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Investigation of Digital Technology Use in the Transition to Parenting: Qualitative Study

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Abstract

Background: The transition to parenting—that is, the journey from preconception through pregnancy and postpartum periods—is one of the most emotionally charged and information-intense times for individuals and families. While there is a developing body of literature on the use and impact of digital technology on the information behaviors of children, adolescents, and young adults, personal use of digital technology during the transition to parenting and in support of infants to 2 years of age is relatively understudied.

Objective: The purpose of this study was to enhance our understanding of the ways digital technologies contribute to the experience of the transition to parenting, particularly the role these technologies play in organizing and structuring emerging pregnancy and early parenting practices.

Methods: A qualitative descriptive study was conducted to understand new parents' experiences with and uses of digital technology during 4 stages—prenatal, pregnancy, labor, and postpartum—of their transition to becoming a new parent. A purposive sampling strategy was implemented using snowball sampling techniques to recruit participants who had become a parent within the previous 24 months. Focus groups and follow-up interviews were conducted using semistructured interview guides that inquired about parents' type and use of technologies for self and family health. Transcribed audio recordings were thematically analyzed.

Results: A total of 10 focus groups and 3 individual interviews were completed with 26 participants. While recruitment efforts targeted parents of all genders and sexual orientations, all participants identified as heterosexual women. Participants reported prolific use of digital technologies to direct fertility (eg, ovulation timing), for information seeking regarding development of their fetus, to prepare for labor and delivery, and in searching for a sense of community during postpartum. Participants expressed their need for these technologies to assist them in the day-to-day demands of preparing for and undertaking parenting, yet expressed concerns about their personal patterns of use and the potential negative impacts of their use. The 3 themes generated from the data included: "Is this normal; is this happening to you?!", "Am I having a heart attack; what is this?", and "Anyone can put anything on Wikipedia": Managing the Negative Impacts of Digital Information.

Conclusions: Digital technologies were used by mothers to track menstrual cycles during preconception; monitor, document, and announce a pregnancy during the prenatal stage; prepare for delivery during labor/birth stage; and to help babies sleep, document/announce their birth, and connect to parenting resources during the postpartum stage. Mothers used digital technologies to reassure themselves that their experiences were normal or to seek help when they were abnormal. Digital technologies provided

mothers with convenient means to access health information from a range of sources, yet mothers were apprehensive about the credibility and trustworthiness of the information they retrieved. Further research should seek to understand how men and fathers use digital technologies during their transition to parenting. Additionally, further research should critically examine how constant access to information affects mothers' perceived need to self-monitor and further understand the unintended health consequences of constant surveillance on new parents.

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KEYWORDS

parenting; digital health; technology; health literacy; information seeking

Introduction

The transition to parenting—that is, the journey from preconception through pregnancy and postpartum periods—is one of the most emotionally charged and information-intense times for individuals and families [1-3]. During this time, resources that are often highly valued by transitioning parents include midwives, physicians, family and friends, and increasingly, internet-based resources [4]. Additionally, the use of digital technologies—broadly defined as devices such as computers, video cameras, and gaming systems; mobile devices such as phones and smartphones; and all applications (eg, internet) that are computer-dependent—constitutes the fastest growing information resource used by families as they negotiate the transition to parenting [5,6]. While there is a developing body of literature on the use and impact of digital technology on the information behaviors of children, adolescents, and young adults [7-10], personal use of digital technology during the transition to parenting and in support of infants to 2 years of age is relatively understudied.

With the rapid proliferation and uptake of digital technologies, it is crucial to understand which technologies are used by parents to make decisions impacting their health and that of their families and how their use contributes to the transition to parenting in both virtual and material spaces and at their intersections [11-13]. Recent information suggests that pregnant women and mothers of young children value information gathered from digital sources [14], with Facebook and other social media being commonly cited resources for new and transitioning mothers [15,16]. In addition to social media outlets, pregnancy-specific mobile apps have an important role in the self-health promotion of women and infants and are often consulted to search for signs or risks of illness [15]. Such apps may also promote well-being by reducing burden and feelings of isolation and improving health outcomes among new parents [16].

Despite the benefits of social media and pregnancy-specific apps, these sources of health information may have negative effects on new and transitioning mothers. For example, mothers who spend considerable time on Facebook after giving birth may experience increased stress, feelings of isolation, and depressive symptoms related to seeking external validation about their parenting practices [16,17]. Additionally, time spent consulting mothering websites was negatively correlated with mothers' intentions to breastfeed [18,19]. Pregnancy-related mobile apps also present challenges to new mothers as these apps present wide variations in the trustworthiness of data and the privacy features [20]. For example, despite the increase in the number of pregnancy-related mobile apps designed to convey information about fetal movement, few apps provide explicit links between or clear instructions on how to interpret and respond to decreased fetal movement, the potential and likelihood of a stillbirth, or other adverse outcomes [21].

Further research is needed to explore parents' use of digital technologies, why parents are going online, what they are viewing, and how it impacts their transition to parenting. Additionally, with issues concerning the access of reliable health information available from diverse digital sources, it is important to investigate parents' use of health information technology to inform safe practices and recommendations for trustworthy and reliable health information access [22]. The purpose of this study was to enhance our understanding of the ways digital technologies contribute to the experience of the transition to parenting, particularly the role these technologies play in organizing and structuring emerging pregnancy and early parenting practices.

Methods

A qualitative descriptive study was conducted to understand new parents' experiences with and uses of digital technology during 4 stages—prenatal, pregnancy, labor, and postpartum—of their transition to becoming a new parent [23].

Recruitment

This study took place during 2019 in an urban setting in Southwestern Ontario, Canada. As of 2016, the median after-tax income of couple families with children in this region of Ontario was CAD $84,608, whereas the median after-tax income of lone-parent families was CAD $45,952 [24]. A currency exchange rate of CAD $1=US $0.78 is applicable. A purposive convenience sampling strategy was implemented to recruit participants who had recently transitioned to becoming a parent within the previous 24 months [25]. Recruitment flyers were posted in locations where new parents were believed to frequent, such as local public health units, daycare centers, family health clinics, and early year play centers. Digital flyers and advertisements were also purchased online through buy-and-sell websites and social media platforms such as Facebook. Interested individuals were eligible to participate if they met the following inclusion criteria: (1) identified as a new parent who had recently undergone the transition to parenting within the last 24 months, (2) were between 16 and 35 years old, and (3) were fluent English speaking. The participant age limit was set to 35 years old as older parents, in particular mothers,

constitute a generationally different cohort in terms of their technology use, health care needs, and health risks. All participants provided written informed consent prior to participating in this study and were provided with a CAD $15 honorarium immediately after signing the consent forms before engaging in data collection.

Data Collection and Analysis

Focus groups and individual interviews were conducted by members of the research team in locations agreed upon between participants and researchers, including public libraries, a shelter, and a children's center. The nature of inquiry within the focus groups was related to participants' use of digital technologies (ie, social media, use of pregnancy or parenting apps, participation in online pregnancy or parenting support groups). If a focus group participant introduced a topic that required additional time or consideration to discuss, an individual follow-up interview was offered. For example, such topics included in vitro fertilization and egg donation. A demographic questionnaire was given to each participant at the outset of the focus group to elicit descriptive characteristics of the participants. Data were digitally recorded and transcribed verbatim with thematic data analysis. Field notes by researchers were also employed to document relevant data not able to be captured by the digital recording, such as nonverbal communication.

Recruitment of focus group participants continued until data saturation was met [26,27]. An iterative, thematic analysis approach was used to coconstruct study findings [28,29]. All members of the research team analyzed each interview transcript, then together cross-compared insights and negotiated emerging themes through face-to-face dialogue during subsequent team meetings. Codes were tracked in a tabular matrix using exemplar quotes from interview transcripts to demonstrate the meaning of the code. Data saturation was achieved once no new themes, patterns, nor codes were identified and when categories being analyzed became repetitive in nature with no new information being generated through additional focus group discussions [28].

Members of the research team included in the analysis process were academics with a diverse range of academic, professional, and personal life experiences as they relate to the transition to parenting. All but one member of the research team were parents, with children between them ranging in age from 7 to 30 years of age at the time the study was undertaken. Team members had varying levels of personal engagement with digital technologies to support parental decision making, and these personal experiences were utilized at times to delve deeper into a particular quote or theme that was emerging. Through dialogue between team members, we engaged in interrelational reflexivity wherein we questioned how our own positional power and social locations shaped our prior assumptions and how our knowing broadened or shifted through interaction with the data collected and emerging themes [30].

Results

Participant Characteristics

A total of 10 focus groups and 3 follow-up individual interviews were completed with 26 participants. There were 2 to 4 participants per focus group. While recruitment efforts targeted parents of all genders and sexual orientations, all participants identified as heterosexual women. Participants ranged in age from 17 to 35 years old, with 8 being 20 years old or younger, 4 being between 21 and 29 years old, and 10 being between 30 and 35 years old. There was a range of formal educational attainment, with 7 in the process of completing secondary school, 1 who had completed high school, 1 who had completed community college or apprenticeship, 10 who had completed a university undergraduate degree, and 2 who had completed a graduate degree. Employment status was reported by 20 participants, of which 9 participants were unemployed, 3 were employed part time, and 7 were employed full time. Participants' household income also varied, as 4 reported a yearly household income of less than $20,000, 3 reported between $20,000 and $50,000, 4 reported between $50,000 and $99,999, and 5 reported household income over $100,000 per year. Half of the participants (13/26, 50%) were married, 7 were single and had never been married, and 1 was separated from her partner. The majority of participants (18/26, 69%) identified as Caucasian, and 3 participants identified as racialized.

Participants were invited to discuss the types of digital technology they used in their day-to-day lives in the context of their transition to parenting. Our analyses identified participants' insatiable need to obtain health information, often through the use of online apps, to support reproduction, to inform their decision making, to validate their parental care practices, and as a means to simply cope with the increasing demands of new parenting. Participants reported prolific use of digital technologies to direct fertility (eg, ovulation timing), for information seeking regarding development of their fetus, to prepare for labor and delivery, and in searching for a sense of community during postpartum. See Table 1 for details regarding technologies used at each stage of the transition to parenting and the reasons participants used them.

Table 1. The types and reported use of technology during the transition to parenting.

Type and reported use	Preconception	Prenatal	Labor and birth	Postpartum
Type of tech use				
Devices	Smartphone	Smartphone, tablet, Doppler	Smartphone	Smartphone, breast pump, baby swing, TV, baby monitor, angel care monitor, scent diffuser
Online sites	Texting, Google	Texting, streaming services (eg, Netflix, YouTube), search engines (eg, Internet Explorer, Google), online registries (eg, BORN Better Outcomes Registry Network), social media (Facebook, Snapchat, Instagram)	Texting, social media (Snapchat)	Texting, The Milk Meg, Google, Motherisk, Pinterest, BORN (Better Outcomes Registry Network), Dr. Jack Newman - International Breastfeeding Centre (IBC)
Apps	Period tracker, fertility tracker, Ovia	Bump, Omama, What to Expect, Baby Centre, 3D ultrasound	Pampers	Facebook groups, FaceTime, White Noise, YouTube, Baby Tracker, Safety First, Baby, Pampers, Let Go (buy and sell app), O Mama
Reason for use	Track menstrual cycle, access information regarding symptoms	Monitoring, documenting, learning about pregnancy, pregnancy announcement, distraction (entertainment when not feeling well)	What to bring to the hospital/what to expect, games to help with labor pains, entertainment, surveys	Help baby sleep (white noise), tracking baby habits (respiratory function), birth announcement, connect to resources for childcare, personal information and social support

Interestingly, participants expressed both the need for these technologies to assist them in the day-to-day demands of preparing for and undertaking parenting, yet simultaneously expressed concerns about their personal patterns of use and the potential negative impacts their use could have on infant development and attachment. The following sections explore how participants navigated these tensions.

"Is This Normal; Is This Happening to You?!"

Participants in this study frequently described using their digital technologies to determine if their preconception, pregnancy, and postpartum experiences were "normal" relative to others. For example, participants regularly used online search engines to access information when feeling anxious:

> *I definitely Googled a lot of stuff. Like, we had been trying for two years and so, I was constantly, "is this supposed to happen?" "is this normal?" "is it supposed to look like this?"* [Transcript 1]

Others would use their technologies to seek validation from friends:

> *I would text my friends that have had babies to say, "is this normal?"* [Transcript 1]

Some participants who had been previously pregnant expressed that they avoided search engines because they assumed their current experiences were normal in relation to their own positive past pregnancy(ies):

> *…then my third [pregnancy], I was just too busy to Google anything, I just assumed everything was normal.* [Transcript 10]

In contrast, participants with prior negative pregnancy experiences frequently consulted resources and reported using search engines for information as a coping mechanism to manage stress and anxiety:

> *…everything went really well while I was pregnant and then… I had a miscarriage and that was really devastating and hard. So, I feel like at the beginning [of my second pregnancy], I was Googling a lot because I was like, "oh my God, what about this symptom?! The last time I felt that pain, this happened." So, with [my second pregnancy], I was more paranoid.* [Transcript 11]

Despite the benefits that search engines potentially offered, some participants acknowledged that the information found through Google or other search engines could lead to false assumptions of normalcy that may endanger their and their fetus' health. One participant described a situation where her husband assumed everything was normal because of her active Google use, while she used the information gleaned from the search engine to conclude that there was a potential abnormal issue developing:

> *I remember reading a lot about if you don't feel your baby move, you should do this… I didn't feel my son move one morning, and I was like, "I'm going to the hospital," and my husband's like, "you're on Google, you're fine." But I wasn't fine, and then I had him an hour later… I obviously found that [being on Google] helped me.* [Transcript 11]

In addition to using search engines to determine if their experiences were normal, participants in this study also used their digital technologies to facilitate communication with their health care providers to determine if their experiences were normal. For example, as one participant described:

> *I wasn't sure what was going on and what would be necessary for the physicians to know, and I'm not really good with explaining things when I'm nervous or upset or have some anxiety about something. So, I took a picture of the spit up [with my phone] and took it to the hospital with me so they knew what I was talking about.* [Transcript 2]

Other participants described using their digital technologies immediately following medical procedures to interpret if the information shared with them by their health care professionals meant that their experiences were normal. For example, one mother described how she verified the normalcy of her daughter's coloration immediately following birth:

> *My daughter was really red when she was born. Her skin was so red and purple, and I'm like, "is that normal?!" They were telling me that it's just because she was just born and probably because she was born so quickly, and then I was just looking that up, "why is she so red?" But I just read that it's pretty much normal for some babies to be really red and purple when they're born.* [Transcript 3]

In a similar sentiment, other participants in this study described how social messaging outlets were a place to share their own expertise to help other mothers when they would ask questions about their experience:

> *I was on boards especially when I was going through the grief stuff... I would share my story about I'm diagnosed with Turner Syndrome and I've gone through all of this, and I had some girls message me that I don't know saying "can you tell me about it? What do I have to look for in store for what my child?"* [Transcript 8]

Finally, participants in this study reported that while their use of digital technology increased, "I think I use technology more now that I have kids…" [Transcript 10], extending most often toward answering the question "Is this normal," their partners' behaviors often remained unchanged: "He uses social media in different ways. He connects with online gaming and those kinds of things, but not so much for parenting." They also described how apps tailored to fathers were hypermasculinized to convey the size of the growing fetus:

> *It was like a daddy app, so it was like relating [the fetus] to a size of a beer or something like that. It was totally like dad style ... And then I think he would come to my pregnancy app to look at it if he wanted it to be a bit more serious.* [Transcript 12]

"Am I Having a Heart Attack; What is This?"

Participants described how digital technologies made it easier and more convenient to communicate with others about their pregnancy and enabled them to find responses to their questions and health information needs more rapidly and during all hours of the day than other communication channels. For example, digital technologies facilitated visual communications between family members and enabled geographically distant family members to interact with the participants' children:

> *[We use] Facetime a lot… Especially like my mom, they go to Florida for the winter, so to keep in contact with them, just we'll Facetime once every couple of days so they can see him [infant].* [Transcript 11]

Digital technologies also offered a convenient means for participants to share health information with health care professionals:

> *In the beginning, during breastfeeding and when I got in touch with the lactation consultant… we communicated via text message, and I would ask her like, and I know it sounds weird, but she would be like "send me a picture of him feeding so I can see." So, I would send her a picture and she would tell me like "adjust his head" or "put your hand this way" or whatever.* [Transcript 11]

Social media websites were often highlighted for the quick responses that participants received from other users when in search of health information about their pregnancy-related experiences:

> *I did a lot of research online about egg donation and in vitro and found out through Facebook through a friend of mine… about her surrogacy journey, so I ended up contacting her online to find out more about the agency that she worked through.* [Transcript 8]

Additionally, the convenience of digital technologies was described as an important element that helped participants address their stress caused by uncertainty around their own health and well-being:

> *I did Google once at like 3 in the morning. I had, like it was heartburn, but that was like – I have never had it before and I was "am having a heart attack?! What is this? This is way more intense than I thought heartburn would be." So, I did Google and read some stories of other people and just "okay, this is pretty intense."* [Transcript 6]

Not all participants found social media to play a positive role in their parenting journey and did not post online out of fear of judgement or concerns about being perceived as not measuring up to the social norms expected of "good mothers." As one participant shared:

> *My house is a mess in the background, and I'm not posting that. Or why are you making that smile or it's blurry? He's [baby] goofy and not wanting to take a picture.* [Transcript 4]

For another participant, the shelter where she was staying forbid the use of digital technologies:

> *I had no phone when I got there, like I was ready to leave. I had no contact with anybody, and I kind of built myself up but, like I said, they had no internet and they refused to get internet because their thing was – it's like a maternity home where like, you know, like kids are there. They're like "you shouldn't be on your phones while you're playing with your kids," and technically that's what you're supposed to be doing all the time.* [Transcript 5]

"Anyone Can Put Anything on Wikipedia": Managing the Negative Impacts of Digital Information

While each participant noted a positive aspect of digital technology usage during the transition to parenting (eg, staying connected to family, obtaining health information, receiving validation from peers), participants also expressed apprehension about the credibility of the digital sources of health information, how to best use these sources, and whether or how to act on information they retrieved. There was some concern about trusting health information on certain websites due to the lack of transparency of the authors' credentials and expertise in the health care field. One participant compared the trustworthiness of health information found on crowd-sourced websites to that found on health-specific websites:

> *Anyone can put anything on Wikipedia. They can change all the information, like you can go on and change it yourself. So, it's like the health websites is usually actually there's a nurse answering your question.* [Transcript 3]

Another participant describes how she lost trust in non-health–specific websites and now only trusts websites published by health care organizations:

> *I did trust it at one point and then people were telling me that people can go in and change the information. So, once I heard that, like multiple times, through growing up, I do not really trust it. Some information, like the health unit website and what not, that is the type of information that I would check.* [Transcript 7]

Participants described how the amount of health information available through digital technologies was often overwhelming, which created uncertainty regarding how to interpret or act on the information they found. One participant described how these feelings during postpartum were alleviated by support to do something with the information she found:

> *Postpartum is just such an intense time that I feel like it—like there's so much information and different information. The support isn't really there.* [Transcript 12]

Due to the amount of health information sources, participants described the need to critically appraise websites to ensure they were obtaining credible information:

> *You have to look at who's sponsoring the article, right? Because you can literally find any information that you want to hear or see. So, you have to really know how to dissect even the science-based studies.* [Transcript 12]

Similarly, participants noted the importance of questioning the credibility of health information they received through social media groups or message boards as some information may inappropriately exacerbate their anxieties:

> *I had a massive bleed at the beginning of my pregnancy, and I thought I was miscarrying. So, of course, I was writing on this and everyone's like, "oh you're probably miscarrying. You should check it out." But then it turned out it was fine… I think it can be like anxiety and comforting at the same time because I feel like any symptom can be put in, it can be either completely abnormal or completely normal.* [Transcript 11]

Some participants described strategies to verify the accuracy of health information they found through digital technologies as a way to ensure they were acting on credible information. For example, checking multiple sites for answers to a question was a common approach to verify health information:

> *If I Google something, I never just go for the first answer. I always have to check out 4 or 5 different websites and, you know, if they all say the same thing then that tells me I think that's a good thing to follow.* [Transcript 2]

Participants did note that consulting some nontechnological resources—such as health care providers, books, pamphlets, prenatal classes, their public health nurse, other moms, family, and friends—was a method to verify information obtained online. Of all nontechnological resources, participants' physicians were viewed as their most trustworthy source for health information and as a credible third-party to verify the accuracy of information found online:

> *So, I thought this Doppler I got like if I could hear the heartbeat that I would feel better. So, I didn't know how to do it, so I had to YouTube it… If I had to, I would go to the doctor, because that's essentially—I would never leave it up to my Googling or my experience with a Doppler to determine if it was valid or not, or if I was doing it right. If I was in doubt, I would go see a professional. I would never take Google's word or YouTube's over mine, but it's helpful.* [Transcript 11]

Discussion

This investigation of digital technology use in the transition to parenting singularly highlights mothers' use of digital technologies across preconception, prenatal, labor, and birth and during postnatal stages. For mother participants in this study, substantial effort was given to understand "Is this normal?" which is consistent with existing literature on mothering [31,32]. Experts argue that the ongoing search for information is generated, in part, by societal norms that prescribe women to parent with the pressure to be perfect, contributing to the toll on mothers' well-being [31,32]. Participants' use of digital technologies in the transition to parenting was accompanied by feelings of constant negotiation, of trade-offs between the relative ease and instantaneous access to answer the question "Is this normal?" with potential downsides, such as the likelihood that the information could be inaccurate or potential judgement. Our study contributes to this literature by demonstrating the ways that participants extended their search for answers beyond the traditional sources. Participants in this study turned to digital technologies, online platforms, apps, forums, and streaming services to answer this relentless question, with most participants accessing information on their cell phones for convenience and because of their ubiquitous presence.

Consistent with other research [33,34], mothers in this study demonstrated an unrelenting drive to obtain health information online: to direct fertility (eg, ovulation timing), for information seeking regarding development of their fetus, to prepare for labor and delivery, and to generate a sense of community during postpartum. In effect, they "Googled" everything. This was especially true for first-time mothers and mothers with a history of negative pregnancy experiences. Across the stages of the transition to parenting, apps, videos, online shopping, and forums were important sources of tangible and intangible information, resources, and services accessible regardless of place and time. In considering increased patient empowerment with the possibilities for greater self-management of prenatal and postpartum care, digital technologies have the potential to enhance efficiency of care and contribute to the revolution in perinatal care [35].

However, our findings suggest that opportunities for enhanced self-management and empowerment may not be equitably obtainable. Consistent with participants in our study, many people feel compelled to search for health information online especially when they experience difficulties in accessing health care services [36]. Despite concerns about the overwhelming amount and questionable trustworthiness of online information, mothers in our study favored the immediacy and convenience of digital information (eg, internet, apps) expressly when health care services were less accessible (ie, middle of the night). In fact, mothers minimized their concerns regarding misinformation and privacy violations in search of validation or direction when the need for information in response to a health concern was perceived as pressing. Our findings add to the research by Amante et al [37], who reported that primary care patients used online information to determine their need for health care services or for self-health management (eg, alter or cease prescribed treatments). Different than the findings reported by El Sherif et al [38], mothers in this study used their digital technologies to communicate with health care providers and online parenting peers (ie, send images of health concerns) to determine if their experiences were normal and the need for additional intervention. In some instances, the online communication mitigated the need for in-person health service consultation (ie, online breastfeeding consult). Additional research is needed to fully understand the antecedents (eg, digital health literacy skills) and consequences (eg, health outcomes, health service inequities) of individuals' use of online health information especially within the context of patient-centered care practices and health service utilization patterns.

As well, some participants in this study conveyed a sensible skepticism regarding the credibility of digital sources of health information. The strategies employed by some of the mothers in this study (eg, assessing the website sponsor, access known government or health organization sites, assess consistency of information across multiple sites or sources) to confirm the trustworthiness of online information resources align with nationally advocated guidelines [39,40]. Yet the online information-seeking challenges for parents remain significant. Despite their awareness of online misinformation, mothers were challenged in their ability to discriminate accurate from false health information. Consider how antivaccination propaganda is amplified online, with life-threatening consequences to infants, children, and the wider community. Researchers have found that information from online sites has influenced decisions whether to vaccinate [41,42], and parents with the greatest need for knowledge about vaccination are seen as most vulnerable to false online health information [43]. Similarly, Ashfield et al [42] reported that parents found online information posted by antivaccination groups as very scientific in appearance (scientific language and academic formatting), making it increasingly difficult to determine credible from misleading information.

Beyond the ability to appraise online health information, we recognized the need for enhanced digital health literacy skills among the mothers in our study. Defined as the ability to seek, find, understand, and appraise health information from electronic resources and apply the knowledge gained to address or resolve health issues, digital health literacy skills involve the mastery of multiple literacies: traditional literacy, health literacy, information literacy, scientific literacy, media literacy, and computer literacy [44]. Like mothers in this study, Lee and Moon [13] found that online health apps were an important source of information for pregnant women. In their study exploring the use of 47 mobile apps for pregnancy, birth, and childcare, they determined that apps have become an important information source for pregnant women, more frequently used when searching for information concerning signs of risk and disease. Concernedly, of the criteria used to evaluate the usability of the apps (eg, information clarity and protection), "the information source" had the lowest score. They concluded that the quick provision of health information was desired and seen as a motivator, but often credible professional information was sorely lacking [13].

Our findings demonstrate that mothers' use of digital health technologies have the potential to move parents beyond self-care practices and into the scope of clinical practice. Furthermore, the "health care work" of mothers has the potential for health-enhancing outcomes, but also dire consequences. Parents' use of digital health technologies without advanced clinical knowledge and skill highlights the risk of adverse events as in the "near miss" described by one mother who noted a lack of fetal movement but was reassured by her partner that the information she found online discounted the need for health care intervention. A false sense of confidence or reassurance when accessing potentially inaccurate health information disseminated online or an inability to appraise and apply the health information to their specific situation may lead to delayed access to health services or increase the need for costly health care services when an emergency arises [38,45]. Additional research is needed to better understand the implications for the health outcomes and health service utilization patterns among individuals in the transition to parenting within the digital health context.

Limitations

While this study provides valuable insight into how new parents perceive the use of digital technologies as they transition to parenthood, it is not without its limitations. Importantly, while this study aimed to recruit all parents, participants reflect a

single type of parenting perspectives: those of heterosexual women. Although this sample presents an in-depth description regarding mothers' use of digital technologies during the transition to parenting, further research is needed to more fully understand this health information–seeking process and its unintended consequences for gendered health work. Future studies that target different types of parents, such as heterosexual men, LGBT men and women, and gender-fluid men and women and parents of different cultures, race, and language other than English may help further elucidate the nature of digital technology use while transitioning to becoming a parent.

Implications for Education, Practice, and Research

This research has implications for enhanced development of parents' digital health literacy skills; parents require the skills to be able to identify misinformation resulting from their online information seeking. Developers of prenatal education programs should consider the importance of digital health literacy skill enhancement and to generate online information to accommodate a range of parental digital health literacy skills. This work also has implications regarding health education and clinical assessment by health care providers; providing credible online resources constitutes an important health education strategy for information-seeking parents.

Further research is needed to understand the nuanced practice and policy impact of mothers' digital technology use and access to health care services. Further studies that create targeted advertisements to different types of parents, such as cis heterosexual men, 2LGBTIA+ families, folks who are gender nonbinary, and parents of different cultures, race, and language other than English may help further elucidate the gendered nature of digital technology use while transitioning to becoming a parent. For example, further research should seek to understand how men and fathers use digital technologies during their transition to parenting. Doing so may illuminate possible avenues to meaningfully engage men in the health information–seeking process and promote a sharing of health work within a parent dyad. Additionally, further research should critically examine how constant access to information affects mothers' perceived need to self-monitor as they transition to parenting. Such research may provide a deeper understanding of the unintended health consequences of constant surveillance on new parents. Together these understandings may provide health care providers and decision makers with information required to appropriately regulate how different health information can be shared with specific populations.

Conclusion

This study provided a descriptive analysis of how new mothers utilize different forms of digital technology as they transition to becoming a new parent. A range of technologies was used at each stage of the transition, with smartphones being ubiquitous across all stages. Digital technologies provided participants with convenient means to access health information from a range of sources such as websites, online support groups, and health care professionals. While digital technologies made health information access more convenient, participants were apprehensive about the credibility and trustworthiness of the information they retrieved due to limited transparency in the authors' expertise and credentials.

Conflicts of Interest

None declared.

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Original Paper

Perspectives of Pregnant and Breastfeeding Women on Participating in Longitudinal Mother-Baby Studies Involving Electronic Health Records: Qualitative Study

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Abstract

Background: Electronic health records (EHRs) hold great potential for longitudinal mother-baby studies, ranging from assessing study feasibility to facilitating patient recruitment to streamlining study visits and data collection. Existing studies on the perspectives of pregnant and breastfeeding women on EHR use have been limited to the use of EHRs to engage in health care rather than to participate in research.

Objective: The aim of this study is to explore the perspectives of pregnant and breastfeeding women on releasing their own and their infants' EHR data for longitudinal research to identify factors affecting their willingness to participate in research.

Methods: We conducted semistructured interviews with pregnant or breastfeeding women from Alachua County, Florida. Participants were asked about their familiarity with EHRs and EHR patient portals, their comfort with releasing maternal and infant EHR data to researchers, the length of time of the data release, and whether individual research test results should be included in the EHR. The interviews were transcribed verbatim. Transcripts were organized and coded using the NVivo 12 software (QSR International), and coded data were thematically analyzed using constant comparison.

Results: Participants included 29 pregnant or breastfeeding women aged between 22 and 39 years. More than half of the sample had at least an associate degree or higher. Nearly all participants (27/29, 93%) were familiar with EHRs and had experience accessing an EHR patient portal. Less than half of the participants (12/29, 41%) were willing to make EHR data available to researchers for the duration of a study or longer. Participants' concerns about sharing EHRs for research purposes emerged in 3 thematic domains: privacy and confidentiality, transparency by the research team, and surrogate decision-making on behalf of infants. The potential release of sensitive or stigmatizing information, such as mental or sexual health history, was considered in the decisions to release EHRs. Some participants viewed the simultaneous use of their EHRs for both health care and research as potentially beneficial, whereas others expressed concerns about mixing their health care with research.

Conclusions: This exploratory study indicates that pregnant and breastfeeding women may be willing to release EHR data to researchers if researchers adequately address their concerns regarding the study design, communication, and data management. Pregnant and breastfeeding women should be included in EHR-based research as long as researchers are prepared to address their concerns.

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KEYWORDS

electronic health records; pregnancy; breastfeeding; maternal-child health; research engagement; mother-infant medical record linkage

Introduction

Attempts to protect pregnant women by labeling them as a vulnerable population have played a role in excluding women, pregnant or not, from clinical research [1]. The difficulties in recruiting pregnant women for clinical trials are well documented [2,3], and tools such as the electronic health record (EHR) hold great potential for longitudinal mother-baby studies, ranging from assessing study feasibility to facilitating patient recruitment to streamlining study visits and data collection to providing data for retrospective observational studies. Longitudinal mother-baby studies are defined as studies that monitor the mother-baby pair beginning in pregnancy through the child's first few years of life. Although the benefits to researchers of using EHR data are well discussed in the literature [4,5], the perspectives of the participants, who are key stakeholders in the clinical research process, are understudied. Understanding the perspectives of pregnant and breastfeeding women in EHR-based research is an important step toward engaging this population in future research studies.

Only a few studies have examined patients' perspectives on the use of EHRs for research. Earlier studies suggested that less than a quarter of patients were willing to share their health records with researchers and even fewer were willing to share when their records contained sensitive information, such as HIV test results [6]. Later studies, likely corresponding with the increasing prevalence of the EHR, found greater willingness of study participants (ranging from 67% to 96%) to share with researchers [7-9]. Patients showed a strong preference for controlling which data would be available to whom [10-12] and were more likely to share deidentified data [6]. Trust in researchers was the strongest determinant of the level of protection desired for medical records and less trust correlated with a stronger desire for a more stringent EHR release process [11]. Patients also expressed concerns about the possibility that their data would fall into the hands of third parties, such as government agencies [6], for-profit organizations [10], and private health insurance companies [6]. Privacy, security, and trust in the research team were factors that impacted the decision to release the EHR to researchers.

Of the few studies to date that have explored the use of EHRs of pregnant women, most have only focused on the adoption of and engagement with EHRs through a patient-friendly portal that allows people to access their personal health information, to message providers, and to schedule appointments with providers [13-15]. Even studies on EHR portal use have largely been conducted in nonpregnant populations, despite indications that pregnant women are interested in web-based access to EHRs [13]. Engaging more pregnant women in longitudinal EHR-based research can help improve the scientific understanding of the developmental origins of health and disease. Successfully engaging this population in clinical research will require an understanding of their perspectives and concerns related to participating in EHR-based mother-baby studies. Therefore, we conducted an exploratory descriptive study of pregnant and breastfeeding women's perspectives on releasing their own and their infants' EHR data for longitudinal research to identify factors affecting their willingness to participate in research.

Methods

Overview

This qualitative study used semistructured, individual interviews with pregnant and breastfeeding women to elicit views about consenting to have their EHRs used for research. The reason for sampling from this population was to understand the perspectives and concerns of people who would be eligible for longitudinal mother-baby studies that use EHRs. The eligibility criteria mirrored those of a larger ongoing longitudinal mother-baby clinical study on the impact of breastfeeding on the infant gut microbiome (NCT03036696).

Individuals were deemed eligible to participate if they were aged between 18 and 40 years and were either pregnant or breastfeeding an infant under 12 months of age. Our study did not include English language fluency as an eligibility criterion. Exclusion criteria included a history of any of the following: inadequate breast milk production, pre-eclampsia, preterm delivery, or substance abuse during pregnancy. Thus, our sample represented those who would be eligible to participate in a real clinical study using EHR data. Participants were recruited through fliers posted at hospitals, restaurants, and grocery stores that detailed the study and included contact information for the research coordinator (MF). Participants were screened for eligibility over the phone, and interviews were scheduled upon confirming the participant's eligibility.

Approach

Trained interviewers (EF and MF) conducted all interviews from September 2017 to December 2018 in private rooms or offices on campus. A semistructured interview guide (Textbox 1) was used to elicit participants' views on research involving the EHR. Participants were asked about their familiarity with the EHR and experience using an EHR patient portal, for example, to communicate with their health care provider.

Questions also explored participants' views about giving researchers access to both their own and their infants' EHRs, length of time of access, and the inclusion of research results in their EHRs. Each interview lasted between 30 and 60 minutes, and the participants received an incentive of US $15. All semistructured interviews were audio recorded and professionally transcribed verbatim (Datagain). Transcribed interviews were stored in REDCap, a secure, web-based database platform. This study was approved by the institutional review board of the University of Florida (IRB201601909). None of the researchers involved had any conflicts of interest.

Textbox 1. Semistructured interview guide questions.

1. How familiar are you with electronic health records and electronic portals?
2. Do you interact with your doctor using the electronic portal?
3. Would you be comfortable with the research team accessing your medical records to collect data related to your pregnancy as part of the longitudinal study?
   - Is there anything you would not want the team to access from your medical record?
   - Can you think of anything that would be off limits either for you personally or in general?
4. Would you be comfortable with the research team accessing your medical records to collect data related to your infant as part of the longitudinal study?
   - Is there anything you would not want the team to access from your infant's medical record?
   - Can you think of anything that would be off limits either for your infant or about your infant's medical records in general?
5. What length of time would you feel comfortable with the research team being able to access your medical records as part of a research study?
6. What length of time would you feel comfortable with the research team being able to access your infant's medical records as part of a research study?
7. Would you want your research results to be included in the electronic health records?

Data Analysis

Transcripts were organized using NVivo 12 software (QSR International). Qualitative and quantitative methods were used to analyze the data. The sample size was determined by reaching thematic saturation [16]. An iterative, inductive approach to thematic analysis was used to examine the data. Two coders (AH and LC) first read all the transcripts line by line and then developed a codebook that reflects both a priori and emergent themes (Table 1). In the first stage of analysis, the 2 coders independently coded each of the transcripts for a priori themes. Frequent discussions to resolve discrepancies in code application occurred between coders until consensus was achieved and emergent themes were identified. The final coded data set was further organized within a spreadsheet for subsequent exploratory analysis. The reliability of findings was enhanced by using a constant comparative method [17] in which coders compared subsequent transcripts with previous transcripts to confirm consistency of themes across data.

