, and sleeping movement. Together, these motions were used to extract ≥75 distinct motion outcomes across the included studies. Most of these outcomes only appeared in one study and were only measured at a single sensor location in each study (per our inclusion criteria). One exception was walking cadence, with different studies measuring it using sensors worn at wrists, ankles, lower back, and chest and in the pants pocket. Additional exceptions were tremor, dyskinesia, and bradykinesia (each measured using sensors placed on the wrists or ankles).

Table 1. Summary of commonly reported outcomes by disease in which the outcome was investigated.

Disease and motor function outcome category	Motor function outcome
Acquired brain injury	
Gross motor impairment or performance and upper body	• Peak upper limb velocity [35] • Upper limb velocity [35]
Alzheimer disease	
Fine motor impairment or performance and continuous motion	• Spiral tracing [82]
Depressive tendencies	
Fine motor impairment or performance and discrete motion	• Finger tap speed [92] • Flight time [92] • Hold time [92]
Healthy participants	
Bradykinesia	• Bradykinesia score [94,100]
Dyskinesia	• Dyskinesia score [100]
Fine motor impairment or performance and continuous motion	• Spiral tracing [82,90]
Fine motor impairment or performance and discrete motion	• Correct finger taps [25,83] • Finger tap accuracy [38,101] • Finger tap count [38,95,101] • Finger tap duration [38,101] • Finger tap interval [38,101] • Finger tap reaction time [38,42,58] • Finger tap rhythm [42,95] • Finger tapping test [102] • Flight time [83,88,103] • Hold time [88]
Gait	• Joint velocity [77] • Step cadence [69,75,81,99] • Step count [40,41,44,74,104] • Step length [44,46,81] • Stride duration [44] • Turning speed [26] • Walking speed [41,69,81]
Gross motor impairment or performance and lower body	• Lower limb velocity [105]
Gross motor impairment or performance and whole body	• Joint velocity [106]
Motor symptom severity	• Rest tremor [102]
Postural stability	• Trunk acceleration [50]
Huntington disease	
Cognitive impairment	• Stroop Color and Word Test [96]
Dyskinesia	• Chorea score [96,107]
Fine motor impairment or performance, discrete motion	• Finger tap speed [96]
Gait	• Step cadence [99]
Mild cognitive impairment	
Fine motor impairment or performance and continuous motion	• Spiral tracing [82]
Multiple sclerosis	

Disease and motor function outcome category	Motor function outcome
Fine motor impairment or performance and discrete motion	• Finger tap count [25]
Gait	• Turning speed [26]
Neurological disorders[a]	
Fine motor impairment or performance and continuous motion	• Spiral tracing [90]
Neuromuscular disorders[b]	
Gait	• Step count [104]
Parkinson disease	
Bradykinesia	• Bradykinesia score [34,48,53,94,97,100,108]
Cognitive impairment	• Stroop Color and Word Test [83]
Dyskinesia	• Dyskinesia score [53,100] • Finger tapping test [56]
Fine motor impairment or performance and discrete motion	• Correct finger taps [83,109] • Finger tap accuracy [38,101] • Finger tap count [38,95,101] • Finger tap duration [38,101] • Finger tap interval [38,101] • Finger tap reaction time [38,42,49] • Finger tap rhythm [42,95] • Finger tapping test [102] • Flight time [88,103,110] • Hold time [88]
Gait	• Freezing of gait [49,54,61,64,93,111,112] • Step cadence [75] • Step count [31,40] • Step length [44,46] • Stride duration [44] • Turning speed [97]
Gross motor impairment or performance and upper body	• Peak upper limb velocity [33]
Gross motor impairment or performance and whole body	• Joint velocity [106]
Motor fluctuations	• On or off state [34,60,62,68,98]
Motor symptom severity	• Rest tremor [49,102] • Tremor test [34,48,97]
Postural stability	• Trunk acceleration [50]
Rapid eye movement (REM) sleep behavior disorder	
Fine motor impairment or performance and discrete motion	• Finger tap reaction time [42] • Finger tap rhythm [42]
Stroke	
Gait	• Step cadence [81] • Step count [41,74] • Step length [81] • Walking speed [41,81]
Transthyretin familial amyloid polyneuropathy	

Disease and motor function outcome category	Motor function outcome
Gait	- Lower limb velocity [78] - Step length [78] - Stride duration [78] - Walking speed [78]
Gross motor impairment or performance and upper body	- Upper limb velocity [78]

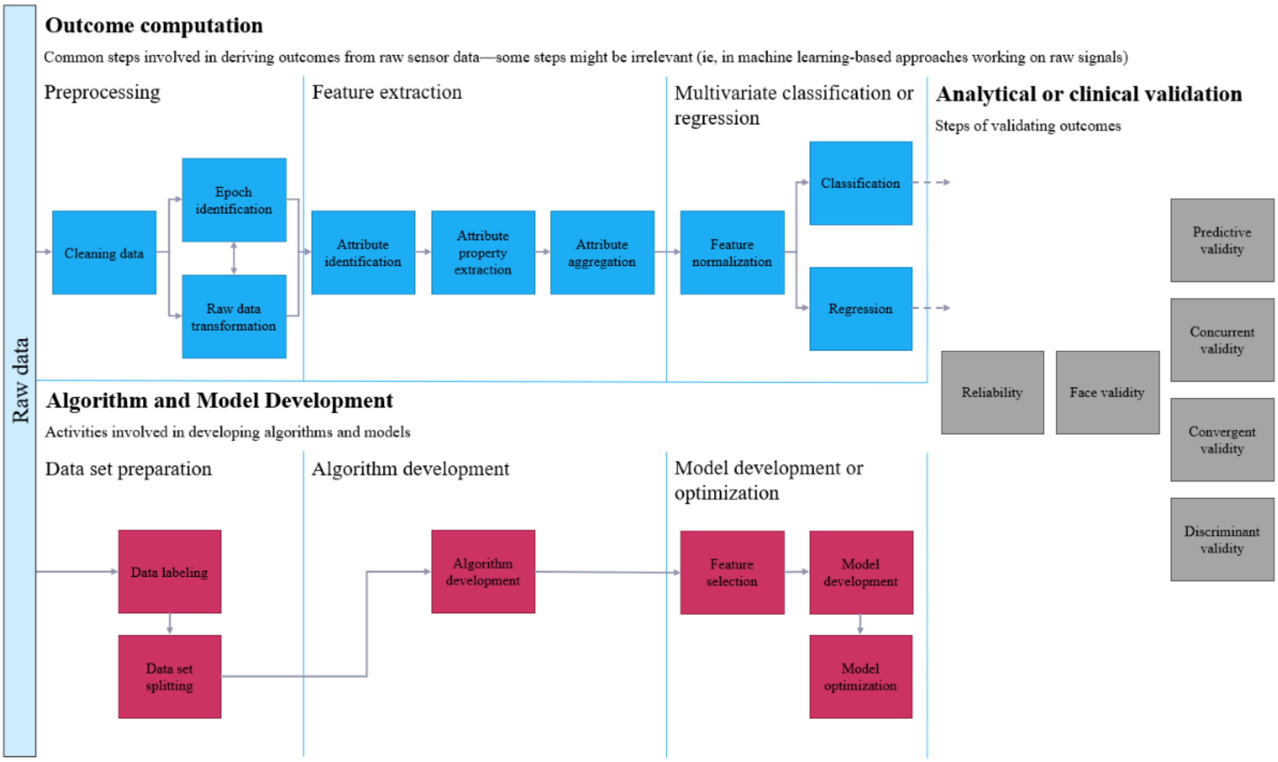
[a] Including Parkinson disease, Huntington disease, early dementia, cerebral palsy, and poststroke.

[b] Including Duchenne muscular dystrophy, limb-girdle muscular dystrophy, and spinal muscular atrophy.

Data Processing and Analysis

The process through which these researchers converted their raw data to validity coefficients is illustrated in Figure 3. On collection of the raw data, 2 parallel processes were typically seen: outcome computation and algorithm or model development. Following the completion of these 2 processes, the model was subjected to either analytical or clinical validation.

Figure 3. Flowchart of the process of converting raw data to validity coefficients.



Outcome Preparation

In ≥90% of the studies, the raw data were first preprocessed before feature processing engineering and analyses. One preprocessing step frequently seen among these studies was the splitting of raw data into temporal epochs or slices. This was done because training an algorithm to detect movement features across long periods greatly reduced the algorithm's validity. Data were trimmed by temporal position (eg, the beginning and ending of the motion recording) or based on extreme values (eg, outliers >4 SDs from the mean). Raw data were subjected to some form of standardization or transformation in ≥90% of the studies.

Although algorithm training (eg, feature selection and threshold determination) typically occurred using data across all participants, several studies took the approach of building the feature detection algorithm using data across all participants but then allowing each participant to vary in latter stages such as feature selection or determining thresholds [34,54,63,68]. Validity estimates from this smaller group of studies were similar in magnitude to those studies that applied the same features and thresholds to the classification of all participants.

Researchers have to decide which of the hundreds of identified candidate features to treat as a signal (by retaining them in the model) and which to dismiss as mostly noise (by excluding them from the model). Relatively few studies clearly described whether they moved all detected features to the next analytic stage (feature selection), but some studies compared prediction based on all extracted features to prediction based on top-performing features [42,49]. These studies reported that the inclusion of additional features did not guarantee a meaningful increase in algorithm performance or validity. One study using smartphones to assess Parkinson disease symptoms found AUC values >0.90 for 998 detected features, with a drop to 0.75 when based on the top 30 features [49]. A second study of participants with Parkinson disease concluded, "Accuracies obtained using the 30 most salient features were broadly comparable with the corresponding sensitivity and specificity values obtained using all 998 features" [42].

Algorithm or Model Development

The included studies showed no clear preference regarding algorithms for feature selection or classification, but the 2 most frequently applied approaches were support vector machines (12/87, 14%) and random forests (4/87, 5%). Authors of these studies were sensitive to the complications of trying to train a classification model with groups of different sizes, as most of the comparative studies included in this review include approximately equal sizes of participants with a disease or disorder and healthy controls.

No consistent pattern emerged from within-study comparisons of feature selection algorithms. A wrist-based sensor was able to detect upper limb movement among participants with pre-Parkinson disease best when using random forests relative to support vector machines and naïve Bayes [55]. A smartphone app testing motor impairment found that both neural networks and boosting outperformed support vector machines and Fisher linear discriminant analysis [90]. Not all motions required feature selection across studies (several needed only to define logic rules to estimate movement angles using geometry), and some studies used proprietary algorithms that were not described in detail. One study that studied freezing of gait among participants with Parkinson disease using a smartphone app found neural networks performed better than other bagging algorithms, including random forest, multilayer perception, decision tree, support vector machine, and naïve Bayes [64]. Another study on motor symptoms among participants with Parkinson disease using ankle-worn sensors found that support vector machines performed better than logistic regression and decision trees [80]. Using smartphone motion data to predict motor impairment among participants with Parkinson disease, another study found that random forests based on Ridge regression outperformed those based on Lasso, or Gini impurity, and that linear support vector machines outperformed logistic regression and boosting [103]. The sole consistent pattern that emerged was that supervised machine learning techniques performed better than unsupervised techniques (eg, naïve Bayes).

Analytical and Clinical Validation

The most common validity criterion was clinical condition (37/87, 43%), which was used in many of these studies to establish known-group construct discriminant validity of sensor-derived motion data by comparing participants with a diseased condition to healthy controls (Table S10 in Multimedia Appendix 1). The second most common validity criterion was the clinical validity established by assessing the convergence or concurrence with traditional standardized clinical assessments (30/87, 34%; eg, Wolf Motor Function Test and Unified Parkinson Disease Rating Scale). Other criteria were clinician ratings (7/87, 8%), research device (9/87, 10%), treatment status (3/87, 3%), and patient-reported outcome (1/87, 1%). Longitudinal studies were more likely to use nonsupervised assessments, whereas cross-sectional studies were more likely to use clinician-supervised assessments.

Across studies, motion data from the sensors identified showed an average Pearson *r* clinical validity coefficient of 0.52 (Figure 4 [27,28,31,35-41,44,47,48,50-53,57,58,66-74,76,77,80-84,86,91,92,95-99,101,102,104,106,108-110,112,113,115]). Among the studies that did not provide sufficient information to calculate a Pearson *r*, the average validity was 0.83 (sensitivity), 0.84 (specificity), and 0.90 (accuracy). These values could be interpreted as very good [123]. The magnitude of validity coefficients did not vary (*P*=.10) between health care–related devices (mean *r*=.47), personal electronic devices (mean *r*=.44), and entertainment consoles (mean *r*=.63). Validity coefficients for motor function generated by healthy adults were higher than those generated by participants with a disease state or impairment (*z* score 3.19; *P*=.001). The only statistical decision that consistently predicted higher validity coefficients was the decision to trim observations during the preprocessing stage based on value (ie, outliers; *z* score 2.10; *P*=.04). There was no difference in validity coefficients across trimming observations based on temporal placement, transforming data, standardizing data, or which feature detection and validation analyses were used. The funnel plot from these studies was asymmetrical in a manner consistent with bias toward higher coefficients (Figure S1 in Multimedia Appendix 1). The magnitude of validity coefficients did not significantly vary across the different device types (Table 2).

Taken as a whole, no consistent pattern emerged from within-study comparisons of the relative analytic validity of any specific motion signal. One study using Kinect found high Pearson *r* validity coefficients (*r*>0.50) for more than 40 distinct motion outcomes but very low validity coefficients for a handful including deflection range roll (measured in degrees), mean sway velocity roll (measured in degrees per second), and up-down deviation (measured in centimeters) [69]. A second study using Kinect found Pearson *r* validity coefficients above 0.50 for variables related to steps taken, distance, and speed but coefficients below 0.50 for variables related to angles (eg, trunk, hips, ankle, trunk, upper limb, and full body) [78]. A third study using a triaxial accelerometer worn on the waist found Pearson *r* validity coefficients above 0.50 for gait, arising from chair, body bradykinesia, hypokinesia, and overall posture and validity coefficients below 0.50 for rigidity of lower and upper extremities axial rigidity, postural stability, legs agility, and tremors in lower or upper extremities [98]. These numbers are in the same range as single items from widely established clinical tools [124-126]. As the validity coefficients for these single motions were moderate, it reinforces the need for future studies and clinical applications to include multiple validated motion signals for any screening or diagnostic tool to achieve adequate levels of composite test validity.

Regarding clinical validation, no clear within-study evidence emerged regarding the relative superiority or inferiority of motion data captured in laboratory settings versus data captured in home settings (Table 1). For example, 1 study comparing typing behavior of participants recently diagnosed with Parkinson disease to the typing behavior of healthy controls found AUC values of 0.76 (when administered at home) versus

0.83 (when administered in clinic) [59]. A second study comparing participants with Parkinson disease to healthy adults on motor function during an activities of daily living task found slightly higher accuracy, sensitivity, and specificity when the task was completed at home [87].

Figure 4. Forest plot of the validity of sensor-derived digital measurements of motor function. Middle points represent the point estimate effect size Pearson r, and the surrounding bars represent 95% CI. Colors indicate the type of validity criteria used.

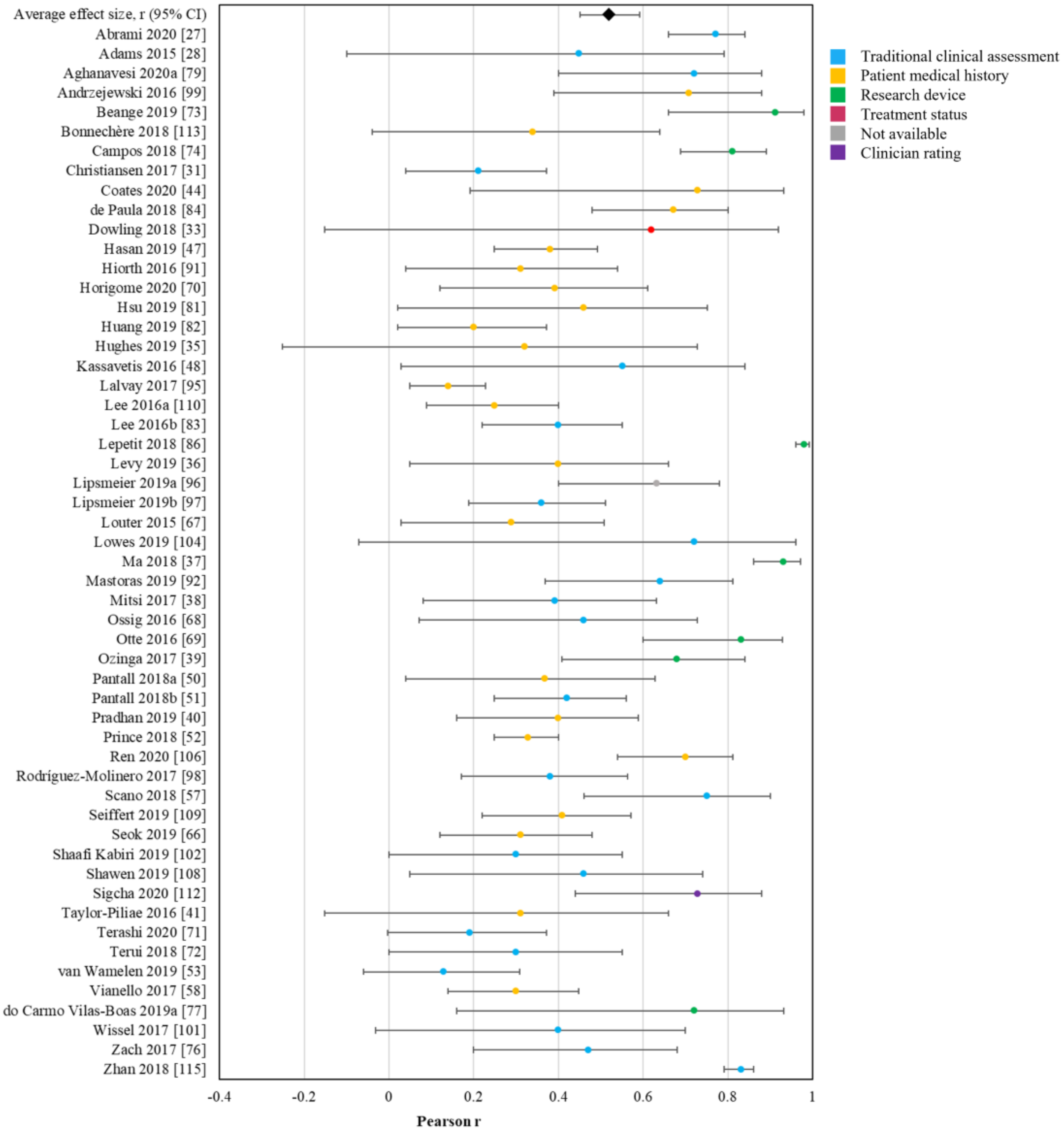


Table 2. Summary table of the between-study and within-study findings on the differences in the validity of sensor-derived measurements of motor function across various groups.