Table 1. A priori and emergent themes.

Themes	Description	Example quote
Concerns about privacy and confidentiality (a priori)	Factors pertaining to limited access of EHR[a] data, including limiting of information related to stigmatizing conditions, deidentification of records, and release without consent to third parties	"I would like to have control over as much of my privacy as I can."
Role of transparency by the research team (a priori)	Factors related to full disclosure about the research being conducted, the purpose for which medical records are being used, and the need for researchers to reobtain consent from participants for future use of EHR data	"Yeah, I don't know the answer. I guess it would have to be I would have to know a little bit more about what the study would be that would require my medical records before I'd say yes or no."
Concerns about surrogate consent (emergent)	Parent or legal representative concerns about consenting for their neonate to participate in clinical research, including the length of access to the child's record and how release of the child's EHR could affect the child later on	"Yes, I guess. That's a hard one for me to answer. Here's why. It's because I'm answering for a child who doesn't have a say..."

[a]EHR: electronic health record.

Results

Participants

Participants included 29 women who were either breastfeeding (n=10) or pregnant (n=19). The demographic characteristics of the participants are presented in Table 2. The age range of participants was from 22 to 39 years, with most (66%) in their 30s. The education level varied from an associate degree to a professional degree, with most (83%) having a bachelor's degree or higher. Most participants described their race as White. The racial characteristics of our sample were similar to those of a local county [18].

Table 2. Participant characteristics (N=29).

Characteristics	Values, n (%)
Age group (years)	
20-29	10 (34)
30-39	19 (66)
Education	
Professional or graduate degree	17 (59)
Bachelor's degree	7 (24)
Associate degree	3 (10)
Tech or vocational degree	2 (7)
Race or ethnicity	
Black	5 (17)
White	20 (69)
Other	1 (3)
Missing	3 (10)
Familiarity with EHR[a] and EHR portals	
Familiar	27 (93)
Not familiar	1 (3)
Not asked	1 (3)
Willingness to release own EHR	
Yes	18 (62)
Ambivalent or conditional yes	11 (38)
Willingness to release infant's EHR	
Yes	18 (62)
Ambivalent or conditional yes	7 (24)
Missing	4 (14)
Length of time of EHR release	
Equal or longer than the length of the study	12 (41)
Others	12 (41)
Missing	5 (17)

[a]EHR: electronic health record.

Familiarity With the EHR

Almost all participants (27/29, 93%) had existing knowledge of and were familiar with the EHR and EHR portals. Participants were coded as being familiar with an EHR portal if they could provide specific examples of how they used it, such as communicating with a physician, checking in for appointments, or viewing test results. Most participants primarily used the EHR to update their health information and view the test results. Notably, 2 participants also had experience interacting with an EHR system for their jobs. Many participants used EHR portals to communicate with providers, including 3 participants who stated that they did this primarily during pregnancy and 1 who stated that it was her preferred method to ask questions because phone calls had a much longer follow-up period. One participant reported preferring to converse with providers in person.

Willingness to Release Records

Most participants were willing to release their own and their infants' EHRs to the researchers. Willingness to provide access to EHR data fell within 2 categories: full EHR release and conditional EHR release. Full EHR release was characterized by participants being completely comfortable and willing to release their own and their infant's EHR for research purposes and without conditions. Conditional EHR release reflected ambivalence about sharing EHR data and was characterized by the participants' willingness to provide restricted or stipulated access to their EHR (eg, "Researchers can access my data as long as they are transparent about its use"). Nearly half of the participants (n=12) were willing to make their own and their infants' EHRs available to researchers for the duration of a research study or longer. More than one-third of the respondents (n=11) expressed conditional agreement about releasing their EHR for research. The salient themes of participants' concerns regarding EHR release are described as follows.

Salient Themes of Participant's Concerns for Releasing EHRs

Although participants were familiar with EHR portals and willing to release their EHRs for research, they articulated several concerns. Concerns centered around 3 themes, including privacy and confidentiality, transparency by the research team, and surrogate consent for infants. Finally, we share patient insights into how the EHR portal may be used for research engagement.

Privacy and Confidentiality

Privacy and confidentiality concerns included whether information would be dispersed without prior consent; the types of personal information that would be used in the study, including access to stigmatized health information; and whether deidentification would be used. Participants were particularly concerned with anonymity and were interested in sharing both their own and their infants' EHRs if the information was deidentified (Textbox 2).

Textbox 2. Quotations representing concerns about privacy and confidentiality when releasing electronic health records for research.

- "That's the only other thing that comes to mind is that maybe it would be deidentified and maybe not use her face along with that if that makes sense." [BIS014]
- "They don't need all of my medical records…I look at the big scale, just the internet today, and how everybody has access to everything, and how there's crazy stuff politically and crazy people, if someone were to ever take advantage, I would like to have control over as much of my privacy as I can." [BIS003A]
- "I'm sure there's people with certain conditions like HIV and stuff like this who wouldn't want that type of stuff to be exposed." [BIS030]
- "I say, this should be like in a secured and it shouldn't be shared with others without permission." [PRG003]
- "I have a very easygoing pregnancy, no complications...So, I'd be comfortable. I don't know if another mom would be if they had some complications or genetic history or whatever." [PRG016]

Participants needed assurance that the EHR data would be secure, with limits on who could access the data. In particular, participants were concerned about the possibility that their data might be shared with third-party institutions, such as health insurance companies:

> *My concern would be if in the research study, anything like if anything came back long term genetic...I don't want connected [to my EHR] because of getting health insurance. [If] I have to get my own plan, how pre-existing conditions will affect it...that would be my biggest concern. Just because I know I don't, I don't trust the state of health care in the country right now.* [PRG010]

Participants also raised concerns about giving researchers unlimited access to EHRs and providing access to stigmatizing health conditions in their EHRs. One breastfeeding woman stated that she was uncomfortable releasing provider notes that included stigmatizing or potentially embarrassing conditions:

> *I think the only way I would maybe not feel comfortable is if I had some sort of alcohol or substance use disorder, if I engaged in an activity that was embarrassing for me, things that are stigmatized, if I had mental health issues. I'm lucky I don't, so I don't have an issue, or if I had HIV or some other infection like that. Yeah, basically any stigmatizing conditions, I might not be open to allowing people to seeing my her.* [BIS001]

In addition to substance use disorders, participants were concerned about general mental health conditions, miscarriages, medical conditions unrelated to pregnancy, HIV, and genetic panels of their infants. People were less willing to share information about medical conditions that were perceived to be more stigmatized.

Transparency by the Research Team

Participants also discussed the importance of transparency by the research team in their decision to release their EHRs (Textbox 3). Transparency is described as full disclosure of the research being conducted and the purpose for which medical records are being used. Research team transparency also includes the need for researchers to reobtain consent from participants for future use of EHR data. Participants expressed fear regarding how the information in the released EHR would be used by the research team. They also wanted the study personnel to clearly explain the specific EHR elements (ie, data points) needed for the study and how the information would be used, with a justification for the length of time records to be accessed.

Textbox 3. Quotations representing the role of transparency by the research team when releasing electronic health records for research.

- "It just depends on how they are going to use that data." [BIS009]
- "I guess it would have to be I would have to know a little bit more about what the study would be that would require my medical records before I'd say yes or no." [BIS013]
- "I guess, I would wanna know and understand why the research team would need continuous access...Throughout the study like what information do you need after like getting my blood type and, you know, my initial like assessment of where I'm at." [PRG016]

One participant remarked on the complexity of conducting research and the possibility of needing to reconsent at a later time point in longitudinal studies:

> *If you're studying developmentally how the child is changing and how good health is affecting that. A lot of times, some of these things aren't diagnosed till later. But do you probe first the parent and then decide whether you're going to collect...I don't know. I don't know if this would just be an open thing where they can do it at any time, but it's like, we're monitoring and then we go, "We're seeing a trend and we want to collect the data on the medical records and this information. Does the parent approve? This is why," and explain to the parent how it could be helpful for future children type of thing.* [BIS003]

Surrogate Consent

Although many participants were comfortable releasing their own and their infant's EHR for research purposes, others expressed uncertainty. In particular, participants were concerned with providing surrogate consent, which was described as a concern over hypothetical situations in which the child may later disagree with the parent's decision to participate in the study. One participant shared:

> *How is this going to impact him when he's older?...You know, where does this information go? Could it ever potentially become something that's limiting or "Mom, why did you release my information to this," you know? Like, "Why do these people keep contacting me? I don't want to participate."* [BIS011]

In one instance, the participant provided a hypothetical example of how her surrogate decision making may intrude on her child's autonomy in deciding who is privy to the child's protected health information:

> *That's a hard one for me to answer. Here's why. It's because I'm answering for a child who doesn't have a say, and maybe they wouldn't, one day, like that information out there. Especially if there's some condition they may end up having later that we don't know, like autism or whatever.* [BIS003]

This discomfort reflects concerns over unpredictable future consequences resulting from their surrogate consenting on behalf of their child to release their child's EHR to researchers. These persons were keenly aware that their decisions may have a lasting, unforeseen impact on their children.

Research Results in EHRs

Researchers can write research notes in an EHR, which become a part of a patient's medical record. Laboratory tests ordered for research instead of clinical care may be included in the EHR. In the final part of the interview, participants were asked, if given the option, whether they would prefer their research results to be included in their EHRs. Participants were overwhelmingly interested in being able to access their research results (eg, laboratory test results conducted as part of a clinical research study) in their EHRs. One participant shared how receiving results would make her feel like she "is a part of a bigger picture...and doing something important" (PRG014). Furthermore, feedback from the research team in the form of research-generated results through the EHR was noted as a strategy to enhance transparency and improve trust in the research process:

> *Yeah, I think it's a good thing because I can't see, so how they use my information, my reports and I'm aware of the process...if they can share some results with me, or at least tell me what they are doing with my records and information, it makes me more happy and confident about the process...And I can trust them.* [PRG011]

A few participants expressed ambivalence about receiving research-generated results through their EHRs. Participants did not want research results to be included if they revealed a stigmatizing condition, such as being a *heroin addict*. Others did not want their research results to be included as part of their permanent record, owing to concerns regarding the physician's ability to interpret research results, which may complicate care.

Discussion

Principal Findings

The aim of this study was to understand the concerns and reservations of pregnant or breastfeeding women about participating in longitudinal mother-baby studies that use EHRs. The participants in our study were largely familiar with the EHR, many gaining familiarity through access to their own EHR. More than half of the pregnant and breastfeeding participants were willing to share their EHR data with the researchers. This finding is similar to that of the research on nonpregnant patients' willingness to share their EHR data [8,9]. In our study, participants wanted to be informed about how researchers were using their EHR data and to retain control over which elements of the EHR were released. In a 2019 study, more than three-fourth of participants who were given a list of EHR data elements to share with researchers chose to withhold

at least one item [9]. This control may be an important component of a person's willingness to participate in a study.

Integrating Research and Health Data

A salient topic discussed by participants was integrating research data with health data in the EHR. Several participants advocated the release and availability of research test results in the EHR. Previous studies support this finding that women overwhelmingly want to actively engage in their health care through the use of EHRs [13,19]. Furthermore, these studies found that pregnant women were significantly more likely to log in to the EHR portal when they could view their personal antenatal health record [20], and the majority of those who created an EHR portal account would use it again for future pregnancies [14]. Moreover, in recent years, organizations such as the National Institutes of Health and the National Academy of Sciences have increasingly demanded that individual research results be shared with participants in biomedical research. This accessibility creates a patient-centric approach to research, which provides a level of transparency that may increase both trust in the research team and future participation in research [21]. Thus, there is an exciting potential for EHRs to encourage research participation. Perhaps a research portal interface with the EHR can help researchers engage populations historically excluded from clinical research. The finding that accessibility to research test results was viewed as improving transparency and trust in the research team suggests that a research portal interface can also help repair the broken trust in research held by certain populations. The integration of health and research uses of the EHR may also be beneficial during a time when participants might have reservations about making additional in-person visits to the hospital, such as during a pandemic. However, using the EHR to encourage active research participation may also perpetuate disparities in research participation as race, education level, and internet access have been shown to affect EHR engagement [22].

Versatility of the EHR

Some participants also recognized that the EHR is a 2-way street: not only are health data going to researchers but research data may also go to health care providers. The integration of research and health data within a health system also highlights the potential for research to help serve those in lower resource settings: research dollars could possibly pay for tests or laboratories that may otherwise be unavailable to patients. Research results also have the potential to serve as a point of health care intervention (eg, screening out a prospective participant because of abnormal results on a laboratory test can serve as an opportunity to refer a patient to an appropriate physician), although participant opinions ranged from doubt (inability of doctors to interpret research results) to objection (in the case of stigmatizing conditions). A frequently cited concern was that a stigmatized condition (eg, substance abuse) discovered during a research study may be shared with health care providers and other third-party vendors, such as health insurance companies. Although perceived stigma led to an emphasis on privacy and confidentiality, we also found that trust was important in mitigating these concerns. Trust in the research team to deliver promises of privacy and confidentiality was an important component of research participation and EHR release. Although all participants hypothetically spoke about having a stigmatized condition, their concerns reflected a real issue of selection bias in EHR research. Patients with stigmatized conditions may be less likely to opt for EHR research studies, which would affect the representativeness of EHR data and compromise generalizability.

Limitations

Although thematic saturation was reached for pregnant or breastfeeding participants' concerns about releasing their own and their infants' EHRs for research, a limitation of our study is the lack of racial and educational diversity and may play a role in the themes identified in the results. For example, previous studies have found that Black participants and those reporting lower education were less trusting of medical researchers and spent more time during the consenting process [8]. Further investigations of racially and ethnically diverse obstetric patients' familiarity with EHR-based research and their perspectives on releasing EHR data to researchers are warranted. Moreover, this study excluded people with previous pregnancy complications, which may have introduced selection bias. Future studies should explore whether this group exhibits different views on sharing EHR data for research. In addition, access to and familiarity with EHRs is predicated on having internet access, which indicates the need to include more educationally and socioeconomically diverse populations.

Conclusions

Previous qualitative studies within the pregnant population have focused on understanding their perspectives on antibiotic use to develop tailored perinatal health education interventions to increase knowledge, particularly using EHRs, to provide additional information on antibiotic use [23]. This is the first qualitative study to explore the perspectives of pregnant or breastfeeding women on participating in EHR research and provides significant insights into their attitudes toward sharing their own and their infants' EHRs. Participants were largely familiar with engagement of their EHR for health care purposes, and most of them were willing to release their EHRs to researchers, provided their concerns for privacy, confidentiality, and transparency were addressed. Participant responses suggested that the EHR may play an underappreciated role in clinical research by providing research-generated test results to participants. This finding marks a departure from a singular focus on only studying the use of the EHR for health engagement toward use for research engagement among pregnant and breastfeeding women. How the EHR can be mobilized to better engage populations traditionally excluded from clinical research is an important topic for future studies.

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Conflicts of Interest

None declared.

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Abbreviations

EHR: electronic health record

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Original Paper

Digital Tools to Support Family-Based Weight Management for Children: Mixed Methods Pilot and Feasibility Study

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Abstract

Background: Family-based behavioral therapy is an efficacious approach to deliver weight management counseling to children and their parents. However, most families do not have access to in-person, evidence-based treatment. We previously developed and tested DRIVE (Developing Relationships that Include Values of Eating and Exercise), a home-based parent training program to maintain body weight among children at risk for obesity, with the intent to eventually disseminate it nationally alongside SafeCare, a parent support program that focuses on parent-child interactions. Currently the DRIVE program has only been tested independently of SafeCare. This study created the "mHealth DRIVE" program by further adapting DRIVE to incorporate digital and mobile health tools, including remotely delivered sessions, a wireless scale that enabled a child-tailored weight graph, and a pedometer. Telehealth delivery via mHealth platforms and other digital tools can improve program cost-effectiveness, deliver long-term care, and directly support both families and care providers.

Objective: The objective of this study was to examine preliminary acceptability and effectiveness of the mHealth DRIVE program among children and parents who received it and among SafeCare providers who potentially could deliver it.

Methods: Study 1 was a 13-week pilot study of a remotely delivered mHealth family-based weight management program. Satisfaction surveys were administered, and height and weight were measured pre- and post-study. Study 2 was a feasibility/acceptability survey administered to SafeCare providers.

Results: Parental and child satisfaction (mean of 4.9/6.0 and 3.8/5.0, respectively) were high, and children's (N=10) BMI z-scores significantly decreased (mean –0.14, SD 0.17; *P*=.025). Over 90% of SafeCare providers (N=74) indicated that SafeCare families would benefit from learning how to eat healthily and be more active, and 80% of providers reported that they and the families would benefit from digital tools to support child weight management.

Conclusions: Pediatric mHealth weight management interventions show promise for effectiveness and acceptability by families and providers.

Trial Registration: Clinicaltrials.gov NCT03297541, https://clinicaltrials.gov/ct2/show/NCT03297541.

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KEYWORDS

parent training; weight loss; telehealth; obesity; SafeCare

Introduction

Obesity affects nearly one in five children and adolescents in the US [1]. The US Preventive Services Task Force [2] and the American Medical Association [3] recommend comprehensive, intensive, family-based weight management programs to treat childhood obesity. Family-based behavioral therapy is efficacious [4], although most children do not have access to evidence-based treatment due to limited availability of programs and trained providers, barriers for travelling to in-person sessions including transportation and time constraints, and cost of participation due to limited or no insurance coverage [5,6].

To overcome barriers to access, evidence-based models that include parent training (eg, SafeCare, Parents as Teachers) can be delivered in the family's home [7]. SafeCare is a parent support program delivered by trained providers that focuses on parent-child interactions to mitigate the risk of abuse or neglect. SafeCare is predominantly delivered in the home, but sessions can also be delivered via technology, over video chat and telephone [8]. SafeCare has been disseminated in more than 25 US states and internationally. Currently, there are approximately 100 SafeCare accredited agencies where providers serve more than 6000 families per year. The underlying principles of SafeCare on improving parent-child interaction, coupled with its broad reach to at-risk and underserved families, make SafeCare an ideal platform for delivery of weight management services.

We developed a parent support focused program to treat childhood obesity that can be delivered in the home called DRIVE (Developing Relationships that Include Values of Eating and Exercise) [9] with the intent to eventually disseminate the program across the SafeCare network. DRIVE incorporates SafeCare principles to promote healthy eating, physical activity, and healthy weight in children by fostering positive parent-child interactions. Previously, we tested the efficacy of DRIVE in a 19-week randomized controlled pilot trial in 16 parent/child dyads (children ages 2-6 years with BMIs ≥ 75th percentile) and found that the change in children's BMI z-scores (BMIz) (Mean –0.1, SE 0.1) was significantly different (P<.01) compared to a health education control group (mean 0.5, SE 0.1) [9].

Although DRIVE was initially developed for in-person delivery, telehealth delivery via mHealth platforms can improve cost-effectiveness, deliver long-term care, and directly support both families and care providers [10]. Identifying alternate avenues for families to access care is increasingly important [10], including for children with obesity during the COVID-19 pandemic when families are unwilling or unable to present in-person for treatment [11]. To this end, the objectives of the studies reported herein were to examine 1) the acceptability of a remotely delivered weight management program (mHealth DRIVE) as determined by the parents and children who used the program, 2) the preliminary effectiveness of this virtual program to reduce child body mass, and 3) the perceived need and willingness to deliver mHealth DRIVE by SafeCare providers.

Methods

Study 1

Participants

Parents were recruited from their children's after-school wellness program. Parents were invited to attend an informational session that explained the purpose of mHealth DRIVE. Eligibility criteria for children included ages 5 to 14 years; be physically capable of exercise; and be free of diseases that affect metabolism, body weight, and food intake, including type 1 or type 2 diabetes, HIV/AIDS, and cancer. Children were excluded if they had significant cardiovascular disease or disorders or other significant medical problems that would prevent them from engaging in regular physical activity. Inclusion criteria for parents included having a smart phone and being willing to use the smartphone for the intervention. Eleven child/parent dyads enrolled, but 1 dyad was excluded from all analyses because of the child's low BMI percentile (4^{th} percentile). Parents provided written informed consent, and children provided assent. Study procedures were approved by the Pennington Biomedical Research Center institutional review board.

Intervention Sessions, Treatment Goals, and Tracking of Weight and Behaviors

Child/parent dyads attended 8 counseling sessions (approximately 30 minutes each) primarily over their internet-connected device (eg, smartphone, tablet, laptop, or desktop computer). Most interactions were via video calls or phone, but email and text communication also occurred. The DRIVE curriculum was shortened to 13-weeks to align with the school semester. A Pennington Biomedical counselor delivered sessions and provided individualized advice and problem-solving strategies for the parent and the child. Each session included an interactive component for the parent and child related to healthy eating and active play, and interactive parenting training. Sessions were based on treatment methods that promote child weight loss that have been sustained for 10 years [12,13]. Although the sessions were remotely delivered, counselors were able to deploy motivational interviewing techniques to address decreases in motivation, which are inevitable in longer-term interventions [14].

The guiding principles of the sessions were 1) weight and activity monitoring; 2) building commitment and overcoming barriers to healthy behavior changes, with a goal of teaching the parent to model appropriate diet and physical activity behaviors for their child; 3) review of progress and problem-solving to address poor adherence to behavioral goals; and 4) food monitoring and goal setting for nutrient intake. Sessions focused on how to motivate the child and manage noncompliance; techniques included praise and reward, positive reinforcement, selective ignoring, contracting, preplanning for meals and physical activity, shaping behaviors, modeling, changes to the home environment, and facilitating social support for behavior change [15,16]. The dietary approach employed food monitoring and goal setting for nutrient intake, and the Traffic Light Diet [12] was included to facilitate remote

modification of dietary changes. The Traffic Light Diet teaches parents and children to categorize foods based on green (low calorie foods to be eaten freely), yellow (moderate-calorie foods to be eaten occasionally), and red (high-calorie foods to be eaten rarely), with the goal to gradually reduce the number of red foods eaten each week. The physical activity approach introduced free or inexpensive activity options that the children enjoyed and addressed barriers to physical activity.

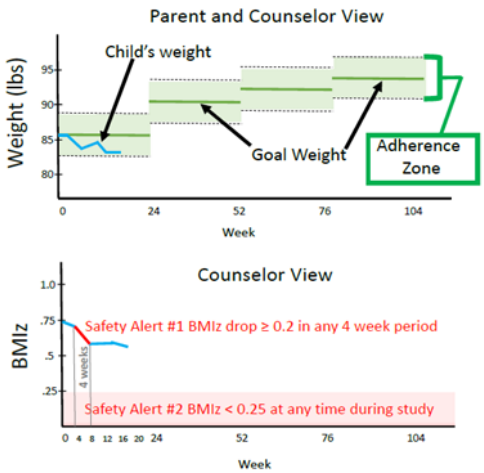
Children's energy requirements were estimated using a physical activity level of 1.4 and the Harris-Benedict equation, which has good accuracy in youth with obesity [17]. The energy intake goal was 250 kcal/d less than estimated energy requirements, which should promote modest weight loss and weight gain attenuation over time. The activity goal of children was to gradually increase physical activity to a goal of approximately 6,000 steps/day above their personal baseline values, which is appropriate as we expected low baseline physical activity [18]. This activity goal was the equivalent of an additional 30 min/day of moderate-to-vigorous physical activity as a gradual increase towards the physical activity guidelines of 60 min/day of moderate-to-vigorous physical activity [18].

The intervention content and parent-training approach were based on DRIVE, and the mHealth aspects of the intervention were based on a successful weight management intervention for adults called SmartLoss [19,20]. Specifically, children's daily physical activity (steps/day) was tracked with a hip-worn Omron HJ-324U pedometer (Omron Healthcare, Inc, Kyoto, Japan), and the parent was asked to document their child's steps daily. The counselor plotted the child's daily step data in relation to their individual goals to help promote adherence to activity goals. The children also received a BodyTrace scale that automatically sent their weights to a website accessible by the counselors. Children were asked to weigh themselves at least weekly, unless contraindicated due to anxiety or other mental health barriers, similar to SmartLoss [19,20]. Weighing at the same time of day and in the same state was encouraged, preferably after getting out of bed in the morning and after voiding. Children's body weight and a weight graph were used to guide intervention delivery to facilitate healthy weight management and avoid unsafe changes in body weight. Specifically, and as detailed in the upper panel of Figure 1, a 6-pound "zone" of acceptable weights or "adherence" was created, and children's individual weights were plotted against this zone. Hence, the zone promotes weight maintenance, but it allows for weight loss of less than 3 pounds if the child's BMI is greater than or equal to the 85th percentile. Further, this approach includes objective safety criteria that are triggered if rapid or excessive weight loss occurs (see Figure 1, lower panel).

The program encourages healthy eating, activity, and weight tracking over time. The counselor and parent utilized the weight graph to modulate intervention intensity and as an objective indicator of the need to change the child's energy intake level. Specifically, the counselor and parent 1) *increase* energy intake if weight loss is excessive, defined as more than 0.2 BMIz reduction within 1 month, which aligns with American Medical Association recommendations for maximum 2 lb/week weight loss in children [3]; 2) *maintain* energy intake if weight maintenance is observed, until the child's BMIz reaches 0.25 (approximately equivalent to the 60th BMI percentile); and 3) *reduce* energy intake if the child is gaining weight at a rate that increases the child's BMIz, unless he/she has reached 0.25 BMIz or approximately the 60th percentile, at which time the child increases body weight over time to maintain 0.25 BMIz or approximately the 60th percentile. The threshold of 0.25 BMIz to begin weight maintenance aligns with the goal of reducing BMIz without promoting energy restriction that could negatively impact growth and development. In a longer-term intervention, the zone would be adjusted every 6 months according to increases in the child's height (see Figure 1), but this pilot study did not adjust the zone due to the study being only 13 weeks in duration. The counselor electronically provided the parent with the child's weight graph during each session (see the upper panel of Figure 1).

Parents who needed help modifying their child's diet had the option of sending their counselor images of how they prepare foods and what foods they provide to their child and family. These were not outcome data but provided the counselor with near real-time data on changes the parents could make to improve their child's diet and health. These images can be captured with any camera-enabled device, and smartphone apps are available to streamline this process (eg, SmartIntake).

Figure 1. Weight graph zone of child's adherence. BMIz: BMI z-score.



Measures and Data Analysis

Parents and children completed an acceptability survey at the end of the intervention that included Likert scales on intervention satisfaction (see Table 1). Children's height and weight (shoes removed, no outer clothing) were collected in duplicate at baseline and end of study by trained assessors, and both assessments occurred in the afternoon. Height was measured with a stadiometer, with the child standing feet flat, with heels, buttocks, upper back, and back of head contacting the stadiometer, and the child's head facing straight ahead. Weight was measured with a digital scale with the child standing in the middle of the scale with arms hanging loosely at their side. Height and weight were recorded to the nearest 0.1 unit (cm or kg, respectively); if the two measures differed by more than 0.5 units, a third measurement was taken and the closest two of three were used in analysis. Mean values and percentages were calculated for satisfaction surveys. Differences in BMI were examined using *t*-tests, with an alpha level of .05. Analyses were conducted using SPSS.

Study 2

A survey of SafeCare providers was conducted across the US to assess 1) the perceived need for diet, physical activity, and weight management services for SafeCare children, and 2) the willingness of SafeCare providers to offer such services.

Participants

Eighty-two SafeCare providers from 14 states provided consent and completed the survey. The sample was predominantly female (n=71), with 5 males, 1 other, and 5 unknown. The mean age of providers was 39.8 years (SD 12.9), with 17 unknown age data. Thirty-eight providers reported delivering care in urban cluster/suburban areas (2500-50,000 people), 22 in urban areas (≥50,000 people), and 18 in rural areas (<2,500 people), with 4 unknown.

Procedures

A recruitment email was sent to the potential participants using the list of contact information for US SafeCare providers. The email contained an anonymous link to the survey conducted through Qualtrics, a secure web-based survey platform that employs high-level security measures to ensure data are protected from malicious data breaches and requires a password in order to download the data. A reminder email was sent 1 week later, reminding participants of the opportunity to complete the survey. The survey was open for 2 weeks.

Measures and Data Analysis

The survey queried demographic data (age, gender, and level of urbanicity where services are delivered) and assessed if SafeCare providers perceive a need for or have experience with additional educational material for child nutrition/weight management. Data were cross-sectional and were analyzed descriptively (ie, percentages were reported for categorical variables; means or percentages were reported for Likert scale items). Reported percentages collapse the "Strongly Agree" and "Agree" responses.

Results

Study 1

Of the 10 children, 6 were girls (60%) and the mean age was 7.8 years (SD 2.3 years; range 6-14 years). Mean BMI percentile and BMIz were 86th (SD 0.17) and 1.4 (SD 0.7), respectively. Four children had obesity, 4 were overweight, and 2 were normal weight. There was a statistically significant reduction in children's BMIz over the 13-week period (mean –0.14, SD 0.17; *P*=.025). There was also a significant BMIz reduction among the 8 children who were overweight or had obesity (mean –0.18, SD 0.15; *P*=.013). The 2 normal weight children did not lose weight. Parental satisfaction (4.9/6.0) and child satisfaction (3.8/5.0) were high (see Table 1).

Table 1. Parent (n=10) and child (n=10) satisfaction survey results.

Survey items	Rating score						
	Mean (SD)	1	2	3	4	5	6
Parent items 1 (responses ranged from 1=Strongly Disagree to 6=Strongly Agree)							
Seeing my child's weight on a graph every week helped me make better food choices for him/her.	4.4 (1.2)	0	0	3	3	1	3
My child was willing to step on the bathroom scale once per week.	5.6 (0.8)	0	0	0	2	0	8
My child was willing to wear a pedometer every day.	4.0 (2.4)	4	0	0	0	0	6
Tracking my child's steps each day helped him/her reach physical activity goals.	4.5 (1.7)	0	2	2	0	1	5
Tracking the foods my child ate helped him/her reach weight goals.	5.1 (1.2)	0	0	2	1	1	6
The healthy tips my child and I received helped me make healthy lifestyle changes for my child.	5.4 (0.9)	0	0	1	0	3	6
The information I received in my health tips helped me make healthy lifestyle changes for my family & myself.	5.3 (0.8)	0	0	0	2	3	5
I enjoyed the individual time talking with my counselor.	5.8 (0.4)	0	0	0	0	2	7
The amount of time talking with my interventionist was enough.	5.7 (0.5)	0	0	0	0	3	7
I would have liked to spend more time talking with my interventionist.	1.8 (0.9)	5	2	3	0	0	0
I enjoyed meeting with my counselor remotely (by phone call or video chat on my smartphone).	5.8 (0.4)	0	0	0	0	2	8
Parent items 2 (responses ranged from 1=Not helpful to 6=Very Helpful)							
Learning about the importance of self-monitoring how much we eat and our activity.	5.7 (0.5)	0	0	0	0	3	7
Learning about portion control.	5.7 (0.5)	0	0	0	0	3	7
Learning about choosing the right foods for you and your child.	5.4 (0.7)	0	0	0	1	4	5
Learning about how to build good social support.	5.4 (0.9)	0	0	1	0	3	6
Learning about fat, protein, and carbohydrates.	5.5 (0.5)	0	0	0	0	5	5
Learning about how to overcome barriers to being healthy.	5.4 (0.9)	0	0	1	0	3	6
Learning about how to make better choices when eating outside the home.	5.7 (0.5)	0	0	0	0	3	7
Learning how to make healthy choices on special occasions such as birthday parties and school functions.	5.3 (0.6)	0	0	0	1	5	4
Learning about healthy eating plans for the whole family, like the Stoplight approach to healthy eating.	5.7 (0.5)	0	0	0	0	3	7
Learning about how much physical activity is recommended for me and my child.	5.5 (0.5)	0	0	0	0	5	5
Taking a closer look at why we eat.	5.3 (0.6)	0	0	0	1	5	4
Learning about healthy beverage choices for me and my child.	5.2 (1.5)	1	0	0	0	3	6
Child items (responses were 1=No; 2=I don't think so; 3=Maybe; 4=I think so; 5=Yes)							
I liked wearing my pedometer.	3.1 (0.4)	2	0	5	1	2	N/A
I liked seeing how many steps I can get each day.	4.3 (0.3)	0	1	1	2	8	N/A
The pedometer was easy to use.	3.5 (0.5)	1	2	1	3	3	N/A
I tried to move more.	3.5 (0.5)	2	0	2	3	3	N/A
I liked talking to [interventionist] about eating healthy foods and being more active.	4.0 (0.4)	1	0	2	2	5	N/A
I tried to eat healthier foods.	4.1 (0.4)	1	0	2	1	6	N/A
I tried new healthy foods that I had not tried before.	3.8 (0.5)	1	1	2	1	5	N/A
I ate less candy.	3.3 (0.5)	2	2	1	1	4	N/A

Survey items	Rating score						
	Mean (SD)	1	2	3	4	5	6
I drank less soda.	3.7 (0.5)	1	1	2	2	4	N/A
I talked with my parents about eating healthier foods.	2.7 (0.5)	3	1	3	2	1	N/A
Getting on the scale once a week was easy.	4.2 (0.4)	1	0	1	2	6	N/A

Study 2

Nearly all respondents indicated that SafeCare families would benefit from learning how to eat more healthily and be more active (71/74, 96% and 68/74, 92%, respectively), and many (57/72, 79%) perceived that families would benefit from a program for child weight management. Most providers indicated that they were interested in learning how to deliver nutrition and physical activity information to their families (70/74, 95% and 60/74, 81%, respectively). About 80% (59/74) of providers reported that they and their SafeCare families would benefit from digital tools to support child weight management (see Table 2).

Table 2. Mean feasibility ratings reported by SafeCare providers (N=74), followed by the number (n) of providers who endorsed each rating from Strongly Disagree (1) to Strongly Agree (4).

Survey items	Mean (SD)	Strongly Disagree, n	Disagree, n	Agree, n	Strongly Agree, n
The parents I work with have regular access to healthy foods.	2.5 (0.7)	7	25	39	3
The parents I work with and their families would benefit from learning more about how to eat healthy.	3.3 (0.6)	1	2	44	27
I would be interested in learning how to deliver nutrition information to the parents I work with.	3.4 (0.6)	0	4	38	32
Most of the parents I work with or their families would benefit from weight loss or better weight management.	2.9 (0.8)	1	25	28	20
I would be interested in learning how to deliver weight management information to the parents I work with.	2.9 (0.9)	5	20	25	24
The parents I work with and their families would benefit from learning more about healthy levels of physical activity and exercise.	3.3 (0.6)	0	6	43	25
I would be interested in learning how to deliver information on physical activity to the parents I work with.	3.1 (0.7)	0	14	38	22
The parents I work with would benefit from a home visiting program designed to improve the body weight and health of young children in the home.[a]	3.0 (0.7)	1	14	40	17
The parents I work with would benefit from mobile health tools (smartphones, online dashboards) designed to improve their diet, activity levels, body weight, and health.	3.1 (0.7)	0	14	42	18
I would be interested in receiving support via mobile health tools (smartphones, online dashboards) to help me deliver health and weight management information to the parents I work with and their families.	3.1 (0.7)	0	15	34	25

[a]N=72 due to missing responses.

Discussion

Principal Findings

In this one-arm small pilot study, an mHealth weight management program significantly reduced children's BMIz, and both parents and children had high levels of satisfaction. These data complement and build upon the prior DRIVE in-person home-based weight management program by integrating digital tools including telehealth counseling sessions, a wireless scale that enabled a child-tailored weight graph, and a pedometer to track child physical activity. Further, the survey of SafeCare providers indicated that providers perceive a need for this type of family-based weight management program and expect that their families will find remotely delivered content and digital tools to be acceptable.

Collectively, these preliminary data suggest that a weight management program delivered to parent/child dyads may be successful when implemented alongside a parenting program, such as SafeCare, via an mHealth platform. These data contribute to the burgeoning evidence that telehealth may be useful as adjunctive to in-person pediatric weight management. A nonrandomized comparative effectiveness study of 100 adolescents participating in a 2-year weight management program compared in-person plus telehealth versus in-person only and observed similar BMI outcomes, attendance rates, and acceptability among families and healthcare providers across the two groups [21]. Digital tools may not only remove barriers to transportation and scheduling for in-person care delivery but also expand reach of interventions to areas that are less likely to have access to multi-disciplinary care, particularly to families who are low income with limited resources such as those served by SafeCare agencies.

A key benefit for the remote delivery of weight management counseling is to increase accessibility to families, especially in more rural areas. However, the family must have the necessary equipment including an internet-enabled device (eg, smartphone, tablet, or computer that is connected to the internet via either a cellular network or WiFi). A recent study of the virtual delivery of SafeCare indicated that many families experienced limited broadband access and technology fatigue, resulting in the need to deliver shorter counseling sessions less than 30 minutes in length [8]. Online interactions may also lessen rapport between the provider and family due to limited ability to see nonverbal cues such as body language. A prior study of a hybrid version of SafeCare, including both face-to-face and virtual sessions, indicated that technology assistance offered efficiencies to the providers in terms of preparation for sessions, but the provider spent more time engaged in rapport-building activities with the family when delivered remotely [22].

Importantly, the parents and children in the pilot study expressed high levels of satisfaction with the remotely delivered program. Children rated satisfaction with talking to their counselor about eating healthier foods as higher than talking with their parent about eating healthier foods, highlighting the effectiveness of remote counseling but the need for further support of the parent-child interaction regarding healthy behavior change. Our findings expand upon a prior study of 360 children and parents randomized to a telehealth family-centered weight management arm in which parents had high levels of engagement and satisfaction with a combination of interactive text messaging and telehealth video calls [23]. A systematic review indicated noninferiority in children's weight status improvement in telehealth versus in-person treatment delivery, with no difference in attrition rates and consistently high parental satisfaction with telemedicine [24].

Further, these findings add information that SafeCare providers report they are willing and interested in being trained in delivering weight management and believe their families would find this approach with digital tools acceptable. The integration of weight management into a previously existing structured parenting program provides an opportunity for large-scale and rapid dissemination. Families who receive services from SafeCare are often experiencing cumulative risk and have many needs, some of which are not directly related to abuse or neglect. Because SafeCare is broadly disseminated, training providers who already have a connection with these vulnerable families can be a vehicle for delivery of prevention programming that targets other public health issues a family may be experiencing. Should DRIVE prove beneficial, it could be offered as a module of additional services that families could receive. As detailed by the survey of SafeCare providers, there is a perceived need for services such as DRIVE, and SafeCare providers are willing to be trained to provide these services.

The pilot study observed a –0.14 reduction in BMIz (–0.18 among youth who were overweight or had obesity) with 8 counseling sessions delivered over a 13-week period. This reduction is greater than a prior 12-month study that observed –0.09 BMIz among children receiving both enhanced standard of care arm and individualized telehealth coaching (text messages 2x/week and telephone/video sessions every other month) [25] and greater than similar family-based weight management interventions according to a recent Cochrane review of interventions that lasted 6 months or longer [26]. However, the total contact hours did not meet the US Preventive Services Task Force recommendations of at least 26 hours to align with prior efficacious interventions [2], and the BMIz reduction did not meet previously suggested threshold of –0.25 for cardiometabolic improvement [27]. Importantly, in the pilot study, only 4 of the 10 children had obesity and an additional 4 were overweight, and it is not known if these children had cardiometabolic dysregulation. Future work should follow children over a longer time course to determine if BMIz reductions are sustained and accrue longer-term health benefits.

Increasing the dosage of telehealth weight counseling may increase weight loss. For example, a prior study of Kurbo, a commercially available weight management program delivered over a mobile app with video coaching sessions, showed that children who engaged in more telehealth coaching sessions over a longer duration had greater weight loss compared to those with less engagement [28], albeit the level of engagement was self-selected by the family and not randomly assigned. Similarly, a three-arm nonrandomized cohort study observed significantly reduced BMIz among children who opted into a multicomponent technology intervention that included family-based behavioral group treatment, a digital tablet with a fitness tracking app, and

individually tailored telehealth coaching sessions, compared to those who received only the group counseling or the group counseling with fitness app [29]. Programs must strike a balance between families' compliance/adherence to counseling sessions and expected weight reduction. The convenience of telehealth and digital tools may enable a sufficient amount of engagement that is both effective and acceptable to families.

Limitations

A limitation of these studies is the one-arm design of the pilot feasibility study without a control or comparator condition and the need for further verification in a larger randomized controlled trial. It is possible that BMIz fluctuations were influenced by maturation bias or regression to the mean [30], though the observed effect size was similar to prior pediatric weight management interventions [26]. Another limitation is the use of BMIz to examine change over time, as researchers have identified concerns with z-score for children with a BMI above the 97th percentile [31]. However, only one child in the sample had a BMI exceeding 97th percentile, so it was determined that this metric was appropriate.

Implications for Research and Practice

Our preliminary work demonstrates that DRIVE is an efficacious childhood weight management program capable of being delivered as a module within existing home-based programs, such as SafeCare, and adaptation of DRIVE to include mHealth would benefit both families and SafeCare providers. Families adhered to and were highly satisfied with the telehealth counseling sessions, the wireless scale and weight graph to track child weight, and the pedometer to track child physical activity. These findings are consistent with emerging research documenting that families are responding well to SafeCare delivery via technology, a delivery approach that was implemented as a result of the COVID-19 pandemic [8]. Integrating the intervention into a comprehensive smartphone app or website may enable a more seamless delivery system of both self-monitoring tools and ongoing remote interaction with the counselor. There are many future areas of investigation for mHealth DRIVE, including measuring the effects on weight-related behaviors including dietary intake and physical activity, examining specific feature utilization of the intervention components (such as sharing photos of food preparation with the counselor) and how this relates to the effectiveness of the intervention, and the extent to which the relationship with the counselor drives health outcomes in the family.

Digital tools may present an opportunity for a hybrid approach to blend in-person care with remotely delivered care, bridging the gap between counseling sessions by equipping parents and children with tools to continue their self-monitoring and assist them in implementing the health lessons into their daily lives. SafeCare providers overwhelmingly indicate the perceived need, and willingness to deliver, such a program. Our future work aims to test the feasibility and effectiveness of the mHealth DRIVE program over a longer term to manage children's weight and improve health-related parenting skills within the context of SafeCare's telehome visit delivery model. The ultimate goal is to package a turn-key weight management program for families of children with obesity, deployed using mHealth tools for wide-scale dissemination.

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Conflicts of Interest

Georgia State University and Pennington Biomedical Research Center and Louisiana State University have an interest in the intellectual property surrounding the curriculum of the DRIVE intervention, and JRS, JWA, and CKM are inventors of the intervention. Although not the focus of the current paper, Pennington Biomedical Research Center and Louisiana State University own the intellectual property surrounding SmartLoss, SmartIntake, and the Remote Food Photography Method, and an author (CM) is an inventor of the technology.

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Abbreviations

DRIVE: Developing Relationships that Include Values of Eating and Exercise

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Original Paper

Days Needed to Characterize the Healthfulness of a Typical Dinner Meal in Direct Observational Research: Mixed Methods Study

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Abstract

Background: Prior research around the home meal environment has demonstrated that family meals are associated with positive health outcomes for children and adolescents. Researchers have begun using direct observational methods to understand key aspects of family meals such as meal healthfulness and family meal frequency to explain the protective nature of family meals. Direct observational research, however, can be resource intensive and also burdensome for participants. Information about the number of days needed to sufficiently characterize typical meal healthfulness using direct observational research methods is needed.

Objective: The current study aimed to produce guidance about the number of meals necessary to approximate typical meal healthfulness at the family dinner meal occasion in a direct observational, mixed methods study of the home food environment.

Methods: Families were recruited between 2012-2013 from primary care clinics in the Minneapolis–St Paul metropolitan area (N=120). A total of 800 meals were collected as part of the Family Meals LIVE! mixed methods study. The Healthfulness of Meal Index was used to evaluate meal dietary healthfulness of foods served at 8 family meal occasions. Participating families were provided an iPad (Apple Inc) and asked to video-record 8 consecutive days of family dinner meals with a minimum of two weekend meals. After the meal, families completed a meal screener, which is a self-reported, open-ended measure of the foods served at the meal.

Results: Weekend and weekday meals differed in their measurement of meal healthfulness, indicating that at least one weekday and one weekend day are necessary to approximate meal healthfulness. Single-day measurement mischaracterized the strength of the relationship between the quality of what was served and intake by almost 50%, and 3 to 4 observation days were sufficient to characterize typical weekly meal healthfulness (r=0.94; P<.001).

Conclusions: Relatively few direct observational days of family meals data appear to be needed to approximate the healthfulness of meals across 1 week. Specifically, 1 weekday and 1 weekend observation are needed, including a total of 3 to 4 days of direct observational meal data. These findings may inform future direct observational study designs to reduce both research costs and participant burden in assessing features of the meal environment.

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KEYWORDS

meal healthfulness; direct observation; family meals; well-being; diet; food

Introduction

Having frequent family dinner meals has consistently been associated with a number of beneficial health outcomes for children, including reduced risk of being overweight [1-3] and healthy diet quality [4-10]. Additionally, quality of the emotional atmosphere [11,12] during family meals and quality of the food served during these meals [13] have been previously characterized as pathways that affect child weight and health outcomes. Direct observational research methods (ie, video recording) are becoming more common in family meals research because they overcome the reporting bias found in commonly used survey-based measures, allowing for a more in-depth and robust picture of the characteristics (eg, interpersonal interactions, meal healthfulness) of family meals that may contribute to child and adolescent health [11,14]. However, the impact of both the timing of the direct observational measurement and duration of the observational measurement period on estimates of meal healthfulness have not been examined.

In the current methodological study, the Healthfulness of Meal Index (HOM), implemented in the Family Meals LIVE! direct observational study [15], was used to answer the research question: how many days of direct observation of the foods served at family dinner meals are needed to characterize "typical" healthfulness of the meal to preserve resources and reduce participant burden? Family dinner meals were defined as an evening meal eaten in the home environment with the majority of family members present. The study further examined if weekends and weekdays influence meal healthfulness and at what number of days the addition of an observation day becomes unnecessary to characterize relationships with child dietary intake. We hypothesized that weekday and weekend day meal healthfulness estimates would differ due to changes in the home meal environment when children are not at school or when parents are not generally at work. We also hypothesized that estimates incorporating fewer days of observations would be weakly correlated with estimates derived from a full week of dinner meals. Results of the current study address a salient public health nutrition research need of providing pragmatic design guidance that could result in improved measurement.

Methods

Sample Population

Data collected from Family Meals LIVE! [15], a direct observational, mixed methods study, were used to measure the healthfulness of foods served at 8 meal occasions. The University of Minnesota's Institutional Review Board Human Subjects Committee approved the study protocol. Families (N=120) were recruited between 2012 and 2013 from 4 primary care clinics in the Minneapolis–St Paul metropolitan area that serve a racially/ethnically diverse, urban population of primarily low-income families. Participating families were provided an iPad (Apple Inc) and asked to video-record 8 consecutive days of family dinner meals with a minimum of two weekend meals. Only dinner meals in the home were recorded because of privacy issues. At the start of each meal, families spoke into the camera to indicate what foods were being served. After the meal, families completed a meal screener, which is a self-reported, open-ended measure of the foods served at the meal. Comprehensive study procedures have been described elsewhere [11,13].

In total, 800 meals were available for analysis [13]. Families were asked to record meal occasions over consecutive days, and recordings were taken every 1.8 days on average (SD 0.89), indicating good participant compliance with data collection procedures and minimal lack of family meals or meals outside of the home. A 1-day washout period was employed to allow families to acclimate to the study procedures and recording equipment.

Direct Observational Research

Previous studies have shown that direct observational research conducted in the home using unstructured observations (eg, play, routines) has more predictive validity and reliability compared to laboratory settings using structured observations (eg, tasks given to participants) and allows participants to acclimate and exhibit less reactivity [16-18]. The lengthened, 8-day observation window has been shown to offer advantages over cross-sectional designs, which include the measurement of weekday and weekend meals, the capture of variability in the healthfulness of weekly meals, and more reliable and objective measurement of family meal occasions [16-18].

Healthfulness of Meal Index

The HOM, created for the Family Meals Live! study and adapted from the Healthy Eating Index 2010 [19], was used to assess family meal healthfulness [13,15,20]. The HOM assesses 7 categories of foods served at meals: fruit, vegetables, dark green vegetables, dairy, protein, high sodium foods (reverse scored), and added sugars (reverse scored). A present-or-absent format is used to score the HOM, the components are summed, and a total of 9 points are available (the fruit and vegetable categories can each receive a total of 2 points). A higher total score is reflective of a more healthful family meal with regards to foods served. To calculate the HOM score, 3 research members (including 2 registered dietitians) watched each video-recorded meal to code the foods present [13]. The self-report meal screener was also used to corroborate the foods seen in the videos. Because the HOM evaluates meal dietary healthfulness, all foods present were coded even if they were not consumed by all family members.

Meal Healthfulness Permutation Measures

Permutations were constructed to evaluate study conditions (timing of measurement and duration of measurement period) that researchers implemented at the design stage of direct observational studies. First, a permutation was calculated to examine how adding observation days affects the HOM relative to a measure that incorporates all observation days. In all, 13 HOM permutations were calculated: a full-week index of average meal healthfulness (this was the primary reference permutation), 6 indices adding 1 additional day on the front end of the observation window (permutation 1: day 1 only; permutation 2: average of days 1 and 2; permutation 3: average of days 1 through 3; etc.), and 6 permutations adding 1

additional day beginning with the last observation day (measure 1: day 7 only; measure 2: average of days 7 and 6; measure 3: average of days 7, 6, and 5 etc). The primary reference permutation was computed assuming that capturing more dinner meals would reduce the random variation in the composition of foods that are served across days to obtain a measure of typical meal healthfulness. Relative to this comprehensive direct assessment of meal healthfulness, a measure containing fewer observation days that is highly correlated with the full measure may sufficiently characterize typical family meal healthfulness without excess resource investment.

Statistical Analysis

Survey estimation procedures were performed for each permutation of the HOM to determine whether the means differed by day of week, with sampling weights being applied to obtain population average meal healthfulness measures generalizable to the 4 clinics from which families were recruited. Effect consistency in the relationship between the HOM and dietary intake and family meal frequency were examined in sensitivity analyses to evaluate the presence of measurement error in permuted variables with a fewer number of observation days. A third correlational analysis was performed to evaluate the strength of the linear relationship between each HOM permutation. Comparisons between each reduced measure and the full reference measure were examined to determine how many days of additional meal recordings were needed to approximate the full reference measure. The intraclass correlation coefficient (ICC; 0.663) was calculated to evaluate consistency across the permutations within families. Pearson correlation coefficients above the ICC were used to visually evaluate at what points the permutations with fewer measurement days approximated the measure incorporating all days. All analysis and data management were performed in Stata 13.1 SE (StataCorp).

Results

The coefficient of variation for the single-day estimate of meal healthfulness was 39.3% (mean 3.3, SD 1.3) and declined to 27.2% as days were added to compute the full reference measure containing all observation days (mean 3.2, SD 0.9). Adding observation days increased the precision of the sample measure, and dispersion around the mean stabilized when 3 observation days were included. The full permutation was overall similar for weekend days (mean 3.1, SD1.4) compared with only weekday observations (mean 3.2, SD 0.9). The permutation variables (day 1 and day 7) which corresponded to measures that would be derived from a 1-day, cross-sectional study design indicated that weekend meal healthfulness was higher in one weekday contrast and less healthy in the other weekday contrast. An evaluation of the noncompliance pattern indicated that meal healthfulness became increasingly difficult to ascertain for more than five meals for the total sample. Specifically, 96.7% of the sample (116/120) provided enough meal recordings to calculate the 5-7–day meal healthfulness permutation, and 78.3% of the sample (94/120) provided a final meal (seventh meal) recording needed to calculate the final meal healthfulness permutation.

The relationships between quality of foods served, dietary intake, and frequency of family meals were examined. The dietary intake association was strongly attenuated when fewer observation days were used to estimate meal healthfulness (Table 1). Compared to the association observed when 4 days were used to compute meal healthfulness, the single-day measure of association was –48% weaker. By 4 days, the observed relationship between meal healthfulness and dietary intake was consistent with associations that included additional observation days. There was no evidence that the association between meal healthfulness and family meal frequency was strengthened or weakened according to how many meal healthfulness observation days were used. There was some evidence that inference would differ when adding observation days (ie, the statistical significance was not met at a *P* value of <.05).

Permutations of HOM were calculated by averaging the HOM scores calculated using 1 to 7 direct observation days. The bivariate associations between each permuted score and the Healthy Eating Index 2010 were examined. Increasing the number of direct observation days used to characterize the healthfulness of foods served (HOM) was positively correlated with healthy dietary intake of the participant child for all permutations (7-day permutation *P*=.001; Table 1). The magnitude of the associations grew as more observation days were included, and they remained similar after 3 or 4 observation days were added, suggesting that about 4 observation days may be sufficient to characterize how the healthfulness of food served at meals is related to child dietary intake.

Effect sizes expressed as correlation coefficient *r* were examined to evaluate the strength of the linear relationship between the permutations using fewer than 7 observation days and the permutation incorporating all observed meals over the observation period (Table 2). Results indicated that the linear relationship between measures (starting with a single day and adding additional days) grew stronger as more observation days were added. A second analysis (removing the first observation day until only the last observation day was used) indicated a consistent pattern. Meals occurring farther apart (ie, the day 1 permutation and the day 7 permutation, each of which use a single observation day), were weakly correlated (*r*=0.36), indicating meal healthfulness may vary across time. Permutations calculated from days closer together were strongly related (day 1 permutation and the permutation including both day 1 and day 2: *r*=0.80; permutation including day 6 and 7 and the day 7 permutation: *r*=0.82). The within-family ICC of all 13 permutations was moderate to strong (ICC 0.663), indicating moderate variation in family meal healthfulness. Four observation days sufficiently characterized the typical weekly meal healthfulness observed in the full measure (*r*=0.94).

Table 1. Association between the number of direct observation days in the healthfulness of meal index permutation and the Healthy Eating Index 2010 and weekly family meal frequency: (N=120) households (caregivers and children) recruited from Minneapolis–St Paul primary care clinics between 2012 and 2013.

Number of HOM[a] permutation observation days	Healthy Eating Index 2010		Weekly family meal frequency	
	Mean response (95% CI)	*P* value	Mean response (95% CI)	*P* value
1 day	1.4 (0.12 to 2.61)	*.03* [b]	0.3 (0.03 to 0.61)	*.03*
2 days	1.9 (0.39 to 3.39)	*.01*	0.4 (0.07 to 0.75)	*.02*
3 days	2.3 (0.74 to 3.77)	*.004*	0.3 (–0.04 to 0.66)	.08
4 days	2.6 (1.15 to 4.13)	*.001*	0.3 (–0.06 to 0.68)	.10
5 days	2.4 (0.86 to 3.84)	*.002*	0.2 (–0.11 to 0.58)	.18
6 days	2.5 (0.97 to 4.01)	*.002*	0.3 (–0.09 to 0.62)	.14
7 days	2.6 (1.05 to 4.07)	*.001*	0.3 (–0.04 to 0.68)	.08

[a]HOM: Healthfulness of Meal Index.

[b]Numbers in italics indicate significance at a *P* value <.05.

Table 2. Family meal healthfulness permutation measures with pairwise Pearson correlations. Correlation coefficients *r* are all significant at *P*<.001.

Permutation variable	Day 1, *r*	Days 1-2, *r*	Days 1-3, *r*	Days 1-4, *r*	Days 1-5, *r*	Days 1-6, *r*	All Days, *r*	Days 2-7, *r*	Days 3-7, *r*	Days 4-7, *r*	Days 5-7, *r*	Days 6-7, *r*	Day 7, *r*
Day 1	—[a]												
Days 1-2	0.80	—											
Days 1-3	0.70	0.90	—										
Days 1-4	0.64	0.83	0.93	—									
Days 1-5	0.60	0.78	0.90	0.97	—								
Days 1-6	0.57	0.76	0.89	0.95	0.98	—							
All days	0.57	0.76	0.88	0.94	0.96	0.98	—						
Days 2-7	0.37	0.63	0.80	0.88	0.92	0.95	0.97	—					
Days 3-7	0.34	0.48	0.70	0.81	0.87	0.90	0.93	0.96	—				
Days 4-7	0.34	0.46	0.56	0.72	0.80	0.84	0.88	0.90	0.94	—			
Days 5-7	0.33	0.48	0.59	0.62	0.73	0.80	0.86	0.88	0.89	0.92	—		
Days 6-7	0.31	0.46	0.52	0.52	0.52	0.65	0.73	0.74	0.74	0.76	0.86	—	
Day 7	0.36	0.43	0.43	0.44	0.45	0.45	0.61	0.61	0.61	0.67	0.72	0.82	—

[a]Not applicable.

Discussion

Principal Findings

Study results were consistent with our hypothesis that a fewer number of direct observation days would be sufficient to characterize typical weekly meal healthfulness. We also found evidence that including both weekday and weekend day family dinner meals differed in healthfulness across a week-long observation period. Single-day and 2-day observations of meal healthfulness may be inappropriate for generalizing about the healthfulness of foods served at dinner meal occasions over the course of a week. In addition, correlational analyses indicated that when using just 2 days of data, the fewer-day permutations were strongly correlated (r>0.70) with the full 7-day measure. This is in part because meal healthfulness was moderately to highly correlated within the family. Thus, it is not surprising that adding a fourth, fifth, and sixth day of observational data provided little additional information about the healthfulness of foods served. Using 3- or 4-day observations of family meal healthfulness appeared to maximize measurement reliability and to minimize the cost of data collection and respondent burden.

Study Limitations and Strengths

The study had several strengths, including the use of direct observational methods, consecutive observation of family meals, and a substantial number of meals (N=800) observed. Practical advantages are also noted, such as assessing measurement variability, providing new information about how to allocate

staff time, and minimizing respondent burden. Replication studies are needed to provide support for the finding that relatively few observation days (ie, 1 weekend day and 1 weekday) are required, with the ideal number of days possibly being as few as 4; to test findings in a population with heterogeneous characteristics; and to assess meal healthfulness in multiple ways to avoid social desirability bias, recall error, and participant reactivity.

Conclusions

Findings from the current study suggest that relatively few direct observational days of family meals data are needed to approximate the healthfulness of meals across 1 week. Specifically, 1 weekday and 1 weekend observation at a minimum, along with 3-4 days of direct observational data, are needed. Findings from the current study may inform future direct observational study designs to reduce both research costs and participant burden.

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Conflicts of Interest

None declared.

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Abbreviations

HOM: Healthfulness of Meal Index
ICC: intraclass correlation coefficient

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Original Paper

Understanding Parents' Experiences When Caring for a Child With Functional Constipation: Interpretive Description Study

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Abstract

Background: Pediatric functional constipation (FC) is a common but serious medical condition. Despite significant effects on children, families, and the health care system, the condition is typically undertreated. Parents carry the primary responsibility for complex treatment programs; therefore, understanding their experiences and needs may offer a critical perspective toward improving clinical care.

Objective: The aim of this study is to understand and give voice to parents' experiences and information needs when caring for a child with FC. The ultimate objective is to build an evidence base suitable for creating a digital knowledge translation tool to better support parents caring for a child with FC.

Methods: This qualitative design used an interpretive description methodology to generate findings aimed at improving clinical care. One-on-one, in-depth interviews were completed either in person or through web-based teleconferencing to explore parents' perspectives. Data collection and analysis occurred concurrently.

Results: Analysis of 16 interviews generated 4 major themes: *living in the shadows*; *not taken seriously*, with a subtheme of *persevering and advocating*; *missing information and misinformation*; and *self-doubt and strained relationships*. One minor theme of *affirmative influences that foster resilience and hope* was identified.

Conclusions: Parents have unmet needs for support and information related to pediatric FC. To address gaps in current care provision, decision makers may consider interventions for clinicians, resources for parents, and shifting care models to better meet parents' needs.

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KEYWORDS

constipation; child; parents; caregivers; qualitative research

Introduction

Background

Constipation among children is common and often mistaken for a mundane nuisance rather than a serious medical condition. More than 95% of pediatric constipation cases are attributed to functional constipation (FC), which occurs without a particular medical, genetic, anatomic, or physiologic cause. Estimates are that at least 1 in 10 children worldwide is affected by pediatric FC [1,2]. FC can present with severe symptoms such as recurrent abdominal pain, painful defecation, fecal incontinence, urinary incontinence, and urinary infections. Pain, toilet avoidance, and stool withholding behaviors worsen the condition by further perpetuating fear of defecation, causing colonic dilation, and

dampening neural feedback about the need to defecate. Despite being very common, pediatric FC is often underrecognized and undertreated [3]. Without effective treatment, most children develop chronic FC, with symptoms continuing through their adult years [4]. In addition, children and families experience psychological, emotional, and social consequences of FC [5-7]. For example, school attendance and peer relationships are understandably compromised by pain and incontinence. Families also report high levels of stress and decreased quality of life [5-7]. Finally, pediatric FC is a financial burden on families and health care systems [8]. Families face inflated expenses such as medications, laundry, and clothing, in addition to indirect effects such as lost income because of caregiving. Similarly, health care systems are burdened with preventable urgent care visits and high usage rates of specialist services [8,9].

Clinical practice guidelines (CPGs) describe a variety of treatment options [10-14]; however, the bulk of responsibility for implementing, monitoring, and adjusting therapies falls to parents. Certainly, clinicians can provide parents with accurate information about the condition and treatments, but improving care also requires that health care professionals move beyond their own perspective of the condition and acknowledge the unique experiences of families living with a child affected by FC. Specifically, parental experiences critically shape their information and support needs [15]. Therefore, an in-depth understanding of parents' experiences and self-identified needs when caring for a child with FC is a necessary step to ensure that clinicians can provide *relevant* education and support. Although parental education is an important part of treatment for pediatric FC [10-12], there is a lack of research about parental perspectives of pediatric FC. A recent systematic review on the topic included only 13 studies examining parents' experiences caring for a child with FC [16]. The primary cited limitation of the review was the small number of included studies [16]. Furthermore, there was a predominance of quantitative studies that focused on quality of life measures, which are helpful in substantiating the familial effects of childhood FC but are not optimal in understanding how health care providers can help mitigate negative experiences and outcomes [16]. Suggestions for future research include a more in-depth exploration of how to best meet parents' information and support needs in light of the dynamic nature of the condition and its profound effects on families [16].

Objectives

The initial aim of this study is to understand and give voice to parents' experiences and information needs when caring for a child with FC. The ultimate objective is to build an evidence base suitable for creating a digital knowledge translation (KT) tool to better support parents caring for a child with FC.

Methods

Design

The study sought to answer the research question: *What are parents' experiences and information needs when caring for a child with FC?* Because our ultimate objective was to develop knowledge that could be used to inform and improve clinical practice, we chose the interpretive description (ID) methodology [17] to foster the applicability of our results. ID methodology was developed specifically for practice-oriented sciences, to generate findings aimed at improving clinical care [17], which aligns with our pragmatic philosophical approach for this research project.

Recruitment

Potential participants were introduced to the study through social media posts shared on child health and parenting groups (eg, Facebook, Twitter). Physical posters were also displayed in locations frequented by families (sports facilities, libraries, health care waiting rooms, etc) in a medium-sized city in Canada. The posts described the purpose of this study and the desire to speak with the parents of children with FC. In addition, we engaged in snowball sampling by asking participants whether they knew other parents who may be interested in contributing to this study. Recruitment was active from May 2019 until data collection was complete in October 2019.

Ethical Considerations

Ethical approval from the relevant research ethics board was granted before the initiation of the study. Each potential participant received an information sheet, which provided details on the purpose of the study, identified the potential risks and benefits, and explained the voluntary nature of their participation. Participants were given an opportunity to ask questions about the research and were free to withhold consent for any reason.

All procedures were in accordance with the ethical standards of the University of Alberta Research Ethics Office (Pro00087548) and the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Data Collection Methods

We used one-on-one, in-depth interviews to explore parents' experiences when caring for a child with FC. The interviews were completed either in person or through web-based teleconferencing, depending on the participant's preference and geographic location. The interviewer (AT) had experience conducting qualitative interviews and providing care as a clinician for children with FC. The interviewer did not have any pre-existing personal or professional relationships with the participants. The interviewer spoke with the participants at the beginning of the interview to discuss the reasons for conducting this research (to understand parental experiences and subsequently develop resources for parents) and to share the interviewer's relevant clinical background—caring for families affected by pediatric FC and noting the challenges they often encountered in managing the condition. The interview style was conversational, and the participants were encouraged to discuss aspects of their experiences they deemed most important. The interviewer also used a semistructured guide (Multimedia Appendix 1) with open-ended questions. Interview questions were developed based on previous research [18-20] and clinical experience of the team. Prompts and spontaneous questions were used to facilitate participant comfort and collection of high-quality data. Interviews were recorded and transcribed verbatim by a professional transcriptionist. Data were

deidentified (ie, removal of identifying data such as city names, people names, institution names) to ensure confidentiality.

Sample

Participants were included if their child met diagnostic criteria for pediatric FC (Multimedia Appendix 2) and were willing to discuss their experiences with the interviewer. Screening was conducted by the interviewer as a preamble to the interview to ensure that participants' stories reflected experiences of childhood FC rather than other conditions. As recruitment was most successful through web-based platforms, participants came from diverse geographical locations across North America.

On the basis of existing literature examining parental perspectives of pediatric FC and methodological recommendations, we anticipated that a sample size between 10 and 20 participants would be adequate to generate clinically significant knowledge [17,21]. The decision to end data collection was an ongoing topic of discussion within the research team and based on the processes of data analysis. Specifically, the occurrence of redundancy within the themes and rich substantiation suggested that data collection could be stopped.

Data Analysis

We followed guidance from the applied methodology of ID [17] throughout data collection and analysis. We conducted data collection and analysis concurrently to promote data immersion as an important step toward a more thorough interpretation of experiences [17]. Interview transcripts were exported into NVivo 12 software to manage the data. Our analytic approach avoided quantification, instead of using thematic and inductive traditions [22,23]. Our analysis followed the processes of engaging with the data, organizing the data, finding patterns within the data, making sense of the patterns, and finally, developing patterns and associations into meaningful findings for applied practice [17]. The process was initiated by the first author, who also conducted the interviews, and then was verified by the author team. Reflexive journaling and field notes were used during data collection and analysis to examine potential bias, build an audit trail, and support rigor.

Rigor

Developers of ID emphasize that the clinical expertise of researchers strengthens the design and rigor of the research [17,24]; therefore, the experiences of clinicians on our research team were seen as a benefit. One member of the research team conducted all the interviews to maintain consistency. The interview guide was reviewed by topic experts and a parent advisory group to enhance credibility and ensure that the questions could elicit meaningful information from participants. A study log was maintained during the research to document and account for methodological decisions. Data were analyzed and findings were collaboratively critiqued by the research team with the intent to develop epistemological integrity, representative credibility, analytic logic, and interpretive authority [17] to ensure high-quality research. Following ID guidance, we did not conduct member checking because of the risks of swaying interpretation and impeding the formation of meaningful clinical implications [17,25]. The study followed the Standards for Reporting Qualitative Research (SRQR) [26] (Multimedia Appendix 3).

Results

Overview

A total of 16 parents of children with FC provided informed consent and participated in this study. Our analysis generated 4 major themes: (1) *living in the shadows*; (2) *not taken seriously*, with a subtheme of (i) *persevering and advocating*; (3) *missing information and misinformation*; and (4) *self-doubt and strained relationships*. We identified one minor theme of *affirmative influences that foster resilience and hope*. The demographic details of the participants are presented in Table 1. All the participants in this study self-identified as caregivers with primary responsibility for managing FC. One of the parents interviewed had more than one child with FC. Participant interviews were randomly assigned a numerical code that was used as a reference marker (eg, P3) for quotes presented to support the themes in our results.

Table 1. Participant characteristics (N=16).

Characteristics	Participants, n (%)
Preferred gender identity	
Female	16 (100)
Number of children	
1	4 (25)
2	8 (50)
3	2 (13)
4 or more	2 (13)
Affected child's age (years)	
3	1 (6)
4	4 (25)
5	4 (25)
6	5 (31)
7	0 (0)
8	0 (0)
9 or older	2 (13)
Education level	
High school	1 (6)
Postsecondary	15 (94)
Yearly family income in Can $ (Can $1.00=US $0.78)	
<20,000 (15,600)	1 (6)
20,000-40,000 (15,600–31,200)	1 (6)
40,000-60,000 (31,200-46,800)	4 (25)
60,000-80,000 (46,800-62,400)	2 (13)
>80,000 (62,400)	8 (50)
Duration of symptoms (years)	
<1	1 (6)
1-2	3 (19)
>2	12 (75)
Number of constipation-related health care visits (total)	
0-5	4 (25)
6-10	6 (38)
More than 10	6 (38)

Living in the Shadows

Parents in this study expressed strong feelings of isolation attributed to living with a condition that is considered taboo. Discussing bowel habits and incontinence was thought to be a difficult or inappropriate topic in social circles and within the health care context. For example, when parents themselves were open to the conversation, most had experienced or anticipated negative reactions from others. One parent related her sense of isolation, "Nobody talks about it…. So, you feel alone… And nobody wants to talk about poop" (P3). Similarly, another parent explained:

> *I think, that for myself...because I don't know a lot of other parents that are – I don't know if people just don't talk about it, so I don't know how common it is.* [P4]

To combat feelings of isolation, parents typically searched for resources without success to meet their social support needs. Parents were surprised about the lack of discussion groups because many described how it seems there is an online forum for almost every rare disease or condition:

> *Something...so you're not alone, right. Because that's the thing and you don't understand why your kid is having so many problems. It's like somebody or*

> *something that explains like oh my kids have this issue, so you don't feel like you're the only one...Just something you can go to whether it's like a chat group or a parent group or something.* [P5]

Another parent described how she would change things to improve other families' experiences with pediatric FC:

> *You know, I think it's one of those things that people could really benefit from a support group because it's something that's so like people don't wanna talk about, they're embarrassed about it.* [P9]

Another parent simply expressed, "I just feel like we were very much left on our own" (P14).

Not Taken Seriously

Parents shared stories of encounters with health care professionals who did not take their concerns about constipation seriously. In some cases, parents were explicitly told that the symptoms were nothing to be concerned about, and in other cases, parents were implicitly given the impression that they were overreacting. One parent shared her care provider's dismissive response to her child's symptoms:

> *I was always told it would pass, it would pass. Probably listen to the patient a little bit better because they know their body, right, and I – me living with her, I know what's going on with her. So, listen a little bit closer and maybe have better options than prune juice.* [P13]

Similarly, another parent said:

> *I wish I had been taken seriously right away. You know, not just like she'll grow out of it, she'll grow out of it. It's normal, she'll grow out of it. It's like this wasn't. I don't know if it ever was.* [P9]

One shared the widespread effects of her child's FC and the trivializing response:

> *I get that pediatricians are really busy with other things that are, you know, more important than constipation, but like now that he's in school, it's affecting his whole class. It's affecting his teacher. It's affecting him and his friends. Like it affects a lot of things and it affects us daily. It takes up our time as parents and his time away from his activities and the only real thing that we hear is, oh don't worry, it'll end soon. Like how?* [14]

One parent reflected on her desire for health care providers to change*:*

> *I guess I wish they would learn – they would take it a bit more seriously and understand how it impacts lives and how it impacts – I mean children's lives.* [P7]

Parallel to instances of health care providers not taking the condition seriously, parents themselves described periods of questioning the legitimacy or validity of their own concerns. For example, one parent shared:

> *I think we could have maybe helped him a lot sooner if I wasn't so scared to start the Lax-A-Day but I also didn't want to make an appointment, take someone else's doctor time...I hate wasting doctors time on what I consider a silly thing...I know it's not the right way to think of it but like to my point, it had to be urgent enough.* [P3]

Similarly, another parent said, "You're like, oh is that normal or not normal and you kind of doubt yourself" (P2).