Are there differences in the validity of sensor-derived measures of motor function as captured	Between-study (ie, meta-analytic) findings	Within-study findings
Using mass market devices vs medical sensors?	- No: digital technology vs mass market digital technologies (*P*=.22); mass market digital technology vs medical devices (*P*=.21); digital technology vs medical devices (*P*=.32)	Insufficient data to evaluate
At specific sensor locations?	- No: wrist vs ankle (*P*=.73); wrist vs chest (*P*=.73); wrist vs hand (*P*=.54); wrist vs thigh (*P*=.59); wrist vs back (*P*=.63); wrist vs pocket (*P*=.78); wrist vs nonwearable (0.31) - No: ankle vs chest (*P*=.46); ankle vs hand (*P*=.38); ankle vs thigh (*P*=.73); ankle vs waist (*P*=.60); ankle vs back (*P*=.49); ankle vs pocket (*P*=.65); ankle vs nonwearable (*P*=.58) - No: chest vs hand (*P*=.30); chest vs thigh (*P*=.39); chest vs waist (*P*=.70); chest vs back (*P*=.82); chest vs pocket (*P*=.50); chest vs nonwearable (*P*=.89) - No: hand vs thigh (*P*=.58); hand vs waist (*P*=.75); hand vs back (*P*=.78); hand vs pocket (*P*=.42); hand vs nonwearable (*P*=.53) - No: thigh vs waist (*P*=.86); thigh vs back (*P*=.73); thigh vs pocket (*P*=.54); thigh vs nonwearable (*P*=.40) - No: waist vs back (*P*=.87); waist vs pocket (*P*=.39); waist vs nonwearable (*P*=.24) - No: back vs pocket (*P*=.45); back vs nonwearable (*P*=.48); pocket vs nonwearable (*P*=.50)	Insufficient data to evaluate
home vs in the laboratory?	- No; *P*=.33	No; 1 study found AUC[a] values of 0.76 (when administered at home) vs 0.83 (when administered in clinic) [59]. A second study found slightly higher accuracy, sensitivity, and specificity when the task was completed at home [87].
In longitudinal vs cross-sectional studies?	- No; *P*=.29	No; One study found high Pearson *r* validity coefficients (r>0.50) for over 40 distinct motion outcomes but very low validity coefficients for a handful, including deflection rage roll (measured in degrees), mean sway velocity roll (measured in degrees per second), and up-down deviation (measured in centimeters) [69]. A second study found Pearson *r* validity coefficients above 0.50 for variables related to steps taken, distance, and speed, but coefficients below 0.50 for variables related to angles (eg, trunk, hips, ankle, trunk, upper limb, and full body) [78]. A third study found Pearson *r* validity coefficients above 0.50 for gait, arising from chair, body bradykinesia, hypokinesia, and overall posture and validity coefficients below 0.50 for rigidity of lower and upper extremities axial rigidity, postural stability, legs agility, and tremors in lower or upper extremities [98].
In healthy vs motor impaired patients?	- Yes; validity higher among healthy adults, z score 3.19, *P*=.001	Insufficient data to evaluate

Are there differences in the validity of sensor-derived measures of motor function as captured	Between-study (ie, meta-analytic) findings	Within-study findings
Using different feature detection algorithms?	• Insufficient data to evaluate	No; One study was able to detect movement best when using random forests relative to support vector machines and naïve Bayes [55]. A second study found that both neural networks and boosting outperformed support vector machines and Fisher linear discriminant analysis [90]. A third study found neural networks performed better than other bagging algorithms including random forest, multilayer perception, decision tree, support vector machine, and naïve Bayes [64]. A fourth study found support vector machines performed better than logistic regression and decision trees [80]. A fifth study found that random forests based on Ridge regression outperformed those based on Lasso, or Gini impurity, and that linear support vector machines outperformed logistic regression and boosting [103]. The sole consistent pattern that emerged was that supervised machine learning techniques performed better than unsupervised techniques (eg, naïve Bayes).
Using particular motion sensor signal types?	• Insufficient data to evaluate	Insufficient data to evaluate
Using all vs a subset of features?	• Insufficient data to evaluate	No; One study found AUC values >0.90 for 998 detected features, with a drop to 0.75 when based on the top 30 features [49]. A second study concluded “Accuracies obtained using the 30 most salient features were broadly comparable with the corresponding sensitivity and specificity values obtained using all 998 features” [42].
With the thresholds held constant across patients vs patient-specific thresholds?	• No; *P*=.48	No; Although algorithm training typically occurred across a sample, several studies took the approach of starting the algorithm (feature detection) using data across all participants but then allowing each patient to vary in later stages such as feature selection or determining thresholds [34,54,63,68]. Validity estimates from this smaller group of studies were similar in magnitude to those studies that applied the same features and thresholds to the classification of all participants.
Using clinically supervised vs nonsupervised assessments of patient clinical status?	• No; *P*=.16	Insufficient data to evaluate
With outliers trimmed vs retained in the feature detection stage?	• Yes; trimming outliers is beneficial, z score 2.10, *P*=.04	Insufficient data to evaluate
With transformed data vs untransformed data?	• No; *P*=.74	Insufficient data to evaluate
With standardized data vs unstandardized data?	• No; *P*=.60	Insufficient data to evaluate

[a] AUC: area under the curve.

Discussion

Principal Findings

To our knowledge, this is the first systematic literature review to evaluate the degree to which wearable and mobile technologies provide clinically valid measures of motor function in mobility-impaired and healthy adults. The identified literature generally consisted of proof-of-concept studies, which aimed to pilot a device and assess whether it could validly measure motor functions. Consequently, most studies used a short follow-up period (<1 week) and had a total sample size of <50 participants. Unsurprisingly, many of the longitudinal studies prioritized nonsupervised measures. Even so, taken together, these studies provide a respectable evidence base supporting the potential these movement sensors have to inform clinical practice.

As the eligibility criteria for our review were inclusive in terms of population, we identified a large range of disease types, which were all but one (chronic obstructive pulmonary disease) nervous system condition (Table 1); however, the most common disease was Parkinson disease, with stroke and Huntington disease coming in a very distant second and third place. The strong focus on Parkinson disease in this literature may be because of its prevalence or perhaps because motor function symptoms are a major characteristic of Parkinson disease for diagnosis and prognosis assessment purposes, making Parkinson disease an ideal model disease for testing the use of mobile technologies [127]. However, it is most probably a mixture of these 2 hypotheses. Parkinson disease is also one of the few diseases with Food and Drug Administration–approved devices (eg, NexStride and Personal KinetiGraph), which assesses motor function to inform treatment decisions. The field would benefit from additional study of mobile technology–assessed motor function among other neurological diseases, including multiple sclerosis, spinal muscular atrophy, amyotrophic lateral sclerosis, and Alzheimer disease. In addition, future studies might consider the advantages of assessing digital devices per neurological impairment (such as difficulties in ambulation or upper limbs) rather than per disease.

Successful integration of wearable-based movement data into clinical tools requires both analytic validation and clinical validation. However, most of the reviewed literature compared wearable sensor-derived motion data to omnibus measures of functioning or disease progression (ie, clinical validation). More studies need to perform analytic validation by comparing wearable sensor-derived motion data to the same motions measured by another source (eg, observer assessment and motion-capture technology). Observed motions may be highly correlated with omnibus assessments of motor skills or disease status (ie, clinical validation), but the foundation of approval as a clinical end point can only be met if the motions identified using the sensor have been shown to be the exact motions that have been approved by the governing or regulatory body. Using as an example the EMA's recent approval of 95% stride velocity as an approved secondary end point in Duchenne muscular dystrophy, appeal to the EMA's approval of wearable sensor stride velocity data as an end point for a given study requires evidence that when the used algorithm claims to measure stride velocity (95th percentile), there be evidence that the algorithm has, in truth, measured stride velocity. Future research in this area should focus their attention on analytic validation.

There was considerable variation in methodological approaches. The review revealed one of the key reasons why this field may still show such inconsistency in analytic approach; it is still developing. Evidence of this is seen in which motion variables could be identified by the algorithms. Despite the hundreds of motion-derived outcome variables identified across these studies, not all theoretically meaningful motions could be recovered. One study of participants with Parkinson disease concluded, "Unfortunately, we failed to find parameters that reflected fatigue (decrement response) and hesitation (intertap irregularity), which are characteristics of motor dysfunction in Parkinson's disease" [110]. Those authors offered that more precise definitions of fatigue and hesitation may be needed to recover them in clinical settings with a smartphone-based tapping test similar to the one used in that study. In addition, the motor functions viewed by some authors as theoretically relevant were occasionally overshadowed by nonmotor signals. The tendency for studies to report diminishing returns after a certain point for additional motion signals is statistically analogous to other clinical efforts to identify causal markers from a multitude of candidates, which revealed many initially flagged markers as spurious [128]. Future studies should include graphical displays to identify inflection points (similar to the scree plot in factor analysis or the elbow plot in latent class analysis) to help show where the statistical signal (or true score) from additional motions becomes outweighed by statistical noise.

The moderate to high validity coefficients reported in the identified literature may support the potential for sensor-derived motor function data from digital health technology tools to eventually contribute to screening, diagnosis, and monitoring of neurological diseases in particular. No significant differences in analytic or clinical validity estimates were found when comparing data generated by mass market devices (eg, smartphones, smartwatches, and Fitbits), game consoles (ie, Nintendo Wii and Microsoft Kinect or Xbox), and marketed motion sensors (eg, ActiGraph, ActivPAL, Axivity, Dynaport, KinetiSense, Opal devices, and PAMSys-X). Furthermore, the motion data provided by these technologies produced equivalent validity estimates in laboratory-based and home-based settings. This further supports the future potential for digital measurement solutions to provide clinically meaningful data and eventually become the gold standard for assessing motor behaviors. The degree and rate of application for motor function data from these devices to clinical practice will depend on how soon clear evidence bases are established for given sensor locations for given movements of interest.

Translation of these motor signals into clinical application is aided by demonstrating sufficient validity outside the scripted protocols of a controlled laboratory setting. The reviewed literature showed that scripted motion tasks were important when only a few minutes of motion data were to be captured. Furthermore, motion data from unscripted everyday living with longer data collection periods were also shown to be adequate and deemed complementary, as episodic scripted assessments of confined tasks might not capture the complex spectrum of potentially altered components of motor function in an unconstrained ecologic setting [129].

As a whole, the reviewed literature revealed several best practices as well as a few cautionary tales for mobile or wearable sensor-based movement data. Although cross-validation techniques all seek to counteract the inflation of validity coefficients that can occur during machine learning techniques, they can produce different results [42]. Despite these best practices, there remained indirect evidence of model overfitting in the form of some abnormally high validity coefficients in the final models (ie, specificity of 1.0, which is perfect) [130,131].

The reviewed literature also highlights areas to consider during the development of any clinical application. One illustration from this review is the critical role of thresholds [132], which require researchers to decide between manual versus automatic thresholds [133] and global versus person specific [134]. Leveraging the strengths of these modeling approaches while keeping them robust and flexible will be important to consider as they are scaled up to create clinical applications [132].

Comparison With Previous Reviews

We identified a number of similar literature reviews during our study selection [12-20]. All identified reviews synthesized their evidence qualitatively, and none provided a quantitative synthesis of the validity of motion data generated from these sensors among patients with neurological conditions. Of the 9 identified reviews, 1 was narrative [16], whereas the remaining were systematic reviews. None of the systematic reviews focused on neurological disorders. Overall, 2 reviews focused specifically on swimming motions [12,13], 2 were focused on older adult patients with no specific disease [15,19], and 2 reviews focused on only upper [14] and lower limb movements [18]. Of the remaining 2 systematic reviews with similar objectives and scope to that of our own, the paper by Díaz et al [17] aimed to review the current literature on the use of wearable sensors in gait, balance, and range of motion analysis. Diseases of participants also varied across their 56 included studies and included a mix of neurological disorders (eg, Parkinson disease, Alzheimer disease, and multiple sclerosis), as well as stroke, amputees, and healthy participants. Similar to our own review, the authors found that most body-worn devices were complex to use and required strong experience in data analysis to interpret the collected information. In addition, the authors pointed out a need for further validation and improvements in sensor systems for them to be used as reliable and accurate clinical devices. A second systematic review conducted by Kristoffersson and Lindén [20] provided a qualitative synthesis of 73 published articles on wearable body sensors used for health monitoring. Similar to our review, the authors found that included studies were generally observational in design and small in sample size. These methodological considerations should be taken into account for future studies testing clinical devices for assessing motor function.

Strengths and Limitations

One strength of this review is that it includes more studies than any other review of similar scope that we identified during our study selection process [12-20]. This review is unique relative to other reviews on this same topic because it summarizes the validity estimates across the included studies instead of simply describing the characteristics of the samples and sensors involved [15-20]. This provides an evaluation of the degree of validity produced by such sensors. An additional strength was that we identified several meaningful patterns in this literature (eg, an absence of consistency in analytic approaches, equivalent validity of motion data collected at home or in a laboratory, and higher validity coefficients for healthy adults), which can help guide future research in this area. A final strength of this review is that it addresses statistical issues in this field. Although most reviews in this research area are silent as to statistical concerns, the findings of this review are consistent with the small group of previous reviews, which have also noted the statistical challenges present in this literature [12-14].

A limitation of this review is insufficient statistical power to address several questions of interest because of the methodological inconsistency and resulting sparseness across studies. A second limitation of this review is that the literature showed some signs of potential bias, which could limit the trustworthiness of the aggregate effect sizes. Examples of potential bias identified during the study quality assessment were that few studies provided a clear description of whether data were available for all participants throughout the study, and no studies corrected for potential selection biases in their analyses. In addition, it is unclear whether the patterns seen in the funnel plot and elsewhere are evidence of publication bias, selective outcomes, or an artifact of the dominant analytic approaches in this field. Much of the reviewed literature has focused on clinical validation, with less than one-fourth of the studies performing analytical validation. As important as clinical validation is for establishing the clinical and real-world utility of sensor-derived motion data, more studies are needed that focus on the fundamental step of analytic validation. An additional limitation may be the fact that some diseases are not as prevalent or well-studied than others, which may have impacted their representation in our analyses. Finally, our review was restricted to publications available in the English language. Therefore, some technologies being investigated for motor function assessment in non–English-speaking countries may have been missed.

Considerations for Future Research

Several questions we initially hoped to answer in this review could not be addressed because of lack of consistency across studies (eg, which technology or sensor is used, where the sensor is placed, which motions are required by participants, preprocessing steps, feature detection and selection algorithms, and number of motion features retained for the prediction algorithm). Even within studies examining the same disease state, there was limited consistency in these characteristics. As a result, we cannot say which movements and motion outcomes produce the most valid indicators of different neurological disease states, or what data preprocessing, feature processing engineering, and analysis should be considered best practices for converting raw sensor-derived motion data into meaningful digital measurements or biomarkers. It was notable that many of the most common movements from the larger clinical literature (eg, reaching, sit-to-stand, tracing, and pointing) appeared so infrequently in this literature. This lack of consistency in the literature could have affected the validity estimates [135-139], and the lack of harmonization across studies limits any inference about methodological or analytic decisions [140].

An earlier review described continuous monitoring using movement-detecting wearable sensors as a potential source of ground truth for motor function data, which were previously available only through participant self-reports [141]. On the basis of the reviewed literature, the field cannot yet provide this type of objective truth. An existing algorithm needs to be applied to multiple samples without additional adjustments or enhancements and show an aggregate performance that approximates the estimates provided by the studies included in this review. No analytic technique will solve this issue; the only true solution is replication attempts in new samples. Researchers should report how many of the detected features were moved to feature selection to give readers a sense of how many features were excluded, a sense of the parsimony of the resultant model, and an awareness of how likely it is that the model may have been overfit. Care must be taken to design the classification algorithm in a way that maximizes the likelihood that it can perform equally well in future samples. This priority needs to be evaluated at each stage of the analysis: data set preparation, preprocessing, feature extraction, algorithm development, model development or validation, and analytical or clinical validation.

Conclusions

In conclusion, sensor-derived motion data can be leveraged to validly predict disease status for a variety of neurological conditions. Future research will elucidate to what extent sensor-derived motion data may yield robust and transformative digital measurements intended to quantify, diagnose, and predict neurological disease state and its longitudinal change.

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Conflicts of Interest

AS, PAC, CM, and SB report employment with Biogen. During completion of the work related to this manuscript, CCG was an employee of Biogen. CCG's current affiliation is ActiGraph, LLC, Pensacola, Florida, US, which was not involved in this work. TS, KH, and MSF report employment with Evidinno Outcomes Research Inc, which was contracted by Biogen to conduct this study.

Multimedia Appendix 1

Supplemental information on the methods and results of the systematic literature review.
[DOCX File , 288 KB-Multimedia Appendix 1]

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Abbreviations

AUC: area under the curve
EMA: European Medicines Agency
PICO: Population, Intervention, Comparator, Outcomes

PRISMA: Preferred Reporting Items for Systematic Reviews and Meta-Analyses
ROBINS-E: Risk Of Bias In Nonrandomized Studies of Exposures

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Original Paper

The Socioeconomic Indicators Linked to Parent Health-Related Technology Use: Cross-sectional Survey

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Abstract

Background: Despite the prevalence of parent health information seeking on the internet and its impact on parenting behavior, there is a paucity of research on parents of young children (ages 3 to 8 years). Given the importance of this developmental period, exploring how family socioeconomic indicators linked to the digital divide and health inequities affect parent proxy- and self-seeking is critical to further understanding variability in health information seeking and associated outcomes.

Objective: This study aimed to explore parental health-related technology use (HTU), the process by which parents engage in support, advice, and information-seeking behavior related to their (self-seeking) and their children's (proxy seeking) health across a range of hardware devices (eg, tablet, wearable, smartphone, laptop, and desktop computer) and sources (eg, search engines, mobile applications, social media, and other digital media).

Methods: A cross-sectional study including 313 parents and guardians of children ages 3 to 8 years recruited through Amazon Mechanical Turk (MTurk) was conducted. Parents were asked to complete a self-administered questionnaire on a broad range of parenting and parent-related constructs, including sociodemographic information, technology device ownership, and engagement in and use, features, and perceptions of HTU. Descriptive and bivariate analyses (chi-square tests) were performed to identify patterns and investigate associations between family socioeconomic indicators and parent HTU.

Results: The overwhelming majority (301/313, 96%) of parents of young children reported engaging in HTU, of which 99% (300/301) reported using search engines (eg, Google), followed by social media (62%, 188/301), other forms of digital media (eg, podcasts; 145/301, 48%), and mobile applications (114/301, 38%). Parents who engaged in HTU reported seeking information about their child's behavior and discipline practices (260/313, 83%), mental or physical health (181/313, 58%), and academic performance (142/313, 45%). Additionally, nearly half (134/313, 43%) of parents reported searching for advice on managing their stress. Among parents who reported using each source, an overwhelming majority (280/300, 93%) indicated that search engines were a helpful online source for proxy- and self-seeking, followed by social media (89%, 167/188), other digital media (120/145, 83%), and mobile apps (87/114, 76%). Among parents who reported using any technology source, approximately one-fifth reported that technology sources were most comfortable (61/311, 20%), most understanding (69/311, 22%), and most influential toward behavior change (73/312, 23%) compared to traditional sources of health information–seeking, including mental health professionals, other health care professionals, school professionals, community leaders, friends, and family members. Indicators of family socioeconomic status were differentially associated with frequency and perceptions of and search content associated with parent HTU across technology sources.

Conclusions: The findings of this study underscore critical considerations in the design and dissemination of digital resources, programs, and interventions targeting parent and child health, especially for families in traditionally underserved communities.

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KEYWORDS

parenting; child; health behavior; information seeking; health-related technology use; health information; digital health; mobile health; socioeconomic status; accessibility

Introduction

In the past decade, researchers have shown an increased interest in parental online health information seeking (OHIS), the process by which parents search for health information using the internet, including search engines, forums, and social networking [1,2]. OHIS has been linked to various aspects of individual and family functioning, including parenting behavior, perceived social support, and health status [3-6]. While parents search for information related to their own health (ie, self-seeking), they are even more likely to use the internet for health information related to their children (ie, proxy seeking). Indeed, data from the past several years revealed that 75% to 90% of parents have searched for health information related to their child [1].

Despite the widespread prevalence of parent health information–seeking on the internet, there is a paucity of research among parents of young children ages 3 to 8 years [1]. Research indicates that up to one-third (15% to 30%) of young children experience social, emotional, and behavioral problems [6-9]. Further, difficulties during this critical developmental period can persist into adolescence and adulthood, increasing the risk for long-term academic, occupational, and physical and mental health difficulties [10,11], especially for children in traditionally underserved communities with less access to quality care [12]. Given the importance of early development in child and family health, exploring how sociodemographic characteristics linked to the digital divide and health inequities affect parent proxy- and self-seeking is critical to further understanding variability in health information–seeking behaviors in the community [13,14].

Accordingly, this study addresses 2 underdeveloped research areas with parents of young children. First, the bulk of work has focused on clinical or treatment-seeking samples of parents with specific presenting issues (eg, attention deficit hyperactivity disorder, hearing loss) or circumstances (eg, after childbirth, during a visit to a pediatric outpatient clinic). However, parents' recognition of health-related concerns outside of the traditional health care system may depend on the extent to which they perceive a mismatch between their child's functioning and the socially and culturally relevant contexts (eg, school, home) in which they engage in daily life. Further, such perceptions may prompt parents to search for health-related content within broader domains (eg, child academic performance, parental discipline) of child and family functioning. Considering the information-seeking behaviors of parents of young children experiencing chronic illnesses or acute health problems may not generalize to other parents, studies with non–treatment-seeking samples are critical to understanding health information needs, seeking behaviors, and outcomes across diverse families.

Second, prior studies investigating parent OHIS have been limited to internet use, defined broadly and inconsistently across studies [1,15]. Considering the increasing adoption and use of other consumer technologies (eg, mobile apps and wearables) for health-related reasons and long-standing disparities in broadband access and connectivity, there is a need to extend current work to account for parent use of a variety of information and communications technologies [16-19]. Accordingly, we refer to parental health-related technology use (HTU) as the process by which parents engage in support, advice, and information-seeking behavior related to their (self-seeking) and their children's (proxy seeking) health across a broader range of devices (ie, tablets, wearables, smartphones, laptops, and desktop computers) and sources (ie, search engines, mobile applications, social media, and other digital media).

Building upon these gaps in the literature, this study aims to describe HTU among parents of young children, including the frequency and perceived usefulness of and search content associated with parent HTU in a non–treatment-seeking sample. In addition, resources (eg, parent access to technology devices) and perceptions (eg, comfortability) that may influence parent engagement in HTU are examined. Finally, whether patterns vary by parent, child, and household-level sociodemographic characteristics is explored.

Methods

Participant Recruitment

Parents and guardians of children ages 3 to 8 years old were recruited through Amazon Mechanical Turk (MTurk) to complete a survey on a broad range of parenting and parent-related constructs. Parents consented online before completing study measures in compliance with university-approved institutional review board (IRB) procedures. Upon confirming eligibility criteria, respondents were asked to select their youngest child in the specified age range to be referred to as the target child throughout the survey. All demographic variables and questionnaires were completed regarding the selected target child.

Additional measures were included to increase confidence in a participant pool that provides responses comparable to traditional samples (eg, [20-22]). To ensure attention to survey responses, 4 attention check questions were included throughout the survey (eg, "For data quality purposes, please select Sometimes") and were assessed as part of the inclusion criteria. Additionally, respondents with duplicate IP addresses, geolocations, and MTurk IDs were excluded from analyses in accordance with recommendations for studies using MTurk samples. As with other crowdsourcing platforms, MTurk duplicates typically reflect multiple entries from the same individual or household or, most prominently, "bot" (ie, computer programs that can automatically complete surveys) or “farmer” respondents (ie, individuals using server farms or commercial data centers to evade MTurk’s screening procedures). Furthermore, these respondents are linked to lower-quality data [20]. Finally, a random numerical code was provided to eligible participants (ie, parents of children ages 3 to 8 years old living in the United States) upon completion of the study to facilitate participant payment of US $2.

Ethics Approval

This study (17-0722) was approved by the institutional review board of the University of North Carolina at Chapel Hill.

Measures

Sociodemographic Characteristics

Parents reported sociodemographic information for their family, including the age, race (eg, White, African American/Black, Asian or Pacific Islander, American Indian/Alaska Native, or multiracial), and ethnicity (eg, Hispanic/Latino) of both the respondent (ie, parent or caregiver) and target child. Multiple indicators of family socioeconomic status were also collected, including annual household income, parent employment status (eg, full-time employment, part-time employment, unemployed but looking for work, nonworking, and retired), parent educational attainment (eg, less than high school or General Education Diploma [GED], high school graduate or GED, some college, associate’s degree, bachelor's degree, master’s degree, and doctorate), and perceived financial difficulty. Finally, parents also reported their household composition, marital status, relationship to the target child, and the target child’s health status (ie, prior diagnosis of or treatment for developmental delays).

Technology Device Ownership, Access, and Use

Parents reported their access to and frequency of using common technology devices (ie, desktop computer, laptop computer, smartphone, tablet, wearable) measured on a 6-point Likert scale ranging from 0 (never) to 5 (more than once daily).

Search Content

Parents reported the content of their search for health-related information, advice, or support, focusing on 3 broad domains of proxy seeking (child academics, behavior, and mental and physical health) and 1 domain of self-seeking (parent stress and stress management).

Frequency and Usefulness of Parent Health-Related Technology Use

Parents indicated their use and perceptions of particular technology-enabled sources (eg, search engines, mobile apps, social media, and other forms of digital media) to search for parenting advice and health-related information for their children. Parents reported the frequency of using each source (ie, “When you are looking for parenting advice, information, and/or support, how often do you turn to each of the following potential sources?”) using a 4-point Likert scale ranging from 0 (never) to 3 (frequently). Although the usefulness of particular sources has been evaluated inconsistently in the literature on parent HTU (eg, [23,24]), researchers often use a single-item measure to capture the construct (eg, “How useful do you feel the internet is in helping you make decisions about your health?”) [25]. Similarly, parents reported the usefulness of a source (ie, “How helpful or useful did you find the parenting advice, information, and/or support you received from these sources?”) using a 4-point Likert scale ranging from 0 (not at all helpful) to 3 (very helpful).

Statistical Analysis

Descriptive statistics were used to summarize family characteristics, parent ownership of or access to consumer technology devices, and parent engagement in and perceptions of HTU. Chi-square tests were conducted to compare proportions of and determine associations between device ownership and characteristics of HTU (eg, search content, frequency of use, and usefulness of technology sources) across groups defined by parent educational attainment (<bachelor’s degree vs ≥bachelor’s degree), perceived financial difficulty (none to mild vs moderate to severe), and low-income status, as determined by the federal poverty level (FPL), which accounts for annual household income and the number of people in the household (<200% FPL vs ≥200% FPL). Importantly, while “low-income” has been defined inconsistently in the literature, the FPL is typically used to determine eligibility for services, including those related to child health and development (eg, Head Start, Children’s Health Insurance Program) [26]. While income eligibility varies by state and service, 200% FPL has been mandated as an upper limit for participation in several government services (eg, Children’s Health Insurance Program, Subsidized Child Care Assistance Program), and incomes below 200% FPL account for a significant proportion of families in the United States who experience increased financial burden and economic insecurity [27]. Indeed, nearly 17% of children in the United States live in poverty, with approximately 7% (New Hampshire) to 56% (Puerto Rico) living in households below 200% FPL across the United States [28,29]. Of note, sociodemographic characteristics were included in analyses based on their theoretical relevance, as indicated in the previous research [13,18]. Missing values were excluded from analyses. Statistical analyses were conducted using SPSS version 26 software.

Results

Participants

Of the 657 respondents who completed the survey, 344 were removed from analyses for screening ineligibility (eg, families without a child in the specified age range or living outside of the United States, n=116), missed attention check questions (n=86), or duplicate IP addresses, geolocations, or MTurk IDs (n=142), yielding a total of 313 for analyses. Parents ranged in age from 19 to 57 years with a mean parental age of 34.19 (SD 7.11) years. Three-fifths (186/313, 59.4%) of parents self-identified as female. Slightly more than half (176/313, 56.2%) of parents obtained a bachelor’s degree or higher (ie, master’s or doctorate), and most were employed full- or part-time (280/313, 89.5%). Most parents were also married (243/313, 77.6%) and the biological parent of the target child (281/313, 89.8%). According to the parent report, approximately half (153/312, 49%) of the target children were female, and their mean age was 4.67 (SD 1.37) years. The racial and ethnic identity of most parents was White and non-Hispanic/Latino (230/312, 73.7%), followed by 8.7% (27/312) African American or Black, 4.5% (14/312) Asian American, 0.6% (2/312) American Indian/Alaska Native, and 3.2% (10/312) multiracial. For 10.5% (33/313) of the children, the parent's self-reported race or ethnicity differed from that of the child. Nevertheless, the majority of children identified as White and non-Hispanic/Latino (207/312, 66.3%), followed by 7.4% (23/312) African American, 3.8% (12/312) Asian American, 0.6% (2/312) American Indian/Alaska Native, and 13.1% (41/312) multiracial. Additionally, 11.8% (37/313) of parents and 15.3% (48/313) of children identified as Hispanic or Latino. The annual combined household income ranged from US $6000 to $380,000 with a median of $60,000 (SD $41,180). Finally, 56.2% (176/313) of families reported living in suburban areas, followed by 25.2% (79/313) in urban areas and 18.5% (58/313) in rural areas. Compared to the general population of parents in the United States, the recruited sample included slightly more college-educated and lower-income participants and a comparable percentage of women and married parents [30,31]. Additionally, parents were less racially and ethnically diverse than the general population of parents in the United States but slightly more so than has been reported in previous studies with parents using MTurk samples.

Device Ownership and Use

Parents reported owning a variety of technology devices, including a smartphone (276/313, 88.2%), laptop (276/313, 88.2%), tablet (243/313, 77.6%), desktop computer (193/313, 61.6%), and wearable device (100/313, 31.9%; Table 1). All (100%, 313/313) parents reported owning or having access to at least 1 technology device at home, and the majority (283/313, 90.4%) of parents reported access to multiple devices. Only 2 (0.64%) parents indicated not having access to a computer (desktop or laptop) at home, and both reported having access at work, school, or another setting (eg, library). Of the 37 (11.8%) participants that reported not having access to a smartphone, 12 (32.4%) reported having access to a smartphone at work, school, or another setting. Chi-square analyses revealed no statistically significant associations between parent educational attainment or perceived financial difficulty and access to technology devices. Families in low-income households were significantly less likely to own or have access to a wearable (χ^2_1=4.7, *P*=.03), but not any other technology device.

Table 1. Technology device ownership and access.

Demographics	Overall (N=313), n (%)	Desktop (n=193)		Laptop (n=276)		Smartphone (n=276)		Tablet (n=243)		Wearable (n=100)	
		Value, n (%)	χ^2(*df*), *P* value	Value, n (%)	χ^2(*df*), *P* value	Value, n (%)	χ^2(*df*), *P* value	Value, n (%)	χ^2(*df*), *P* value	Value, n (%)	χ^2(*df*), *P* value
Parent educational attainment			0.120 (*1*), .729		0.005 (*1*), .945		0.178 (*1*), .673		0.010 (*1*), .921		0.849 (*1*), .357
<Bachelor's degree	137 (43.8)	83 (60.6)		121 (88.3)		122 (89.1)		106 (77.4)		40 (29.2)	
≥Bachelor's degree	176 (56.2)	110 (62.5)		155 (88.1)		154 (87.5)		137 (77.8)		60 (34.1)	
Household income			0.246 (*1*), .620		2.676 (*1*), .102		0.147 (*1*), .701		0.075 (*1*), .785		4.744 (*1*), .034[a]
<200% FPL[b]	95 (30.4)	57 (60.0)		88 (92.6)		85 (89.5)		73 (76.8)		22 (23.2)	
≥200% FPL	216 (69.0)	136 (63.0)		186 (86.1)		190 (88.0)		169 (78.2)		77 (35.7)	
Perceived financial difficulty			0.028 (*1*), .868		0 (*1*), .998		0.591 (*1*), .442		0.127 (*1*), .721		1.563 (*1*), .211
None to mild	220 (70.3)	135 (61.4)		194 (88.2)		196 (89.1)		172 (78.2)		75 (34.1)	
Moderate to severe	93 (29.7)	58 (62.4)		82 (88.2)		80 (86.0)		71 (76.3)		25 (26.9)	

[a] *P*<.05.

[b] FPL: federal poverty level.

Among parents who reported access to a computer at home or another setting, 100% reported using their laptop or desktop device at least once monthly, with most reporting using their desktop computer (104/155, 67.1%) or laptop (117/177, 66.1%) more than once daily. Over half (31/55, 56.4%) of the parents reported using a wearable device multiple times during the day, and only 7% (4/55) reported using their wearable less than every 3 days. Approximately 31.1% (28/90) of parents who reported access to a tablet at home or another setting reported using their device more than once daily, and nearly a quarter (22/90, 24.4%) reported using their tablet once weekly or less. Most (177/196, 90.3%) parents reported using their smartphone multiple times per day, and no parents reported using their smartphone less than every 3 days.

The frequency of smartphone use was significantly lower for families in low-income households (χ^2_2=9.8, *P*=.007) and with parents reporting moderate to severe financial difficulty (χ^2_2=7.8, *P*=.021). Additionally, parents experiencing moderate to severe financial difficulty used their desktop computer (X^2_5=11.5, *P*=.042), laptop (X^2_5=12.4, *P*=.015), and tablet (X^2_5=23.9, *P*<.001) less frequently than their peers. The frequency of using any technology device did not vary significantly by parent educational attainment. Notably, parent age was not significantly correlated with the frequency of using any technology device.

Parent HTU

Most parents (301/313, 96.2%) reported using technology sources to search for parenting advice and health-related information for their children. Parents who engaged in health-related technology use reported using search engines (eg, Google; 300/301, 99.7%), social media (188/301, 62.5%), other forms of digital media (eg, podcasts; 145/301, 48.2%), and mobile applications (114/301, 37.9%). Approximately one-third (91/301, 30.2%) of parents reported using all 4 sources for proxy- and self-seeking.

There were no significant differences between parents who did and did not report engaging in HTU via mobile apps, social media, and other digital media across parent educational attainment (Table 2). Parents in low-income households were significantly more likely to report using mobile apps (χ^2_1=4.7, *P*=.030) and social media (X^2_1=4.9, *P*=.026) for health-related reasons, but not other forms of digital media. Parents reporting moderate to severe financial difficulty were significantly more likely to report using mobile apps (X^2_1=5.5, *P*=.019), social media (X^2_1=4.2, *P*=.040), and other digital media (X^2_1=7.3, *P*=.007) in comparison to their peers. Given that most participants reported using search engines, the technology source was not included in the chi-square analyses.