Persevering and Advocating

As a result of symptoms and concerns not being taken seriously, parents demonstrated perseverance and became stronger advocates for their child's health. One parent described her feelings about health care encounters:

> *I had talked to my doctor about it. Like our doctor and the doctor said like, oh you know, she's still really young. She'll grow out of it, all that kind of stuff...eventually after lots of kind of like advocating, I ended up – I was like I need another opinion on this.* [P9]

Similarly, another parent stated:

> *We found that we've gone to the doctor a couple of times now and they haven't been super helpful...and then we wound up back at the doctor because we're still – she's still having accidents.* [P16]

Parents returned to health care providers repeatedly and asked for referrals to other providers because their child's condition was worsening without adequate treatment. For example, "I'd asked many times for her to be seen by somebody else just because I need this figured out" (P13). Parental frustration frequently became the catalyst for advocacy. One parent expressed:

> *They don't take it serious enough...it would just be nice if there was a doctor that would take you a little more serious. I know lots of kids have it and I get that, but when they get to be older and it's a school issue, I think like we push. I think we asked – my doctor was out of town so we asked the stand in and then we asked the walk-in clinic and then we asked my doctor.* [P5]

Missing Information and Misinformation

Parents caring for a child with FC frequently have unanswered questions about the condition, causes, symptoms, prognosis, and treatment. One parent said:

> *Maybe I wouldn't have been so upset about it or, you know, it wouldn't have been such an overly concern for me if I'd had a little bit more information.* [P2]

Similarly, another parent explained the lack of teaching provided about pediatric FC:

> *I'm saying like you go into the doctor and you're like this is an issue and they don't give you...like there's nothing, they give you nothing. My doctor was just very much like, oh it's super common and...like not giving you any further advice or resources.* [P6]

Parents frequently questioned whether there was an underlying medical cause for constipation. For example, one parent stated:

> *Maybe something else medically. Like maybe she's lactose intolerant – we thought well maybe there's some issues with milk or dairy which, of course, would not be constipation...but we were convinced it was something she was eating. Maybe it was gluten, maybe it was this, maybe it was that.* [P1]

Episodes of incontinence often cause parents to question the underlying reason. One parent wondered, "I don't know if it's medical or constipation or is it just laziness?" (P5). Similarly, another parent stated:

> *We had no idea whether she actually like did she have control, did she not have control. Could she feel it, could she not feel it? Was she just ignoring it? Did she need to pay more attention? Like all of these huge question marks.* [P9]

Questions about the treatment for pediatric FC were also common. A parent shared concerns about medication use:

> *You read the Lax-A-Day thing it says, "Adults only", blah, blah, blah. So, I'm like 'Are you sure?' Like it feels wrong...But then, again we're trying to cut back now on the Lax-A-Day because you can't be on Lax-A-Day forever, can he? Like I don't know.* [P3]

In addition to having questions about pediatric FC, parents shared instances of misinformation that was detrimental to their child's care. As explored above in the theme of not being taken seriously, parents were often incorrectly told that the condition would resolve on its own. One parent shared the common false reassurances she received:

> *It was very much like, no, no, no, he's fine. And it's just constipation and he'll grow out of it and like I feel like everybody I talked to said, he'll grow out of it. He'll grow out of it. He'll grow out it. And now, two years later, he's not growing out of it.* [P14]

Parents were also commonly given misinformation about dietary changes as treatment. "We were just told to increase fibre, increase water, skip the junk food, but we eat all whole foods anyways" (P4). Similarly, another parent shared, "The doctor said, it'll get better. You know, just make sure she's eating healthy, which she does, and it'll get better. It'll get better" (P16). Dietary misinformation was problematic because it was ineffective, difficult for families to manage, and delayed further treatment:

> *The nurse said don't give her any dairy. And so, we were off dairy for a while and then we were off wheat for a while and it was just like a – none, none of that seemed to make much difference.* [P9]

Similarly, another parent reported, "Cut [cheese] out and try to increase the fruits, the vegetables, take away the bread. It was like a constant diet struggle" (P3).

Within this theme, there was one divergent case of a parent who conveyed confidence and felt that they had adequate knowledge about caregiving for a child with FC. The case had minimal health care encounters because the parent felt further support or intervention was not required. Unfortunately, the parent's knowledge was inferred from personal experience with medical care of an unrelated population and condition, which does not align with current evidence for pediatric FC. Thus, although the participant expressed a divergent view of her experience, the data further substantiated the theme of missing information and misinformation.

Self-Doubt and Strained Relationships

Perhaps the most resounding theme from parents' stories was the overarching sense of frustration that developed while caring for a child with FC. One parent shared the emotional fragility that pediatric FC had created for her as a parent:

> *It's pretty terrible actually. Like I should know how to deal with this. I'm a nurse. Like I was a pediatric nurse. (crying). I should know and everything that I've tried didn't work and I didn't have any guidance or any help. Like I called the doctor, well it's you know, the pediatrician – it's six months to get into her, so I, you know. I'm just trying things on my own. I'm googling how do you deal with this and you know, information and none of it is working and it makes me feel like – I don't know. Like I should know how to do this, and I don't.* [P14]

Self-doubt and conflict were strongly tied to the previous themes of living in the shadows, not being taken seriously, and missing information and misinformation. One parent clearly expressed the situation stating:

> *It was just like extremely frustrating because I felt like I wasn't getting – I wasn't getting enough support or information from the medical – like the health professionals we were dealing with...Like it's so frustrating. I'm like if this is so common, why does no one have answers? – it's just so, so frustrating.* [P9]

Symptoms and physiology of pediatric FC were further sources of emotional turmoil for parents:

> *We are very frustrated and, again, the accidents, I don't know if it's because of this issue or because she's lazy or like because she's so constipated...it's the accidents that are driving us crazy.* [P5]

Another parent explained:

> *We'll tell him fifteen times to go to the bathroom and he won't and then he'll have an accident and you feel like – you just get to your boiling point sometimes and you don't want to yell and get angry, but sometimes you do.* [P14]

Finally, relationships frequently became strained as a result of pediatric FC:

> *It impacts a whole family dynamic, you know. Like our world, it seems like I mean this might sound dramatic, but our world has literally revolved around her bathroom habits for the last three years.* [P16]

Another parent expressed the strain related to behavioral interventions, "Like it's always a fight to get her on the toilet" (P6). Another parent stated, "There's been lots of fights. Lots of fights. Lots of I hate yous" (P10). Emotional burden related

to pediatric FC also sparked conflict between parents and eroded parental self-efficacy:

> *We're both feeling – neither one of us are confident in our parenting. So, we're frustrated, and we can argue about it, for sure...I really felt like a failure as a mom. (pause). I don't know and I still don't know what to do. I don't feel like we're making progress and I don't feel like I have the confidence to fix it. And then I feel like that kind of –permeates, I guess, into our whole situation. Like into everything. Like if I can't figure out constipation, how can I figure out big things?* [P14]

Affirmative Influences Foster Resilience and Hope

Despite the predominantly despondent themes that were reflected in parents' stories, there were small but significant moments of affirmation that helped bolster parents' confidence. This is a minor theme of our analysis because the occurrence of positive encounters and resources was unfortunately infrequent. After episodes of misinformation, accurate and understandable explanations of the condition and symptoms were critically important for parents:

> *They explained the encopresis is like the fact that like you know, when she did get constipated, the accidents would just be like the new poop coming around the old stuff that's not coming out...it's just like your muscles are just weak because like they've been holding it for so long. Yeah, and I was just like – at first, it just kinda blew my mind and I'm like, why the hell has no one told me about this?* [P9]

Validation came from a variety of sources and was always highlighted as an important event within the caregiving experience. For example, one parent found support through the school system:

> *And it was really just brushed off and it's still being brushed off until like finally – now that he's taking up so much time from his teacher, the principal has become involved and she has been our only real advocate and our only – like the principal of the school. Like she's not a health care provider. You know, like she's the only person that has really like tried to help at all.* [P14]

Parents identified encounters that met their support and informational needs as turning points that rekindled hope and buoyed their confidence. Unfortunately, affirmative influences were meaningful but scarce in parents' experiences. Specifically, many parents did not relate any positive encounters or support at all throughout their caregiving journey. One parent explained:

> *I told them this has been an ongoing issue. This isn't getting any better. This isn't an issue we've had for six months. This is an issue we've had for over three years now.* [P16]

Discussion

Principal Findings

Findings from our exploration of parents' experiences with pediatric FC parallel and expand upon results from previous research in the field. In a 2003 study, researchers examined parents' health care encounters related to childhood constipation and found similar themes of "dismissed and fobbed off, asserting the need for action, and validation and acknowledgment" [21]. The continuity of these findings with ours suggests that parents' perceptions of encounters with health care providers related to pediatric FC have not improved significantly over the last 17 years. Despite the widespread prevalence of the condition [1,2] and advances in understanding childhood FC [3,4], parents' concerns continue to be minimized and clinicians' treatment discussions lag behind or are incongruent with symptom severity. In other words, when health care providers acknowledge that pediatric FC requires treatment (which in itself may occur belatedly, if at all), the level of intervention is often inadequate for the advanced nature of symptoms described by parents.

Similar to exploring patient and family experiences, measuring quality of life is considered an important way to understand the effects of a health condition or treatment on "patients' lives, rather than just on their bodies" [27]. Numerous studies have highlighted the diminished quality of life of parents and families living with pediatric FC [28-32]. For example, 3 studies found that increased family conflict, impaired family functioning, and increased parental worry or stress were related to the presence of fecal incontinence [29,30,32]. Furthermore, Wang et al [31] found that the caregivers of children with FC gave lower ratings of their daily activities and family relationships, in addition to reporting lower physical, emotional, social, cognitive, and communication scores compared with those of the caregivers and families with healthy children. Although quality of life data provide a broad assessment of the effects of a health condition and are a central contribution to the field, qualitative methods are helpful in adding important context by exploring why and how families are affected. In this study, parental perspectives provide insights into the significant physical, emotional, and psychological burden on caregivers. Parents' feelings of isolation and frustration were related to incontinence and further compounded by nonsupportive interactions and misinformation. Parents' experiences of being told erroneously that pediatric FC would resolve, feeling blamed for the condition or lack of treatment success, and struggling to talk about the condition may help explain the widespread and profound impairments in quality of life for families affected by pediatric FC [28-32].

A 2019 study examining the prevalence of defecation disorders in children concluded that childhood constipation is likely underestimated by parents who may not consider symptoms sufficient to be labeled a medical condition [2]. The findings seem to be in contrast to our data, which found that parents were more frequently dismissed by health care providers rather than they being dismissive of the child's symptoms. One potential explanation for this difference could be the relative disease severity of the parents surveyed in the 2 studies. Specifically,

the cross-sectional study included a random selection of parents from the general population and was, therefore, more likely to include parents with early or mild manifestations compared with parents included in this study whose children all met full diagnostic criteria for pediatric FC. The findings from this study offer a relevant counterpoint, meaning that although parents and families may underestimate early symptoms, once the magnitude of the condition becomes evident, health care providers may be more of a barrier to recognition and diagnosis than parents.

Clinical Implications

Our exploration of parents' experiences of caring for a child with FC provides important insights toward improving clinical care for this difficult condition. CPGs, which are intended to support clinicians and optimize care, identify family education about pediatric FC as a key component of treatment [10-12]. Unfortunately, our results suggest that this step is commonly missing in health care encounters and that some providers even contribute to misinformation. As our data were focused on parental perspectives, we cannot report the reasons for CPG deviations. Given the time-consuming nature of consultations to provide emotional support and education, it is possible that care providers may be tempted to defer, rush through, or simply struggle to fit these practices into already-busy schedules. On the basis of parents' reluctance to initiate discussions about bowel concerns, it may be prudent for professionals to recognize that effects may be more severe and have persisted for a significant duration by the time these issues are brought to their attention. In contrast to the temptation to offer hasty reassurance, clinicians may need to reframe their thinking toward acknowledgment, education, and active treatment. For example, explaining that the condition is common can be a method of validating parents' concerns and mitigating parental feelings of guilt but should not be conflated by suggesting that the symptoms are normal or do not require treatment. Improving the quality of health care encounters may require education or interventions to improve the responses and treatment knowledge of health care providers. Similar to findings from a previous study about medication adherence [33], parents commonly expressed a lack of information about medication use; therefore, discussions about dosing, duration of use, side effects, and safety are likely to be well received by parents. Finally, clinicians should be attuned to inquiring about parental experiences of isolation and lack of social support during assessment and include these factors as part of treatment plans [10-12]. In addition to the existing system constraints that disincentivize lengthy consultations, it is unlikely that specialty care providers or primary care clinicians alone can adequately meet complex parental needs. Consideration of alternative care models, such as integration of nursing and allied health members, may be helpful to more accurately and consistently meet parents' support needs when caring for a child with FC [34-36].

Future Steps

The results of this study are an important foundation for creating resources that directly address parents' experiences and self-identified needs when caring for a child with FC. Developing support such as digital KT tools that target parents' information needs may improve families' experiences of living with pediatric FC. For example, parents seek answers to concrete questions about medication dosing, titration, side effects, safety, and long-term use. Sharing information with parents about digestive physiology, including how constipation can contribute to fecal incontinence, may be helpful in empowering parents' caregiving when faced with the uncertainty and frustration that arise from a child's stool accidents. In addition, the emotional toll of pediatric FC on families was often underacknowledged, wherein parents' caregiving abilities were hindered because of self-doubt and guilt. Creating resources that validate parental concerns and experiences can be an important contribution to meeting the support needs of parents caring for a child with FC. Finally, in light of our findings related to health care providers, future research exploring health care professionals' knowledge of pediatric FC and their experiences working with affected families can clarify the challenges and barriers to improving care provision for this condition.

Limitations

Although the recruitment was open to all parents, we only received interest from mothers. The interviewer asked whether any other caregivers from each family would be interested in sharing their perspective; however, we did not successfully recruit any further participants. Therefore, our results may not reflect the experiences of fathers and nonprimary caregivers. Parents who shared their story for this study were typically from higher education and income levels; therefore, experiences of parents with lower levels of education or income may not be adequately captured in our findings. In addition, the sample may reflect bias because of the self-selection nature of the recruitment process.

Conclusions

Understanding parents' experiences when caring for a child with FC is an important and often overlooked step toward improving care for this difficult condition. Our findings indicate that parents have significant unmet needs for support and information related to pediatric FC. To address gaps in current care provision, decision makers may consider interventions for clinicians, resources for parents, and shifting care models to better meet parents' needs.

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Authors' Contributions

AT, SM, EW, and SS conceived and designed this study. AT was responsible for data acquisition. All authors contributed to the analysis of the data. AT drafted the preliminary version of the manuscript. SM, EW, and SS critically revised the work for important intellectual content. All authors approved the final manuscript and agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. SS obtained the research funds through which this research was conducted.

Conflicts of Interest

None declared.

Multimedia Appendix 1
Draft interview guide.
[PDF File (Adobe PDF File), 76 KB - pediatrics_v4i1e24851_app1.pdf]

Multimedia Appendix 2
ROME IV diagnostic criteria for pediatric functional constipation.
[PDF File (Adobe PDF File), 14 KB - pediatrics_v4i1e24851_app2.pdf]

Multimedia Appendix 3
Standards for Reporting Qualitative Research (SRQR) checklist.
[PDF File (Adobe PDF File), 158 KB - pediatrics_v4i1e24851_app3.pdf]

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Abbreviations

CPG: clinical practice guidelines
FC: functional constipation
ID: interpretive description
KT: knowledge translation

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Original Paper

A Theory of Change for Web-Based Therapy and Support Services for Children and Young People: Collaborative Qualitative Exploration

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Abstract

Background: Web-based counseling and support has become increasingly commonplace for children and young people (CYP). Currently, there is limited research that focuses on the mechanisms of change within complex telepsychology platforms, a factor that makes designing and implementing outcome measures challenging.

Objective: This project aims to articulate a theory of change (ToC) for Kooth, a web-based therapy and support platform for CYP.

Methods: A collaborative qualitative research design involving professional staff, academic partners, and young people was used to develop the ToC. The following three major reflective phases were engaged: a scoping workshop involving professional staff and academic partners, a series of explorative projects were completed to inform the development of the ToC, and the draft ToC was reviewed for coherence by key stakeholders (young people, online professionals, and service managers).

Results: A collaboratively developed ToC was presented. This was divided into the conditions that lead to individuals wanting to access web-based therapy and support (eg, individuals wanting support there and then or quickly), the mode of service delivery (eg, skilled and experienced professionals able to build empathetic relationships with CYP), and the observed and reported changes that occur as a consequence of using the service (eg, individuals being better able to manage current and future situations).

Conclusions: Developing the ToC helps to shed light on how web-based therapy and support services aid the mental health and well-being of CYP. Furthermore, it helps to understand the development of *positive virtual ecosystems* and can be used to devise evaluative tools for CYP telepsychology providers.

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KEYWORDS

telepsychology; digital mental health; online therapy; young people; Kooth; Theory of Change; positive virtual ecosystems

Introduction

This paper reports a study conducted as a collaboration between professionals working for Kooth and academic researchers working within the United Kingdom. Kooth is a web-based counseling and support service for young people and young adults (aged 11-25 years). It is anonymous at the point of access, with young users typically finding out about the service through educational providers and only having to provide limited information on registration. In 2019, it received approximately 1700 log-ins per day, a figure that increased to just over 3000 log-ins on the day in which the COVID-19 pandemic caused schools to be shut down and the government to restrict movement of the UK population (March 20, 2020). Of these

log-ins, just under 1000 were a result of new registrations, which represented an increase of over 50% in the trend for registrations earlier in that week.

During the evolution of the Kooth service, it became clear that existing tools for measuring outcomes did not prove fit for purpose, with both the format of the tools and the concepts that they measure proving to be problematic [1]. In this regard, further scrutiny of the work that was undertaken on the website proved to be necessary. As a first step, it was considered necessary to review the work of the service as a whole and develop a theory of change (ToC) so as to consider what might be the most appropriate means of evaluating the work that the service engages in.

Reflexive Statement by the Authors

Before continuing, it is important to note that most of the team members contributing to this paper have been involved in the development of web-based therapeutic services for several years. As such, they view developments in this arena as both necessary and inevitable. Three of the team members have actively researched web-based therapeutic provision (TH, JP, and AS), and 2 of the team members have been involved in the development of such services (TH and AS). One team member provides consultancy related to the development of theories of change (JG), and one team member is a trainee counseling psychologist with an interest in developing accessible therapeutic services (AE).

Therapeutic Provision for Children and Young People

There is a global acknowledgment that children and young people (CYP) would benefit from additional support to improve their mental health and well-being [2,3]. In the United Kingdom, where this study has been completed, research indicates that as many as 1 in 8 young people will experience mental health issues between the ages of 5 and 19 years [4]. Furthermore, it is believed that a large number of young people belonging to this group will continue to experience mental health difficulties as they become adults, thus impacting wider issues such as individuals' future employment [3]. Although there is evidence to suggest that CYP can benefit from therapeutic interventions provided by a wide variety of services [5], access to support varies greatly depending on the geographical location.

In the United Kingdom, there is a wide range of ways in which young people might access psychological support. These include statutory health services in the form of child and adolescent mental health services (CAMHS), school-based counseling and psychology services, community-based services, and mediated services (web-based and telephone) [6-8]. Recent statistics indicate that the average wait time for statutory support is 56 days, with the shortest time being 49 days and the longest being 65 days [9]. Furthermore, the waiting times assume that young people will have their referral accepted to CAMHS. Owing to the emphasis on meeting specific service criteria, approximately three-quarters of young people with a diagnosable mental health condition will have their referral rejected. Overall, this is reflective of the disparities between adult and young people's mental health provision, with it being estimated that, despite making up 20% of the population, only 10% of the mental health budget is spent on CYP [10]. The limited resources available are also the main reason that additional NGOs have developed services to fill the gaps within this provision.

Young People, Mental Health and Well-being, and the Internet

Young people have been described as *digital natives* [11], notably individuals who have never lived in a world without the internet. Consequently, within countries that have widespread access to the internet, young people and young adults are now viewed as the biggest users of the internet and social media [12]. Furthermore, research examining help-seeking behaviors of this group suggests that they use the internet as their first point of call for support for issues related to mental health and well-being [13,14]. The type of support available on the web includes informational support (eg, websites that include information about particular issues) and emotional support (eg, social connections and connections with professionals) [15,16]. More specifically, this can include websites that include (1) informative content, (2) web-based question and answer sessions, (3) online forums [16], (4) stand-alone therapeutic programs or apps [17], and (5) web-based contact with professionals (eg, web-based therapists) [18].

Kooth, the service that this project has worked alongside, is one such service that offers a suite of web-based support options to CYP. It is a free web-based therapy and support service that supports over half a million CYP in the United Kingdom primarily through text-based support [19]. It has grown swiftly during this period and, at the time of writing, is funded by 115 of the 135 National Health Service Clinical Commissioning Groups in England. It differs from many web-based services, as it aims to explicitly integrate itself into existing local services and works alongside community-based professionals such as teachers, doctors, psychologists, and community health teams. The CYP who access the service remain anonymous and can tailor the support they receive, with individuals choosing between support that involves direct contact with professionals or not. For instance, some individuals may only read web-based content in the form of psychoeducational articles or online forums. Others may directly communicate with professionals by using synchronous and asynchronous chat options. This decision is led by the individuals accessing the service rather than the professionals offering support, and many individuals choose to use a combination of the above. This is in keeping with the organization's humanistic value base [20,21], an underpinning that is positively focused and prizes the agency of the individuals seeking support [22,23], and the professionals who offer support adopt a pluralistic goal-directed therapeutic approach [24,25]. These professionals include counselors, psychologists, psychotherapists, and social workers, and they work closely alongside those with more specific remits to write content and manage the technical side of the website.

The provision of anonymous support to CYP is a contentious arena. For some, the delivery of such services can be viewed as risky or dangerous. This proves particularly the case where an individual may be at risk of serious harm to themselves or another person, with some countries insisting that such support can only be provided with the consent of parents or caregivers.

However, in the United Kingdom, there is a long tradition of providing anonymous telephone support, with ChildLine, an anonymous telephone helpline for CYP, which was set up in 1986 following a public campaign focusing on the cruelty and abuse affecting CYP [26]. This service purposefully offered anonymity to its users with a view to provide the much-needed support that would most likely not have been accessed if only offered face-to-face. A number of web-based services now work in this way and the young people who access these services often highlight the importance of being able to access support anonymously [27]. Individuals who access such services do so with a wide range of complex needs [1] and typically these anonymous services work with individuals regardless of the issue disclosed. Risk is not ignored; however, professionals will support individuals to access additional services if needed. In doing so, they will attempt to work at the pace of the person obtaining support and not remove support if individuals choose not to provide further identifiable information.

Measuring Outcomes in Web-Based Therapy and Support Services

The current climate of mental health provision requires service providers to demonstrate the benefits of their work to those commissioning them. Commonly, these take the form of aggregated scores collated from self-report outcome measures reflecting upon the reduction of negative symptoms or the success of goals. There are numerous accepted processes and protocols for doing so, with organizations such as the Child Outcomes Research Consortium, creating specific guidance and recommendations. However, such measures have proven to be difficult to transfer into virtual environments. Difficulties have included practical issues such as transferring measures into web-based formats and navigating copyright issues and, more significantly, considering whether the measure itself is used in the same way as it would be in face-to-face relationships [1]. Therefore, there are suggestions that individuals accessing web-based services, particularly anonymous web-based services, do so for different reasons than those accessing face-to-face therapy. Indeed, exploration around the nature of web-based therapeutic relationships with CYP suggests that specific web-based issues, such as the safety afforded by the anonymous relationship, help to enhance the work entered into [27,28]. As a consequence of these differences, it can be argued that it is necessary to develop evaluation tools that take into account the complex environment more fully and make the best use of the technology available to ensure that any tools that are adopted are user friendly.

Developing a ToC

A ToC is an outcomes-based approach that can be used to identify and evaluate how programs and services achieve their stated goals of change. ToC has been defined as "the description of a sequence of events that is expected to lead to a particular desired outcome" [29]. Previous reviews indicate that ToCs should be flexible, be reflective, and ensure that any assumptions are explicitly stated [30]. When designing a ToC, Kail and Lumley [31] identified the following 5-step process:

1. Identify a realistic and definite goal.
2. Work backward from the goal to work out the intermediate outcomes.
3. Establish links between outcomes and their order by working out causes and effects.
4. Work out which activities lead to which outcomes.
5. Identify what else is needed for the intervention to work.

Developing a ToC can have several benefits for key stakeholders in a service. Staff members typically play an active role in the development of the ToC, owing to their perspective on what works are valued. This collaborative process can help to identify hidden perspectives while also helping to motivate staff by showing them how their contributions fit into the service-level goals [32]. In addition, a ToC clarifies what needs to be measured, as the assumptions about what makes the intervention work are identified. A ToC can also be used as a heuristic to help guide the creation of ToC for similar programs [33]. For example, this could be for applying the lessons learned from a ToC for a web-based CYP service to the development of ToC for a web-based service for adults. Finally, there is the added benefit that a ToC can be both *retrospective* by evaluating the efficacy of a service to date and *prospective* when used as a tool to support the planning of service development [34,35]. ToCs have been used in a range of contexts for young people, including sports programs within the youth justice service [34], school-based interventions for students and their families [36], and well-being programs for those unable to access mainstream education [37].

Aims, Rationale, and Research Questions

Given the complex nature of the web-based therapeutic environment, the aims of this study are to develop a ToC and map out the ways in which CYP access support on the Kooth platform. As such, the research question for this project is "what do key stakeholders identify as the core elements of a ToC for an anonymous online therapy and support service?"

Methods

Design

Working *with* organizations, instead of examining them from afar, is advocated to create more ecologically relevant pieces of research [38,39]. This project therefore reflects upon a collaborative piece of research between professionals from the Kooth service and academics with an interest in web-based therapeutic resources and theories of change. The close working between researchers and professional staff members was purposefully egalitarian in nature so as not to prize one set of knowledge or interpretation over the other. As such, the professionals involved in the project were both coresearchers [40] and practitioner researchers [41] responsible for constructing the ideas presented in this paper.

In keeping with the exploratory perspective adopted, the project has a social constructionist epistemology [42] and proved to be primarily inductive in nature [43]. However, it was acknowledged that this blank canvas approach was influenced by the theoretical perspectives of the individuals involved. For instance, as noted in the background of this paper, the principles of humanistic psychology were greatly valued by both the

service and the researchers involved. As such, a critically reflexive approach to the research was adopted for the conceptual presentations [44,45]. In practical terms, the researchers have purposefully engaged in critical discussions and sought external dialog to inform the development of the proposed theory. Here, it is noteworthy that ideas from good practice guidelines for qualitative research that advocate coherence checks with a variety of partners [46,47] have been used to enhance the overall trustworthiness of the synthesis that is presented.

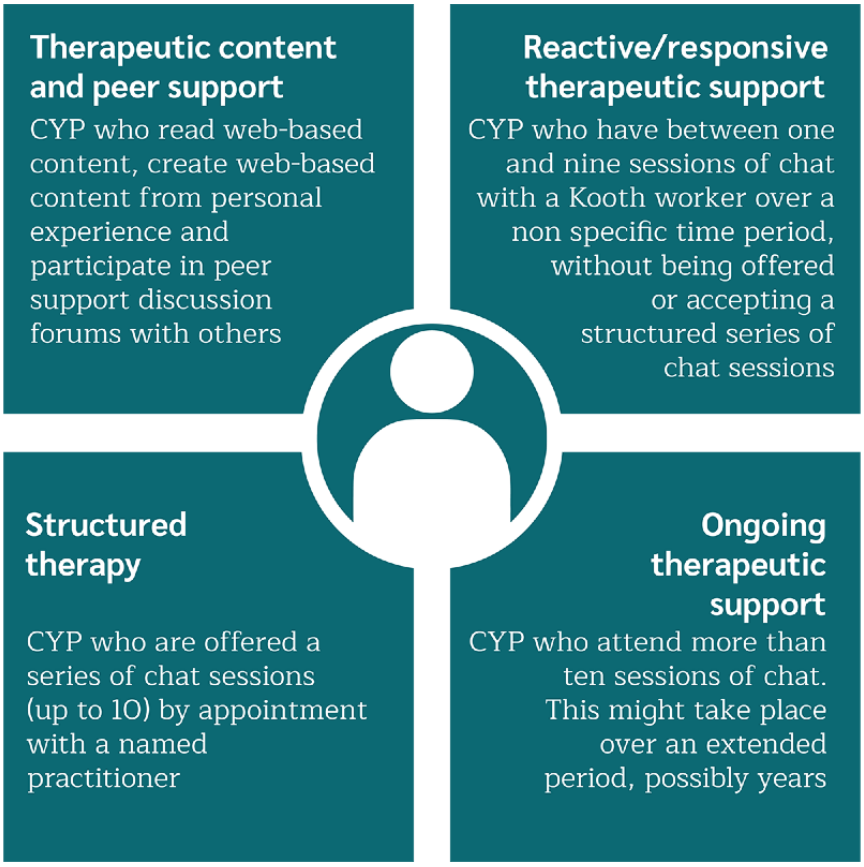
As described briefly in the background section of this paper, the study made use of ToC methodologies to help devise a deeper understanding of the work that the Kooth service enters with CYP. In particular, the ToC methodology focused on gaining insights into the specific conditions that lead individuals to use the service in question, the mode of delivery that services are offered, and the change people report or observe as a consequence of using the service [31]. These methods are purposefully collaborative in their approach and are primarily inductive at the starting point of the project.

The following sections outline this collaborative approach. Initially, the three phases of the project are briefly described: (1) a scoping workshop activity is described; this is followed by (2) the facilitation of a series of practitioner researcher activities and (3) a final coherence checking process. Each phase refers to the individuals involved, the process of generating data, and the data analysis procedures that are engaged.

Phase 1: Scoping Workshop

A workshop was held by combining 11 Kooth staff (5 therapists, 2 emotional well-being practitioners, 1 community engagement worker, 1 learning and development coordinator, and 2 research staff) and academic partners (TH and JG). As the staff members possess intimate knowledge regarding the way CYP make use of and benefit from the service, this workshop proved to be a vital starting point to direct the project. During the workshop, a series of presentations were given to provide information about the purpose of the project and the notion of a ToC. Focused conversations [48] were then held around the different ways in which CYP engaged with the Kooth service. These focused on the following 3 aspects: (1) the way in which CYP use the service (specifically highlighting any conditions present that mean CYP wish to use a web-based therapy and support service); (2) the ways in which CYP engage with the service (modes of delivery and the activities of key players); and (3) the impacts observed or reported while using the service (identified outputs and observed change). At the end of the workshop, seeds were sown about the ToC itself, with a rough draft being presented, and 4 support pathways were identified that represent the way in which individuals use the Kooth service. Figure 1 provides a description of these pathways.

Figure 1. Descriptions of the 4 service pathways for Kooth. CYP: children and young people.



Phase 2: Practitioner Researcher Pathway Explorations

Following the initial scoping workshop, 6 practitioners (4 therapists, 1 emotional well-being practitioner, and 1 community engagement worker) working for Kooth were invited to become practitioner researchers [41]. These individuals worked in groups to develop an understanding of the four agreed service pathways previously noted. The practitioner researchers involved were invited based on their experience and prior training in research methods, and their understanding of the pathways employed in the Kooth service.

The practitioner researcher teams were assigned to specific pathways and instructed to explore these in detail to develop an understanding of the ToC for that particular pathway. To facilitate the exploration of the pathways, individuals were provided with a series of anonymized transcripts of therapeutic interactions that occurred in 2018. Depending on the pathway, the transcripts were either taken from sessions with professionals or from interactions on the online forum. To ensure confidentiality and privacy, the content of the transcripts was managed within the service evaluation limitations of the organization's clinical audit guidelines.

Following a brief training session introducing qualitative research methods and the process of conducting thematic analysis [49], the following instructions were provided to each group of practitioner researchers:

- Inductively analyze a series of anonymized transcripts using the protocols of thematic analysis.
- Develop an overarching thematic map to provide a summary of the analysis in a common format of themes, subthemes, and codes.

Research supervision and advice were provided during this process. The findings of these exploratory studies were presented in a second workshop event and helped the working group understand how the different pathways worked and interacted. Following the presentations, all of the individuals who had been involved in examining the specific pathways worked collaboratively to articulate a ToC for the whole organization. This process involved deductively working to identify the agreed core elements of the ToC. These included the 3 overarching elements: (1) the *conditions* present for individuals wanting to access anonymous web-based therapy and support (including the CYP characteristics); (2) the mode of delivery (including the service inputs, worker activities, CYP activities, and the associated outputs); and (3) the reported and observed change (including the desired outcomes and the associated impact). The draft of this ToC was developed by an expert in the ToC methodology (JG) in collaboration with all other working group members.

Phase 3: Coherence Checking the ToC

Once the draft ToC had been agreed upon by all members of the working group, the ToC was opened up for consultation with other core stakeholders of the Kooth community. This was a multifaceted strategy that sought comments about the coherence and plausibility of the ToC formulation of the overall presentation [50]. The wider stakeholder group that was consulted included 7 young people, 9 service managers, and 29 practitioners working for the Kooth service. All individuals were asked to review and comment on the draft ToC using a web-based questionnaire. The questionnaire specifically sought comments about the core elements of the ToC. Following the consultation process, the draft ToC was reviewed once again by the working group and revised to accommodate the learning from these new viewpoints. The ToC was broadly accepted by all members of the wider stakeholder group, but a number of minor changes were made to the language of the document to enhance the clarity of the theory presented.

Ethical Considerations

The initial stages of this project were conducted as consultation exercises between academic partners and professionals involved in Kooth. The final phase of the work, which involved producing a public facing output, was approved by the University Research Ethics Committee of the fourth author (JP).

Results

This project set out to develop a ToC for web-based therapy and support services, Kooth. A sustained period of reflexive exploration, which combined academic expertise and professional wisdom, led to the description of the ToC reported and discussed below. Textbox 1 provides a summary of the specific ToC that was arrived at following the three phases of reflection. The ToC includes elements common to ToC methodology (ie, focusing on the conditions present for individuals to use such a service, the mode of delivery, and the change that was reported or observed) and has been adjusted in areas to fit the needs of the Kooth service. For instance, the activities of Kooth workers and young service users have been separated to delineate the different ways in which people are involved in the change process.

Textbox 1. The theory of change for Kooth.

Young person characteristics

- Want support then and there or quickly
- Do not have or want family and friends to turn to—may be in a marginalized group
- Curious, exploring, or looking for information and reassurance
- Unable or unwilling to access face-to-face services
- Comfortable with a preference for web-based communication
- Seeking a nonjudgmental space on the web
- Seeking a different connection with others

Triage or decision to offer service

- Delivery
  - Service inputs
    - Skilled and experienced professionals
    - Flexible access platform available out of hours, and written information or articles available 24/7
    - Robust clinical governance and risk managed through clinical oversight
  - Worker activities
    - Building an empathetic relationship
    - Drawing on professional understanding of child and adolescent developments
    - Assessing distress and risk and tailoring responses
    - Giving information and signposting
    - Cocreating goals and solutions with young people
    - Identifying what has helped before
    - Encouraging reflection and taking responsibility
    - Exploring the young person's relationship and support systems
  - Young person activities
    - Offloading their worries
    - Opening up, articulating, and sharing their story
    - Learning about mental health so they can understand their experiences
    - Exploring their thoughts and feelings
    - Building a trusted connection with the worker
    - Identifying coping skills and testing approaches
  - Outputs
    - Feels heard and has feelings validated
    - Gets information
    - Changes perspective or sees new options
    - Has experienced opening up to someone and built a relationship with a professional
    - Takes ownership of an issue
    - Starts to engage or has information about face-to-face services
    - Builds connections and a safe online and offline community
- Change
  - Desired outcomes

- Safer or crisis reduced
- Able to reflect on thoughts, feelings, and perceptions
- Able to consider future strategies
- Greater self-awareness and emotional regulation
- Acknowledges a reduction in stress
- Achieves personal goals and recognizes progress made
- Feels a sense of community

- Impact
  - Better able to manage current and future situations
  - Is able to demonstrate ambition and hope for the future
  - Increased confidence, personal responsibility, and ability to make decisions
  - Sets personal goals for change
  - Is aware that ongoing support is available—is not alone
  - Has a positive experience of a web-based space

Discussion

Principal Findings

In the following sections, we reflect on the major elements of ToC in chronological order, notably the conditions that are present for CYP to access such support, the mode of delivery offered, and the change reported and observed. The presentation refers to the different elements of the ToC; however, due to restrictions of space, it is not possible to refer directly to each component in detail.