While the frequency of engagement in HTU varied across sources, parents reported more frequent use of search engines on average, followed by social media, mobile apps, and other digital media (Figure 1). The frequency of parent use was not significantly associated with self-reported parent educational attainment. The mean frequency of social media use (χ^2_3=16.4, *P*<.001) was significantly greater for parents in low-income households, and the use of social media (χ^2_3=11.9, *P*=.008) and other digital media (χ^2_3=10.4, *P*=.016) was also increased for parents reporting moderate to severe financial difficulty.

Among parents who reported using each source, an overwhelming majority (280/300, 93.3%) indicated that search engines were a useful online source for proxy- and self-seeking, followed by social media (167/188, 88.8%), other digital media (120/145, 82.8%), and mobile apps (87/114, 76.3%). Parents in low-income households also rated other digital media as more useful than their peers (χ^2_3=9.19, *P*=.027). Perceived financial difficulty and parent educational attainment were not significantly associated with the perceived usefulness of any technology source.

Table 2. Parent engagement in health-related technology use (HTU).

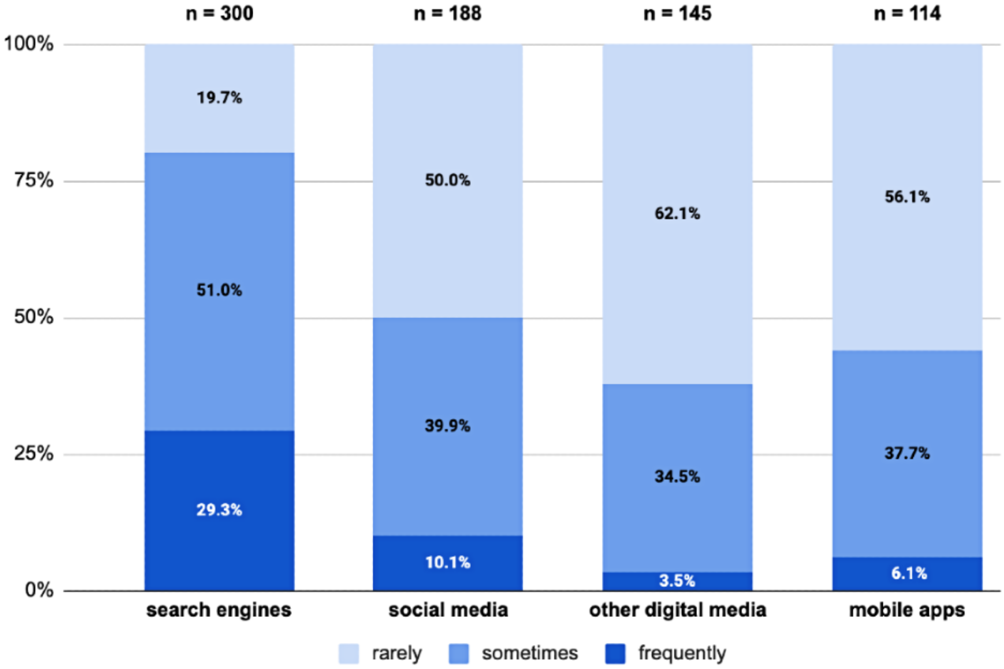
Demographics	Overall (N=313), n (%)	Social media (n=188)		Other media (n=145)		Mobile apps (n=114)	
		Value, n (%)	χ^2(*df*), *P* value	Value, n (%)	χ^2(*df*), *P* value	Value, n (%)	χ^2(*df*), *P* value
Parent educational attainment			0.027 (*1*), .868		1.560 (*1*), .212		0.001 (*1*), .981
<Bachelor's degree	137 (43.8)	83 (60.6)		58 (42.3)		50 (36.5)	
≥Bachelor's degree	176 (56.2)	105 (59.7)		87 (49.4)		64 (36.4)	
Household income			4.983(*1*), .026[a]		0.982 (*1*), .322		4.714 (*1*), .030[a]
<200% FPL[b]	95 (30.4)	66 (69.5)		48 (50.5)		43 (45.3)	
≥200% FPL	216 (69.0)	121 (56.0)		96 (44.4)		70 (32.4)	
Perceived financial difficulty			4.226 (*1*), .040[a]		7.332 (*1*), .007[c]		5.504 (*1*), 0.019[a]
None to mild	220 (70.3)	124 (56.4)		91 (41.4)		71 (32.3)	
Moderate to severe	93 (29.7)	64 (68.8)		54 (58.1)		43 (46.2)	

[a] *P*<.05.

[b] FPL: federal poverty level.

[c] *P*<.01.

Figure 1. Frequency of parent health-related technology use (HTU) across technology sources.



Search Content

Parents who engaged in HTU reported seeking information about their child's behavior and discipline practices (260/313, 83.1%), mental and physical health (181/313, 57.8%), and academic performance (142/313, 45.4%). Additionally, 42.8% (134/313) of parents reported searching for advice on managing their stress. Parents in low-income households were significantly less likely to search for health-related information or advice about their child's physical and mental health (χ^2_1=5.0, *P*=.025) and more likely to search for content about parent stress and stress management (χ^2_1=12.2, *P*<.001; Table 3). Parents reporting moderate to severe financial difficulty were also more likely to search for the latter (χ^2_1=4.2, *P*=.041). Parent educational attainment was not significantly associated with any search content.

Table 3. Parent proxy and self-seeking content areas.

Demographics	Overall (N=313), n (%)	Child behavior/discipline (n=260)		Child academic performance (n=142)		Child physical/mental health (n=181)		Parent stress/stress management (n=134)	
		Value, n (%)	χ^2(*df*), *P* value	Value, n (%)	χ^2(*df*), *P* value	Value, n (%)	χ^2(*df*), *P* value	Value, n (%)	χ^2(*df*), *P* value
Parent educational attainment			0.059 (*1*), .808		0.424 (*1*), .515		1.452(*1*), .228		3.695 (*1*), .055
<Bachelor's degree	137 (43.77)	113 (82.48)		65 (47.45)		74 (54.01)		67 (48.91)	
≥Bachelor's degree	176 (56.23)	147 (83.52)		77 (43.75)		107 (60.80)		67 (38.07)	
Household income			0.085 (*1*), .770		1.013 (*1*), .314		5.018 (*1*), .025[a]		12.231 (*1*), <.001[b]
<200% FPL[c]	95 (30.35)	80 (84.21)		39 (41.05)		46 (48.42)		55 (57.89)	
≥200% FPL	216 (69.01)	179 (82.87)		102 (47.22)		134 (62.04)		79 (36.57)	
Perceived financial difficulty			0.061 (*1*), .805		0.629 (*1*), .428		0.309 (*1*), .578		4.186 (*1*), .041[a]
None to mild	220 (70.29)	182 (82.73)		103 (46.82)		125 (56.82)		86 (39.09)	
Moderate to severe	93 (29.71)	78 (83.87)		39 (41.94)		56 (60.22)		48 (51.61)	

[a] *P*<.05

[b] *P*<.01

[c] FPL: federal poverty level.

Perceptions

Among parents who reported using any technology source, approximately one-fifth reported that technology sources were the most comfortable (61/311, 19.6%), most understanding (69/311, 22.2%), and most influential toward behavior change (73/312, 23.4%) compared to traditional sources, including mental health professionals, other health care professionals, school professionals, community leaders, friends, and family members. For perceived understanding, the majority of parents (48/69, 69.6%) referenced search engines, followed by social media (19/69, 27.5%) and other digital media and mobile apps (both less than 1/69, 2%). Similarly, for perceived comfortability, most parents listed search engines (42/61, 68.9%) and social media (18/61, 29.5%), and fewer mentioned mobile apps (1/61, 1.6%) and other digital media (0/61, 0%). Finally, in terms of parenting behavior change, search engines accounted for 73.97% (54/73), followed by social media (16/73, 21.91%), other digital media (2/73, 2.74%), and mobile apps (1/73, 1.36%). Perceived financial difficulty, but not any other socioeconomic status (SES) indicator, was significantly associated with perceptions of technology sources for health information seeking, such that parents experiencing moderate to severe difficulty were more likely to perceive engagement in HTU as the most understanding ($\chi^2_1=14.2$, $P<.001$), most comfortable ($\chi^2_1=7.9$, $P=.005$), and most likely to lead to behavior change ($\chi^2_1=7.3$, $P=.007$) compared to traditional sources (Table 4).

Table 4. Parent perceptions of health-related technology use (HTU).

Demographics	Overall (N=313), n (%)	Most understanding (n=69)		Most comfortable (n=61)		Most parenting behavior change (n=73)	
		Value, n (%)	χ^2(*df*), *P* value	Value, n (%)	χ^2(*df*), *P* value	Value, n (%)	χ^2(*df*), *P* value
Parent educational attainment			2.773 (*1*), .096		0.528 (*1*), .468		0.735 (*1*), .391
<Bachelor's degree	137 (43.8)	36 (26.7)		29 (21.5)		35 (25.7)	
≥Bachelor's degree	176 (56.2)	33 (18.8)		32 (18.2)		38 (21.6)	
Household income			0.978(*1*), .323		1.144 (*1*), .285		0.074 (*1*), .785
<200% FPL[a]	95 (30.4)	24 (25.5)		22 (23.4)		23 (24.2)	
≥200% FPL	216 (69.0)	44 (20.5)		39 (18.1)		49 (22.8)	
Perceived financial difficulty			14.169 (*1*), <.001[b]		7.851 (*1*), .005[b]		7.298 (*1*), .007[b]
None to mild	220 (70.3)	36 (16.4)		34 (15.5)		42 (19.2)	
Moderate to severe	93 (29.7)	33 (35.9)		27 (29.3)		31 (33.3)	

[a] FPL

[b] $P<.01$

Discussion

Principal Findings

Given the increased prevalence of parent health-related technology use in recent years, this study aimed to explore family socioeconomic factors associated with this parenting behavior in a diverse sample of parents of young children. Considering that several developmental, socioemotional, and behavioral problems emerge in early childhood, understanding parent HTU use during this period has numerous clinical and public health implications. Indeed, children from lower SES households are more likely to experience reduced health quality and are less likely to have access to traditional health care services than children from higher SES households, and the relationship between these disparities and long-standing structural barriers is well established [32]. Further, research suggests similar barriers persist in access to technology devices and broadband, which may also challenge recent efforts to leverage technology to address health disparities [17,33,34]. Thus, understanding patterns and perceptions of parental HTU is critical for efforts to democratize digital health for parents of young children.

In the past decade, there has been a significant increase in technology device ownership in the United States, most substantially among smartphones and tablets [19]. Recruited families displayed a slightly higher percentage of smartphone, computer, tablet, and wearable device ownership and access in comparison to recent surveys of US adults [16,19], which may be reflective of our focus on parents (rather than adults in general), recruitment methods (eg, telephone interviews vs Amazon Mechanical Turk), or the inclusion of families in analyses with access to devices in other settings (eg, work, school, or library). Over three-fifths of parents endorsed ownership or access to a smartphone, tablet, and desktop or laptop computers, which did not vary across educational attainment, perceived financial difficulty, or household income. However, fewer than a third of parents reported access to a wearable device, and families with a lower income were significantly less likely to own a wearable (23% vs 36%).

Importantly, the majority of parents of young children reported using their laptop (150/177, 85%) or desktop computer (130/155, 84%) and wearable devices (45/55, 82%) daily, and the overwhelming majority of parents reported using their smartphone more than once per day. In contrast, only half of the parents reported using their tablet daily. Some, but not all, indicators of SES were significantly associated with how often parents used their smartphone (income and perceived financial difficulty), tablet (perceived financial difficulty), and desktop computer (parent educational attainment), with parents without a bachelor's degree, those experiencing moderate to severe financial difficulty, and those in lower-income households using their technology devices less frequently than their peers.

Regarding engagement in HTU among parents of young children, our findings were congruent with the high rates observed in previous studies of parent health information seeking via the internet [1]. However, these results extended the existing research by examining differential engagement across technology sources (eg, search engines, mobile apps, social media, and other digital media) in general and across sociodemographic groups. Consistent with previous research, nearly all parents in our study endorsed the use of search engines. In general, fewer parents reported using social media for health-related reasons in comparison to estimates of general social media use by parents (62% v 75%) [35]; however, existing work has primarily examined parents of infants, toddlers, or children under 18 years of age broadly [15]. Findings also indicate that less than half the parents of young children currently use mobile apps (38%) and other digital media (48%) to search for health-related information, advice, or support.

Additionally, there have been inconsistent findings regarding the relationship between family SES and parent HTU. For objective dimensions of SES, this is partly attributable to the underreporting of household income, household composition, and parent educational attainment in studies (40% did not report the education level of participants in a recent meta-analysis), as well as the recruitment of predominantly highly educated (over 50% to 75% with an academic degree) and higher-income parents among remaining studies [1]. Furthermore, to our knowledge, no studies to date have included subjective dimensions of SES in analyses (eg, perceived financial hardship, subjective social status), despite their distinct effects on parenting behavior and family health [36-41]. In contrast to studies observing higher rates of health-seeking behavior via the internet with increased parent educational attainment [1,42,43], our findings suggest no significant associations between parent educational attainment and engagement in or frequency of health-related technology use across sources. However, parents in lower-income households and those experiencing greater financial difficulty were significantly more likely to use social media (69% vs 56% for both) and mobile apps (45% vs 32% and 46% vs 32%, respectively) for health-related reasons. Parents who reported greater financial difficulty were also more likely to use other forms of digital media (58% vs 41%). Moreover, parents experiencing moderate to severe financial difficulty used social media less frequently than their peers. In terms of search content, both lower income and increased perceived financial difficulty (52% vs 39%) were associated with increased self-seeking behavior related to parent stress and stress management, and lower income was additionally associated with a decreased likelihood of parent engagement in proxy seeking related to their child's mental and physical health. Finally, parent perceptions of health-related technology use broadly did not vary by any objective dimensions of SES; however, parents experiencing moderate to severe financial difficulty were significantly more likely to perceive technology sources as the most comfortable (29% vs 15%), understanding (36% vs 16%), and likely to influence behavior change (33% vs 19%) compared to traditional sources. These findings support early research suggesting that SES indicators have differential impacts on health behavior and outcomes, providing a basis for further exploration of the underlying mechanisms contributing to outcomes in parent HTU.

Taken together, the results of this study underscore potential considerations for clinicians, researchers, and public health practitioners engaged in the design and dissemination of digital resources, programs, and interventions targeting family health and well-being. For instance, our findings suggest that digital health tools developed with greater attention to the types of technology sources parents prefer for health-related information, their frequency of engagement with these sources (eg, daily or weekly), and the availability of technology devices required to access these sources may yield increased uptake. Further, our results suggest practical considerations for efforts striving to optimize effectiveness (eg, which commercial devices and sources have the necessary features and functionality?), scalability (eg, what are the current estimates of, trends in, and barriers to adoption of these devices, especially in historically excluded communities?), and sustainability (eg, how acceptable and usable are both the devices and sources for the target population?). For example, digital resources, programs, and interventions requiring devices compatible solely with mobile operating systems (eg, mobile apps for Android, Apple iOS, and iPadOS) may call for a consideration of parent access to, familiarity with, and perceptions of smartphones and tablets, as well as their perceptions of mobile apps as a source for health-related information and support. Importantly, the success of these efforts hinges on broader attention to policies that address the structural information, infrastructure, and implementation barriers to diverse parents' safe and effective engagement in HTU, such as access to technology devices and reliable internet (eg, [44]) and threats to online safety (eg, health misinformation and disinformation [45-47]).

Limitations and Future Directions

Despite its strengths, this study has some limitations. First, primarily descriptive analyses were conducted to explore associations between sociodemographic factors and outcome variables. Second, inadequate representation of all racial and ethnic groups precluded our ability to examine how the diversity of social context and experiences across and within groups influence health-related technology use, which is a critical step in future research given the well-established disparities in digital adoption, health outcomes, and access to care among racially and ethnically minoritized children and their families [48-51]. Third, sources were grouped into technology (ie, search engines, mobile apps, social media, and other digital media) and traditional (ie, family, friends, mental health care providers, other health care providers, school professionals, and community leaders) categories for the analyses of perceptions of HTU, despite their potential interconnections in daily life (eg, use of social media to connect with family members about child-related health concerns, use of telemedicine apps for remote health care services). Future research should explore these complex relationships, which are likely linked to other relevant individual (eg, parent and child psychosocial factors and attitudes) and environmental (eg, social support, discrimination) factors associated with engagement in HTU and outcomes (eg, specific parent behaviors, family health outcomes, subsequent HTU). Finally, survey data were collected in late 2018 (prior to the COVID-19 pandemic), spotlighting the importance of future work examining potentially evolving trends in technology adoption and parent HTU.

Conclusion

In summary, this study investigated engagement in support, advice, and information-seeking behavior among parents of young children across technology devices and sources. It also examined resource access and perceptions that may influence engagement and explored patterns across family SES. Overall, this study supports the growing body of evidence demonstrating the potential for digital technologies to disseminate health-related information, support, and resources to young children and families facing structural socioeconomic barriers. Furthermore, it may inform future research necessary to advance understanding on how to more optimally tailor and deliver supports that benefit the health and well-being of all children.

Acknowledgments

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Conflicts of Interest

None declared.

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Abbreviations

FPL: federal poverty level
GED: General Education Diploma
HTU: health-related technology use
IRB: institutional review board
MTurk: Amazon Mechanical Turk
OHIS: online health information seeking
SES: socioeconomic status

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Review

Interactive Remote Patient Monitoring Devices for Managing Chronic Health Conditions: Systematic Review and Meta-analysis

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Abstract

Background: Telemedicine is an expanding and feasible approach to improve medical care for patients with long-term conditions. However, there is a poor understanding of patients' acceptability of this technology and their rate of uptake.