The Conditions Present for CYP to Want to Access Web-Based Therapy and Support: CYP-Directed Mental Health and Well-being Support

Web-based therapy and support have become important resources for individuals who cannot access traditional face-to-face support [14]. This includes those who cannot physically access support and includes those that might struggle psychologically to access it too [51]. During the COVID-19 pandemic, web-based psychological support became an important resource for individuals within countries that were placed in enforced lockdown periods where self-distancing and self-isolation measures became commonplace. The ToC described here reflects some of these benefits and highlights that CYP may use these resources as points of informational and emotional support [15,16]. As a consequence, individuals may choose to access a combination of resources, with informative static webpages about particular issues, peer discussion forums, and professional support and guidance. The desire to obtain different types of support has previously been observed in the goals articulated by clients in web-based therapy [52] and the way that individuals use online forums [53]. Therefore, psychological services that offer anonymous therapy and support on the web need to be prepared to offer a variety of resources to accommodate the informational and emotional needs for which individuals access services.

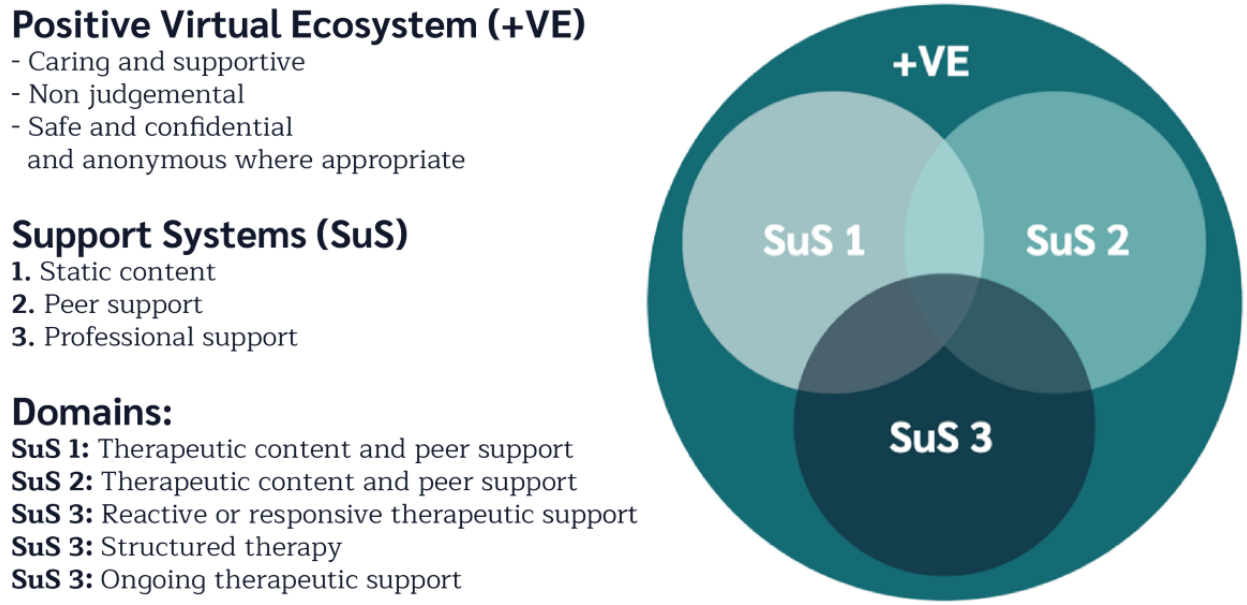
Another area of development in this project was the view that anonymous web-based resources offer a novel means of CYP accessing and directing their support. Young users of web-based therapy have previously expressed the importance of not providing identifiable materials [27,54]. What is evident here however is that this stretches much further. Unlike face-to-face support services that describe themselves as child-centered [55] or prize shared decision making [56], anonymous web-based environments provide CYP with the opportunity to take ownership of their support packages and direct their engagement as a default. This position might be aligned to active client theories in therapy [57], with individuals being active both in directing the support they access and within the relationships with supporters themselves. As a consequence, they can be a means of leveling the power differentials between adult professionals and CYP accessing services. Such a position is likely to prove challenging for some professionals who might see their professional experience being undermined or not accounted for. As such, anonymous psychological support arguably recalibrates how professionals might define child-centered support by extending definitions to allow CYP to direct their support as a default position.

The Mode of Delivery: A Positive Virtual Ecosystem

In discussing and devising the service-level ToC, it was apparent that specific interventions are not offered in isolation. This may seem obvious, but many efficacy and effectiveness research designs often overlook the broader systems that impact the lives of individuals. Within the analysis process noted above, it was therefore apparent that the professionals developing the ToC were considering the way that individuals made use of a variety of resources on the Kooth website. For instance, the individuals discussed obtaining informational and emotional support from different pathways separately and in combination. As such, it may be possible to separate these elements into specific support systems in which individuals might access static support that does not change (eg, web-based content), peer support (eg,

online forums), or professional support (eg, web-based therapy), but there was a general view that "the whole was greater than the sum of its parts," with many reflecting that the young service users often evaluated their relationship with Kooth as a whole, rather than its specific components. As such, the Kooth service may be viewed as a broader ecosystem, involving a variety of resources that specifically aims to provide a safe and anonymous space for CYP in which they felt accepted to explore the issues that they encounter in life. The environment might therefore be viewed as akin to that advocated by person-centered therapists [22] or humanistic educationalists [58,59], in which individuals aim to develop a caring and supportive nonjudgmental environment for individuals to grow constructively. The Kooth service might therefore be viewed as a *positive virtual ecosystem* (+VE; Figure 2).

Figure 2. A positive virtual ecosystem (+VE).



Psychological support services are relatively underdeveloped in terms of keeping up with technological advances [60]. Given this, it is common for services to attempt to replicate face-to-face ways of working when developing web-based therapeutic resources and to judge them using the same criteria [61]. The concept of +VE offers a more systemic perspective of how internet-based support might be more helpfully conceptualized, particularly given the current data-rich environments that are being created. Thus, rather than seeing specific interventions in isolation of one another, they might be viewed as part of a larger system in which all elements play an important role. Thus, if you take a section of the service away, such as the web-based static content, Kooth would have a different feel to it, and this would impact upon the other resources on offer.

Finally, when considering +VEs, the importance and value of static content and peer support needs to be acknowledged. These themselves can cater to individuals in need of informational support and, with regard to forums, emotional support from peers [62]. Given the large number of CYP who might benefit from additional support, and the limited finances often allocated to CYP services [10], appropriately curated content and moderated peer-support resources can be a positive way in which services might provide appropriate help to larger numbers of individuals.

Understanding Change in Web-Based Therapy and Support for CYP

The desired outcomes noted in the ToC come as a consequence of young service users proactively engaging with the service inputs and activities of the professionals. They might be reduced to those activities that provide informational support (eg, giving information and signposting) and those that provide emotional support (eg, building an empathic relationship) [15]. These support types have already been well described within peer-support contexts on the web [62,63], and this frame can also be extended to describe the professional support offered. In accounting for this, when identifying whether CYP obtained what they wanted from the service, considering whether they received the information that they wanted and/or the engagement in supportive relationships proved important to consider.

Notably, the changes described in the ToC are broad and idiographic in nature. Furthermore, they did not reflect specific diagnostic criteria. As such, the changes identified might appear more aligned to the humanistic psychology perspective that underpins the service under scrutiny [20,23] and consolidates the view that traditional tools are unlikely to be relevant for all of those using these types of services [1]. In a similar way to the activities offered, one member of the working group (TH) suggested that the impact of the service might be reduced to a simplistic form. Specifically, they may be divided into those that have an impact on the intrapersonal world of the individuals (eg, able to reflect on thoughts, feelings, and perceptions) and those that impact their interpersonal worlds (eg, feeling a sense of community). This frame resonated strongly with other members of the working group. Considering the types of activities offered and the types of outcomes worked toward, a high-level outcome matrix might therefore be used to consider what a successful therapeutic engagement might look like for

an anonymous web-based therapy and support service. Table 1 presents this matrix and provides an example of the types of response outcomes that CYP might articulate.

Table 1. High-level outcome matrix for Kooth services.

Outcome	Emotional support	Informational support
Intrapersonal	"I understand myself more"	"I can identify with something important to me"
Interpersonal	"It helps me relate to others"	"I have some skills I want to try with others"

Implications for Professionals

The ToC that has been developed has numerous implications for mental health and well-being practitioners working in these environments. These include broad benefits, such as the ToC itself being an informative training tool for professionals working in this environment, to narrower elements, such as the need to develop bespoke tools to capture the outcomes of humanistic web-based support. These 2 areas were briefly considered.

It is important that professionals seriously consider the implications of transferring work to web-based formats. The ToC developed here helps to demonstrate the complexity inherent in working in this way. First, it highlights the need to be aware that some young people who access web-based support have different needs from their counterparts who access face-to-face support. Furthermore, the need for skilled and experienced professionals who are competent web-based communicators also comes to the fore. Although there are many transferable skills from face-to-face work, web-based resources, such as Kooth, are multifaceted and highlight the need for alternative or extended ways of thinking about support. In particular, it is important for professionals to consider the full offer on the web that is available to young people seeking help. The +VE that services provide can be adapted by service users in a multitude of ways. Such a position is in keeping with a pluralistic therapeutic approach that advocates that support should be led by the person seeking support [23] and that "one size can never fit all" [25]. Systemic thinking, which involves consideration of the different support systems being offered, therefore needs to be incorporated into the training of those supporting CYP on the web.

As previously indicated, capturing and measuring the change that individuals experience as a consequence of using a web-based counseling and support service proves to be challenging. Previous research has shown that standardized self-report measures developed for face-to-face support may not be transferable to web-based settings [1]. This seems particularly relevant when evaluating +VE. Given various ways in which individuals may tailor the support they are accessing, it is suggested that a flexible means of evaluating support is needed. As such, it is recommended that therapeutic outcomes are considered using self-report measures that make use of the outcome matrix previously noted. On the basis of this framework, an idiographic satisfaction measure can be used to complement other sources of personalized data collection, such as goal-based outcome measures.

Strengths, Limitations, and Future Research

This is the first ToC to be created, focusing on a telepsychology resource for CYP. It provides a descriptive account of the types of people who might use these services, the resources needed to offer them, the activities that individuals engage in, and the outcomes and impacts that might be expected. The main strength of this conceptualization is its closeness to the organization that it reflects upon. Working alongside professionals from the organization to devise the project has helped to retain a direct currency for the partner organization. Such a position differs from other models that might adopt top-down approaches that ultimately lack ecological validity in application [38,39]. In contrast, it is acknowledged that this frame of understanding is underpinned by research groups' underpinning in humanistic psychological principles [20,21]. As such, the synthesis of findings might be viewed as reflecting this more holistic positioning, and it is likely that others would see value in creating a more reductive frame that directly examines assessments focusing on pathological symptomology.

Going forward, it will be important to scrutinize the ideas presented in this paper in depth. In particular, we consider the following three areas to be core areas in need of further investigation:

- It is clear that anonymous web-based psychological support can prove to be liberating to some CYP. However, such freedom poses numerous challenges for professionals and services and therefore warrants further investigation. This can include reflecting upon practical issues, such as responding to risky behaviors, and relational issues, such as how disinhibition in web-based communication changes the therapeutic relationship.
- The +VE is currently unchartered territory. Numerous web-based services offer a variety of resources. In contrast to the creativity in the packages on offer, much evaluation remains focused on specific elements of the services (eg, web-based therapy). Given the data-rich nature of web-based resources, considering a more holistic picture becomes possible and analysis will arguably reflect the work of the services more fully. Furthermore, the systematic evaluation should not be limited to virtual ecosystems, and the CYP mental health and well-being support ecosystem should be extended to *in-person* work as well.
- Finally, a high-level outcome matrix needs to be considered alongside real-world activities. It will be possible to develop easy-to-complete satisfaction measures that are CYP friendly to capture information based on this framework. The utility of doing so however needs to be examined in depth.

Conclusions

This project adds to the view that telepsychology directed toward CYP is diverse and complex to understand. This highlights the need to consider a broader virtual ecosystem and the interactions between different resources. More specifically, it describes the concept of the +VE in which a variety of interrelated resources are provided in a safe and caring overarching package. In doing so, it highlights the way in which services can support CYP to take ownership of and direct the support they obtain if they so desire. Such a position might be viewed as pluralistic in nature and differs greatly from the professionally led mental health and well-being services that are commonly offered face-to-face.

It is argued that web-based services available to CYP on the web should not solely aim to transplant face-to-face services on the web. Given the likelihood of telepsychology becoming more commonplace following the COVID-19 crisis, a variety of supportive approaches can be used to realize the full potential of the technology available. These resources can offer informational and emotional support in a variety of guises and include providing informative content, moderated online forums, stand-alone therapeutic programs, and professional psychological support such as therapy. By offering a variety of support options, individuals are able to tailor the support they access, and services can helpfully respond more fully to the differing needs and wants of the CYP seeking support.

Finally, the evaluation of the telepsychology activities offered on the web needs to be fit for purpose, with face-to-face resources arguably having limited transferability. The evaluation of interventions might focus on specific interventions, but by doing so, evaluation strategies can neglect the various ways in which individuals access and use web-based therapy and support services. Where resources are evaluated in a more holistic manner and include consideration of the broader virtual ecosystem, a higher level of assessment is needed to accommodate the complexity inherent in the variety of therapeutic resources on offer. Here, it is recommended that outcomes might be assessed around a matrix examining whether individuals received the informational or emotional support that they were seeking in conjunction with whether this was directed to supporting intrapersonal or interpersonal change. This simplistic frame might be presented in a palatable form so as to be appropriate for young individuals to use and be easily adopted in a web-based environment. Such a frame needs further exploration going forward but has scope to provide a useful means of assessing whether +VEs meet the specific needs of those accessing the services.

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Conflicts of Interest

AS is an employee of Kooth.

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Abbreviations

CAMHS: Child and Adolescent Mental Health Service
CYP: children and young people
ToC: theory of change
+VE: positive virtual ecosystem

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Original Paper

Factors Contributing to Adolescents' and Young Adults' Participation in Web-Based Challenges: Survey Study

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Abstract

Background: Web-based challenges, phenomena that are familiar to adolescents and young adults who spend large amounts of time on social media, range from minimally harmful behaviors intended to support philanthropic endeavors to significantly harmful behaviors that may culminate in injury or death.

Objective: This study aims to investigate the beliefs that lead adolescents and young adults to participate in these activities by analyzing the amyotrophic lateral sclerosis (ALS) ice bucket challenge, representing nonharmful behaviors associated with web-based challenges, and the cinnamon challenge, representing web-based challenges that lead to harmful behaviors.

Methods: A retrospective quantitative study was conducted with a total of 471 participants aged between 13 and 35 years who either had participated in the ALS ice bucket challenge or the cinnamon challenge, or had never participated in any web-based challenge. Binomial logistic regression models were used to classify those who participated in the ALS ice bucket challenge or cinnamon challenge versus those who did not engage in either challenge using the integrated behavioral model's beliefs as predictors.

Results: The findings showed that participants of both the cinnamon challenge and the ALS ice bucket challenge had significantly greater expectations from the public to participate in the challenge they completed in comparison with individuals who never participated in any challenge (P=.01 for the cinnamon challenge and P=.003 for the ALS ice bucket challenge). Cinnamon challenge participants had greater value for the outcomes of the challenge (P<.001) and perceived positive public opinion about the challenge (P<.001), in comparison with individuals who never participated in any challenge. In contrast, ALS ice bucket challenge participants had significantly greater positive emotional responses than individuals who never participated in any challenge (P<.001).

Conclusions: The constructs that contribute to the spread of web-based challenges vary based on the level of self-harm involved in the challenge and its purpose. Intervention efforts could be tailored to address the beliefs associated with different types of web-based challenges.

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KEYWORDS

web-based challenges; self-injurious behavior; behavior; integrated behavioral model; social media; challenge; adolescent; young adult; participation; survey

Introduction

Background

More than 70% of Americans use social media platforms to post personal information, engage with posted content, and connect with others [1-4]. Adolescents and young adults were among the earliest internet and social media adopters and continue to use these websites at high levels [2,5,6]. Web-based challenges, or social media challenges, are popular phenomena, especially among adolescents and young adults, perhaps because of their frequent use of social networks. In these challenges, participants record themselves engaging in specific activities and share their experiences through social media platforms [6,7]. These challenges are ubiquitous and can be found on many social media platforms, including YouTube, Instagram, Facebook, and WhatsApp [8,9]. Although the activities involved in web-based challenges can vary from fun to fatal [10-13], they can generally be classified into 2 categories: (1) minimal harm challenges, which in some cases support a philanthropic cause such as the amyotrophic lateral sclerosis (ALS) ice bucket challenge [14], or (2) harmful challenges, which entail self-injurious behavior such as the cinnamon challenge [15]. Although the ALS ice bucket challenge has faced criticism (eg, safety concerns and waste of water), it is the most successful and influential fund-raising event to date [14]. In addition to raising more than US $115 million for ALS research [16], it is also credited for increasing public awareness about the disease [17].

In contrast, the cinnamon challenge involves swallowing a teaspoon of ground cinnamon without drinking any liquid for 60 seconds. The problem is that cinnamon does not dissolve or biodegrade in the lungs, as evidenced by animal-based laboratory studies, which experienced symptoms ranging from mild multifocal granulomatous inflammation to alveolar lipoproteinosis and alveolar cell hyperplasia [15,18-20]. The consequences are just as serious for humans because swallowing a large amount of cinnamon can cause pulmonary inflammation, allergic and irritant reactions, and even more serious situations, such as hypersensitivity-induced asthma attacks, which can be fatal [15]. However, none of these potentially fatal consequences have stopped adolescents and young adults from participating in the cinnamon challenge. As of 2013, there are more than 51,100 public YouTube clips of someone accepting this challenge, with some videos garnering more than 19 million views globally [15].

Given the significant amount of controversy concerning these web-based challenges, there is little research on the factors that lead individuals to participate in such challenges. For example, the extant literature on self-harm focuses primarily on a single challenge and its effect on public health and safety [15,21-23] or on how viewing content showing self-harm could lead to intentional self-harm by modeling the behavior of those we observe [24-26]. Furthermore, the literature on adolescent web-based risk focuses on the effects of engaging in web-based sexual and aggressive risk exposure [27,28]. To our knowledge, no quantitative research has comprehensively investigated the phenomenon of web-based challenges and why adolescents and young adults engage in these activities.

In this study, quantitative data were collected to explore adolescents' and young adults' exposure to web-based challenges and the determinants of their engagement with them through direct participation. The integrated behavioral model (IBM) [29] was used to investigate its generalizability to these behaviors on the web. It is important to reassess that and other existing behavioral theories concerning behaviors on the web because what may be true about traditional human behaviors may not apply to web environments [30].

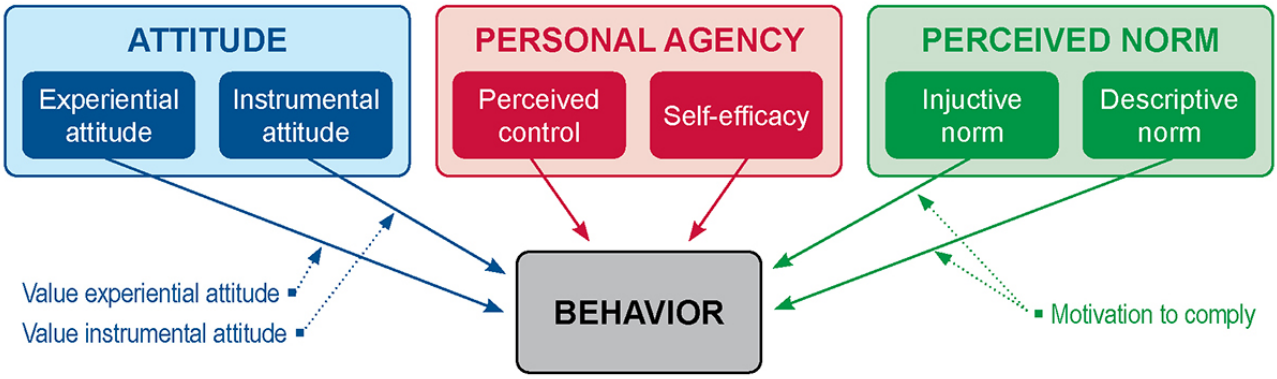
IBM as the Underlying Framework

As seen in Figure 1, IBM suggests that the intention to perform a behavior is driven by 3 factors: attitude, perceived norms, and personal agency regarding behavior. Attitude, defined as an individual preference for a certain behavioral performance, is composed of 2 dimensions: experiential attitude and instrumental attitude [31-33]. Experiential attitude is an individual's emotional reaction to a behavior. For example, an individual with a positive emotional response toward a specific social media challenge is more likely to engage in it than an individual with a negative emotional response. Instrumental attitude is cognitively based, meaning that it is affected by a person's beliefs about the outcomes of the behavior depending on the value of those outcomes.

Perceived norms regarding behavior and the social pressure to perform it are composed of injunctive and descriptive norms. Injunctive norms refer to the normative beliefs about others' opinions toward participating in a challenge and the motivation to comply (if others approve or disapprove of the behavior). Descriptive norms refer to common patterns of behavior that lead to the expectations of people behaving according to that pattern.

Personal agency consists of 2 constructs: perceived control and self-efficacy. Perceived control refers to personal beliefs about the degree of control over performing the behavior. These beliefs are based on individual perceptions of how environmental factors will make the performance of the behavior difficult or easy. Self-efficacy is the individual's certainty in their ability to perform the behavior in addition to their belief that they can overcome each prohibitive condition or obstacle [29].

Figure 1. Integrated behavior model attitude.



Objectives

The purpose of this study is to use IBM quantitatively to enhance our understanding of how each belief in IBM contributes to adolescents' and young adults' willingness to participate in web-based challenges. Another purpose of this study is to discern which beliefs are more influential than others. The findings from this study can be used to guide the development of interventions to reduce participation in harmful social media challenges among adolescent and young adult populations. Specifically, this research addressed the following research question: what is the effect, if any, of attitudes, perceived norms, and personal agency beliefs on adolescents' and young adults' willingness to participate in the cinnamon challenge and the ALS ice bucket challenge?

To explore our research question, we applied the IBM developed by Montano and Kasprzyk [29] depicted in Figure 1 to our hypotheses listed in Textbox 1.

Textbox 1. Research hypotheses.

Research hypotheses developed based on integrated behavioral model:

- Hypothesis 1: The experiential attitude is positively related to cinnamon challenge and amyotrophic lateral sclerosis (ALS) ice bucket challenge participation.
- Hypothesis 2: The instrumental attitude is positively related to cinnamon challenge and ALS ice bucket challenge participation.
- Hypothesis 3: The value assigned to experiential attitude items moderates the relationship between experiential attitude and cinnamon challenge and ALS ice bucket challenge participation.
- Hypothesis 4: The value assigned to instrumental attitude items moderates the relationship between instrumental attitude and cinnamon challenge and ALS ice bucket challenge participation.
- Hypothesis 5: The injunctive norm is positively related to cinnamon challenge and ALS ice bucket challenge participation.
- Hypothesis 6: The descriptive norm is positively related to cinnamon challenge and ALS ice bucket challenge participation.
- Hypothesis 7: The motivation to comply moderates the relationship between the injunctive norm and cinnamon challenge and ALS ice bucket challenge participation.
- Hypothesis 8: The motivation to comply moderates the relationship between the descriptive norm and cinnamon challenge and ALS ice bucket challenge participation.
- Hypothesis 9: Perceived control is positively related to cinnamon challenge and ALS ice bucket challenge participation.
- Hypothesis 10: Self-efficacy is positively related to cinnamon challenge and ALS ice bucket challenge participation.

Methods

Study Overview

A survey-based study was used to investigate the application of IBM in the prediction of social media challenge behavior among adolescents and young adults. The developed survey included measures of the IBM constructs, similar to the studies reported in the literature [34-36]. The survey was pilot tested and modified accordingly. Finally, the survey was deployed to a larger sample to explore the reasons for participation in these challenges, retrospectively.

Measures

The dependent variable—social media participation—was collected at the beginning of the survey. The participants were asked whether they participated in the cinnamon challenge only, the ALS ice bucket challenge only, or never participated in any social media challenge. The classes for the dependent variable were balanced, with approximately one-third of the participants being in each class. The survey was then structured to include 3 main sections to assess the independent variables: a demographic section, a section related to participation in the cinnamon challenge, and a section related to participating in the ALS ice bucket challenge. The demographic section included questions about the participant's age, gender, race or ethnicity,

education, internet use, and social media challenge participation. The second and third sections assessed the following theoretical constructs related to the cinnamon challenge and the ALS ice bucket challenge: attitude, perceived norm, and personal agency. Note that the scale score for each construct was obtained by computing the mean of the relevant items.

Attitude was measured using 2 subconstructs: experiential attitude and instrumental attitude. Experiential attitude was measured using 4 items, each using a 7-point Likert scale. Instrumental attitude was measured using 2 items, each using a 7-point Likert scale. The value assigned to each item for both instrumental attitude and experiential attitude was measured using a 7-point bipolar scale.

Perceived norm was measured using 2 subconstructs: injunctive norm and descriptive norm. Each was measured using 7 items on a 7-point Likert scale. The motivation to comply construct assessed the participants' willingness to comply with other individuals and their beliefs. This construct was measured using 7 items, each using a 7-point bipolar scale.

Personal agency was assessed using 2 subconstructs: perceived control and self-efficacy. Perceived control was assessed using 6 items measured on a 7-point Likert scale, whereas self-efficacy was assessed using 4 items measured on a 7-point Likert scale.

The items for these constructs were developed using the strategy suggested by Glanz et al [37] in two stages. First, a team of researchers used the data from a previous qualitative study on this topic to develop the initial set of items that measured each of the subconstructs [38]. The survey was then pilot tested using a sample of 20 participants. The results of the pilot testing were used to delete the questions that had little to no variance [29] and to improve the clarity of the remaining questions. Internal consistency reliability was calculated for each scale using Cronbach alpha (Table 1). Examples of the specific items that comprise each construct for the cinnamon challenge and the ALS ice bucket challenge are reported in Multimedia Appendix 1.

Table 1. Construct reliability measured using Cronbach alpha.

Construct	Cronbach alpha for Cinnamon challenge items	Cronbach alpha for ALS[a] ice bucket challenge items
Experiential attitude	.81	.67
Instrumental attitude	.87	.69
Value assigned to experiential attitude	.85	.67
Value assigned to instrumental attitude	.92	.91
Injunctive norm	.94	.92
Descriptive norm	.91	.88
Motivation to comply	.88	.88
Perceived control	.70	.85
Self-efficacy	.66	.78

[a]ALS: amyotrophic lateral sclerosis.

Participants

Qualtrics Research Suite [39] was used to deploy the surveys to the participants. Inclusion criteria for the participants were participating in either the cinnamon challenge or the ALS ice bucket challenge (not both) or no participation in any social media challenge and age within the range of 13 to 35 years at the time of the study (adolescents or young adults only). A total of 471 participants completed the study. Approximately half of the participants—234 out of 471—were aged under 18 years (adolescents), and the rest—237 out of 471—were aged between 18 and 35 years (young adults), with approximately 82.6% (389/471) being females. Approximately one-third (n=153) of the respondents had participated in the cinnamon challenge only, one-third (n=155) had participated in the ALS ice bucket challenge only, and the remaining (n=163) had not participated in any social media challenge. More information about the participants is provided in Table 2.

Table 2. Participants' demographics.

Variable	Values, n (%)
Gender	
Female	389 (82.6)
Male	78 (16.6)
Prefer not to answer	4 (0.8)
Education	
Some high school	171 (36.3)
High school or GED[a]	155 (32.9)
2-year college degree	26 (5.5)
Some college	58 (12.3)
4-year college degree	33 (7.0)
Master's degree	21 (4.5)
PhD degree	2 (0.4)
Professional degree (eg, Juris doctor or Doctor of medicine)	5 (1.1)
Race	
White	220 (46.7)
African American	132 (28.0)
Native American	4 (0.8)
Asian	32 (6.8)
Pacific Islander	2 (0.4)
Hispanic or Latino	55 (11.7)
Other	26 (5.5)
Employment	
Full-time	86 (18.3)
Part-time	87 (18.5)
Student	236 (50.1)
Retired	2 (0.4)
Unemployed	60 (12.7)
Age (years)	
<18	234 (46.7)
18-35	237 (50.3)
Social media participation	
Cinnamon challenge	153 (32.5)
ALS[b] ice bucket challenge	155 (32.9)
None	163 (34.6)
Internet use per day (hours)	
<1	16 (3.4)
1-2	30 (6.4)
2-3	63 (13.4)
3-4	98 (20.8)
>4	264 (56.1)

[a]GED: General Educational Development.

[b]ALS: amyotrophic lateral sclerosis.

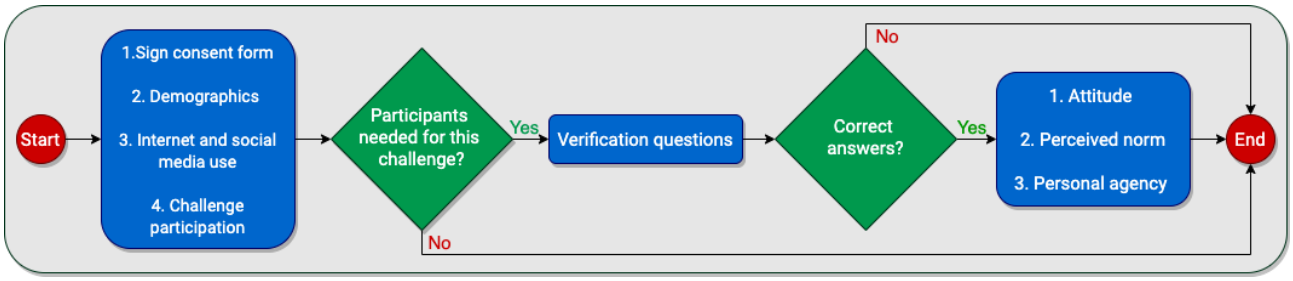
Procedure

First, the participants read and signed the informed consent form, read the introduction to the study, and answered questions about their demographics and social media and internet use, followed by a set of screening questions. None of the participants who met the inclusion criteria based on the screening questions were ineligible to participate in the study. The screening questions were as follows:

- "Have you participated in any online challenges?"
- "Which of the following challenges did you participate in?"
- "Of the following statements, which one matches what you did in this challenge?"

For the group of participants who never participated in any challenges, they had to state that in the first screening question. For the other 2 groups, if the challenge and the description did not agree, the participant was not eligible for the study. In addition, the number of participants for each challenge was restricted to having at least 75 adolescents and 75 young adults to ensure having participants from each group. The participants then answered questions to assess the constructs reported in the *Measures* section (attitude, perceived norms, and personal agency) about the cinnamon challenge and the ALS ice bucket challenge. The order of the cinnamon challenge and the ALS ice bucket challenge sections was randomly assigned to the participants. Figure 2 shows the flowchart of the study procedure. The study was approved by the Institutional Review Board of Clemson University, and all participants read and signed an informed consent form before beginning the study. Each participant was given a US $10 gift card as compensation for their time.

Figure 2. Procedure flow chart.



Data Analysis

Binomial logistic regression was used to understand whether participation in a social media challenge (ie, either the cinnamon challenge or the ALS ice bucket challenge) can be predicted from people's attitudes, perceived norms, and personal agency beliefs. Participation in a social media challenge is a dichotomous dependent variable (ie, 1=*participated* or 0=*did not participate*), justifying the use of the binomial logistic regression [40]. The binomial logistic regression analysis was performed using SPSS 24.0 to predict cinnamon challenge participation first with 7 predictors: age group, experiential attitude, instrumental attitude, injunctive norm, descriptive norm, perceived control, and self-efficacy. Four interaction predictors were also added to the model: experiential attitude by the value of experiential attitude, instrumental attitude by the value of instrumental attitude, the injunctive norm by the motivation to comply, and the descriptive norm by the motivation to comply. Only the interaction terms recommended by IBM were included in the analysis [29] to ensure greater power to detect significant findings within our multivariate analysis [41]. Participants who had completed the cinnamon challenge and those who did not participate in any challenge were included in this model (n=316).

A second binomial logistic regression model was used to predict ALS ice bucket challenge participation using similar predictors assessing the participants' perception of the ALS ice bucket challenge. Participants who had completed the ALS ice bucket challenge and those who did not participate in any challenge were included in the second model. For each model, fit indices, McFadden pseudo R^2, effect size estimates, estimated regression coefficients and their significance, and corresponding odds ratios and their confidence intervals were calculated.

All the data were checked to confirm the independence of observations; the existence of a linear relationship between an independent variable and the logit transformation of the dependent variable; and the absence of any multicollinearity, significant outliers, high leverage points, and highly influential points [40].

Results

Cinnamon Challenge

The results from the direct logistic regression model predicting cinnamon challenge participation are presented in Table 3. A test of the full model with all predictors against a constant-only model was statistically significant (χ^2_{11}=221.8; n=316; P<.001), indicating that the predictors, as a set, reliably distinguished between people who had participated in the cinnamon challenge and those who had not. The deviance in participating in the cinnamon challenge accounted for by these predictors was large, with R^2_L=0.5. To test each predictor's significance, each variable was removed from the model, and the change in χ^2 was examined to determine if the removal of a variable led to a worsening of the model fit [42,43]. Independent removal of 4 of the 11 predictors significantly harmed the model fit, specifically instrumental attitude ($\Delta\chi^2_1$=11.5; P<.001), injunctive norm ($\Delta\chi^2_1$=30.4; P<.001), descriptive norm ($\Delta\chi^2_1$=6.6; P=.01),

and the interaction term injunctive norm by the motivation to comply ($\Delta\chi^2_1$=8.8; *P*=.003). Figures 3-5 illustrate the form of these relationships.

To interpret the significant interaction for injunctive norm by motivation to comply, simple slopes were calculated from the regression coefficients at the mean of motivation to comply and 1 SD above and below the mean of motivation to comply [42]. This analysis found the slope of injunctive norm and probability to participate at the mean of motivation to comply to be β=1.01, and at 1 SD above and below the mean of motivation to comply to be β=1.80 and β=0.23, respectively. Figure 6 illustrates the form of this interaction.

Table 3. Results of binomial logistic regression model predicting cinnamon challenge participation.

Predictor[a]	*B* (SE)	$\Delta\chi^2$*(df)*	Odds ratio (95% CI)
Constant (Intercept)	−0.68 (0.29)	N/A[b]	N/A
Age group[c]	0.12 (0.35)	0.1 (1)	1.12 (0.57-2.26)
Experiential attitude	−0.31 (0.16)	4.0 (1)	0.73 (0.53-1.00)
Instrumental attitude	0.42 (0.13)	11.5 (1)**	1.52 (1.19-1.96)
Injunctive norm	1.15 (0.24)	30.4 (1)**	3.15 (2.04-5.15)
Descriptive norm	0.57 (0.23)	6.6 (1)*	1.78 (1.15-2.80)
Perceived control	0.53 (0.28)	3.9 (1)	1.71 (1.00-3.00)
Self-efficacy	0.25 (0.19)	1.7 (1)	1.30 (0.89-1.87)
Experiential attitude × value of experiential attitude	−0.09 (0.12)	0.7 (1)	0.91 (0.72-1.14)
Instrumental attitude × value of instrumental attitude	0.10 (0.07)	2.4 (1)	1.11 (0.97-1.27)
Injunctive norm × motivation to comply	−0.48 (0.16)	8.8 (1)**	0.62 (0.45-0.85)
Descriptive norm × motivation to comply	−0.12 (0.15)	0.7 (1)	0.88 (0.64-1.17)

[a]Model χ^2_{11}=151.05; n=318; R^2_L=0.34; null −2 Log likelihood=440.63; model 2 Log likelihood with predictors=289.58.

[b]N/A: not applicable.

[c]Age group was a dummy variable where 0=under 18 years old and 1=from 18 to 35 years old.

*P<.01; **P<.001.

Figure 3. The relationship between instrumental attitude and probability of cinnamon challenge participation.

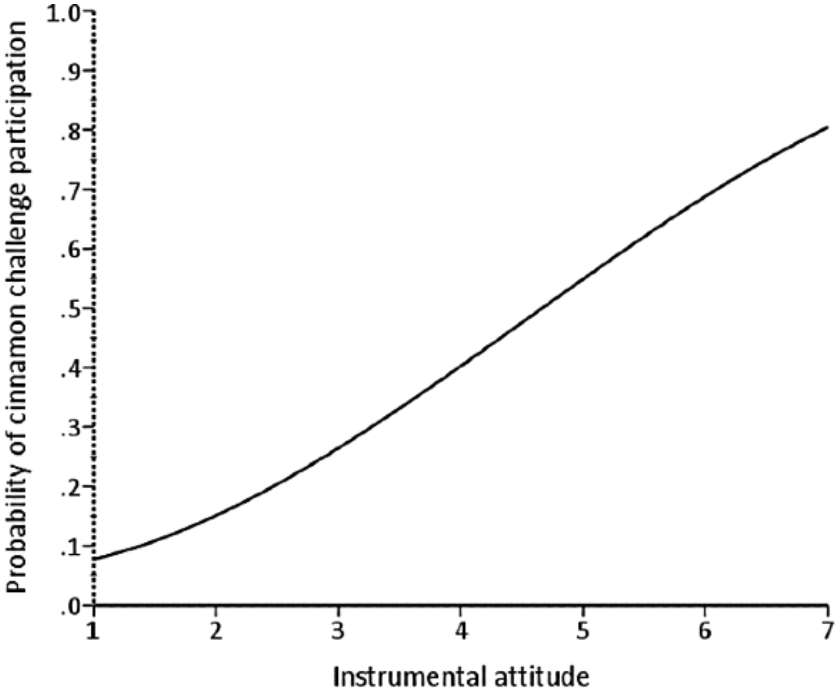


Figure 4. The relationship between injunctive norm and probability of cinnamon challenge participation.

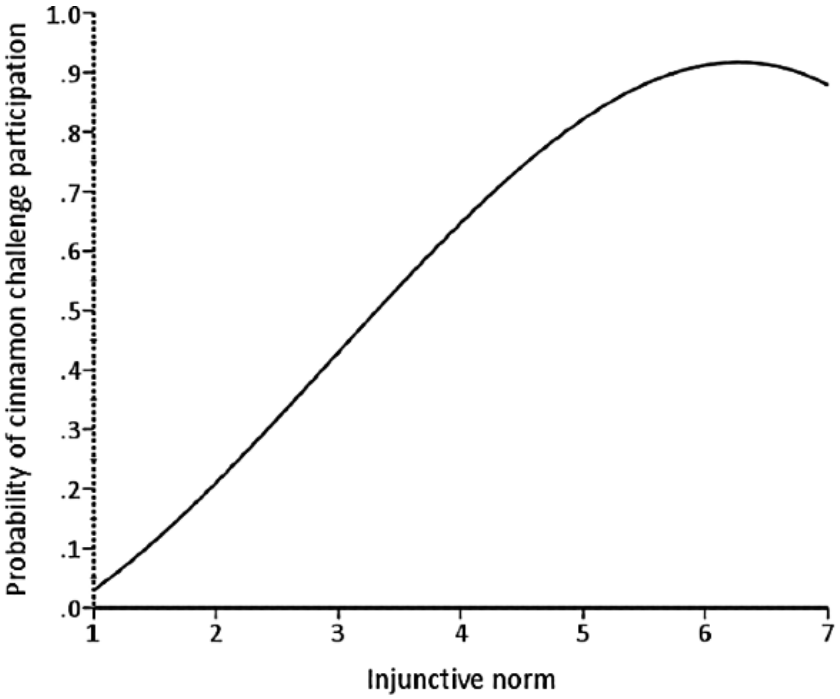


Figure 5. The relationship between descriptive norm and probability of cinnamon challenge participation.

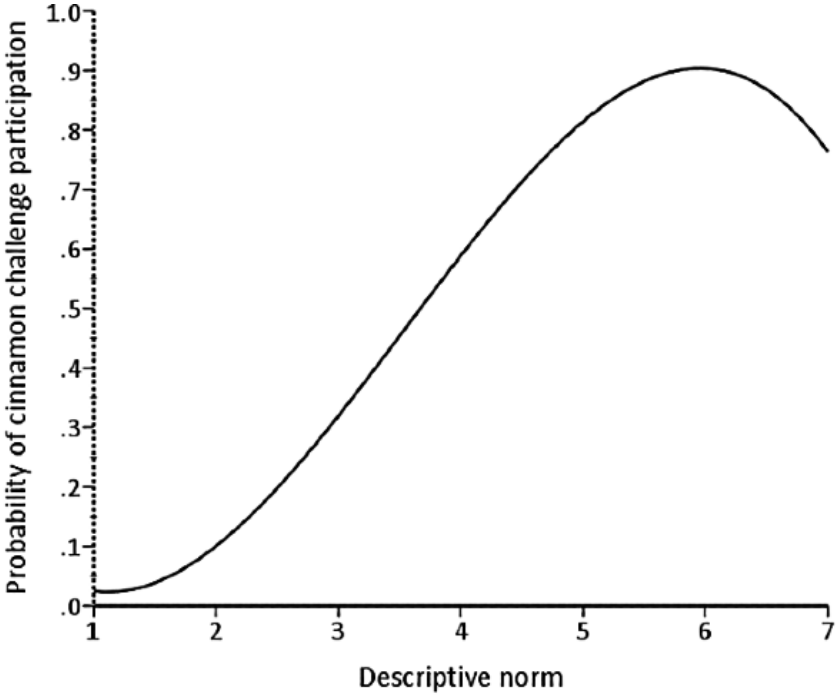
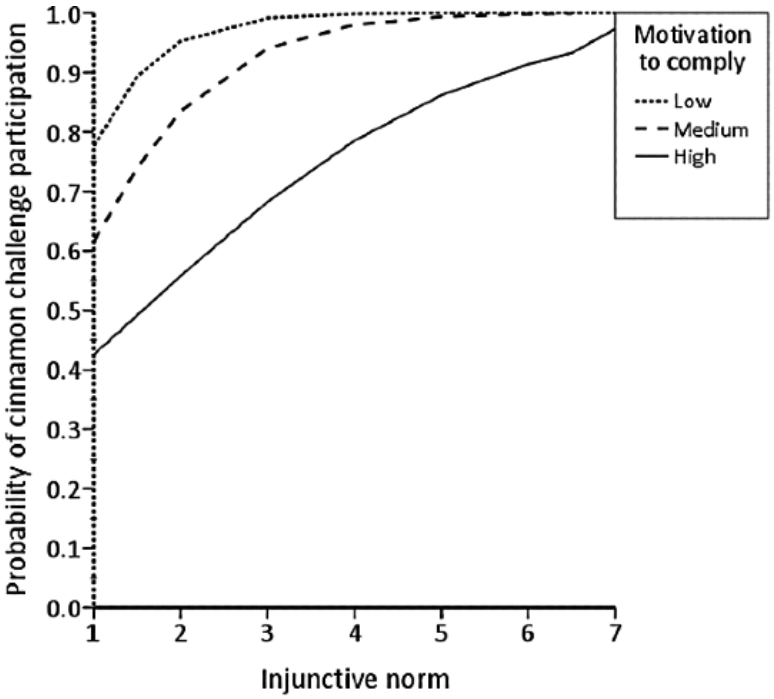


Figure 6. The relationship between injunctive norm and probability of cinnamon challenge participation moderated by motivation to comply.



Ice Bucket Challenge

A similar approach was used to predict ALS ice bucket challenge participation, with the results presented in Table 4. A test of the full model with all predictors against a constant-only model was statistically significant (χ^2_{11}=151.1; n=318; *P*<.001; R^2_L=0.34), meaning that, as a set, the predictors reliably distinguished between people who had participated in the ALS ice bucket challenge and those who had not. Independent removal of 2 of the 11 predictors significantly harmed the model fit, specifically experiential attitude ($\Delta\chi^2_1$=20.4; *P*<.001) and descriptive norms ($\Delta\chi^2_1$=9.6; *P*=.003). Figures 7 and 8 illustrate the form of these relationships.

Table 4. Results of binomial logistic regression model predicting amyotrophic lateral sclerosis ice bucket challenge participation.

Predictor[a]	*B* (SE)	$\Delta\chi^2$*(df)*	Odds ratio (95% CI)
Constant	0.05 (0.23)	N/A[b]	N/A
Age group[c]	−0.26 (0.30)	0.7 (1)	0.78 (0.43-1.40)
Experiential attitude	0.66 (0.16)	20.4 (1)**	1.94 (1.44-2.69)
Instrumental attitude	−0.25 (0.13)	4.0 (1)	0.78 (0.60-1.00)
Injunctive norm	0.28 (0.18)	2.4 (1)	1.33 (0.93-1.91)
Descriptive norm	0.61 (0.20)	9.6 (1)*	1.84 (1.25-2.79)
Perceived control	−0.05 (0.25)	0.1 (1)	0.95 (0.58-1.56)
Self-efficacy	0.15 (0.20)	0.6 (1)	1.16 (0.79-1.72)
Experiential attitude × value of experiential attitude	−0.15 (0.14)	1.1 (1)	0.86 (0.65-1.13)
Instrumental attitude × value of instrumental attitude	0.05 (0.07)	0.6 (1)	1.05 (0.93-1.20)
Injunctive norm × motivation to comply	0.06 (0.14)	0.2 (1)	1.06 (0.80-1.40)
Descriptive norm × motivation to comply	−0.24 (0.14)	3.0 (1)	0.79 (0.60-1.03)

[a]Model χ^2_{11}=151.05; n=318; R^2_L=0.34; null −2 Log likelihood=440.63; model 2 Log likelihood with predictors=289.58.

[b]N/A: not applicable.

[c]Age group was a dummy variable where 0=under 18 years old and 1=from 18 to 35 years old.
*P<.01; **P<.001.

Figure 7. The relationship between experiential attitude and probability of amyotrophic lateral sclerosis ice bucket challenge participation.

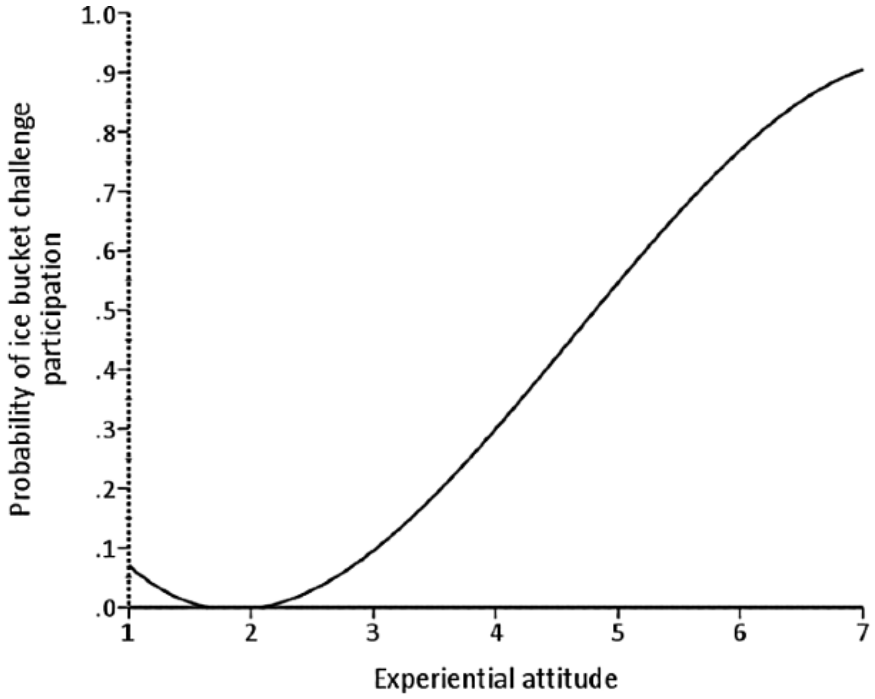
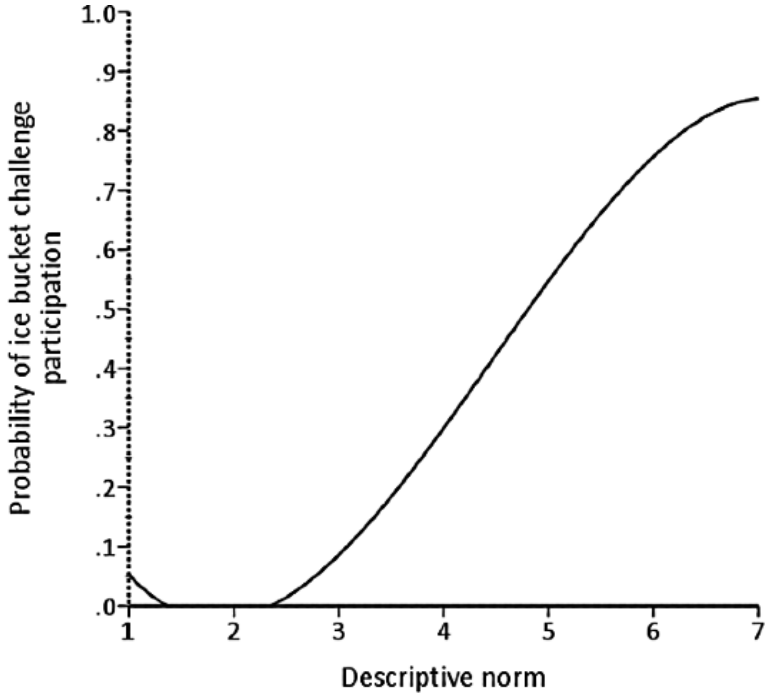


Figure 8. The relationship between descriptive norm and probability of amyotrophic lateral sclerosis ice bucket challenge participation.



Discussion

Overview

To our knowledge, this is the first study to quantitatively investigate the theoretical constructs for predicting participation in web-based challenges using data provided by actual participants. This study aimed to investigate the behavioral beliefs of people who have participated in these challenges and compare them with the beliefs of people who did not. Thus, we identified potential factors that were critical to the participants' final decision. The results showed the attitude subconstructs, the perceived norm subconstructs, and the interaction between injunctive norm and motivation to comply to be good predictors of cinnamon challenge participation. In addition, the experiential attitude and the descriptive norm are good predictors of ALS ice bucket challenge participation.

Cinnamon Challenge

The analysis showed that attitude and perceived norm subconstructs (hypotheses 2, 5, and 6) are strong predictors of cinnamon challenge participation. This finding is consistent with other studies that used IBM to predict other behaviors such as condom use, which also found these 2 constructs to be the

strongest predictors [34,44]. As seen in the *Results* section, the relationship between instrumental attitude, injunctive norm, and descriptive norm and probability of participating in the cinnamon challenge is proportional. The positive relationship between instrumental attitude and probability of participation indicates that the more people perceive enjoyment and rewards involved in the cinnamon challenge, the more willing they were to engage in the challenge. This shows that those people thought the challenge was easy, with minimal harmful consequences. In addition, the positive relationship between injunctive norms and the probability of participation shows that the more perceived attention paid to the challenge by the public, the higher the probability of participants engaging in the cinnamon challenge because they believe their videos will receive more views. In addition, our findings suggest that there is a positive relationship between descriptive norms and the probability of participating in the cinnamon challenge. This relationship means that the less attention participants received from people around them, warning them about participating in the challenge, the higher the chance they would engage in the challenge. Consequently, it appears that the more the peers were engaging in the challenge, the higher the likelihood that participants would engage in the challenge, as they may have believed it is a common behavior that is *okay* to do. These findings are similar to a previous study on criminal behavior, highlighting the significant role that culture plays in committing crime or violent behavior [45]. In other words, in a culture where crimes occur frequently, there is a higher chance of more people committing more crimes and violent behaviors in the future.

In addition, testing hypothesis 7 showed that there is a significant interaction between injunctive norms and motivation to comply. The interaction implies that there is a positive relationship between injunctive norms and the probability of participation in the cinnamon challenge. However, this relationship is stronger for those with low motivation to comply with scores. This means that people with low motivation to comply with predominant social norms are more likely to participate in the cinnamon challenge than those with high motivation to comply. This finding is different from most of the literature on human behavior, which suggests the opposite of our findings. This is mainly because of the negative nature of the behavior that this study investigates, which involves self-harm. For example, a person with low motivation to comply with their parents is more likely to commit a self-harm behavior than someone with high motivation to comply with their parents.

Analysis of the change in model fit after removing each of the significant predictors indicated that the injunctive norm explains most of the variability in the probability of cinnamon challenge participation, followed by instrumental attitude and descriptive norm. Thus, interventions to reduce participation in similar challenges in the future should focus on these constructs, with greater emphasis on the injunctive norm, as it is the stronger predictor. This could be done by having people adolescents trust send persuasive messages highlighting the consequences of challenge behavior and explaining why they should not engage in these activities [46,47]. In addition, as there is a significant interaction between injunctive norms and motivation to comply, intervention development should consider both of these factors simultaneously. Changing only 1 of these 2 factors may lead to an undesired or unintended effect on other's impact on challenge participation. The intervention should specifically mention the disapproval of such behaviors from those around us, even those who say they do not comply or *care* about what others say.

Ice Bucket Challenge

Unlike cinnamon challenge participation, only the experiential attitude and descriptive norm significantly predicted ALS ice bucket challenge participation (hypotheses 2 and 6). In other words, adolescents and young adults primarily participated in this challenge for two reasons (1) enjoyment or popularity (getting more views and likes on social media) and (2) a sense of obligation due to the large number of participants completing the challenge, which made them feel that it's the norm to do so [48-51]. We believe that other factors were not significant because of the positive nature of the challenge. For example, even people who did not participate in the challenge generally rated it as easy to perform and believed that they were capable of completing it. These beliefs may explain why perceived control and self-efficacy factors were not found to be significant predictors of ALS ice bucket challenge participation. In addition, as this challenge, in particular, was very popular, even people who chose not to participate generally indicated that everyone around them would approve of their participation. This explains why the injunctive norm factor was not found to be a significant predictor of ALS ice bucket challenge participation.

Among the significant predictors of ALS ice bucket challenge participation, experiential attitude explained the largest amount of variability, followed by the descriptive norm. These findings can help develop or market other philanthropic challenges by focusing on making them enjoyable with obvious direct rewards and emphasizing the attention given to them by the public. By developing a challenge that targets these beliefs more than the others in IBM, one could potentially create a philanthropic challenge that goes viral and leaves a health-promoting impact on society with minimal harmful consequences.

Limitations and Future Work

This study has several limitations. Only 2 challenges were used to represent all other similar challenges. This could limit the generalizability of the findings; hence, future work could investigate the applicability of these findings to other challenges. Moreover, this study was retrospective and cross-sectional in nature, making it difficult to draw conclusions about causal relationships between the predictors and outcomes. Future work could study the impact of the constructs in controlled settings by developing interventions and examining their effects on people's willingness to participate in social media challenges.

Conclusions

A theoretical framework was used to guide the study design and to inform the development of theory-driven intervention efforts to change social media challenge participation intention and behavior. The cinnamon challenge was used to represent challenges with a harmful impact, and the ALS ice bucket challenge was used to represent positive-impact challenges. The constructs that were critical to the participants' decision to participate were identified. This study provides a good

theoretical model to understand the phenomenon of social media challenges. In addition, the findings provide information about which constructs should be the focus of intervention efforts. The content and thrust of those intervention efforts must be based on knowledge of how the specific items making up each construct apply specifically to social media (eg, the desire to get likes and affirmation and the social norms that are portrayed via media, videos, and images).

Acknowledgments

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Conflicts of Interest

None declared.

Multimedia Appendix 1
Items used to measure research constructs.
[PDF File (Adobe PDF File), 122 KB - pediatrics_v4i1e24988_app1.pdf]

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Abbreviations

ALS: amyotrophic lateral sclerosis
IBM: integrated behavioral model

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Original Paper

Accuracy and Monitoring of Pediatric Early Warning Score (PEWS) Scores Prior to Emergent Pediatric Intensive Care Unit (ICU) Transfer: Retrospective Analysis

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Abstract

Background: Current approaches to early detection of clinical deterioration in children have relied on intermittent track-and-trigger warning scores such as the Pediatric Early Warning Score (PEWS) that rely on periodic assessment and vital sign entry. There are limited data on the utility of these scores prior to events of decompensation leading to pediatric intensive care unit (PICU) transfer.

Objective: The purpose of our study was to determine the accuracy of recorded PEWS scores, assess clinical reasons for transfer, and describe the monitoring practices prior to PICU transfer involving acute decompensation.

Methods: We conducted a retrospective cohort study of patients ≤21 years of age transferred emergently from the acute care pediatric floor to the PICU due to clinical deterioration over an 8-year period. Clinical charts were abstracted to (1) determine the clinical reason for transfer, (2) quantify the frequency of physiological monitoring prior to transfer, and (3) assess the timing and accuracy of the PEWS scores 24 hours prior to transfer.

Results: During the 8-year period, 72 children and adolescents had an emergent PICU transfer due to clinical deterioration, most often due to acute respiratory distress. Only 35% (25/72) of the sample was on continuous telemetry or pulse oximetry monitoring prior to the transfer event, and 47% (34/72) had at least one incorrectly documented PEWS score in the 24 hours prior to the event, with a score underreporting the actual severity of illness.

Conclusions: This analysis provides support for the routine assessment of clinical deterioration and advocates for more research focused on the use and utility of continuous cardiorespiratory monitoring for patients at risk for emergent transfer.

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KEYWORDS

pediatric intensive care unit; cardiorespiratory monitoring; hospital transfer; clinical deterioration; monitoring; ICU; intensive care unit; pediatric; retrospective; detection; deterioration; child; accuracy; cohort

Introduction

Events of clinical deterioration leading to emergent pediatric intensive care unit (PICU) transfer can have dire consequences for children [1,2]. Children who have events of clinical deterioration while on the acute care floor can have a 13-fold increased risk of hospital mortality; increased morbidity, and longer ICU and overall hospital lengths of stay (LOS) [1,3-5]. Current approaches to identify children at risk for clinical deterioration on the acute care floor include the use of early warning scoring systems, such as the Pediatric Early Warning Score (PEWS), to offer a “triggering” threshold based on

physiological severity of illness parameters leading to escalations in care or the use of rapid response teams [6-10].

Despite the widespread use of PEWS, there has been variability in implementation and standard use. In a retrospective study conducted by Akre and colleagues [7], 85.5% of children with a rapid response team or code event leading to emergent ICU transfer had a PEWS score in the critical range documented many hours (median 11 hours, 36 minutes) prior to the event of interest, suggesting there may be challenges with routine assessments, incomplete observations, lack of standardized scoring between clinicians, establishing situational awareness of changing risk scores, or uncertainty in how to initiate an appropriate proactive clinical action [7,11-14]. Further, children likely deteriorate for many different reasons, and a single score is unlikely to detect them all equally well [15]. These reasons may be why the PEWS score has not been shown to decrease hospital mortality despite its utility in initiating rapid response team intervention [10,16].

Further complicating early warning assessment is the unresolved and debated utility of continuous cardiorespiratory monitoring for children on the acute care pediatric floor [17-19]. One specific exemplar where the guidance on continuous cardiorespiratory monitoring is not clear includes hospitalized children with bronchiolitis who have been recently deescalated from supplemental oxygen. The American Academy of Pediatrics Clinical Practice Guideline for this population suggests that the potential benefits of forgoing continuous respiratory monitoring include shorter LOS, decreased alarm fatigue, and decreased cost, whereas the potential harms include delayed detection of hypoxemia and a delay in appropriate weaning of oxygen. The overall continuous monitoring recommendation for hospitalized children with bronchiolitis in the absence of oxygen therapy is labeled a weak recommendation [20]. When McCulloh and colleagues [21] conducted a randomized controlled trial to assess outcomes associated with intermittent versus continuous pulse oximetry for nonhypoxemic infants admitted for bronchiolitis, they found that there was no difference in LOS or use of therapeutic measures between the 2 groups. Parents of children hospitalized for bronchiolitis perceive that the presence of continuous pulse oximetry monitoring is reassuring [22]. Physiological deterioration can happen to a child between routine 8-hour vital sign assessments, and further refinement on who could benefit the most from continuous cardiorespiratory monitoring for early detection of clinical deterioration is an area of much needed clarification.

There is still much to be learned about how PEWS and continuous cardiorespiratory monitoring are used in routine practice environments. We were also particularly interested in how both PEWS and continuous respiratory monitoring were used prior to clinical deterioration in a sample that required emergent PICU transfer and initiation of therapy escalation (ie, “rough” PICU transfer). The purpose of our study was to (1) determine the clinical reason for emergent transfer, (2) quantify the frequency of physiological monitoring prior to transfer, and (3) assess the values, timing, and accuracy of the PEWS scores 24 hours prior to transfer.

Methods

We conducted a retrospective cohort study of patients ≤21 years of age transferred emergently from the acute care pediatric floor to the PICU due to clinical deterioration from January 2011 to July 2019 in the University of Virginia Children’s Hospital. Emergent transfers with clinical deterioration were defined as those children or adolescents requiring (1) emergent intubation and mechanical ventilation, (2) initiation of vasopressors, (3) stat transfusion of more than one blood product, or (4) transfer following cardiac arrest on the acute care floor. For all transfers not associated with cardiac arrest criteria (items 1-3), they had to be initiated either prior to transfer or within 12 hours of PICU transfer. Bonafide and colleagues [3] previously developed a clinical deterioration metric using the initiation of mechanical ventilation or vasopressors within 12 hours of transfer. We added the criterion of rapid transfusions to capture deteriorating postsurgical cases and unstable hematology-oncology conditions. Clinical notes and orders were adjudicated for each eligible child to ensure that the PICU transfer was due to clinical deterioration and not for planned procedures or postoperative transfers. Children and adolescents also had to be admitted to the acute care floor long enough to have routine care established (at least 6 hours).

To determine the indication for transfer, all available notes for the admission of interest were reviewed by RLK following adjudication definitions used by Blackwell and colleagues [15]. Reasons for clinical deterioration included respiratory distress (leading to emergent intubation or mechanical ventilation), concern for or worsening infection, bleeding or anemia requiring transfusion, cardiac arrest, seizure, stroke, unplanned surgery, or other reasons. These categories were not mutually exclusive, and a child or adolescent could have more than 1 reason for the transfer. Determination of physiological monitoring status was obtained by reviewing the order sets (a medical order for continuous telemetry or pulse oximetry monitoring) prior to the time of transfer to determine if the child or adolescent had an order for continuous telemetry or pulse oximetry monitoring or intermittent vital sign monitoring (every 1, 2, 4, or 8 hours).

Finally, the 3 PEWS scores documented in the electronic medical record (EMR) prior to transfer were abstracted. The University of Virginia implemented a modified PEWS score (Table 1) beginning in 2012 as a part of routine clinical care with the expectation that it was to be completed at every routine nursing assessment and vital sign acquisition. The modified PEWS score closely resembles the Monaghan PEWS score [6] and the automated PEWS (AutoPEWS) score that has been tested for integration within EMR [23]. To determine accuracy, we used the time-concordant heart rate and respiratory rate to determine if any of the categories were underscored or overscored. Of note, capillary refill could not be reliably adjudicated so the scores were only compared for correctness with the available vital sign parameters. Other clinical variables abstracted include age and overall LOS. Descriptive statistics were calculated using R (2019; R Core Team, Vienna, Austria).

Table 1. Modified Pediatric Early Warning Score (PEWS).

Category	Score			
	0	1	2	3
Behavior	Playing, appropriate, at patient's baseline	Inappropriately sleepy *or* fussy but consolable	Irritable *or* inconsolable	Lethargic/confused *or* reduced response to pain
Cardiovascular	Pink *or* capillary refill <2 seconds	Pale *or* capillary refill 3-4 seconds *or* mild tachycardia *or* single ventricle shunted (BT[a] shunt or Norwood/Sano)	Gray/dusky *or* capillary refill 4-5 seconds *or* moderate tachycardia	Gray/mottled *or* capillary refill >5 seconds *or* severe tachycardia *or* new onset bradycardia
Respiratory	Within normal parameters, no retractions	Mild tachypnea *or* using accessory muscles *or* >30% FiO_2 or >0.5 L/min/kg	Moderate tachypnea *or* retractions *or* >40% FiO_2 or >1 L/min/kg	Severe tachypnea *or* RR[b] < normal for age *or* >50% FiO_2 or >1.5 L/min/kg

[a]BT: Blalock-Taussig.

[b]RR: respiratory rate.

Results

We found 72 cases of emergent PICU transfer due to clinical deterioration. The median age was 2.3 years (25% 7.6 months, 75% 11.4 years), and the majority of the children were less than 12 months of age.

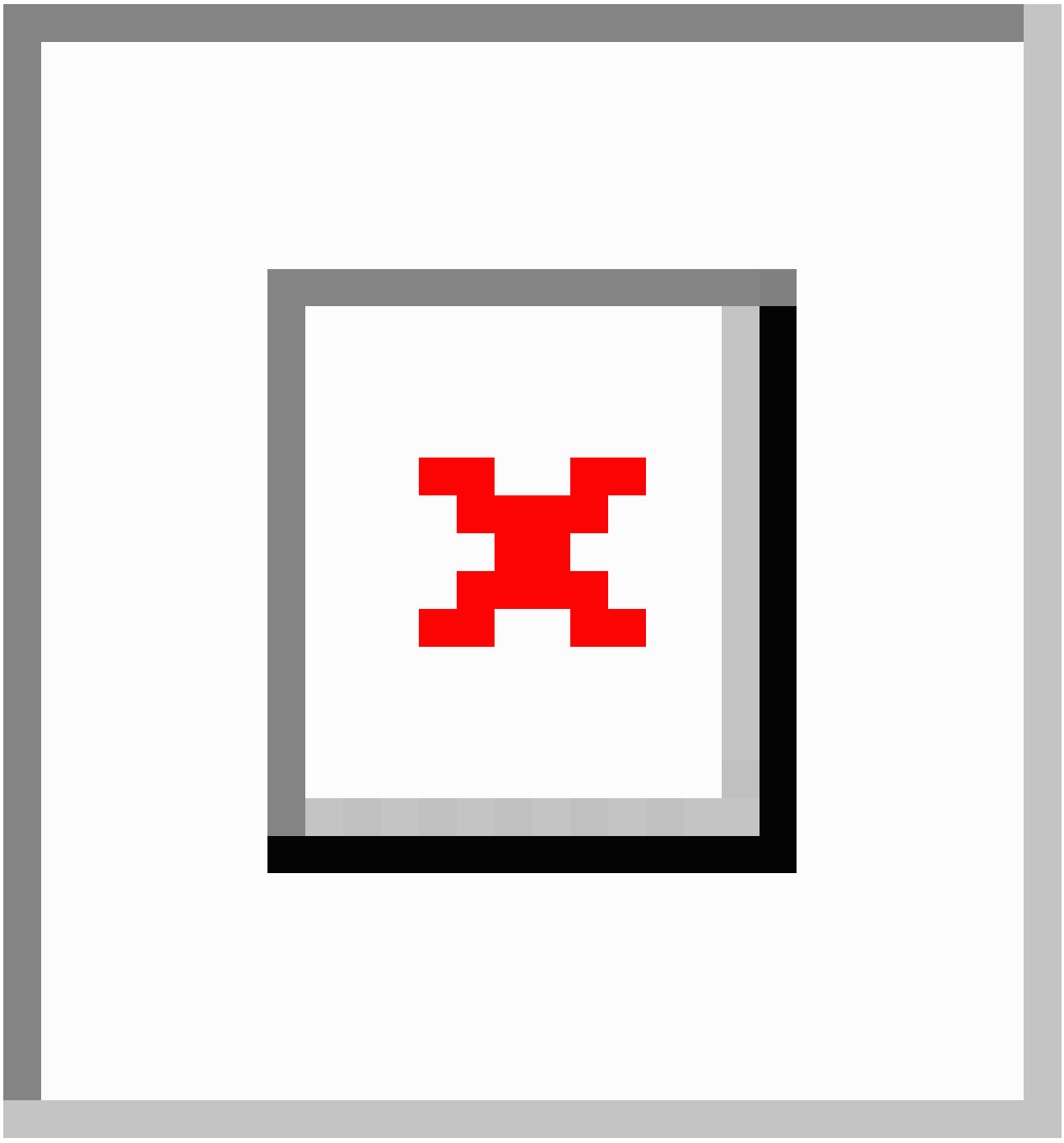
The median LOS on the acute care floor prior to transfer was 1.4 days (25% 0.5 days, 75% 2.8 days); 31 children and adolescents (31/72, 43%) transferred within 24 hours of arrival, and 44 (44/72, 66%) transferred within 48 hours of arrival to the acute care floor. The children were severely ill at the time of transfer. Within 6 hours of transfer to the PICU, 54 (54/72, 75%) of patients were emergently intubated, 15 (15/72, 21%) were rapidly transfused, 9 (9/72, 13%) were given vasopressors, and 1 (1/72, 1%) patient experienced a cardiac arrest while on the acute care floor.

Respiratory distress was the most common indication for transfer (36/72, 50%), followed by infection (28/72, 39%), bleeding or anemia requiring transfusion (8/72, 11%), uncontrolled seizure (4/72, 6%), stroke (2/72, 3%), unplanned surgery (2/72, 3%), and other reasons (14/72,19%). Prior to the PICU transfer, only 25 (25/72, 35%) of patients were continuously monitored; 33 (33/72 46%) of the patients had vital signs ordered every 4 hours, 10 (10/72, 14%) had vital signs ordered every 8 hours, and 3 (3/72, 4%) had vital signs ordered every 1 or 2 hours. The overall LOS was long, and the median time in the hospital was 24 days for the sample (25% 10.8, 75% 45.8 days). The mortality of children who were emergently transferred was high (15/72, 21%).

Only 56 of the 72 patients (78%) had documented PEWS scores prior to emergent PICU transfer, and the mean time of the last PEWS score documented prior to transfer was 3.0 hours (SD 3.2 hours). Patients who were not being continuously monitored had higher documented PEWS scores across all 3 time points. In the last recorded PEWS score prior to transfer, those in the group that were continuously monitored had nearly a full point lower average PEWS score (mean 2.2, SD 2.4) compared to those who were not being continuously monitored (mean 3.2, SD 2.2), indicating an increased severity of illness prior to transfer in the noncontinuously monitored group (P=.15). There were no clinically nor statistically significant differences in the last recorded PEWS score prior to PICU transfer between children who died and those who did not.

Figure 1 shows several elements relating to the 3 PEWS scores before transfer. The major finding is that nearly half of the sample (26/72, 47%) had at least 1 incorrectly recorded score in the 24 hours prior to emergent PICU transfer, and all of the errors were underscored PEWS values, meaning that the recorded score in the EMR was less than what it should have been if calculated accurately.

Figure 1. Pediatric Early Warning Score (PEWS) scores prior to emergent transfer. The ordered sequence of PEWS scores in the electronic medical record prior to PICU transfer are shown, indexed in the order they occurred, and the size of the data points is proportional to the score itself. Correct: score was recorded and accurate; Incorrect: score was recorded, but lower than the recalculated value; N/A: score was not recorded.



Discussion

This analysis presents a description of the accuracy of the documented PEWS score and continuous monitoring presence prior to events of clinical deterioration. Early warning scores like PEWS intend to enable clinicians to act early in recognizing clinical deterioration in children. Faced with an already complex workflow, clinicians need to be able to systematically calculate accurate scores, trust the scores, and develop standard practices for proactive care [11,24]. Some of that trust will lie in their availability, accuracy, and ability to determine trends over time. In this retrospective review of emergent PICU transfers, we found that more than 20% of cases had no PEWS recorded, and nearly half of those recorded were underscored, thereby underestimating the actual risk of the child for deterioration. Our finding was similar to the work of Chapman and colleagues [14] who found that only 36% of their sample had adequate vital signs documented to calculate a PEWS score, and when documented, nearly 20% of the PEWS scores contained an error. In their sample, underscoring was more common than overscoring, and 9% of the inaccuracies were deemed clinically significant [14]. Further, when Trubey and colleagues [25] conducted a systematic review of the validity and effectiveness of pediatric early warning systems, they found that the completeness of documentation and interrater reliability of the

score varied widely, with some studies only achieving 67% agreement. While an evaluation of the differential accuracy of higher PEWS scores versus lower PEWS scores has not been delineated, it may be that higher respiratory and heart rates (thus higher PEWS scores) may have greater variability in accuracy. This is an area of needed further inquiry.