Objective: The aim of this study was to systematically review the current evidence on telemonitoring in the management of patients with long-term conditions and evaluate the patients' uptake and acceptability of this technology.

Methods: MEDLINE, Scopus, and CENTRAL (the Cochrane Central Register of Controlled Trials) were searched from the date of inception to February 5, 2021, with no language restrictions. Studies were eligible for inclusion if they reported any of the following outcomes: intervention uptake and adherence; study retention; patient acceptability, satisfaction, and experience using the intervention; changes in physiological values; all-cause and cardiovascular-related hospitalization; all-cause and disease-specific mortality; patient-reported outcome measures; and quality of life. In total, 2 reviewers independently assessed the articles for eligibility.

Results: A total of 96 studies were included, and 58 (60%) were pooled for the meta-analyses. Meta-analyses showed a reduction in mortality (risk ratio=0.71, 95% CI 0.56-0.89; *P*=.003; I^2=0%) and improvements in blood pressure (mean difference [MD]=−3.85 mm Hg, 95% CI −7.03 to −0.68; *P*=.02; I^2=100%) and glycated hemoglobin (MD=−0.33, 95% CI −0.57 to −0.09; *P*=.008; I^2=99%) but no significant improvements in quality of life (MD=1.45, 95% CI −0.10 to 3; *P*=.07; I^2=80%) and an increased risk of hospitalization (risk ratio=1.02, 95% CI 0.85-1.23; *P*=.81; I^2=79%) with telemonitoring compared with usual care. A total of 12% (12/96) of the studies reported adherence outcomes, and 9% (9/96) reported on satisfaction and acceptance outcomes; however, heterogeneity in the assessment methods meant that a meta-analysis could not be performed.

Conclusions: Telemonitoring is a valid alternative to usual care, reducing mortality and improving self-management of the disease, with patients reporting good satisfaction and adherence. Further studies are required to address some potential concerns regarding higher hospitalization rates and a lack of positive impact on patients' quality of life.

Trial Registration: PROSPERO CRD42021236291; https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=236291

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KEYWORDS

chronic condition; telemonitoring; telemedicine; eHealth; self-monitoring; systematic review; meta-analysis

Introduction

Background

In the United Kingdom, 15 million people live with at least one long-term condition [1], with their care accounting for 70% of the National Health Service budget [1]. Those with long-term conditions have significantly reduced quality of life (QoL) as well as an increased risk of morbidity and mortality [2,3]. Cardiovascular disease, diabetes mellitus, and chronic obstructive pulmonary disease (COPD) are the most common chronic conditions worldwide [4]. Lack of care coordination [5,6] and care planning consultation [5,6] are among the common barriers that patients with long-term conditions face. In addition, the restrictions induced by the COVID-19 pandemic have amplified the challenges that people living with chronic diseases experience in terms of managing their health and accessing health care [7].

Advances in technology have the potential to support patients with long-term conditions in managing their health at home, making the provision of remote health care more accessible and efficient [8]. Web-based health care and telemedicine include the remote delivery of care using communication technology (eg, videoconference software, web-based applications, and home-based health measurement) to enable consultations between patients and their care team, providing continuous monitoring of relevant health parameters. This allows health care professionals to promptly respond to changes in patient health status and adapt their clinical management in real time [9].

Objectives

Recent evidence has deemed telemedicine feasible for patients with long-term conditions and effective in terms of improving medical care [10]. As telemedicine is a rapidly expanding and changing field, recent umbrella reviews [10,11] that consider older primary studies have potentially made conclusions based on noncontemporary data. Therefore, the aim of this systematic review was to update and expand the current literature on telemonitoring by better defining the interventions included to encompass the role that interactive, 2-way communication devices play in improving the care of patients with long-term conditions, as well as evaluate patient uptake and acceptability of this technology.

This review aimed to address the following research questions: (1) What is the rate of uptake, patient retention, and patient satisfaction when using 2-way (patient-health care provider) remote patient monitoring devices to manage chronic health conditions? (2) What factors are associated with patient retention and satisfaction when using 2-way (patient-health care provider) remote patient monitoring devices to manage chronic health conditions? (3) Does the use of 2-way (patient-health care provider) remote patient monitoring devices for the management of chronic health conditions affect patient outcomes (eg, changes in physiological measurements, QoL, all-cause and cardiovascular-related hospitalizations, and all-cause and disease-specific mortality)?

Methods

Overview

This systematic review was registered on PROSPERO (International Prospective Register of Systematic Reviews; CRD42021236291) and conducted in accordance with the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines [12].

Criteria for Considering Studies to Include in the Review

Studies carried out in any setting aiming to evaluate telemonitoring interventions for participants with at least one chronic condition among the following—cardiovascular disease, COPD, or diabetes mellitus—were eligible for inclusion. All randomized controlled trials (RCTs) and nonrandomized trials, before-and-after (pre-post) studies, and interrupted time series were considered for inclusion. Cross-sectional studies and case reports were excluded. Qualitative studies were included to assess participant satisfaction. Ongoing studies (if any) were also considered and presented in a dedicated table.

Participants

Adult participants (aged ≥18 years) were eligible for inclusion in this review if they reported one or more of the following chronic health conditions: cardiovascular diseases (eg, coronary artery disease, atrial fibrillation, stroke, heart failure, and hypertension), COPD, or diabetes mellitus.

Intervention

Interventions designed to remotely collect health information from patients using digital technologies and electronically transfer the information to health care professionals for monitoring and assessment were eligible for inclusion. Only interventions where the participant received a digital device for remote patient monitoring and the participant or their caregiver took physiological measurements and either input the information into the device or the device automatically uploaded the data were included. Health devices suitable for inclusion had to transmit data to the participant's health care team, and the participant's health care team had to monitor the information received, assessing it and making appropriate changes to the participant's treatment accordingly. A 2-way exchange of information was required for a study to be included.

Comparator

Studies in which usual care or a different intervention was used as control or comparator were also considered as eligible for inclusion, as were studies that did not have a control group.

Outcomes

The primary outcomes of interest were (1) intervention uptake (number of people willing to participate in the intervention) and adherence (level of commitment of the patient to the prescribed intervention); (2) study retention (number of people who completed the intervention); and (3) patient acceptability (level of acceptance of the intervention by the participants), satisfaction (number of participants pleased with the intervention), and experience using the intervention. Secondary outcomes included (1) changes in physiological measurements (eg, oxygen saturation, blood pressure [BP], and blood glucose level); (2) all-cause and cardiovascular-related hospitalizations; (3) all-cause and disease-specific mortality; (4) patient-reported outcome measures (eg, mental well-being, depression, and anxiety questionnaires); and (5) QoL, quality-adjusted life years, and any other health economic outcomes reported in the studies. All the studies that reported one or more of these outcomes were considered eligible for inclusion.

Search Strategy

The search strategy was developed by the review team, which agreed on the key terms. Medical Subject Headings terms and synonyms for the different terms, such as “telemedicine,” “digital monitoring,” and “e-health” (Table S1 in Multimedia Appendix 1 [13-163]), were used and combined with Boolean operators, proximity operators, truncations, and wildcards. MEDLINE, Scopus, and CENTRAL (the Cochrane Central Register of Controlled Trials) were searched from the date of inception to February 5, 2021, for relevant studies. There were no language restrictions, but the availability of the full text was a requirement for inclusion. Search results were managed using EndNote (version X9.3.3; Clarivate Analytics).

Study Selection

Two reviewers (MC and DGL) independently screened the titles and abstracts of the studies retrieved from the databases against the search criteria. Additional screening of the preliminary results was independently undertaken by 3 other reviewers (BB, SH, and MI). The full texts of all potentially relevant articles were retrieved and independently assessed by the reviewers in duplicate. Any disagreement was resolved through discussion with the senior author (DL).

Data Extraction

Data extraction was conducted independently by 2 reviewers (DGL and MC). The following information was extracted: (1) authors, year, country, and reference; (2) study aim; (3) study characteristics (study design and sample size); (4) participant characteristics (age, sex, and ethnicity); (5) health condition; (6) intervention (type of telemedicine device, input of the data [manual or automated], delivery of the intervention, staff involved, duration and frequency of the intervention, and follow-up points); (7) comparators (usual care, different intervention, or no intervention); and (8) outcomes (primary and secondary, as reported in the study).

Risk of Bias Assessment

Six authors (DGL, MC, BB, SH, MI, and DL) independently assessed the individual studies for risk of bias in duplicate, and any discrepancies were resolved via discussion or referral to a third reviewer, as required. For RCTs, the Cochrane Risk of Bias version 2 tool [164] was used. For nonrandomized studies, the Risk Of Bias In Non-randomized Studies of Interventions [165] was used.

Data Synthesis

Meta-analyses were conducted on comparable studies. Primary and secondary outcome effect measures with 95% CIs were pooled using the RevMan software (The Cochrane Collaboration) [166]. The results are presented visually using forest plots. Where continuous data were not homogeneous, an estimate of the standardized mean difference (MD) with 95% CIs was calculated. For studies in which quantitative data were too few or too heterogeneous, a narrative synthesis approach was used.

Dichotomous analyses were conducted using the number of events and total sample size as reported in the included studies. The results of the selected studies were combined using the Mantel-Haenszel method. Effect sizes are expressed as relative risk and 95% CIs. Random effect models were applied to all meta-analyses owing to heterogeneity in study characteristics and populations. Heterogeneity was quantitatively assessed using the Higgins index (I^2).

For the analysis of QoL, the postintervention scores, as reported in the included studies, were used. Where the SD was not reported, it was calculated using the calculator function available in RevMan. For analysis of changes in physiological parameters (BP and glycated hemoglobin [HbA_{1c}]) and QoL, the results of the selected studies were combined using the generic inverse variance method. Effect sizes are expressed as the MD and SD.

Findings from the included qualitative studies will be synthesized elsewhere using a meta-aggregative approach to data synthesis.

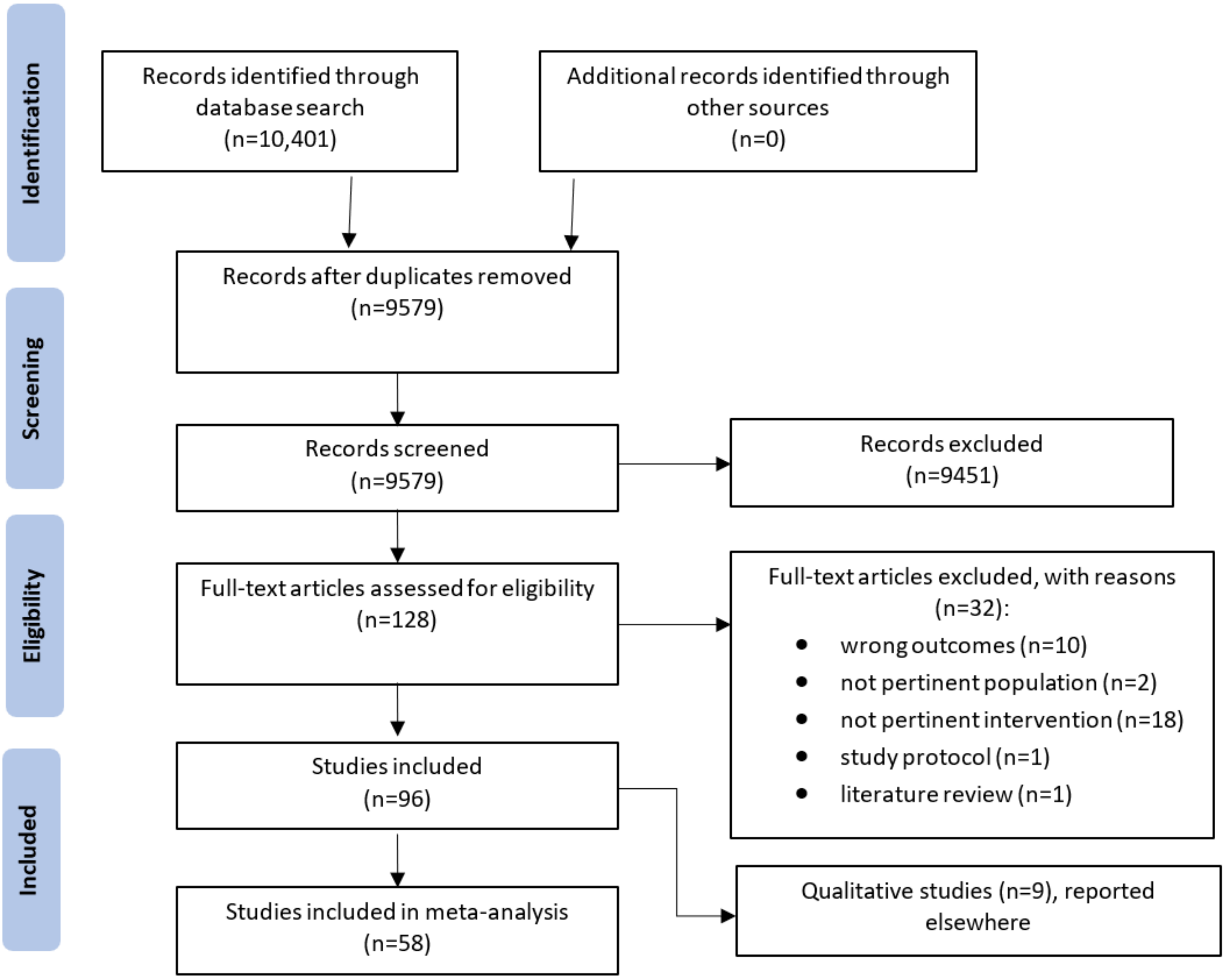
Results

Overview

The database searches identified 10,401 papers. After independent screening of titles and abstracts by 2 study authors, 98.77% (10,273/10,401) of papers were determined to be duplicates or not eligible. After screening against the inclusion and exclusion criteria, of the remaining 128 papers, 96 (75%) were included. No ongoing studies were found (Figure 1). A full list of the excluded studies with reasons for exclusion is provided in Table S2 in Multimedia Appendix 1. Full texts of all 96 included papers [13-109] were retrieved.

No study reporting outcomes related to intervention uptake, study retention, and patient acceptability were identified in our search and, therefore, these outcomes could not be analyzed. The following analyses and results concern only patient adherence and satisfaction as well as clinical and patient-reported outcomes.

Figure 1. PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) diagram depicting the screening and study selection process.



Characteristics of the Included Studies

The included studies were published between 1998 and 2020, with sample sizes ranging from 20 [36,99] to 3562 [102] participants and a total sample of 26,167 participants. The mean age ranged from 44 [22] to 78 [107] years, and the proportion of men varied from 25% [51] to 76% [91]. Most of the included studies were conducted in the United Kingdom (21/96, 22%) and the United States (29/96, 30%), with additional studies conducted in Belgium (2/96, 2%), Canada (4/96, 4%), Denmark (5/96, 5%), Poland (2/96, 2%), Singapore (2/96, 2%), South Korea (2/96, 2%), Spain (9/96, 9%), Germany (4/96, 4%), and Italy (6/96, 6%; Multimedia Appendix 2 [13-109,136]). In addition, the following countries had 1% (1/96) of the studies each: Australia [37], China [99], Finland [106], Greece [49], Hong Kong [28], Israel [14], Japan [66], Malaysia [67], the Netherlands [25], and Taiwan [29] (Multimedia Appendix 2).

Populations in the included studies comprised patients with diabetes (27/96, 28% of the studies), cardiovascular disease (stroke, atrial fibrillation, hypertension, and heart failure; 52/96, 54% of the studies), COPD (12/96, 12% of the studies), and mixed chronic conditions (diabetes, hypertension, and COPD; 5/96, 5% of the studies; Multimedia Appendix 2).

Types of Interventions

The studies varied in their design, type of telemonitoring system used, and method of delivery (Multimedia Appendix 2). Most (64/96, 67%) were RCTs, with 4% (4/96) being nonrandomized controlled studies, 2% (2/96) being cluster randomized studies, 10% (10/96) being longitudinal studies, 4% (4/96) being retrospective analyses, 3% (3/96) being pre-post analyses, and 9% (9/96) having a mixed methods or qualitative design. Most studies (88/96, 92%) used telemonitoring systems that collected patient information via computers, tablets, or dedicated devices (eg, modem) and transferred these data to a web-based server. Some studies collected patient data via SMS text message (3/96, 3%) or by telephone (4/96, 4%). A total of 4% (4/96) of the studies provided educational videos to increase the patients' knowledge of the disease. The length of the intervention was highly variable, with 5% (5/96) of the studies assessing it over a short period (7-45 days), 21% (20/96) assessing it over a 2- to 4-month period, and most interventions (76/96, 79%) lasting 6 to 12 months. The follow-up periods were inconsistent among the studies and, where present, ranged from 3 to 18 months.

Types of Comparators

Most studies (79/96, 82%) compared the intervention with usual care, which consisted of routine visits (outpatient clinics) and in-person consultations with general practitioners or the hospital care team (Multimedia Appendix 2). A total of 10% (10/96) of the studies did not have a control group. A total of 1% (1/96) of the studies asked the control group to manually record their data in a diary. In total, 2% (2/96) of the studies used educational videos in the control group to improve patients' knowledge of the disease, another 2% (2/96) compared the intervention with another telemonitoring device, and 1% (1/96) compared the intervention (telemonitoring device) with telephone communication. A total of 1% (1/96) of the studies used a similar intervention as the control group comparing patients with and without heart failure.

Types of Outcomes

In total, 12 studies reported adherence to the intervention, including 9 (75%) in patients with cardiovascular disease, 2 (17%) in patients with diabetes, and 1 (8%) in patients with COPD (Multimedia Appendix 2). Patient satisfaction with the intervention was assessed in 9% (9/96) of the studies (2/9, 22% in patients with cardiovascular disease; 3/9, 33% in patients with diabetes; 2/9, 22% in patients with COPD; and 2/9, 22% in a mixed population; Multimedia Appendix 2).

Most studies (31/96, 32%) reported changes in physiological parameters, which varied depending on the population observed, with 39% (12/31) of these studies reporting BP values for patients with cardiovascular disease, 55% (17/31) reporting HbA_{1c} values for patients with diabetes, and 6% (2/31) reporting multiple physiological values in mixed populations (Multimedia Appendix 2).

Hospital admission during the intervention was recorded in 29% (28/96) of the studies (21/28, 75% in patients with cardiovascular disease; 4/28, 14% in patients with COPD; and 3/28, 11% in a mixed sample), and death was noted in 18% (17/96) of the studies (14/17, 82% in patients with cardiovascular disease; 2/17, 12% in patients with COPD; and 1/17, 6% in a mixed population; Multimedia Appendix 2).

QoL before and after the intervention was recorded in 22% (21/96) of the studies (11/21, 52% in patients with cardiovascular disease; 2/21, 10% in patients with diabetes; 6/21, 29% in patients with COPD; and 2/21, 10% in a mixed population; Multimedia Appendix 2).

Excluded Studies

A total of 25% (32/128) of the studies assessed for eligibility [110-141] were excluded. A summary of these studies can be found in Table S2 in Multimedia Appendix 1. Most (18/32, 56%) were excluded as they were not related to a telemonitoring intervention, 6% (2/32) included disease populations not covered in this review, 31% (10/32) reported outcomes outside the scope of this review, 3% (1/32) were literature reviews, and 3% (1/32) were study protocols.

Risk of Bias Assessment

A summary of the risk of bias assessment of the included studies can be found in Tables S3-S5 in Multimedia Appendix 1. Overall, most RCTs (48/66, 73%) and non-RCTs (17/20, 85%) included in this review showed either some concerns or a high risk of bias. Most RCT studies (45/66, 68%) showed either some concerns or a high risk of bias in the randomization process as well as in the selection of the reported results. Some RCTs (18/66, 27%) showed either some concerns or a high risk of bias in missing outcome data. Few RCTs (17/66, 26%) showed either some concerns or a high risk of bias in the measurement of the outcomes.

Most of the non-RCTs (18/20, 90%) showed either some concerns or a high risk of bias in the *bias due to confounding* category. A total of 50% (10/20) of the studies showed either some concerns or a high risk of bias in the *bias in measurement of outcomes* category. Few of the non-RCTs (9/20, 45%) showed either some concerns or a high risk of bias in the *bias due to missing data* category as well as in the *bias due to deviations from the intended intervention* category.

The studies included in the meta-analyses were assessed for publication bias. Funnel plots and Egger tests were performed only where ≥10 studies were available [167].

Funnel plots for the outcomes of systolic BP (SBP), HbA_{1c}, and mortality can be found in Figures S1-S6 in Multimedia Appendix 1. The Egger test results revealed no evidence of publication bias for SBP, HbA_{1c}, or mortality.

Ongoing Studies

The database search did not return any protocols for ongoing studies. Searches on ClinicalTrials.gov (updated to February 5, 2021) identified 22 ongoing studies [142-163] (n=14, 64% on patients with cardiovascular disease; n=4, 18% on patients with diabetes; and n=4, 18% on patients with COPD), which are reported in detail in Table S6 in Multimedia Appendix 1.

Primary Outcomes

Adherence

Adherence was assessed in 12 studies at different time points: 1 month (n=3, 25%) [51,66,84], 6 weeks (n=2, 17%) [58,103], 2 months (n=1, 8%) [13], 3 months (n=1, 8%) [30], 6 months (n=4, 33%) [42,48,59,92], and 12 months (n=1, 8%) [36]. Of the 12 studies, 7 (58%) [13,36,42,48,58,59,92] demonstrated a benefit of telemonitoring on patient adherence when compared with a comparator, whereas 4 (33%) [30,51,66,84] showed no difference when compared with a comparator. A total of 8% (1/12) of the studies [103] compared 2 telemonitoring systems and showed that educational support combined with telemonitoring positively influenced adherence compared with telemonitoring alone. Owing to variations in how adherence was defined in the studies, a meta-analysis was not performed. A summary of these studies is presented in Table 1.

Table 1. Studies examining the impact of telemonitoring interventions versus comparator on adherence (N=12).

Study type and authors, year, and country	Study population, N	Condition	Intervention type, number of participants, age (years), men (n [%])	Comparator, number of participants, age (years), mean (n [%])	Outcomes	Follow-up	Impact of telemonitoring
Randomized controlled trials							
Ong et al [84], 2016, United States	1437	CHF[a]	Automated upload of data on dedicated device or software, 715, mean 73 (SD not reported), men: 382 (53.4); women: 333 (46.6)	Usual care, 722, mean 73 (SD not reported), men: 382 (53.4); women: 333 (46.6)	Adherence electronically recorded; 82.7%	1 month	=[b]
Gallagher et al [51], 2017, United States	40	HF[c]	Manual upload of data on dedicated device or software, 20, median 68 (IQR 49-79), men: 15 (75); women: 5 (25)	Usual care, 20, median 62 (IQR 52-75), men: 15 (75); women: 5 (25)	Adherence recorded electronically; 81% in both groups	1 month	=
Kotooka et al [66], 2018, Japan	183	CHF	Automated upload of data on dedicated device or software, 93, mean 67.1 (SD 12.8), men: 51 (56); women: 39 (44)	Usual care, 91, mean 65.4 (SD 15.6), men: 56 (61); women: 35 (39)	Adherence recorded electronically; 90% at 12 months	12 months	=
Varon et al [103], 2015, United Kingdom	534	HF	Docobo system (telemonitoring only), 135, mean 69.1 (SD 12.6), not reported	Motiva system (telemonitoring+ educational videos), 399, mean 69.1 (SD 12.6), not reported	Adherence assessed by the amount of missing data during the telemonitoring period	6 weeks	–[d]
Kardas et al [58], 2016, Poland	60	Type 2 diabetes	Automated upload of data on dedicated device or software, 30, mean 59.9 (SD 5.31), men: 17 (57); women: 13 (43)	Usual care, 30, mean 59 (SD 8.9), men: 19 (63); women: 11 (47)	Adherence expressed as medication taken vs medication prescribed; 92.9%	6 weeks	+[e]
Cho et al [30], 2009, South Korea	69	Type 2 diabetes	Mobile app, 35, mean 51.1 (SD 13.1), 26 men; 74 women[f]	Web-based telemonitoring system, 34, mean 51.1 (SD 13.1), 26 men; 74 women[f]	Adherence, self-reported; >70% in both groups	3 months	=
Seto et al [92], 2012, Canada	100	CHF	Automated upload of data on dedicated device or software, 50, mean 55.1 (SD 13.7), men: 41 (82); women: 9 (18)	Usual care, 50, mean 52.3 (SD 13.7), men: 38 (76); women: 12 (24)	Adherence registered electronically; 80%	6 months	+
Evans et al [48], 2016, United States	441	HF and healthy	Disease group: automated upload of data on dedicated device or software, 421, mean 71.8 (SD 8.8), 46 men; 54 women[f]	Healthy group: automated upload of data on dedicated device or software, 20, mean 72.2 (SD 4.3), 50 men; 50 women[f]	Adherence checking the amount of data against the participants' time spent in the study; between 71% and 81%	6 months	+
Nonrandomized studies							
Agboola et al [13], 2013, United States	30	Hypertension	Web-based device, 15, mean 61.9 (SD not reported), 20 men; 80 women[f]	Mobile blood pressure device, 15, mean 61.6 (SD not reported), 20 men; 80 women[f]	Adherence recorded electronically based on frequency of data transmission	2 months	+
Domingo et al [42], 2012, Spain	97	HF	Automated upload of data on dedicated device or software, 46, mean 66.5 (SD 11.5), men: 14 (30); women: 32 (70)	Usual care, 51, mean 66.5 (SD 11.5), men: 15 (30); women: 36 (70)	Adherence based on the number of educational videos watched; between 67% and 85%	6 months	+

Study type and authors, year, and country	Study population, N	Condition	Intervention type, number of participants, age (years), men (n [%])	Comparator, number of participants, age (years), mean (n [%])	Outcomes	Follow-up	Impact of telemonitoring
Karg et al [59], 2012, Germany	36	COPD[g]	Automated upload of data on dedicated device or software, 36, mean 67.9 (SD 6.9), men: 27 (75); women: 9 (25)	N/A[h]	Adherence: use of the device for at least two-thirds of working days; full compliance	6 months	+
De Lusignan et al [36], 2001, United Kingdom	20	CHF	Manual upload of data on dedicated device or software, 10, mean 75.2 (SD not reported), not reported	Usual care, 10, mean 75.2 (SD not reported), not reported	Adherence based on the frequency of the uploaded data; 90%	12 months	+

[a] CHF: congestive heart failure.

[b] No differences between telemonitoring and usual care.

[c] HF: heart failure.

[d] Negative impact of telemonitoring over comparator.

[e] Positive impact of telemonitoring over comparator.

[f] Absolute value not reported in the paper.

[g] COPD: chronic obstructive pulmonary disease.

[h] N/A: not applicable.

Satisfaction

Patient satisfaction with the intervention was assessed in 9 studies (n=2, 22% in patients with cardiovascular disease; n=3, 33% in patients with diabetes; n=2, 22% in patients with COPD; and n=2, 22% in a mixed population; Table 2). A total of 56% (5/9) of the studies [22,28,42,78,91] demonstrated a benefit of telemonitoring on patient satisfaction when compared with a comparator, whereas 44% (4/9) [30,43,44,95] showed no difference when compared with a comparator. Owing to variations in how satisfaction was defined in the studies, a meta-analysis was not performed. A summary of these studies is provided in Table 2.

Table 2. Studies examining the impact of telemonitoring interventions versus comparator on satisfaction (N=9).

Study type and authors, year, and country	Study population, N	Condition	Intervention type, number of participants, age (years), mean (n [%])	Comparator, number of participants, age (years), mean (n [%])	Outcomes	Follow-up	Impact of telemonitoring
Randomized controlled trials							
Bergenstal et al [22], 2005, United States	47	Type 2 diabetes	Automated data transmitted via modem, 24, mean 44 (SD 17), 37 men; 63 women[a]	Data transmitted via telephone, 23, mean 45 (SD 13), 39 men; 61 women[a]	Satisfaction: 5-point questionnaire; 4.30 in the phone group and 4.52 in the modem group	4 weeks	=[b]
Chau et al [28], 2012, Hong Kong	40	COPD[c]	Manual upload of data on dedicated device or software, 22, mean 73.5 (SD 6), men: 21 (95); women: 1 (5)	Usual care, 18, mean 72.2 (SD 6), men: 18 (100); women: 0 (0)	Satisfaction: 10-item questionnaire based on a 5-point system; 91%	2 months	+[d]
Edmonds et al [44], 1998, Canada	35	Type 2 diabetes	Mobile phone data transmission, 16, not reported, not reported	Usual care, 19, not reported, not reported	Satisfaction: patient questionnaire	3 months	Further studies required
Cho et al [30], 2009, South Korea	69	Type 2 diabetes	Mobile app, 35, mean 51.1 (SD 13.1), 26 men; 74 women[a]	Web-based telemonitoring system, 34, mean 51.1 (SD 13.1), 26 men; 74 women[a]	Satisfaction: questionnaire, internet vs phone; 81% vs 79%, respectively	3 months	=
Sicotte et al [95], 2011, Canada	46	COPD	Manual upload of data on dedicated device or software, 23, mean 73.7 (SD 9.6), men: 13 (56); women: 10 (44)	Usual care, 23, mean 75.4 (SD 9.7), men: 13 (56); women: 10 (44)	Satisfaction: 5-point questionnaire; 4.50 score	3 months	=
Domingo et al [42], 2012, Spain	97	HF[e]	Automated upload of data on dedicated device or software, 46, mean 66.5 (SD 11.5), men: 14 (30); women: 32 (70)	Usual care, 51, mean 66.5 (SD 11.5), men: 15 (30); women: 36 (70)	Satisfaction: 10-point questionnaire; 8.4 score	6 months	+
Nonrandomized studies							
Schoenfeld et al [91], 2004, United States	59	CHF[f]	Manual upload of data on dedicated device or software, 59, mean 64 (SD 14), men: 45 (76); women: 14 (24)	N/A[g]	Satisfaction: 3-point questionnaire; 98.1% indicating ease of use of the device	7 days	+
Donate-Martinez et al [43], 2016, Spain	74	Chronic conditions (COPD, type 2 diabetes, and HF)	Manual upload of data on dedicated device or software, 74, mean 67.95 (SD 11.14), men: 49 (66); women: 25 (44)	N/A	Satisfaction: 11-item questionnaire with 10-point score; 8.63 score overall	12 months	=
Mira-Solves et al [77], 2014, Spain	410	Chronic conditions (type 2 diabetes, hypertension, CHF, and COPD)	Automated upload of data on dedicated device or software, 410, not reported, 64 men; 36 women[a]	N/A	Satisfaction: questionnaire, 89.4% were satisfied with the ease of use.	24 months	+

[a] Absolute value not reported in the paper.

[b] No differences between telemonitoring and usual care.

[c] COPD: chronic obstructive pulmonary disease.

[d] Positive impact of telemonitoring over comparator.

[e] HF: heart failure.

[f] CHF: congestive HF.

[g] N/A: not applicable.

Secondary Outcomes

QoL Measurement

Studies included in the meta-analyses were pooled by comparable scales (eg, the Short Form 36 Health Survey Questionnaire) and end points (eg, 6 or 12 months), with 8% (8/96) of the studies [16,31,33,35,47,96,101,104] included in the meta-analyses.

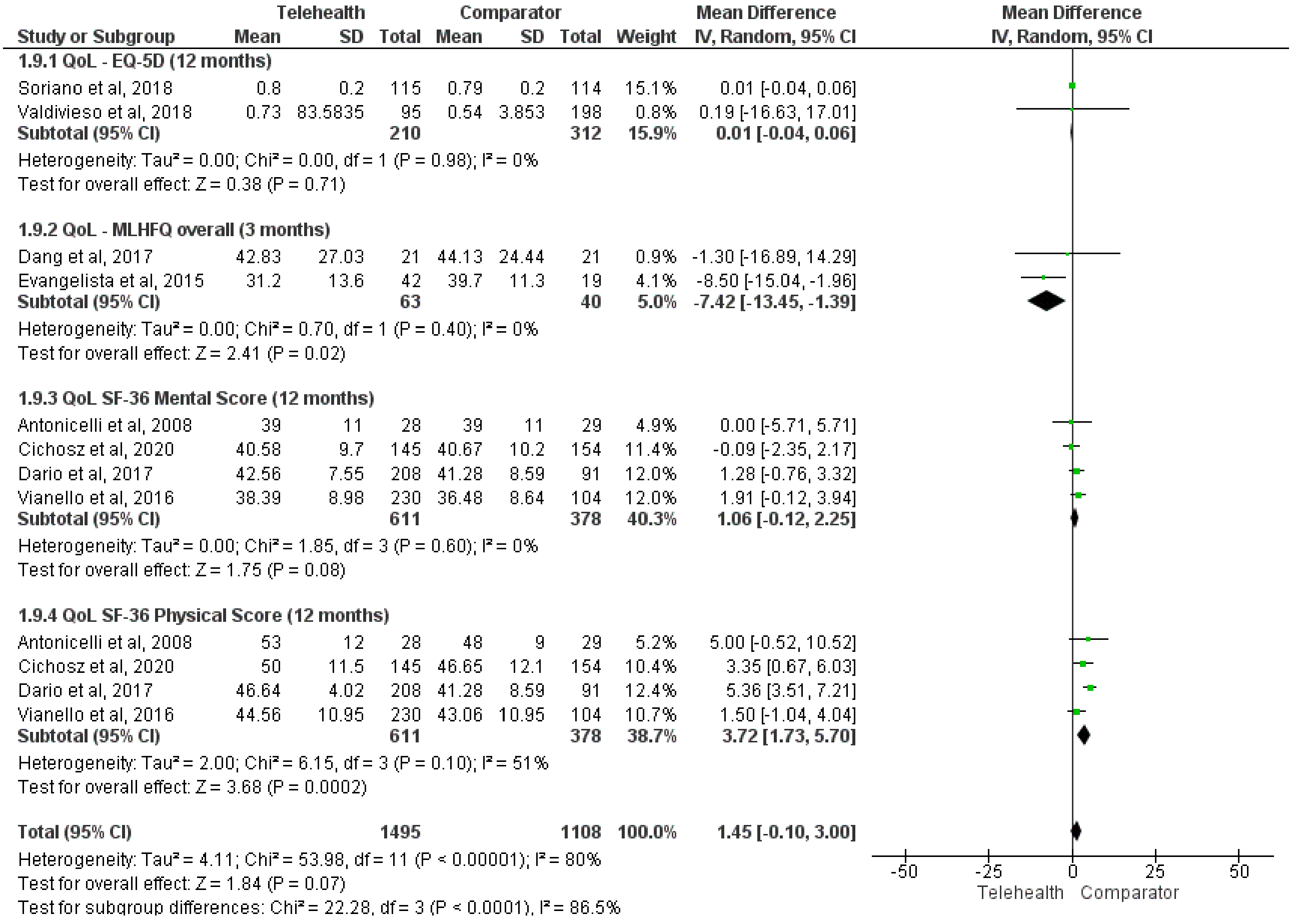
A total of 50% (4/8) of these studies [16,31,35,104] reported the Short Form 36 Health Survey Questionnaire scores (mental and physical) at comparable end points (12 months) and were included in the meta-analyses (Figure 2 [15,31,35,47,96,101,104,136], subgroups 1.9.3 and 1.9.4). From the meta-analysis, telemonitoring showed greater improvements compared with usual care on physical component scores (weighted MD=3.72, 95% CI 1.73-5.70; $P<.001$; I^2=51%; Figure 2) compared with the comparator but no difference in mental component scores (weighted MD=1.06, 95% CI −0.12 to 2.25; P=.08; I^2=0%; Figure 3 [15,39,40,50,60,64,84,96,101,105,107]).