In addition to the incomplete documentation and inaccuracies in reporting, we found that the PEWS scores were often documented many hours prior to the actual PICU transfer event, indicating incomplete assessments in the hours immediately preceding the transfer when the scores could have been the most helpful in providing early warning of clinical deterioration. Further, the majority of children demonstrated clinical deterioration within 48 hours of arrival to the acute care floor. This finding emphasizes the known challenges with prognostication, defining clinical acuity, and determining the appropriate level of care [26,27]. We found many clinical reasons for deterioration, supporting the notion that there is unlikely to be a single early warning score that adequately captures all types of decompensation [15]. Further, there is substantial heterogeneity in ages represented in any pediatric sample. When Spaeder and colleagues [28] developed a machine learning model to predict early onset of pediatric sepsis, they found that parameters performed differently in the model given the age of the pediatric patient, again indicating that no one model likely performs equally well in all age ranges represented in pediatric care (neonate, infant, child, adolescent).

We note that very few of our patients were continuously monitored with telemetry or pulse oxygenation prior to their emergent PICU transfer. Additionally, those without a continuous monitoring order had higher recorded PEWS scores prior to transfer than those with continuous monitoring, indicating that clinicians may be missing important changes in the underlying physiology when relying on intermittent vital sign assessment alone. Continuous monitoring in children can be challenging to implement because it can be difficult to keep continuous monitoring leads and probes on mobile children, and previous estimates have demonstrated as few as 1% of alarms in children are clinically meaningful [29]. We speculate that there can be clinical benefit to shifting the clinical monitoring paradigm away from its use only as a means of responding to critical physiological alarms and towards a means for early detection of clinical deterioration using continuous predictive analytics monitoring so clinicians can initiate proactive clinical actions [28,30,31]. To avoid medical overuse and further contribution to false alarms, there is a defined need to determine the populations that could benefit the most from continuous cardiorespiratory monitoring in the acute care pediatric setting while also determining the correct “dose” of continuous cardiorespiratory monitoring for those at risk of clinical deterioration.

PEWS, like all point scores of its kind including the Pediatric Rothman Index [32], takes snapshots of clinical status at the time of nurse vital sign assessments. Much can happen clinically between these intermittent events—here, there is a role for continuous physiological monitoring as a means for detecting clinical deterioration. In adults at risk for ICU transfer, metrics extracted from advanced mathematical analyses of monitoring data added information to vital signs and lab tests in early detection of clinical deterioration [31]. Further, continuous predictive analytics monitoring does not rely on arbitrary thresholds of risk cutoff and can incorporate small changes in vital signs, electrocardiogram changes, and laboratory findings, which may cumulatively present a different and more accurate representation of overall risk and represent various clinical etiologies for decompensation [15,33,34].

There are several limitations of this analysis that must be noted. Data collection was limited to 1 tertiary academic children’s hospital with a high proportion of children with medical complexity, including complex cardiac surgical cases; therefore, the results may not be generalizable to other settings. Additionally, the sample size was small because we chose a strict classification of emergent PICU transfer with clinical deterioration. Finally, the clinical abstraction was limited to what was available as documentation in the medical record, and there were many instances of a lack of documentation of the PEWS score. Further, there may be changes in continuous monitoring practices without documentation of a written order based on clinical severity at the time of presentation.

This analysis provides support for the routine assessment of clinical deterioration and advocates to extend current monitoring paradigms with the development of continuous predictive analytics monitoring for patients at risk of clinical instability and emergent transfer. It also suggests that more study is needed to determine “who, when, and how much” continuous telemetry or pulse oximetry monitoring should be used and may be the most beneficial for higher risk children and adolescents.

Conflicts of Interest

JRM is Chief Medical Officer and stockholder in AMP3D, Charlottesville, VA and a shareholder in Medical Predictive Science Corporation, Charlottesville, VA.

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Abbreviations

EMR: electronic medical record
LOS: length of stay
PEWS: Pediatric Early Warning Score
PICU: pediatric intensive care unit

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Original Paper

Parents' and Students' Perceptions of Telepractice Services for Speech-Language Therapy During the COVID-19 Pandemic: Survey Study

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Abstract

Background: The ongoing COVID-19 pandemic has resulted in the suspension of face-to-face classes and a considerable increase in the use of telepractice services in speech-language pathology. However, little is known about parents' and students' satisfaction with telepractice services and their preferences for different service delivery modes. These factors may affect therapy effectiveness and the future adoption of telepractice.

Objective: We evaluated students' and parents' perceptions of telepractice efficacy and their preferences for different service delivery modes (ie, on-site practice vs telepractice). We also identified factors that affect parents' and students' preferences for different service delivery modes during the COVID-19 pandemic.

Methods: A 19-question survey on telepractice satisfaction and preferences was administered to 41 Hong Kong Chinese students and 85 parents who received telepractice services from school-based speech-language pathologists during the COVID-19 class suspension period. In addition to providing demographic information and data on the implementation of telepractice services, all participants were asked to rate their perceptions of the efficacy of telepractice services and compare on-site practices to telepractice on a 5-point Likert scale (ie, 1=strongly disagree/prefer the use of on-site speech-language therapy services and 5=strongly agree/prefer the use of telepractice services).

Results: Despite the fact that telepractice efficacy was highly rated by parents (95% CI 3.30-3.66) and students (95% CI 3.21-3.76), both groups believed that telepractice was less effective than on-site practices (parents: 95% CI 2.14-2.52; students: 95% CI 2.08-2.65). Moreover, parents preferred on-site practices over telepractice (95% CI 2.04-2.43), whereas students did not prefer one mode of practice over the other (95% CI 2.74-3.41). A significant association between telepractice efficacy and a preference for telepractice services was found only among the students (τ=.43, P<.001), not the parents (τ=.07; P=.44).

Conclusions: Although telepractice is an acceptable alternative service delivery option for providing speech and language therapy services to school-aged individuals, speech-language therapists and parents must play a more proactive role in telepractice services to facilitate effective communication between clinicians and parents.

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KEYWORDS

eHealth; telepractice; speech and language pathology; user satisfaction; COVID-19; school-based service

Introduction

As of January 2021, over 90 million people have been infected with the SARS-CoV-2 virus. This has necessitated social distancing and school closures worldwide. As a result, telehealth (ie, the use of audio or videoconferencing technology to provide health care services) has received increasing attention. Telehealth care has been regarded as an alternative to

face-to-face care in many countries [1,2]. Furthermore, speech-language pathologists have engaged in telepractice over the past 2 decades in various countries [3-6]. The efficacy of telepractice has been supported by scientific research on speech, language, voice, and fluency disorders across different age groups [7-9]. Additionally, telepractice has been deemed valid and effective by different professional organizations [10,11]. With the COVID-19 pandemic seriously disrupting the provision of speech and language therapy services, telepractice services have been increasingly adopted and regarded as the best option for delivering speech and language therapy during the pandemic [12,13].

Despite the increasing adoption of telepractice in schools, various stakeholders have held different beliefs about telepractice. Although several surveys have shown that school-based speech-language pathologists doubt the efficacy of telepractice, others have revealed a positive attitude after using telepractice services [12,14,15]. However, parents' and children's perceptions of telepractice are not well understood. A few studies have examined parents' and students' satisfaction with telepractice programs, but the findings have been mixed. In a pilot survey, 13 teachers and 8 parents from a remote school were highly satisfied with the progress brought about by telepractice [8]. Positive findings were also noted in parents' and students' responses to a survey on web-based speech and language interventions that were conducted by university clinics [8,16]. In contrast, an interview study of 5 parents raised concerns about poor telepractice engagement by students and ineffective communication between parents and clinicians in telepractice services [17]. These factors may lower people's acceptance of school-based telepractice services [17]. Given the high rate of telepractice adoption in school settings during the pandemic [12,13], a survey study on parents' and students' satisfaction with telepractice could reveal the perceived efficacy of these services.

Perceived efficacy is an important measure in speech and language therapy for both on-site practices and telepractice, because it reflects the effectiveness of the therapy and students' and parents' motivations for undergoing the therapy [18,19]. The Davis' Technology Acceptance Model also argues that perceived efficacy, which is based on perceived usefulness and convenience, influences the future adoption of technology [20]. Perceived efficacy can be reflected by people's engagement with therapy sessions, which correlates with children's treatment outcomes [21]. Moreover, the amount of therapeutic skills that families practice during their daily routine and the collaboration between clinicians and parents affect the generalization of treatment [22]. Therefore, investigating parents' and students' perceptions of telepractice efficacy and their involvement with telepractice and daily therapeutic practices are critical for evaluating treatment fidelity.

Previous studies have largely focused on students' and parents' satisfaction with research-oriented telepractice, but none have investigated clients' and parents' preferences for different modes of practice. Since service delivery modes have expanded during the pandemic, students' and parents' preferences for different delivery models are critical for designing a future service delivery model for school-based speech and language therapy services. Thus, in this study, we examined how clients' therapy characteristics, including age, comorbidity, and parent support, influence their preferences for different modes of service. This information may inform speech-language pathologists about selecting appropriate students for telepractice services [10].

In summary, the following 3 research questions were addressed in this satisfaction survey study: (1) what are parents' and students' perceptions of telepractice efficacy; (2) do parents and students prefer on-site practices or telepractice; and (3) what are the critical factors that affect parents' and students' preferences for different service delivery modes?

Methods

Survey Design and Development

Survey Summary

We developed a web-based survey for both parents and students to evaluate school-based speech and language therapy practices in Hong Kong (see Multimedia Appendix 1). To meet internal clarity, construct, and content validity criteria, all survey questions were independently reviewed by 3 school-based speech-language pathologists. This review ensured that the survey's wording, content, and question order were clear and appropriate. The survey questions were revised and finalized in accordance with the speech-language pathologists' suggestions. All respondents completed the survey in about 10 minutes. Ethics approval was granted by the Human Research Ethics Committee of University of Hong Kong, and participants signed consent forms before completing the survey.

The survey for parents and students consisted of 4 sections, including (1) the implementation of telepractice, which consisted of 2 items; (2) telepractice efficacy, which consisted of 7 items for parents and 4 items for students; (3) the comparison between telepractice and on-site practice, which consisted of 6 items for parents and 5 items for students; and (4) demographics, which consisted of 5 items. All responses for sections 2 and 3 were based on Likert-type scale scores, which ranged from 1 (ie, strongly disagree) to 5 (ie, strongly agree).

Section 1: Implementation of Telepractice

The 2 items in this section assessed the amount of therapy students received and how frequently students used telepractice services during the COVID-19 class suspension period.

Section 2: Telepractice Efficacy

The 7-item survey for parents included questions about whether telepractice was effective in enhancing their child's language skills, meeting their child's needs, engaging with their child, and providing satisfaction with the amount of therapy their child received (Cronbach α=.94). The 4-item survey for students included questions about whether telepractice services met their needs and whether they enjoyed telepractice services (Cronbach α=.84).

Section 3: Comparison Between Telepractice and On-site Practice

The 6-item parent survey included questions about whether telepractice services for speech therapy provided better

communication than on-site speech and language therapy. There were also questions regarding the implementation of home therapy practices (Cronbach α=.89). The 5-item student survey included questions about whether students learned better language skills and exhibited better engagement with on-site practices than with telepractice (Cronbach α=.88).

Section 4: Demographics

The 4 items in this section were used to collect information on each student's grade, gender, special education needs status, and family income.

Participants

From July to August 2020, 85 parents (ie, 75 mothers and 10 fathers) and 41 students (ie, 7 girls and 34 boys) participated in our web-based survey. Based on the last 4 digits of participants' telephone numbers, 27 families participated in both the parent and student surveys. These 27 families accounted for the 31% (27/85) of parents and 65% (27/41) of students who participated. The families who responded to both the parent and student questionnaires represented students from Grades 1-7 (parents' questionnaire: median=Grade 3; students' questionnaire: median=Grade 4). In terms of students' comorbidities in the parent survey, the most prevalent special educational needs subtype was autism spectrum disorder (53/85, 62%), followed by attention deficit/hyperactivity disorder (33/85, 38%), specific learning difficulties (20/85, 23%), intellectual disabilities (3/85, 3%), hearing impairment (2/85, 2%), visual impairment (1/85, 1%), and physical disabilities (1/85, 1%). Additionally, 12% (11/85) of students had no comorbidities except for speech and language disorders. In terms of students' comorbidities in the student survey, the most prevalent special educational needs subtype was autism spectrum disorder (24/85, 58%), followed by attention deficit/hyperactivity disorder (15/41, 36%), specific learning disorders (6/41, 14%), intellectual disabilities (1/41, 2%), and visual impairment (1/41, 2%). Additionally, 21% (9/41) of students had no comorbidities except for speech and language disorders. Around half of the participants (parents' survey: 42/85, 49%; students' survey: 22/41, 53%) had an average monthly family income that fell below the median for average household income (ie, around US $3290).

To achieve a Cronbach α value of .05 and a moderate effect size (ie, Cohen *d*=0.5), a statistical power of .99 and .86 was needed for 85 parents and 41 students, respectively. This was determined by using G*Power 3 software (G*Power Team) [23]. In addition, good quality results can be obtained by performing a factor analysis on samples with at least 50 people or samples with a factor loading value of >.60 [24].

Results

Implementation of Telepractice

Most students reported that they had fewer than 5 telepractice sessions during the pandemic (parents' survey: 73/85, 85%; students' survey: 31/41, 75%). In terms of session frequency, the most common amount of therapy was 1 session per month (parents' survey: 35/85, 41%; 36%; students' survey: 15/41, 36%), followed by 1 session per 2 weeks (parents' survey: 25/85, 29%; students' survey: 15/41, 37%), and 1 session per week (parents' survey: 21/85, 24%; students' survey: 12/41, 29%).

Telepractice Efficacy

Parents and students had positive views of the efficacy of telepractice with respect to their understanding of the treatment goals (parents: mean 3.48, SD 0.84; 95% CI 3.30-3.66; students: mean 3.49, SD 0.87; 95% CI 3.21-3.76) and the ability of telepractice services to meet the needs of students (parents: mean 3.24, SD 1.03; 95% CI 3.01-3.46; students: mean 3.49, SD 0.84, 95% CI 3.22-3.75). Based on the parents' responses, parents had positive views of students' enjoyment of telepractice services (mean 3.29, SD 1.14; 95% CI 3.05-3.54) and the ability of telepractice services to enhance students' language abilities (mean 3.33, SD 1.01; 95% CI 3.11-3.55). Based on the students' responses, students had a neutral view of telepractice efficacy with regard to (1) enjoyment (mean 3.32, SD 1.08; 95% CI 2.98-3.66) and (2) language ability enhancement (mean 3.29, SD 0.96; 95% CI 2.99-3.59). Independent 2-tailed sample *t* tests revealed that there were no significant differences in the above views between parents and students (enjoyment: *P*=.92; understanding of treatment goals: *P*=.97; meeting students' needs: *P*=.18; language ability enhancement: *P*=.85). In addition, parents held a positive view of the progress that students made during telepractice services (mean 3.35, SD 0.96; 95% CI 3.15-3.56) and a neutral view of the amount of therapy that students received (frequency: mean 2.99, SD 1.04; 95% CI 2.76-3.21; amount of therapy: mean 3.21, SD 1.03; 95% CI 2.99-3.43).

Factors That Affected Telepractice Efficacy

Our Spearman rank-order correlation analysis showed that there were no significant correlations between student grade and perceived telepractice efficacy (parents: ρ=0.03; *P*=.76; students: ρ=0.07; *P*=.65). The Bayes factor (BF) was computed to evaluate whether the evidence supported the null hypothesis over the alternative hypothesis. BF_{01} values of >3 and >10 indicated moderate and strong support, respectively, for the null hypothesis [25]. Strong evidence that supported the null hypothesis (ie, no correlation between grade and telepractice efficacy) was found in the parent group (BF_{01}=11.34), whereas moderate evidence that supported the null hypothesis was found in the student group (BF_{01}=7.84).

Comparison Between Telepractice and On-site Practice

Students' enjoyment of telepractice services and on-site services was comparable, based on the students' responses (mean 2.93, SD 1.06; 95% CI 2.59-3.26). However, students' enjoyment of telepractice services was lower in the parents' responses (mean 2.76, SD 1.02; 95% CI 2.54-2.98). Furthermore, telepractice was rated lower than on-site practice in terms of treatment effectiveness. The aspects of treatment effectiveness included the acquisition of speech and language skills (parents: mean 2.47, SD 0.92; 95% CI 2.27-2.67; students: mean 2.46, SD 0.93; 95% CI 2.17-2.76), communication with speech-language pathologists (parents: mean 2.52, SD 0.88; 95% CI 2.33-2.71; students: mean 2.32, SD 0.82; 95% CI 2.06-2.58), and treatment efficacy (parents: mean 2.33, SD 1.89; 95% CI 2.14-2.52; students: mean 2.37, SD 0.92; 95% CI 2.08-2.65). An

independent 2-tailed sample *t* test revealed no significant differences in these aspects between parents and students (enjoyment: *P*=.41; acquisition of speech and language skills: *P*=.97; communication with speech-language pathologists: *P*=.22; treatment efficacy: *P*=.83). In addition, parents rated telepractice lower than on-site practice, in terms of the implementation of therapy practices at home via telepractice services or on-site services (mean 2.46; 95% CI 2.27-2.65).

Parents had a significant negative view of telepractice, with regard to whether they preferred telepractice over on-site practice (mean 2.24; 95% CI 2.04-2.43), whereas students had a neutral view (mean 3.07; 95% CI 2.74-3.41). An independent 2-tailed sample *t* test revealed a significant difference in preferences for telepractice and on-site practice between parents and students (t_{124}=4.59; *P*<.001; *d*=0.87; 95% CI 0.48-1.26).

Factors That Affected Preferences for Telepractice and On-site Practice

Grade

Our Spearman rank-order correlation analysis showed no significant correlations between student grade and participants' preferences for the 2 service delivery modes (parents: ρ=0.07; *P*=.52; students: ρ=0.03; *P*=.85). The BF analysis showed strong evidence that supported the null hypothesis (ie, no correlation between grade and preferences for the mode of practice) in the parent group (BF_{01}=10.89), whereas moderate evidence that supported the null hypothesis was found in the student group (BF_{01}=8.17).

Treatment Efficacy

To examine the relationship between treatment efficacy and preferences for the 2 service delivery modes, we created a composite score based on the factor scores that were obtained from our exploratory factor analysis, by performing principal axis factoring extraction. As shown in Table 1, we obtained a factor score that accounted for 73% and 69% of the variance in the parent and student groups, respectively. All factor loadings were greater than .55.

Table 1. Principal axis factoring analysis of questions on telepractice efficacy. The pattern matrix for parents and students is shown.

Item	Parents[a], factor loading value	Students[b], factor loading value
Student enjoyment	.857	.552
Understanding of treatment goals	.798	.941
Meeting the needs of students	.926	.776
Enhancing speech and language abilities	.903	.819
Understanding treatment progress	.914	N/A[c]
Appropriate session frequency	.726	N/A
Appropriate session duration	.670	N/A

[a]The factor score for the parent group accounted for 73% of the variance in the items. Each item had an eigenvalue of 5.13.

[b]The factor score for the student group accounted for 69% of the variance in the items. Each item had an eigenvalue of 2.79.

[c]N/A: not applicable. These items only appeared in the parent questionnaire.

The Kendall rank correlation coefficient, τ, was computed based on the factor scores for telepractice efficacy and preferences for the mode of practice. No significant correlation was found in the parent group (τ=.07; *P*=.44); the BF for this correlation (BF_{01}=8.53) moderately supported the null hypothesis (ie, there is no correlation between telepractice efficacy and preferences for the mode of practice). A significant correlation between telepractice efficacy and preferences for the mode of practice was found in the student group (τ=.43; *P*<.001).

Discussion

Principal Findings

Unlike previous telepractice studies, which have largely focused on clinicians' attitudes, our study examined parents' and students' perceptions of telepractice efficacy and their attitudes toward telepractice during the COVID-19 pandemic. We found that students and parents were satisfied with the efficacy of treatments that were provided through telepractice services. Although students and parents had similar preferences for telepractice and on-site practice, parents preferred on-site practices. These findings are discussed in terms of telepractice efficacy and factors that affect engagement with telepractice services.

Perceived Efficacy of Telepractice

One important finding of this study was that students and parents who engaged in telepractice services expressed satisfaction with these services, as evidenced by their ratings for telepractice services in school settings. These ratings show that telepractice services not only improved students' speech and language abilities, but also increased students' engagement with speech-language therapy and their motivations for learning. These results extend the findings of client satisfaction studies that focused on the evaluation of telepractice treatment programs [16,26,27]. These results also suggest that telepractice services help with retaining user satisfaction in real-life school service settings. Users' satisfaction with telepractice is supported by compelling evidence concerning telepractice services for

school-aged students with various disorders [7,28,29]. This evidence suggests that students with special education needs can benefit from treatments that are provided through telepractice services.

Preference for Telepractice and On-site Practice

Despite students' and parents' satisfaction with telepractice efficacy, students did not prefer one mode of practice over the other, whereas parents preferred on-site practice over telepractice. However, there was no significant correlation between telepractice efficacy and parents' preference for on-site practice (P=.44). This indicates that other concerns may have influenced parents' preferences. Interestingly, compared to parents' views of on-site practice, parents expressed a negative view of telepractice in terms of treatment effectiveness, the implementation of therapy practices at home, and communication with speech-language pathologists. This negative opinion can be explained by the lack of effective communication in telehealth. Due to the lack of personal interaction that occurs in telehealth services, extra communication and visual features for communication are needed to build a rapport between clinicians and parents [29]. For example, when discussing sensitive topics (eg, diagnosis, comorbidity, and prognosis) on web-based platforms, parents may feel a sense of depersonalization [29,30]. In addition, face-to-face communication has been indicated as a preferred mode of communication in various studies, as face-to-face communication allows for the better observation of visual cues, such as facial expressions and body language [31-33]. Another explanation for parents preferring on-site practices over telepractice is that parents need to provide extra effort and input in telepractice services. In telepractice sessions, parents need to solve technological problems and control students' behaviors throughout the session. Therefore, parents must allocate more time and energy in telepractice sessions than they do in on-site sessions [33,34].

In this study, the students did not prefer one mode of practice over the other. This could be explained by their satisfaction with telepractice and the significant correlation between their perceptions of telepractice efficacy and their preferences for modes of practice (P<.001). Given that the students had fewer practical concerns than parents, and the fact that students acknowledged the effectiveness of both on-site practice and telepractice, they did not have a preference for the 2 service delivery modes.

Our findings also show that student grade was not significantly associated with telepractice efficacy (parents: P=.76; students: P=.65) or preferences for telepractice and on-site practice (parents: P=.52; students: P=.85). These results reflect the efficacy of telepractice and show that preferences did not differ considerably across different ages. This is consistent with other scientific studies, which have suggested that telepractice is suitable for school-aged students [7-9].

Study Strengths

To our knowledge, this study is the first to investigate parents' and students' satisfaction with telepractice services for a school-aged population during the COVID-19 pandemic. Evaluating parents' and students' perceptions of the efficacy of telepractice is critical. This information not only helps speech-language therapists understand clients' perceptions of telepractice, but also informs educational policy makers about the implementation and adoption of telepractice services beyond the pandemic period. Our study clearly demonstrates that users' satisfaction with telepractice helps to promote evidence-based telepractice. Based on our analysis of both parents' and students' attitudes toward telepractice, we believe that both stakeholders acknowledged the efficacy of telepractice. This is a positive indicator for the future adoption of telepractice as another possible service delivery method, which is needed due to the potential psychosocial challenges of the COVID-19 pandemic. Such challenges include disrupted clinical routes, school closures, and reduced educational and medical support [35].

Limitations and Future Research

This study focused on a limited sample size with a restricted age range (ie, Grades 1-7), even though school-based speech therapy services cover students in Grades 1-12. In addition, the small sample size restricted our investigation of the effect of comorbidity on telepractice efficacy, as communication and literacy characteristics can potentially affect telepractice efficacy.

Future research should consider investigating the effect of comorbidity on telepractice efficacy and satisfaction, by testing a larger sample that includes students of different ages and children with different types of special educational needs. For example, parental involvement is lower in the adolescent population than in the younger student population. Furthermore, in the adolescent population, treatment is focused on academic success. It is important to see whether the acceptance of telepractice services among adolescents differs from the acceptance among young, school-aged children. It should also be noted that our study focused on parents' and students' satisfaction with telepractice after a relatively short-term telepractice session. Future research should extend this study by investigating parents' and students' perceptions of telepractice efficacy and their attitudes toward telepractice after a long-term telepractice session. Our suggestions for future research may elucidate the long-term benefits and sustainability of telepractice, and provide guidance for telepractice strategy development. This information is needed to enhance the quality of digital medical approaches and psychological benefits for children and their families [36].

Implications

The results of this study indicate that telepractice efficacy was well acknowledged by parents and students, and that students in Grades 1-7 had similar preferences for telepractice and on-site practice. The use of telepractice is supported not only by scientific evidence, but also by students' and parents' satisfaction. These results suggest that telepractice is a possible service delivery option for school-aged students.

The findings of our study are in line with those of existing literature, which suggests that telepractice is a suitable service delivery method [7-9]. Our study provided supporting evidence for schools and speech-language pathologists to adopt

telepractice in real-life situations. In addition, our results suggest that speech-language pathologists and parents should be more proactive in telepractice services. Given that the parents had a negative view of treatment effectiveness and communication with speech-language pathologists during telepractice sessions, clinicians should consider engaging more effectively with both students and their parents. Speech-language pathologists can regularly update and inform parents and students about treatment effectiveness to increase their confidence during the transition to telepractice. In addition, clinicians should directly address parents' concerns to build a therapeutic relationship [17]. The engagement and participation of parents is highly important in telepractice services. The importance of parent involvement is well noted in the literature [37,38], and the behavioral management of students during telepractice sessions relies on parents. Moreover, the role of the parent in telepractice services extends to providing technical support and troubleshooting [10]. Clinicians can pay attention to potential technical problems and provide relevant support to parents. If clinicians participate in and engage with telepractice services more often, it is expected that parents will have a better rapport with clinicians, which will facilitate the promotion and acceptance of telepractice [37].

Conclusions

This study showed that both Hong Kong Chinese parents and students believed that telepractice was satisfactory and effective. Although students did not prefer one speech therapy delivery mode over the other, parents preferred on-site speech and language therapy. The perceived efficacy of telepractice was associated with students' preferences for service delivery modes, but it was not associated with parents' preferences. This could be explained by inadequate communication between clinicians and parents. Our findings suggest that it is necessary for speech-language pathologists to play a more proactive role by integrating telepractice into service delivery and explaining the efficacy of telepractice to parents and students.

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Conflicts of Interest

None declared.

Multimedia Appendix 1
Study questionnaire.
[DOCX File , 16 KB - pediatrics_v4i1e25675_app1.docx]

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Abbreviations

BF: Bayes factor

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Review

Telemedicine in Pediatrics: Systematic Review of Randomized Controlled Trials

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Abstract

Background: Telemedicine modalities, such as videoconferencing, are used by health care providers to remotely deliver health care to patients. Telemedicine use in pediatrics has increased in recent years. This has resulted in improved health care access, optimized disease management, progress in the monitoring of health conditions, and fewer exposures to patients with illnesses during pandemics (eg, the COVID-19 pandemic).

Objective: We aimed to systematically evaluate the most recent evidence on the feasibility and accessibility of telemedicine services, patients' and care providers' satisfaction with these services, and treatment outcomes related to telemedicine service use among pediatric populations with different health conditions.

Methods: Studies were obtained from the PubMed database on May 10, 2020. We followed the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines. In this review, we included randomized controlled trials from the last 10 years that used a telemedicine approach as a study intervention or assessed telemedicine as a subspecialty of pediatric care. Titles and abstracts were independently screened based on the eligibility criteria. Afterward, full texts were retrieved and independently screened based on the eligibility criteria. A standardized form was used to extract the following data: publication title, first author's name, publication year, participants' characteristics, study design, the technology-based approach that was used, intervention characteristics, study goals, and study findings.

Results: In total, 11 articles met the inclusion criteria and were included in this review. All studies were categorized as randomized controlled trials (8/11, 73%) or cluster randomized trials (3/11, 27%). The number of participants in each study ranged from 22 to 400. The health conditions that were assessed included obesity (3/11, 27%), asthma (2/11, 18%), mental health conditions (1/11, 9%), otitis media (1/11, 9%), skin conditions (1/11, 9%), type 1 diabetes (1/11, 9%), attention deficit hyperactivity disorder (1/11, 9%), and cystic fibrosis–related pancreatic insufficiency (1/11). The telemedicine approaches that were used included patient and doctor videoconferencing visits (5/11, 45%), smartphone-based interventions (3/11, 27%), telephone counseling (2/11, 18%), and telemedicine-based screening visits (1/11, 9%). The telemedicine interventions in all included studies resulted in outcomes that were comparable to or better than the outcomes of control groups. These outcomes were related to symptom management, quality of life, satisfaction, medication adherence, visit completion rates, and disease progression.

Conclusions: Although more research is needed, the evidence from this review suggests that telemedicine services for the general public and pediatric care are comparable to or better than in-person services. Patients, health care professionals, and caregivers may benefit from using both telemedicine services and traditional, in-person health care services. To maximize the potential of telemedicine, future research should focus on improving patients' access to care, increasing the cost-effectiveness of telemedicine services, and eliminating barriers to telemedicine use.

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KEYWORDS

telemedicine; telehealth; pediatrics; COVID-19; coronavirus; pandemic; digital; eHealth; mHealth; mobile health

Introduction

Telemedicine is a broad term that describes the use of technology in health services for patients and families [1-3]. Such services include teleeducation, telecounseling, and telecommunication platforms that enhance the effectiveness and reach of health care [1,2]. Physicians and other health care providers mainly use telemedicine technology to conduct remote patient visits [1]. This is especially true in the field of pediatrics, given that patients and families frequently face obstacles such as a limited number of pediatric specialists and barriers to long-distance travel [4-7]. Recent advances in pediatric telemedicine have made it possible to deliver pediatric services to medically underserved regions and low-income countries [2,8,9]. Overall, this has led to improved access to health care and the fast assessment, monitoring, and treatment of patients [2,10]. Numerous studies have reported that these benefits, along with the cost-effectiveness of videoconferencing visits (ie, compared to that of in-person visits), have improved the quality of life of patients and their caregivers [8-12]. However, even with new telemedicine technology, barriers to telemedicine access still exist, including the need for strong internet connections, software, and equipment [3,8,10]. Furthermore, studies have shown that the maintenance of telemedicine software is costly, especially in rural areas where such software can be especially useful [8]. The professional and ethical challenges that come with internet-based health care affect patients and physicians [3,13]. Patients and their caregivers can be hesitant to partake in telemedicine encounters due to their desire to see a physician in person, the need for insurance reimbursement, or their attitudes toward technology [1].

Due to the many benefits that telemedicine encounters can provide to patients and physicians, telemedicine services have been used more frequently in recent years [1]. The COVID-19 pandemic has highlighted several important benefits, challenges, and barriers in health care delivery [5,14-18]. Stay-at-home orders, reductions in the number of elective procedures, the loss of jobs, and people's avoidance of hospitals and emergency rooms have made it increasingly difficult for patients to maintain their health care needs during the pandemic [14,17,19,20]. Telemedicine technologies can be especially beneficial during the pandemic, as they can be used to minimize people's exposure to patients with illnesses and provide an on-demand alternative to traditional, in-person visits [15,17,21-23]. Although children who test positive for COVID-19 typically exhibit mild symptoms, routine health services are still an important aspect of a child's well-being [24]. Patients with chronic conditions or those who exhibit risk factors for severe disease (eg, asthma or allergies) can be evaluated via telemedicine modalities for ensuring proper disease management [24].

The future uses of telemedicine technology may include remote patient monitoring, triage, and the implementation of telemedicine services in rural settings or low-income countries [1,8,10]. The goals of telemedicine research include reducing the cost of telemedicine services and optimizing the use of telemedicine technology across different settings [2,8]. These goals are achievable, especially with the growing amount of evidence that supports the feasibility, acceptability, and efficacy of many digital interventions (eg, telehealth approaches) [25-29].

The unique challenges resulting from the COVID-19 pandemic, limited accessibility of pediatric health care in rural areas, management of childhood chronic illnesses, lack of pediatric specialists (ie, compared to the number adult care specialists), and difficulties in traveling with children have highlighted the usefulness and importance of telemedicine modalities for the pediatric population [4-7]. Recent studies and reviews have suggested that telemedicine is a cost-effective, feasible, and beneficial mode of delivering health care for a variety of medical conditions, such as diabetes, heart disease, and depressive disorder [30-33]. Telemedicine's beneficial role in neonatal intensive care unit patient monitoring and pediatric obesity management have also been noted in reviews [10,34]. This review aims to compare the use of telemedicine modalities to that of standard care modalities and determine whether telemedicine procedures can replace standard, face-to-face care procedures. Specifically, the objective of this review is to systematically evaluate the most recent evidence on the feasibility and accessibility of telemedicine services, patients' and care providers' satisfaction with these services, and treatment outcomes related to the use of telemedicine among pediatric populations with different health conditions.

Methods

Study Design

We followed the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines to report on evidence from the studies that were included in this systematic review [35-37]. The PRISMA checklist is shown in Multimedia Appendix 1. We conducted a literature search on the PubMed database on May 10, 2020. The following four keywords were used to conduct the PubMed database search: "telemedicine pediatrics," "telehealth pediatrics," "telemedicine kids," and "telehealth kids." These search terms accounted for related Medical Subject Headings terms, which allowed us to capture a broad range of relevant articles from the database. The "randomized control trial" and "last ten years" filters were applied to all four searches, which were based on each keyword. All articles from the literature search were collected, and duplicate articles were excluded from this review. Titles and abstracts were independently screened based on the eligibility criteria. Articles that did not meet the inclusion criteria were excluded from this review. Afterward, full texts were retrieved and independently screened based on the eligibility criteria. Disagreements were settled by discussion.

Eligibility Criteria

Original randomized controlled trials that were published after 2010 and used telemedicine modalities for different pediatric populations were eligible for this review. No restrictions were placed on the language, condition, setting, or country of a trial. The inclusion criteria included original research papers, randomized controlled trials, pediatric populations (ie, general pediatric care or a subspecialty of pediatric care), and a focus on telemedicine as a study intervention. This review was limited to randomized controlled trials so that we could assess studies with the highest quality of evidence. In order to focus on recent telemedicine advances and the current uses of telemedicine technology, eligible studies were limited to those that were published within the last 10 years.

Data Extraction and Synthesis

A standardized form was used for data extraction. The data items in this form included the following: publication title, first author's name, publication year, participants' characteristics, study design, the technology-based approach that was used, intervention characteristics, study goals, and main study findings. Synthesized data were qualitatively analyzed. ACS conducted the data extraction and SMB conducted a review of the final data.