In total, 25% (2/8) of the studies [96,101] reported EQ-5D scores at comparable end points (12 months) and were included in the meta-analysis (Figure 2, subgroup 1.9.1). There was no difference in QoL between the groups (weighted MD=0.01, 95% CI −0.04 to 0.06; P=.71; I^2=0%)

A total of 25% (2/8) of the studies [33,47] using the Minnesota Living with Heart Failure Questionnaire overall scores at 3 months were included in the meta-analysis (Figure 2, subgroup 1.9.2), demonstrating that the telemonitoring group showed greater improvements in QoL (weighted MD=−7.42, 95% CI −13.45 to −1.39; P=.02; I^2=0%) compared with the comparator.

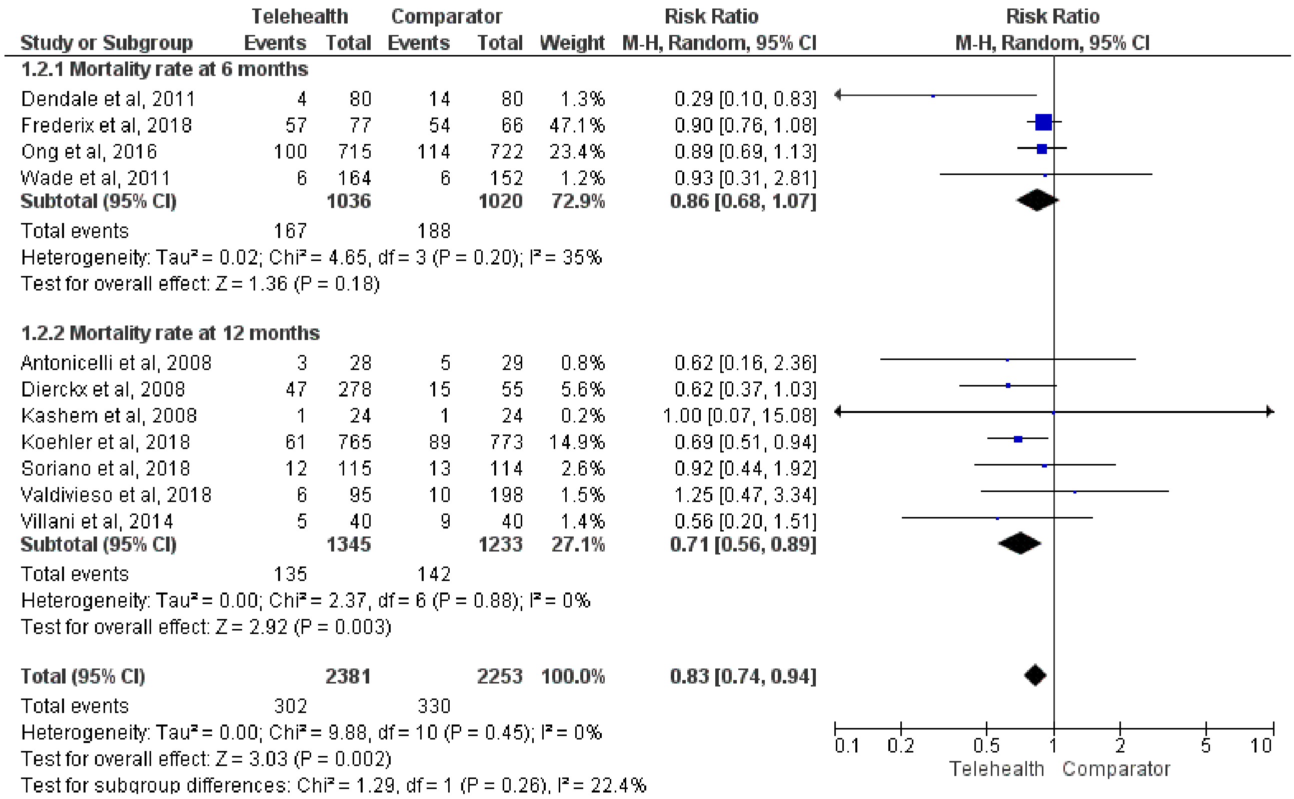
A total of 14% (13/96) of the studies [20,23,36,43,58,62,65,70,92,100,103,107,108] could not be included in the meta-analysis because they reported different time points and used different questionnaires to assess QoL. Of these 13 studies, 4 (31%) reported a significant improvement in QoL in the telemonitoring group compared with usual care at 6 weeks [58], 6 months [92,100], and 12 months [43] measured using a variety of questionnaires (Minnesota Living with Heart Failure Questionnaire [92], EQ-5D [43,58], and 15D [100]), whereas 9 (69%) reported no difference in QoL between telemonitoring and usual care at 4 weeks [70], 6 weeks [65,103], 7 weeks [70], 3 months [36], 6 months [23,62,107], 9 months [108], and 12 months [36]. A total of 8% (1/13) of the studies [20] reported significant improvement in QoL in the usual care group compared with telemonitoring at 2 and 6 months using the St George's Respiratory Questionnaire.

Figure 2. Impact of telemonitoring versus comparator on quality of life (QoL). 1.9.1: EQ-5D; 1.9.2: Minnesota Living with Heart Failure Questionnaire (MLHFQ); 1.9.3: SF-36 mental score; and 1.9.4: SF-36 physical component [15,31,35,47,96,101,104,136].



Study or Subgroup	Telehealth Mean	Telehealth SD	Telehealth Total	Comparator Mean	Comparator SD	Comparator Total	Weight	Mean Difference IV, Random, 95% CI
1.9.1 QoL - EQ-5D (12 months)								
Soriano et al, 2018	0.8	0.2	115	0.79	0.2	114	15.1%	0.01 [-0.04, 0.06]
Valdivieso et al, 2018	0.73	83.5835	95	0.54	3.853	198	0.8%	0.19 [-16.63, 17.01]
Subtotal (95% CI)			**210**			**312**	**15.9%**	**0.01 [-0.04, 0.06]**
Heterogeneity: Tau² = 0.00; Chi² = 0.00, df = 1 (P = 0.98); I² = 0%								
Test for overall effect: Z = 0.38 (P = 0.71)								
1.9.2 QoL - MLHFQ overall (3 months)								
Dang et al, 2017	42.83	27.03	21	44.13	24.44	21	0.9%	-1.30 [-16.89, 14.29]
Evangelista et al, 2015	31.2	13.6	42	39.7	11.3	19	4.1%	-8.50 [-15.04, -1.96]
Subtotal (95% CI)			**63**			**40**	**5.0%**	**-7.42 [-13.45, -1.39]**
Heterogeneity: Tau² = 0.00; Chi² = 0.70, df = 1 (P = 0.40); I² = 0%								
Test for overall effect: Z = 2.41 (P = 0.02)								
1.9.3 QoL SF-36 Mental Score (12 months)								
Antonicelli et al, 2008	39	11	28	39	11	29	4.9%	0.00 [-5.71, 5.71]
Cichosz et al, 2020	40.58	9.7	145	40.67	10.2	154	11.4%	-0.09 [-2.35, 2.17]
Dario et al, 2017	42.56	7.55	208	41.28	8.59	91	12.0%	1.28 [-0.76, 3.32]
Vianello et al, 2016	38.39	8.98	230	36.48	8.64	104	12.0%	1.91 [-0.12, 3.94]
Subtotal (95% CI)			**611**			**378**	**40.3%**	**1.06 [-0.12, 2.25]**
Heterogeneity: Tau² = 0.00; Chi² = 1.85, df = 3 (P = 0.60); I² = 0%								
Test for overall effect: Z = 1.75 (P = 0.08)								
1.9.4 QoL SF-36 Physical Score (12 months)								
Antonicelli et al, 2008	53	12	28	48	9	29	5.2%	5.00 [-0.52, 10.52]
Cichosz et al, 2020	50	11.5	145	46.65	12.1	154	10.4%	3.35 [0.67, 6.03]
Dario et al, 2017	46.64	4.02	208	41.28	8.59	91	12.4%	5.36 [3.51, 7.21]
Vianello et al, 2016	44.56	10.95	230	43.06	10.95	104	10.7%	1.50 [-1.04, 4.04]
Subtotal (95% CI)			**611**			**378**	**38.7%**	**3.72 [1.73, 5.70]**
Heterogeneity: Tau² = 2.00; Chi² = 6.15, df = 3 (P = 0.10); I² = 51%								
Test for overall effect: Z = 3.68 (P = 0.0002)								
Total (95% CI)			**1495**			**1108**	**100.0%**	**1.45 [-0.10, 3.00]**
Heterogeneity: Tau² = 4.11; Chi² = 53.98, df = 11 (P < 0.00001); I² = 80%								
Test for overall effect: Z = 1.84 (P = 0.07)								
Test for subgroup differences: Chi² = 22.28, df = 3 (P < 0.0001), I² = 86.5%								

Figure 3. Impact of telemonitoring versus comparator on the mortality rate at 6 and 12 months. The study by Mortara et al [80] was not included in the mortality meta-analyses because of the use of a composite outcome of mortality and hospitalization where absolute mortality results were not available. The study by Seto et al [92] was not included in the mortality meta-analyses because of 0 events in the control group [15,39,40,50,60,64,84,96,101,105,107].



Study or Subgroup	Telehealth Events	Telehealth Total	Comparator Events	Comparator Total	Weight	Risk Ratio M-H, Random, 95% CI
1.2.1 Mortality rate at 6 months						
Dendale et al, 2011	4	80	14	80	1.3%	0.29 [0.10, 0.83]
Frederix et al, 2018	57	77	54	66	47.1%	0.90 [0.76, 1.08]
Ong et al, 2016	100	715	114	722	23.4%	0.89 [0.69, 1.13]
Wade et al, 2011	6	164	6	152	1.2%	0.93 [0.31, 2.81]
Subtotal (95% CI)		**1036**		**1020**	**72.9%**	**0.86 [0.68, 1.07]**
Total events	167		188			
Heterogeneity: Tau² = 0.02; Chi² = 4.65, df = 3 (P = 0.20); I² = 35%						
Test for overall effect: Z = 1.36 (P = 0.18)						
1.2.2 Mortality rate at 12 months						
Antonicelli et al, 2008	3	28	5	29	0.8%	0.62 [0.16, 2.36]
Dierckx et al, 2008	47	278	15	55	5.6%	0.62 [0.37, 1.03]
Kashem et al, 2008	1	24	1	24	0.2%	1.00 [0.07, 15.08]
Koehler et al, 2018	61	765	89	773	14.9%	0.69 [0.51, 0.94]
Soriano et al, 2018	12	115	13	114	2.6%	0.92 [0.44, 1.92]
Valdivieso et al, 2018	6	95	10	198	1.5%	1.25 [0.47, 3.34]
Villani et al, 2014	5	40	9	40	1.4%	0.56 [0.20, 1.51]
Subtotal (95% CI)		**1345**		**1233**	**27.1%**	**0.71 [0.56, 0.89]**
Total events	135		142			
Heterogeneity: Tau² = 0.00; Chi² = 2.37, df = 6 (P = 0.88); I² = 0%						
Test for overall effect: Z = 2.92 (P = 0.003)						
Total (95% CI)		**2381**		**2253**	**100.0%**	**0.83 [0.74, 0.94]**
Total events	302		330			
Heterogeneity: Tau² = 0.00; Chi² = 9.88, df = 10 (P = 0.45); I² = 0%						
Test for overall effect: Z = 3.03 (P = 0.002)						
Test for subgroup differences: Chi² = 1.29, df = 1 (P = 0.26), I² = 22.4%						

Mortality

Meta-analyses for mortality were conducted at the 6- and 12-month follow-up (Figure 3). Sensitivity analyses were conducted at the 6- and 12-month follow-up excluding studies at high risk of bias and at 12 months excluding non-RCTs (Figure S1 in Multimedia Appendix 1). A sensitivity analysis with the exclusion of non-RCTs at 6 months was not conducted as all the studies included were RCTs.

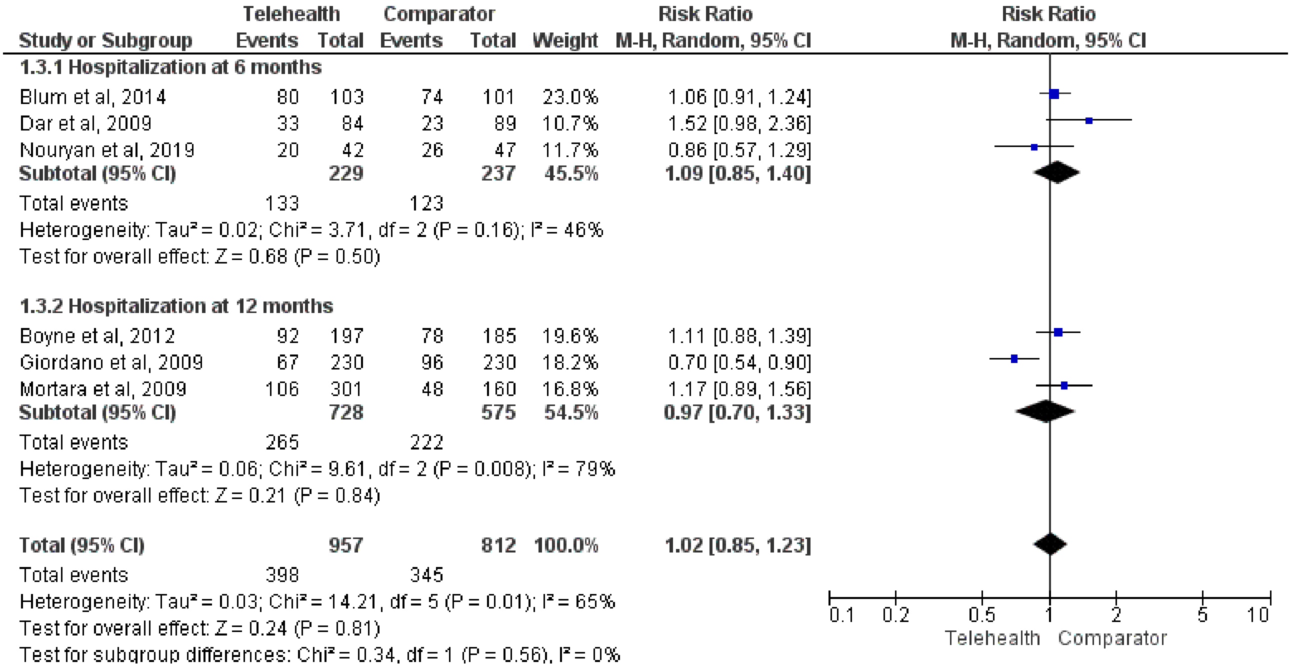
A total of 11 studies contributed to the all-cause mortality meta-analysis: 4 (36%) [39,50,84,107] (N=2056) provided data at 6 months, and 7 (64%) [16,40,61,64,96,101,105] (N=2578) provided data at 12 months. There was no significant difference in all-cause mortality between telemonitoring and the comparator at 6 months (risk ratio [RR]=0.86, 95% CI 0.68-1.07; *P*=.18; I^2=35%; Figure 3). This finding was consistent when studies evaluated as having a high risk of bias were excluded (Figure S1 in Multimedia Appendix 1). There was a significantly lower risk of all-cause mortality with telemonitoring than with the comparator at 12 months (RR=0.71, 95% CI 0.56-0.89; *P*=.003; I^2=0%; Figure 3). This finding was consistent following the exclusion of non-RCTs and studies evaluated as having a high risk of bias (Figure S1 in Multimedia Appendix 1).

Hospitalization

Meta-analyses for hospitalization at the 6- and 12-month follow-up were conducted (Figure 4 [23,25,34,52,80,83]), with sensitivity analyses excluding studies classified as having a high risk of bias (Figure S2 in Multimedia Appendix 1) and a subgroup analysis including only studies on patients with heart failure (12/96, 12%). Subgroup analyses for studies on patients with COPD and multiple chronic conditions were not possible because of a lack of absolute values or comparator [29,85].

A total of 8 studies contributed to the all-cause hospitalization meta-analyses: 3 (38%) [23,34,83] (n=466) provided data at 6 months, and 5 (62%) [25,52,80,96,101] (n=1825) provided data at 12 months. There was no significant difference in the risk of all-cause hospitalization between the groups at 6 months (RR=1.09, 95% CI 0.85-1.40; *P*=.50; I^2=46%) or 12 months (RR=0.97, 95% CI 0.70-1.33; *P*=.84; I^2=79%; Figure 4). This result was also consistent after the exclusion of studies evaluated as having a high risk of bias (Figure S2 in Multimedia Appendix 1). The meta-analysis that included only patients with heart failure showed no difference in the risk of hospitalization between the telemonitoring and comparator groups (RR=0.99, 95% CI 0.81-1.22; *P*=.94; I^2=69%; Figure S2 in Multimedia Appendix 1).

Figure 4. Impact of telemonitoring versus comparator on hospitalization at 6 and 12 months [23,25,34,52,80,83].



Study or Subgroup	Telehealth Events	Telehealth Total	Comparator Events	Comparator Total	Weight	Risk Ratio M-H, Random, 95% CI
1.3.1 Hospitalization at 6 months						
Blum et al, 2014	80	103	74	101	23.0%	1.06 [0.91, 1.24]
Dar et al, 2009	33	84	23	89	10.7%	1.52 [0.98, 2.36]
Nouryan et al, 2019	20	42	26	47	11.7%	0.86 [0.57, 1.29]
Subtotal (95% CI)		**229**		**237**	**45.5%**	**1.09 [0.85, 1.40]**
Total events	133		123			
Heterogeneity: Tau² = 0.02; Chi² = 3.71, df = 2 (P = 0.16); I² = 46%						
Test for overall effect: Z = 0.68 (P = 0.50)						
1.3.2 Hospitalization at 12 months						
Boyne et al, 2012	92	197	78	185	19.6%	1.11 [0.88, 1.39]
Giordano et al, 2009	67	230	96	230	18.2%	0.70 [0.54, 0.90]
Mortara et al, 2009	106	301	48	160	16.8%	1.17 [0.89, 1.56]
Subtotal (95% CI)		**728**		**575**	**54.5%**	**0.97 [0.70, 1.33]**
Total events	265		222			
Heterogeneity: Tau² = 0.06; Chi² = 9.61, df = 2 (P = 0.008); I² = 79%						
Test for overall effect: Z = 0.21 (P = 0.84)						
Total (95% CI)		**957**		**812**	**100.0%**	**1.02 [0.85, 1.23]**
Total events	398		345			
Heterogeneity: Tau² = 0.03; Chi² = 14.21, df = 5 (P = 0.01); I² = 65%						
Test for overall effect: Z = 0.24 (P = 0.81)						
Test for subgroup differences: Chi² = 0.34, df = 1 (P = 0.56), I² = 0%						

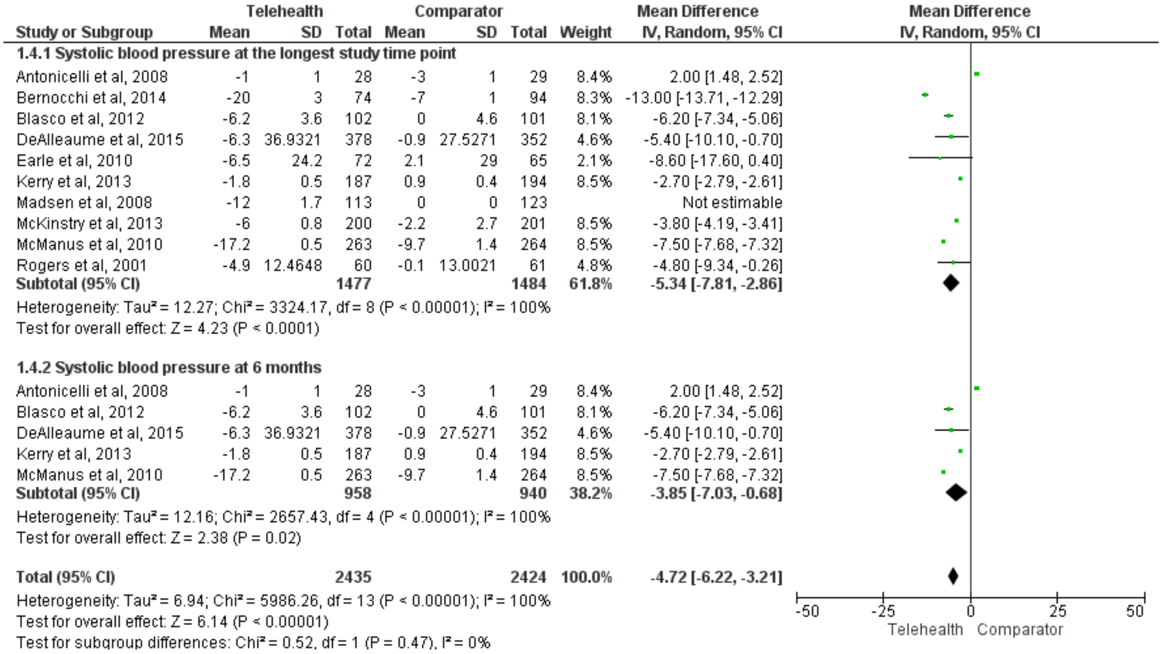
Changes in BP

A total of 10% (10/96) of the studies [16,17,24,38,45,62,72,75,77] reporting on the change in SBP and 8% (8/96) of the studies [15,17,24,45,62,72,75,77,90] reporting on the change in diastolic BP (DBP) between a telemonitoring intervention and usual care were included in the meta-analyses. Further details on the analyses of BP are provided in Multimedia Appendix 1.

Changes in SBP

SBP was significantly reduced in the telemonitoring group (n=1477) compared with that in the usual care group (n=1484; weighted MD=−5.34 mm Hg, 95% CI −7.81 to −2.86; *P*<.001; I^2=100%; Figure 5 [15,17,24,38,45,62,72,75,77,90]). In the subgroup analysis according to study time points, similar results were observed for SBP at 6 months (weighted MD=−3.85 mm Hg, 95% CI −7.03 to −0.68; *P*=.02; I^2=100%; Figure 5) and 12 months (weighted MD=−3.85 mm Hg, 95% CI −7.03 to −0.68; *P*=.02; I^2=100%; Figure S3 in Multimedia Appendix 1) in favor of telemonitoring.

Figure 5. Impact of telemonitoring versus usual care on changes in systolic blood pressure (mean difference) at the longest study time point and at 6 months [15,17,24,38,45,62,72,75,77,90].



Study or Subgroup	Telehealth Mean	Telehealth SD	Telehealth Total	Comparator Mean	Comparator SD	Comparator Total	Weight	Mean Difference IV, Random, 95% CI
1.4.1 Systolic blood pressure at the longest study time point								
Antonicelli et al, 2008	-1	1	28	-3	1	29	8.4%	2.00 [1.48, 2.52]
Bernocchi et al, 2014	-20	3	74	-7	1	94	8.3%	-13.00 [-13.71, -12.29]
Blasco et al, 2012	-6.2	3.6	102	0	4.6	101	8.1%	-6.20 [-7.34, -5.06]
DeAlleaume et al, 2015	-6.3	36.9321	378	-0.9	27.5271	352	4.6%	-5.40 [-10.10, -0.70]
Earle et al, 2010	-6.5	24.2	72	2.1	29	65	2.1%	-8.60 [-17.60, 0.40]
Kerry et al, 2013	-1.8	0.5	187	0.9	0.4	194	8.5%	-2.70 [-2.79, -2.61]
Madsen et al, 2008	-12	1.7	113	0	0	123		Not estimable
McKinstry et al, 2013	-6	0.8	200	-2.2	2.7	201	8.5%	-3.80 [-4.19, -3.41]
McManus et al, 2010	-17.2	0.5	263	-9.7	1.4	264	8.5%	-7.50 [-7.68, -7.32]
Rogers et al, 2001	-4.9	12.4648	60	-0.1	13.0021	61	4.8%	-4.80 [-9.34, -0.26]
Subtotal (95% CI)			**1477**			**1484**	**61.8%**	**-5.34 [-7.81, -2.86]**
Heterogeneity: Tau² = 12.27; Chi² = 3324.17, df = 8 (P < 0.00001); I² = 100%								
Test for overall effect: Z = 4.23 (P < 0.0001)								
1.4.2 Systolic blood pressure at 6 months								
Antonicelli et al, 2008	-1	1	28	-3	1	29	8.4%	2.00 [1.48, 2.52]
Blasco et al, 2012	-6.2	3.6	102	0	4.6	101	8.1%	-6.20 [-7.34, -5.06]
DeAlleaume et al, 2015	-6.3	36.9321	378	-0.9	27.5271	352	4.6%	-5.40 [-10.10, -0.70]
Kerry et al, 2013	-1.8	0.5	187	0.9	0.4	194	8.5%	-2.70 [-2.79, -2.61]
McManus et al, 2010	-17.2	0.5	263	-9.7	1.4	264	8.5%	-7.50 [-7.68, -7.32]
Subtotal (95% CI)			**958**			**940**	**38.2%**	**-3.85 [-7.03, -0.68]**
Heterogeneity: Tau² = 12.16; Chi² = 2657.43, df = 4 (P < 0.00001); I² = 100%								
Test for overall effect: Z = 2.38 (P = 0.02)								
Total (95% CI)			**2435**			**2424**	**100.0%**	**-4.72 [-6.22, -3.21]**
Heterogeneity: Tau² = 6.94; Chi² = 5986.26, df = 13 (P < 0.00001); I² = 100%								
Test for overall effect: Z = 6.14 (P < 0.00001)								
Test for subgroup differences: Chi² = 0.52, df = 1 (P = 0.47), I² = 0%								

The sensitivity analysis, excluding studies where the SD was not reported directly [38,45,90], did not materially change the results (weighted MD=−5.19 mm Hg, 95% CI −8.01 to −2.37; $P<.001$; $I^2=100\%$; Figure S3 in Multimedia Appendix 1). The sensitivity analysis was also performed excluding studies with a high risk of bias (Figure S3 in Multimedia Appendix 1); the results remained in favor of telemonitoring (weighted MD=–2.84 mm Hg, 95% CI −4.22 to −1.46; $P<.001$; $I^2=98\%$).

Changes in DBP

A meta-analysis including the longest time point demonstrated a significant reduction in DBP in favor of telemonitoring (n=1218) compared with the comparator (n=1255; weighted MD=−2.83 mm Hg, 95% CI −3.98 to −1.68; $P<.001$; $I^2=99\%$; Figure S4 in Multimedia Appendix 1). In the subgroup analysis, a similar result was observed for DBP reduction at 6 months (weighted MD=−5.44 mm Hg, 95% CI −9.00 to −1.87; $P=.003$; $I^2=100\%$; Figure S4 in Multimedia Appendix 1) in favor of telemonitoring but not for DBP at 12 months (weighted MD=−1.09 mm Hg, 95% CI −4.76 to 2.57; $P=.56$; $I^2=97\%$; Figure S4 in Multimedia Appendix 1). Sensitivity analyses at the longest time point excluding studies with high risk of bias (Figure S4 in Multimedia Appendix 1) showed no significant reduction in DBP in the telemonitoring group (weighted MD=−1.07 mm Hg, 95% CI −2.58 to 0.44; $P=.16$; $I^2=98\%$) compared with usual care.

Changes in HbA_{1c}

A total of 19% (18/96) of the studies reported on HbA_{1c}, and all the studies (18/18, 100%) compared telemonitoring with usual care, with 61% (11/18; n=3277) included in the meta-analysis [27,30,35,46,49,58,63,87,89,94,109]. Further details on the excluded studies for the meta-analysis are provided in Multimedia Appendix 1.

The duration of the interval before and after varied, with 18% (2/11) of these studies reporting a 6-week assessment [58,87], 45% (5/11) [27,30,46,49,63] reporting 3-month assessments, 9% (1/11) reporting 9-month assessments [109], and 27% (3/11) [35,89] reporting 12-month assessments. A sensitivity analysis was performed excluding studies with a high risk of bias [58,94].

The overall mean change in HbA_{1c} is shown in Figure S5 in Multimedia Appendix 1. The pooled estimate showed a reduction in the mean change in HbA_{1c} in the telemonitoring group (n=1703; weighted MD=−0.33, 95% CI −0.57 to −0.09; $P=.008$; $I^2=99\%$; Figure S5 in Multimedia Appendix 1). The results did not materially change after the sensitivity analysis excluding studies at high risk of bias [58,87] (Figure S5 in Multimedia Appendix 1). Subgroup analyses according to study time points showed no significant difference in the change in HbA_{1c} values between telemonitoring and the comparator (Figure S5 in Multimedia Appendix 1).

Discussion

Principal Findings

Our results suggest that telemonitoring interventions are associated with good patient adherence and satisfaction. Although this review did not demonstrate improvements in QoL with telemonitoring, there was evidence to suggest reductions in all-cause mortality and improvements in BP and blood glucose control. Conversely, there was evidence to suggest that telemonitoring interventions may be associated with a higher rate of hospitalizations, which could be interpreted as a positive role of telemonitoring in detecting patients' health issues more than usual care.

Comparison With Prior Work

Our review showed improvements in physiological parameters (BP and blood glucose) in patients receiving telemonitoring interventions. These findings demonstrate the positive role of telemonitoring in improving patients' self-management of their conditions. This is in line with other reviews that have shown similar improvements in hypertension [168] and type 2 diabetes self-management [169] after telemonitoring interventions.

The studies included in this review consistently showed that patients receiving telemonitoring interventions had lower all-cause mortality compared with patients receiving usual care. A recent umbrella review [170] examining the effects of telemonitoring on mortality in several clinical populations (cardiovascular, COPD, and neurological) reported similar findings for the cardiovascular population, where the mortality rate was either reduced in the telemedicine users or remained unchanged compared with usual care. The same review [170] did not find any difference in mortality between telemonitoring and usual care in patients with COPD. The impact on death is an important outcome when considering the administration of remote interventions over in-person visits, and the reduced mortality rate with telemonitoring reported in our review suggests the effectiveness of telemonitoring for patients with chronic conditions.

Surprisingly, the overall results of our review showed a higher risk of hospitalization among patients undergoing telemonitoring interventions. There is inconsistency in the previous literature on the role that telemonitoring plays in reducing the risk of rehospitalization, with some studies reporting no differences compared with usual care [171] and others concluding that telemonitoring is an effective tool to reduce all-cause hospitalization in adults with heart failure [172]. Thurmond et al [173] noted the importance that the type of telemonitoring intervention has on its acceptability by patients and, consequently, their adherence to it, which, when poor, may influence the rate of rehospitalization. This would suggest the need to identify common characteristics of effective telemonitoring interventions (or "active ingredients") that facilitate patient acceptability. It may also be possible that increased hospitalizations with telemonitoring is a positive finding (ie, reasons for hospitalization may be identified earlier by telemonitoring, and hospitalization may be initiated earlier than with usual care, averting serious outcomes and death). Hypothetically, this could have contributed to the reduced mortality at 12 months; however, future research is needed to substantiate this.

The results of this review are in line with those of previous systematic reviews assessing patient satisfaction with telemonitoring interventions [174,175]. From qualitative reports, the convenience of decreased travel time and costs and the reassurance of being monitored are the most likely reasons for patients preferring telemonitoring over usual care [176]. It is important to note that patient satisfaction may differ with the type of telemonitoring device used; indeed, available evidence suggests that higher patient satisfaction is reported for videoconferences and devices that allow for automated data transmission [174].

The included studies did not report significant improvements in the QoL of patients receiving a telemonitoring intervention compared with usual care. Our findings confirm previous reviews [177,178] while expanding the results to populations outside care homes [178] and including study designs other than RCTs [177]. Although telemonitoring does not seem to improve QoL compared with usual care, previous findings [178] have shown important benefits of telemonitoring in improving patients' confidence in accessing health care services.

Strengths and Limitations

This review used a strict definition of telemonitoring, only including studies that used a device to collect health measures and facilitated 2-way communication or action between the patient and health care team. Despite the inclusion of studies with low methodological quality, sensitivity analyses were conducted where appropriate, reducing the potential for bias to affect the results of this review. The studies included in this review presented a wide range of telemonitoring interventions that differed in the personnel involved, administration of the intervention, and technology used and that were examined in a variety of populations with different long-term conditions, thus making the results highly generalizable. A robust methodology was used, with independent screening and data extraction by 2 reviewers and risk of bias assessment in duplicate.

Several limitations are noteworthy. First, despite our initial plans to investigate uptake, patient retention and satisfaction, and associated factors when using 2-way (patient-health care provider) remote patient monitoring devices to manage chronic health conditions, no studies reported uptake and retention outcomes and, therefore, these outcomes could not be reported in this review. Most of the included studies assessed similar outcomes but used different measurement tools, thus making comparison difficult, particularly in studies investigating patient adherence [13,30,36,42,48,51,58,59,66,84,92,103] and satisfaction [22,28,30,42-44,78,91,95] with the intervention. Second, despite our efforts to define the best search strategy to identify all relevant articles for our review, the possible omission of papers because of the heterogeneity in the key terms used by the authors cannot be ruled out. We did not conduct any searches for gray literature. Third, most outcomes analyzed in this review have been infrequently investigated in the literature (eg, mortality was reported only in 17/96, 18% of the included studies; adherence was reported in only 12/96, 12% of the studies; and satisfaction was reported in only 9/96, 9% of the studies), and further research is required to properly assess the effects of telemonitoring on these outcomes. Moreover, some conditions (eg, COPD) were underrepresented as few studies investigating the effects of telemonitoring interventions on these populations were available; thus, we could not conduct a separate meta-analysis for each condition. The type and quality of usual care also varied throughout the included studies, which may have influenced the results in favor of or against telemonitoring.

Conclusions

Telemonitoring is a promising tool to manage long-term conditions, with the potential to reduce the associated costs and alleviate patient difficulties in accessing primary health care. Patient satisfaction and adherence to telemonitoring appear, overall, to be promising. Although telemonitoring resulted in improvement in physiological parameters and reduced all-cause mortality compared with usual care, there was no improvement in QoL and an increased risk of hospitalization with telemonitoring. Although the latter may be a positive finding indicating earlier detection of health issues and action (resulting in hospitalization), this result warrants further investigation. Telemonitoring is expanding rapidly, more so since the COVID-19 pandemic, and has been shown to be a viable alternative to usual care for the management of patients with long-term health conditions.

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Conflicts of Interest

BJRB has received research funding from the Bristol Myers Squibb (BMS)-Pfizer Alliance. SLH has received an investigator-initiated grant from BMS. GYHL has been a consultant and speaker for the BMS-Pfizer Alliance, Boehringer Ingelheim, and Daiichi-Sankyo. No fees were received personally. DJW has been a consultant and speaker for Medtronic and Boston Scientific. DAL has received investigator-initiated educational grants from BMS; been a speaker for Boehringer Ingelheim, Bayer, and the BMS-Pfizer Alliance; and consulted for Boehringer Ingelheim, Bayer, and the BMS-Pfizer Alliance, all outside the submitted work.

Multimedia Appendix 1

Supplementary figures and tables that were not included in the main manuscript.
[DOC File , 1432 KB-Multimedia Appendix 1]

Multimedia Appendix 2

Summary of the included studies (N=96).
[DOCX File , 57 KB-Multimedia Appendix 2]

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Abbreviations

BP: blood pressure

COPD:	chronic obstructive pulmonary disease
DBP:	diastolic blood pressure
HbA$_{1c}$:	glycated hemoglobin
MD:	mean difference
PRISMA:	Preferred Reporting Items for Systematic Reviews and Meta-Analyses
PROSPERO:	International Prospective Register of Systematic Reviews
QoL:	quality of life
RCT:	randomized controlled trial
RR:	risk ratio
SBP:	systolic blood pressure

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