Quality and Strength of Evidence

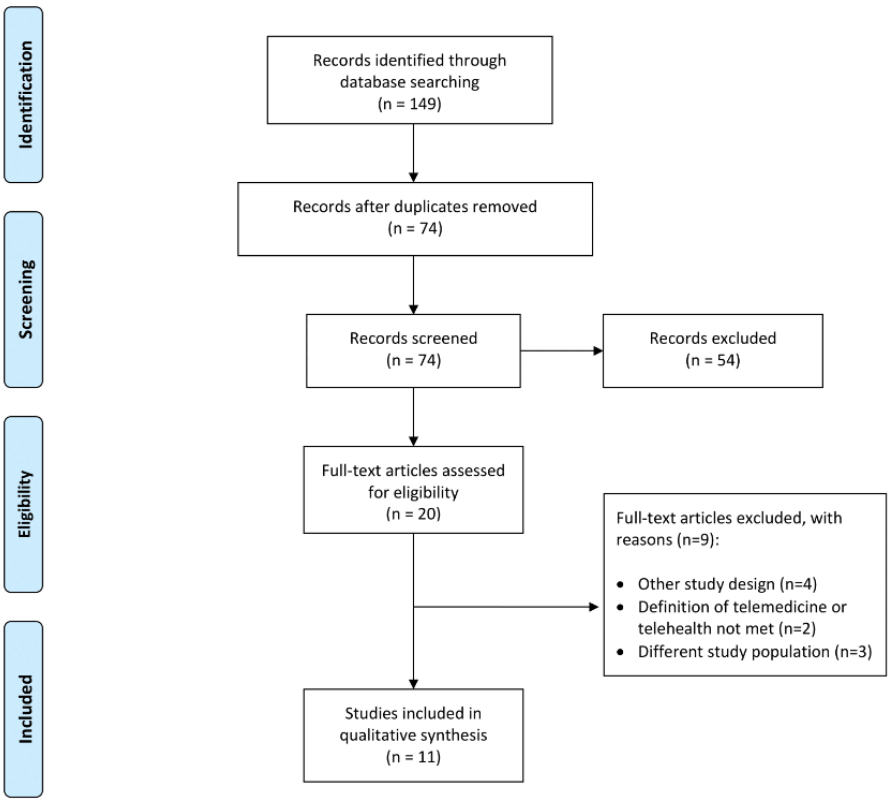
The quality of evidence from the studies that were analyzed in this review was independently evaluated by using the Grading of Recommendations, Assessment, Development, and Evaluation (GRADE) approach [38]. This approach involves assigning an initial quality level rating to a study based on the study design. Randomized controlled trials were all assigned an initial quality level rating of high. The quality level of a study can then be upgraded or downgraded based on the various factors listed in the GRADE guidelines. Factors for downgrading a study's quality level included limitations in the study design and the execution of a study, indirect evidence, inconsistent results, imprecise results, and bias. Quality levels could be upgraded if a study had large effect sizes or dose gradients. Disagreements on GRADE quality levels were settled by discussion.

Results

Literature Search

We conducted a literature search on the PubMed database on May 2020, and this initial literature search yielded a total of 149 references. The "randomized control trial" and "past ten years" filters were applied to all four searches. After excluding duplicates, 74 references remained. The titles and abstracts of all 74 articles were screened, and of these 74 articles, 20 met all the predefined inclusion criteria. Full texts were retrieved from these 20 articles. Afterward, 9 articles were excluded. A total of 11 articles were included in this review [39-49]. The reasons for excluding full-text articles are stated in the PRISMA study flowchart (Figure 1).

Figure 1. Flow diagram of the study inclusion and exclusion process.



Study Characteristics

The characteristics of all included studies are reported in Tables 1 and 2. The studies in this review involved a broad range of health conditions, including asthma (2/11, 18%) [46,47], obesity (3/11, 27%) [40,41,44], mental health conditions (1/11, 9%) [48], otitis media (1/11, 9%) [49], skin conditions (1/11, 9%) [43], type 1 diabetes (1/11, 9%) [42], attention deficit hyperactivity disorder (ADHD) (1/11, 9%) [45], and cystic fibrosis–related pancreatic insufficiency (1/11, 9%) [39]. Of the 11 included studies, 9 (82%) were conducted in the United States of America [39-41,43-48], 1 (9%) was conducted in Italy [42], and 1 (9%) was conducted in Finland [49]. All studies were published in English. Studies' sample sizes ranged from 22 participants [44] to 400 participants [46]. Of the 11 included studies, 4 (36%) had a small sample size (ie, <50 participants) [41,43,44,49], and another 4 (36%) had a sample size of >200 participants [45-48]. The average or median age of participants ranged from 21 months [49] to 17.7 years [42]. Of the 11 studies, 1 (9%) reported that the median age of participants was <3 years [49], and 2 (18%) reported that the average age of participants was >13 years [41,42]. Most trials (7/11, 64%) had a greater proportion of male participants than female participants [39,42,45-49]. All study designs were classified as either randomized controlled trials (8/11, 73%) [39,41-46,49] or cluster randomized controlled trials (3/11, 27%) [40,47,48], as per the inclusion criteria of this review. Follow-up periods ranged from 60 days [49] to 5 years [46]. Of the 11 included studies, 8 (73%) had a follow-up period that ranged between 6 months and 12 months [40-42,44-48], and 1 (9%) did not conduct a participant follow-up [43]. Based on the GRADE criteria, the quality of evidence from most studies was low (4/11, 36%) [41,43,47,48] or moderate (6/11, 55%) [40,42,44-46,49]. Of the 11 studies, only 1 (9%) had a quality rating of high [39]. The telemedicine techniques that were used in the studies included patient and doctor telemedicine visits (5/11, 45%) [40,41,45-47], telemedicine-based screening visits (1/11, 9%) [48], smartphone-based interventions (3/11, 27%) [42,43,49], and telephone counseling (2/11, 18%) [39,44]. Detailed descriptions of the telemedicine techniques that were used in the included studies are discussed in the "Telemedicine Approaches" section. The primary and secondary outcome measures of each study are included in Table 2. Most primary outcomes focused on changes in patients' symptoms (8/11, 72%) [35,39-42,44-47], the time effectiveness of telemedicine (1/11, 9%) [48], or the concordance between in-person and telemedicine diagnoses (2/11, 18%) [43,49].

Table 1. Characteristics of participants in all included studies.

Source (year, country)	Number of participants	Mean age of participants	Female participants, %
Cocker et al (2019, United States) [48]	• Total: 342 • Control: 178 • Intervention: 164	8.6 years	38.3
Erkkola-Anttinen et al (2019, Finland) [49]	• Total: 41 • Immediate group: 20 • Delayed group: 21	21 months[a]	42
Perry et al (2018, United States) [47]	• Total: 363 • Control group: 183 • Intervention group: 180	9.6 years[a]	44
Halterman et al (2018, United States) [46]	• Total: 400 • Control group: 200 • Intervention group: 200	7.8 years	38.25
O'Connor et al (2017, United States) [43]	• Total: 40 • Control group: 20 • Intervention group: 20	6.96 years	55
Di Bartolo et al (2017, Italy) [42]	• Total: 182 • Control group: 90 • Intervention group: 92	17.7 years	48.9
Fleischman et al (2016, United States) [41]	Beginning of study: • Total: 40 • Control group: 21 • Intervention group: 19 End of study: • Total: 33 • Control group: 19 • Intervention group: 14	14.3 years	77.5
Rhodes et al (2017, United States) [44]	• Total: 22 • Low GLb group: 11 • Low-fat group: 11	Low GL group: 8.1 years Low-fat group: 8.2 years	Low GL group: 54.5 Low-fat group: 63.6
Stoep et al (2017, United States) [45]	• Total: 223 • Control group: 112 • Intervention group: 111	9.23 years	29.9
Davis et al (2016, United States) [40]	• Total: 103 • Control group: 61 • Intervention group: 42	9.14 years	55.34
Powers et al (2015, United States) [39]	• Total: 78 • Control group: 42 • Intervention group: 36	3.8 years	43

[a]Median used instead of mean.

[b]GL: glycemic load.

Table 2. Summary of study characteristics and the quality of evidence from all included studies.

Source (year, country)	Health condition	Study design	Telemedicine approach	Outcome measures	Follow-up period	Quality of evidence[a]
Cocker et al (2019, United States) [48]	Mental health	Cluster RCT[b]	Video orientations and videoconferencing screening visits with a mental health clinic	• Primary: completion of screening visit • Secondary: time from referral to screening visit and completion of intake visit	6 months	Low
Erkkola-Anttinen et al (2019, Finland) [49]	Otitis media	RCT	At-home otoscopy videos via smartphone	• Primary: exclusion of otitis media • Secondary: diagnostic quality of videos and effects of teaching interventions	60 days	Moderate
Perry et al (2018, United States) [47]	Asthma	Cluster RCT	Asthma education and monitoring via a telemedicine approach	• Primary: number of symptom-free days • Secondary: peak flow meter use, medication adherence, quality of life, self-efficacy, lung function, and asthma knowledge	6 months	Low
Halterman et al (2018, United States) [46]	Asthma	RCT	School-based telemedicine visits	• Primary: number of symptom-free days • Secondary: number of days with symptoms, use of rescue medication, and number of days with limited activity	7-9 months for intervention and up to 5 years after enrollment	Moderate
O'Connor et al (2017, United States) [43]	Skin condition	RCT	Parents used a smartphone to photograph their child's skin condition for direct patient-to-physician telemedicine.	• Primary: Concordance between in-person and photograph-based diagnoses • Secondary: parents' willingness, image quality, and effect of photograph instructions	None	Low
Di Bartolo et al (2017, Italy) [42]	Type 1 diabetes	RCT	Glucose meters were able to sync with a phone app, which can directly send information to health care workers. Patients were able to contact physicians via email, SMS text messaging, or telephone.	• Primary: changes in hemoglobin A_{1c} levels • Secondary: number of patients who self-monitored their blood glucose levels and patients' quality of life	12 months	Moderate
Fleischman et al (2016, United States) [41]	Obesity	RCT	Televisits with obesity specialists and teleconsults between physicians and specialists	• Primary: changes in BMI • Secondary: waist circumference, triceps skinfold, blood pressure, dietary glycemic load, and physical activity	12 months	Low
Rhodes et al (2017, United States) [44]	Obesity	RCT	Dietary counseling via telephone	• Primary: changes in glycemic load and total number of calories in fat • Secondary: total energy intake	12 months	Moderate
Stoep et al (2017, United States) [45]	Attention deficit hyperactivity disorder	RCT	Telepsychiatry sessions via video counseling	• Primary: changes in distress, as measured by a variety of questionnaires • Secondary: patient health, caregiver strain, parenting stress, and family empowerment	25 weeks	Moderate

Source (year, country)	Health condition	Study design	Telemedicine approach	Outcome measures	Follow-up period	Quality of evidence[a]
Davis et al (2016, United States) [40]	Obesity	Cluster RCT	Physicians delivered behavioral group interventions to families via a telemedicine approach.	• Primary: BMI *z* score • Secondary: feasibility measures, parents' BMIs, 24-hour dietary recall, behavioral checklist scores, feeding assessment scale scores, and accelerometer data	8 months	Moderate
Powers et al (2015, United States) [39]	Cystic fibrosis and pancreatic insufficiency	RCT	Parts of both treatments were delivered via telephone.	• Primary: changes in energy intake • Secondary: changes in weight *z* scores and changes in height *z* scores	18 months	High

[a]Quality ratings are based on the Grading of Recommendations, Assessment, Development and Evaluation criteria.

[b]RCT: randomized controlled trial.

Telemedicine Approaches

Telemedicine approaches widely varied across all included studies. Several studies (5/11, 45%) involved traditional patient and doctor visits [40,41,45-47]. These studies conducted videoconferencing visits instead of in-person physician visits [40,41,45-47]. Of the 11 studies, 3 (27%) used telemedicine interventions that involved the use of a smartphone [42,43,49], and 2 (18%) required parents to perform a task with their smartphone prior to the doctor visit [43,49]. One of these tasks required a parent to perform an at-home smartphone otoscopy of a patient's ear [49], and another required a parent to take a picture of a patient's skin condition in the clinic waiting room [43]. Another smartphone telemedicine approach involved using a new blood glucose meter, which synced data from patients' phones with an app that was able to notify their physicians [42]. Furthermore, two studies used telephone counseling as their principal telemedicine approach [39,44]. In the first study, telephone dietary consultations were made available to participants [44]. The second study involved telephone nutrition counseling and telephone-based education on child behavior management for parents [39]. Additionally, one study used videoconferencing and telemedicine methods in the intervention group and telephone communication methods in the control group [40]. Another study conducted a screening visit via a telemedicine approach [48]. In this study, a mental health clinic conducted an initial screening visit via videoconferencing instead of a traditional, in-person visit [48]. Detailed descriptions of telemedicine approaches are included in Textbox 1.

Textbox 1. Summary of the telemedicine approaches that were used in all included studies.

Cocker et al (2019) [48]

- This was a study on mental health.
- A community mental health clinic conducted an initial screening visit via videoconferencing instead of via telephone.
- After receiving a mental health referral from the primary care physician, parents watched an introduction video about the community mental health clinic.
- Parents returned to the health center and connected with the community mental health clinic coordinator via videoconferencing to determine their eligibility for a screening visit.

Erkkola-Anttinen et al (2019) [49]

- This was a study on otitis media.
- Patients were randomized into either the immediate and delayed teaching groups.
- The immediate teaching group received instructions on how to use a smartphone otoscope before the study began.
- The delayed teaching group received instructions after the first week of the study.
- Parents performed a bilateral smartphone otoscopy on their child for a minimum of 5 days during the first week.
- After the first week, bilateral otoscopy was performed (1) once per week if the child was not experiencing symptoms; (2) every day if child was experiencing respiratory symptoms; (3) every day for 1 week following a diagnosis of acute otitis media; (4) any day the child was experiencing ear pain; and (5) on days of physician visits.
- Bilateral otoscopy videos were sent to the study physician via iMessage, email, or WhatsApp.

Perry et al (2018) [47]

- This was a study on asthma.
- Students participated in five age-appropriate asthma education telemedicine sessions with an allergist, respiratory therapist, or asthma educator.
- These sessions involved the use of a standard, prewritten script.
- Parents or caregivers participated in two telemedicine asthma education sessions that were conducted at a school.
- Nurses participated in two telemedicine asthma education sessions that were conducted at a school.
- If 3 or more sessions were missed, education was delivered via telephone, and education materials were mailed ahead of time.
- Patients were assessed via telemonitoring on months 0 and 3, and asthma medication information was provided by parents on months 3 and 6.
- Caregiver-reported outcomes were measured via telephone interviews on months 0, 3, and 6.

Halterman et al (2018) [46]

- This was a study on asthma.
- Initial asthma assessments for patient and caregivers were conducted via a telemedicine approach.
- A telemedicine assistant entered baseline patient data into the electronic health record system, and a clinician completed the visit within 3 days (ie, from the office or via real-time videoconferencing).
- Afterward, the clinician contacted patients' caregivers by phone or videoconference to discuss initial patient symptoms, treatment plans, and asthma education.
- If a patient's primary care physician did not conduct telemedicine visits, another physician was assigned as the patients' primary physician during the study. Information was forwarded to the original primary care physician.
- Follow-up assessments were conducted via a telemedicine approach every 4-6 weeks.
- All telemedicine visits were reviewed by a nurse to ensure that proper guidelines were followed.

O'Connor et al (2017) [43]

- This was a study on skin conditions.
- Parents took photographs of their child's skin condition with their smartphone in the examination room.
- In this study, 50% of parents received photography instructions and the other 50% did not.
- Photographs were uploaded to electronic medical records.

Di Bartolo et al (2017) [42]

- This was a study on type 1 diabetes.
- Patients who were allocated to the IBGStar (Sanofi US) group received training on how to use the IBGStar machine.
- These patients were able to measure their blood glucose levels with the IBGStar machine at home and sync the readings to an app on their smartphone.
- Data on the app could be directly shared with health care providers.
- All participants in this study were able to contact their physician via email, SMS text messaging, or telephone.

Fleischman et al (2016) [41]

- This was a study on obesity.
- All participants attended in-person visits with their primary care physician every 3 months.
- All participants' primary care physicians conducted a teleconsultation with an obesity specialist 1 week before the visit to discuss obesity treatment.
- Group 1 attended obesity specialist televisits and primary care physician visits for the first 6 months of the study. In the following 6 months, participants only visited their primary care physician in person.
- Group 2 only visited their primary care physician in person for the first 6 months of the study. In the following 6 months, primary care physician visits were supplemented with obesity specialist televisits.

Rhodes et al (2017) [44]

- This was a study on obesity.
- All participants received weekly dietician telephone consultations for 5 consecutive weeks.
- Consultation sessions were recorded, and several sessions were screened to ensure that they adhered to the study protocol.
- This study had a standardized procedure for addressing any missed consultations.

Stoep et al (2017) [45]

- This was a study on attention deficit hyperactivity disorder.
- Families in the telemedicine group underwent a total of 6 combined telemedicine and in-person treatment sessions.
- Videoconferencing was used to deliver child psychiatry treatment and therapy.
- Therapists provided parents with education on attention hyperactivity disorder at the end of each telepsychiatry session.
- All of the sessions were recorded, and a subset of sessions was reviewed to ensure that they were accurate and guideline compliant.
- Therapists were provided with asynchronous telehealth training modules on how to most effectively deliver attention deficit hyper activity education to caregivers.
- These telehealth modules involved viewing recordings of interventions on an asynchronous website.
- Recordings were obtained from volunteer families.
- The control group received 1 telepsychiatry session at the beginning of the study.
- The telepsychiatrist recommended treatment to patients' primary care physicians based on this visit.
- Primary care physicians recommend this treatment, along with any other treatment that they felt would be beneficial, to their patients.

Davis et al (2016) [40]

- This was a study on obesity.
- The schools in this study were randomly allocated into either the telephone or telemedicine groups.
- Telephone and telemedicine sessions were held at schools and focused on family-based cognitive behavioral therapy.
- The telephone group sat around a speakerphone, which was used to connect with the research team during the sessions.
- Speakerphones were provided if the school did not already have one.
- The telemedicine group used the audio and video functions of a television screen to communicate with the research team.

Powers et al (2015) [39]

- This was a study on cystic fibrosis and pancreatic insufficiency.
- The behavioral and nutritional treatment group received individualized nutritional counseling and parent education on child behavioral management.
- Treatment/education sessions and data collection were conducted via an in-person approach or a telehealth approach (ie, telephone).

- If a family did not consistently report on their child's dietary data, a nurse would contact the family via telephone in order to retrieve data.
- The education and attention control group were given educational resources that were related to cystic fibrosis and pancreatic insufficiency. Individualized counseling was not provided to this group. In-person visits and telehealth (ie, telephone) techniques were used to conduct appointments and collect data.

Study Outcomes

Summary of Study Outcomes

Descriptions of study outcomes are reported in Textbox 2. Additional details on these study outcomes are included in Multimedia Appendix 2 [39-49].

Textbox 2. Summary of the main findings and outcomes of all included studies.

Cocker et al (2019) [48]

- This was a study on mental health.
- The initial screening visit was completed by a greater proportion of patients in the telemedicine group (132/164, 80.49%) than in the control group (114/178, 64.04%).
- Patients in the telemedicine referral group required more days to complete the initial screening visit (mean 23.6 days) than patients in the control group (mean 17.1 days).
- No significant difference was observed in the proportion of patients who completed the recommended intake visit after the screening visit between the two groups (telemedicine group: 93/116, 80.2%; control group: 81/97, 83.5%; *P*=.51).
- Based on the adjusted analysis, no significant difference was observed in the time from referral to the screening visit between the two groups (*P*=.62).
- Compared to parents in the control group, those in the telemedicine group reported higher satisfaction with the referral system and the care that they received.
- No significant differences were observed in patients' quality of life (ie, after 6 months) between both groups (*P*=.82).

Erkkola-Anttinen et al (2019) [49]

- This was a study on otitis media.
- A video or image was obtained during 98% (1472/1500) of all parent-performed examinations (median video length=18 seconds).
- In total, 67% (867/1293) of all videos were of sufficient diagnostic quality.
- Diagnoses could be made for 56% (486/867) of videos that were of sufficient diagnostic quality.
- Diagnoses could only be made for 8% (35/426) of the videos that were of insufficient diagnostic quality.
- Diagnoses could be made for 40% (521/1293) of all videos.
- Acute otitis media diagnoses could be confirmed or excluded for 87% (609/699) of all videos that were obtained during respiratory infection.
- In total, diagnoses could be confirmed or excluded with 99% (495/501) of the videos that were of sufficient diagnostic quality.
- In total, diagnoses could be confirmed or excluded with 58% (114/198) of the videos that were of insufficient diagnostic quality.
- During week 1 of the intervention, the immediate teaching group was taught how to perform otoscopy and the delayed teaching group was not. There were significantly more videos that were of sufficient diagnostic quality in the immediate teaching group (95/152, 62%) than in the delayed teaching group (39/179, 22%) (*P*<.001).
- One week after the delayed teaching group received their education session, 64% (85/133) of their videos were of sufficient diagnostic quality.
- In total, 24% (10/41) of families believed that smartphone otoscopy was a burden.
- In total, 83% (34/41) of families considered conducting smartphone otoscopies on a daily basis.

Perry et al (2018) [47]

- This was a study on asthma.
- No significant difference was observed in the number posttreatment symptom-free days between the intervention and usual care groups (*P*=.51).
- Patients in both groups still had uncontrolled asthma at the end of treatment.
- Compared to the intervention group, the usual care group had significantly higher scores in the family activity domain of the Child Health Survey for Asthma (*P*=.02).
- Compared to the usual care group, the intervention group had a significantly greater percentage of patients that used a peak flow meter (*P*<.001).
- Compared to the usual care group, the intervention group had a significantly greater percentage of patients who were compliant with posttreatment asthma medication (*P*=.03).
- There was no significant difference in the baseline quality-of-life scores between both treatment groups (*P*=.06).

Halterman et al (2018) [46]

- This was a study on asthma.
- Children in the telemedicine group had significantly more postintervention symptom-free days (mean 11.6 days) than children in the control group (mean 10.97 days) (*P*=.01).
- The intervention group had fewer symptom days, symptom nights, and limited activity days than the control group.
- Compared to the control group, the telemedicine group had a greater proportion of patients who were prescribed preventive medication (control group: 132/196, 67%; telemedicine group: 181/199, 91%).

- In the final follow-up longitudinal visit, the telemedicine group had 0.85 more symptoms than the control group, and a significant correlation was observed between treatment efficacy and time (P<.02).
- Decreases in exhaled nitric oxide levels were greater in the telemedicine group than in the control group (mean difference=−5.54).
- Caregivers' quality of life improved in both groups; there was no significant difference in caregivers' quality of life between both groups (95% CI −0.08 to 0.37).
- In total, 95.7% (361/377) of patients reported that the program was helpful, and 96.5% (365/367) reported that they would partake in another similar program.

O'Connor et al (2017) [43]

- This was a study on skin conditions.
- The median photograph quality rating score was 9.
- The concordance between photograph diagnosis and in-person diagnosis for all photographs was 83% (33/40).
- The mean quality rating score for photographs with a diagnosis was 8.9, whereas the mean quality rating score for photographs with no diagnosis was 7.0.
- The group that received photography instructions had a higher average image quality score and a higher mean number of images than the group that did not receive instructions, but this was not statistically significant.
- No significant difference was observed in the concordance of diagnosis between the group that received photograph instructions and the group that did not receive instructions (P=.68).
- Parents' willingness to use teledermatology services was measured on a scale of 1 (ie, not willing) to 10 (ie, very willing). The median response score was 8.

Di Bartolo et al (2017) [42]

- This was a study on type 1 diabetes.
- The telemedicine and control groups exhibited reduced hemoglobin A_{1c} levels after treatment; there was no significant difference between the two groups (P=.051).
- Patients who self-monitored their blood glucose levels exhibited reduced hemoglobin A_{1c} levels at 6 months posttreatment.
- Patients who did not self-monitor their blood glucose levels only exhibited minor changes in hemoglobin A_{1c} levels at 6 months posttreatment.
- Patients in the telemedicine group exhibited greater decreases in hemoglobin A_{1c} levels at 6 months posttreatment than the control group (P=.25).
- The control group started using the experimental telemedicine meter at 6 months posttreatment. At 12 months posttreatment, the control group exhibited decreases in hemoglobin A_{1c} levels (P=.24).
- At 12 months posttreatment, the experimental group's hemoglobin A_{1c} levels remained stable (ie, compared to their hemoglobin A_{1c} levels at 6 months posttreatment).
- There were no significant differences in quality-of-life measures between both groups at 6 months and 12 months posttreatment (P=.23).

Fleischman et al (2016) [41]

- This was a study on obesity.
- Group 1 (ie, patients who attended primary care physician visits and specialist televisits) exhibited greater decreases in BMI z scores after 3 months than Group 2 (ie, patients who only attended primary care physician visits) (P=.049).
- The BMIs in group 1 significantly decreased after 6 months (P<.001), while the BMIs in Group 2 did not (P=.08). No significant differences were observed in BMIs between the two groups (P=.23).
- After 6 months, group 1 only attended primary care physician visits and Group 2 attended primary care physician visits and specialist televisits.
- The baseline BMIs in group 1 were significantly different from those after 9 months (P.004) and 12 months (P=.03).
- The baseline BMIs in group 2 were significantly lower than those after 12 months (P=.03).
- If given the opportunity to choose between obesity specialist televisits or in-person visits, 14 patients would choose televisits and 7 had no preference.

Rhodes et al (2017) [44]

- This was a study on obesity.
- There were no significant differences in dietary fat content (ie, before and after treatment) between or within the two groups (P=.68).
- After treatment, the low glycemic load group had lower glycemic loads than the low-fat group (P=.003).

- There were no significant differences in posttreatment glycemic loads between both groups (*P*=.06).
- The low glycemic load group exhibited a significant decrease in total energy intake levels after treatment (*P*<.005).
- The low glycemic load group had significantly lower posttreatment total energy intake levels than the low-fat group (*P*=.001).
- There were no significant differences in changes in total energy intake levels (ie, from baseline to after treatment) between both groups (*P*=.06).

Stoep et al (2017) [45]

- This was a study on attention deficit hyperactivity disorder.
- Caregivers in both the Children's Attention Deficit Hyperactivity Disorder Telemental Health Treatment Study (CATTS) and augmented primary care groups showed improvements in caregiver distress by the end of the study.
- Caregivers in the CATTS group had significantly lower Parenting Stress Index (*P*<.01; Cohen *d*=0.59), Patient Health Questionaire-9 (*P*<.05; Cohen *d*=0.27), and Cognitive Skills Quotient (*P*<.001; Cohen *d*=0.45) scores after 25 weeks of treatment compared to those at baseline.
- Caregivers in the CATTS group also had significantly higher Falls Efficacy Scale scores after 25 weeks of treatment (*P*<.01; Cohen *d*=−0.44).

Davis et al (2016) [40]

- This was a study on obesity.
- The satisfaction scores between the telemedicine and telephone groups were not considerably different.
- There were no significant differences in changes in patients' BMIs (ie, pretreatment to posttreatment) between the telemedicine and telephone groups (*P*>.05).
- There were no significant differences in changes in parents' BMIs (ie, pretreatment to posttreatment) between the telemedicine and telephone groups (*P*>.05).

Powers et al (2015) [39]

- This was a study on cystic fibrosis and pancreatic insufficiency.
- After treatment, the control group had significantly lower energy intake levels than the behavioral and nutritional treatment group (*P*<.001).
- After treatment, there were no significant differences in weight *z* scores between the two groups (*P*=.25).
- After treatment, the control group exhibited greater decreases in height *z* scores than the behavioral and nutritional treatment group (*P*=.49).
- During the follow-up, the behavioral and nutritional treatment group had greater average energy intake levels than the control group (*P*=.02).
- At follow-up, there were no significant differences in weight *z* scores between the two groups (*P*=.61).

Effects of Telemedicine on Asthma Symptoms

Perry et al [47] and Halterman et al [46] used a school-based telemedicine approach to aid patients with managing their asthma symptoms. Perry et al [47] reported that there were no significant differences in the number of symptom-free days (SFDs) between the telemedicine and usual care groups (*P*=.51), while Halterman et al [46] reported a significant increase in the number of SFDs in the telemedicine group compared to that in the control group (*P*=.01). Perry et al [47] reported that there was a significant increase in medication adherence (*P*=.03) and peak flow meter use (*P*<.001) in the telemedicine group compared to those in the usual care group. Furthermore, Halterman et al [46] reported that the telemedicine group had a greater proportion of patients who were prescribed preventative medicine (181/199, 91%) compared to the control group (132/196, 67%). The telemedicine group also had lower hospitalization rates (14/199, 7%) than the control group (29/196, 15%). Additionally, patients in the telemedicine group had a significantly higher number of SFDs in the follow-up longitudinal visit than the control group (*P*<.02) [46]. Both Perry et al [47] and Halterman et al [46] reported no significant differences in quality-of-life scores between the groups at the end of their studies. In terms of satisfaction, most parents in the Halterman et al study [46] stated that they found the program helpful (361/377, 95.7%) and would partake in another similar program (365/377, 96.5%). Furthermore, parents in the telemedicine group were more likely to learn more about asthma medication (152/193, 78.8%) than parents in the control group (111/184, 60.3%) [46].

Effects of Telemedicine on Weight Management and Energy Intake

Fleischman et al [41], Rhodes et al [44], and Davis et al [40] investigated the role of telemedicine in weight management by conducting specialist televisits, telephone dietary counseling, and physician telemedicine interventions, respectively. In the Fleischman et al study [41], obesity specialists found that each group's BMIs significantly decreased 6 months after the telemedicine phase of the study (group 1: *P*=.006; group 2: P=.03). Rhodes et al [44] showed that a low–glycemic index diet significantly decreased the posttreatment total energy intake levels of both groups (*P*<.005). Furthermore, the low glycemic load group exhibited greater decreases in total energy intake levels than the low-fat diet group (*P*=.001). However, there were no significant differences in changes in total energy levels (ie, from the beginning of treatment to the end of treatment) between the two groups (*P*=.06) [44]. Similarly, Davis et al

[40] reported that there were no significant differences in changes in patients' and parents' BMIs (ie, from baseline to after treatment) within (P>.05) and between (P>.05) the two groups. In the Fleischman et al study [41], most patients (14/21, 67%) stated that they prefer televisits over in-person specialist visits, and patients in the telemedicine group found the program more helpful than patients in the control group (P=.06). Alternatively, Davis et al [40] did not observe a significant difference in satisfaction scores between the telemedicine and telephone groups [40]. Powers et al [39] tracked the effects that telehealth-based nutritional counseling and education had on patients with cystic fibrosis–related pancreatic insufficiency. Powers et al [39] reported that the control group had significantly lower posttreatment energy intake levels (P<.001) and greater decreases in height z scores (P=.49) than the treatment group. No significant differences were observed in posttreatment weight z scores between the two groups (P=.25) [39].

Effects of Telemedicine on Diabetes Management

Di Bartolo et al [42] measured changes in patients' blood glucose levels by using a traditional blood glucose meter and the IBGStar blood glucose meter (Sanofi US). This study showed that both groups exhibited reductions in hemoglobin A_{1c} (HbA_{1c}) levels. There were no significant differences in HbA_{1c} levels between the two groups at the end of treatment (P=.051) [42]. The number of patients who self-monitored their blood glucose levels was comparable between the two groups (P=.85) [42]. The self-monitoring of blood glucose levels was associated with decreases in HbA_{1c} levels [42]. The telemedicine group used the experimental IBGStar meter and reported greater decreases in HbA_{1c} levels at 6 months posttreatment than those who used the traditional meter (P=.25) [42]. Even at 12 months posttreatment, the experimental group's HbA_{1c} levels were stable (ie, compared to their HbA_{1c} levels at 6 months posttreatment) [42]. There were no significant differences in quality-of-life measures between both groups at 6 and 12 months posttreatment [42]. Participants in the telemedicine group contacted their physician (ie, via SMS text messaging, telephone call, or email) more frequently than the control group [42].

Effects of Telemedicine on Screening Efficiency

Cocker et al [48] compared the efficiency of telemedicine mental health screening visits to that of in-person screening visits. Although screening visits were completed by a greater percentage of patients in the telemedicine group (132/164, 80%) than in the in-person group (114/178, 64%), patients in the telemedicine group required longer times to complete the screening visit (telemedicine group: mean 23.6 days; in-person group: mean 17.1 days) [48]. The mode of delivery for the screening visit did not have a considerable effect on the percentage of patients who completed the in-person intake visit [48]. Patients' quality of life did not differ between the two groups, but patients in the telemedicine group reported higher satisfaction with the screening process than the in-person group [48].

Effects of Telemedicine on Patients' and Caregivers' Quality of Life

Stoep et al [45] assessed the effects of ADHD therapy and caregiver education (ie, both were provided via a telemedicine approach) on parents' quality of life (ie, parents from the Children's ADHD Telemental Health Treatment Study). After 25 weeks, parents in the telemedicine group exhibited significant decreases in their Parenting Stress Index (P<.01), Patient Health Questionaire-9 (P<.05), and Client Satisfaction Questionnaire (P<.001) scores, as well as significant increases in their Falls Efficacy Scale scores (P<.01) [45]. At the end of the study, parents experienced improvements in different domains of caregiver distress, including parenting stress (41%), caregiver depression (48%), caregiver strain (43%), and family empowerment (26%). These percentages refer to the effects of treatment on caregiver outcomes (ie, changes in children's symptoms/roles) [45]. Reductions in the number of patient's oppositional defiant disorder symptoms correlated with decreased levels of caregiver distress [45].

Effectiveness of Parent Telemedicine Education

Erkkola-Anttinen et al [49] and O'Connor et al [43] conducted studies that required parents to learn telemedicine techniques for documenting their child's health condition. Erkkola-Anttinen et al [49] provided caregivers with education on performing a smartphone otoscopy of a patient's ear. O'Connor et al [43] instructed parents to take a photograph of a patient's skin condition. Erkkola-Anttinen et al [49] showed that acute otitis media diagnoses that were confirmed or excluded based on videos from parents who received smartphone otoscopy instructions (495/501, 99%) were more accurate than those based on videos from parents who did not receive instructions (114/198, 58%). In the Erkkola-Anttinen et al study [49], a considerable difference was observed in the quality of videos from the teaching and nonteaching groups. However, O'Connor et al [43] reported that there was no significant difference in the concordance of photograph-based and in-person diagnoses between parents who received instructions and parents who did not receive instructions (P=.68). The mean quality rating score of photographs from which a diagnosis could be made (8.9) was higher than that of photographs from which a diagnosis could not be made (7.0) [43]. Similarly, Erkkola-Anttinen et al [49] reported that a diagnosis could be made with 56% (486/867) of otoscopy videos that were of sufficient diagnostic quality. However, a diagnosis could only be made with 8% (35/426) of videos that were not of sufficient diagnostic quality. In the O'Connor et al study [43], parents' willingness to use teledermatology services was measured on a scale of 1 (ie, not willing) to 10 (ie, very willing). The median rating was 8 [43].

Discussion

Principal Findings

The evidence from this review suggests that telemedicine visits for pediatric care may be comparable to and occasionally more beneficial than in-person visits. In this review, 11 studies that met all listed inclusion criteria were identified. All included studies were randomized controlled trials that assessed the use of telemedicine in pediatrics. The following eight health

conditions were assessed: asthma, obesity, otitis media, mental health conditions, skin conditions, ADHD, type 1 diabetes, and cystic fibrosis–related pancreatic insufficiency. According to the GRADE criteria, the quality of evidence from almost all studies (10/11, 91%) was either low or moderate. Most low or moderate ratings were due to limitations in study design and implementation and the indirectness of evidence. The quality of evidence from one study was high. Most studies conducted videoconferencing visits instead of traditional, in-person physician visits. Other telemedicine interventions that were used included smartphone-based apps, telephone counseling, and web-based screening visits.

Overall, although the impact of telemedicine on pediatric health care was modest, telemedicine interventions showed promise. Studies on school-based telemedicine interventions for asthma had contradictory results for the effects of telemedicine on asthma SFDs [46,47]. However, parents were satisfied with these interventions and noticed improvements in outcome measures, such as asthma education, medication adherence, and the number of preventative medicine prescriptions [46,47]. Similarly, although studies about the impact of telemedicine on weight management had mixed results, patients reported that they preferred televisits over in-person visits or had no preferences for the two methods [39-41,44]. Patients also reported that they were more satisfied with telemedicine approaches than with mental health screening visits [48]. Furthermore, parents' (ie, those of children with ADHD) quality of life improved after attending web-based therapy and education sessions [45]. This suggests that telemedicine services can be used to supplement in-person visits. Studies have also reported that parent education on telemedicine techniques for monitoring and documenting children with health conditions is a feasible approach that is acceptable to caregivers [43,49]. Additionally, patients who use telemedicine-based blood sugar monitoring devices have reported that they contact their physicians more frequently. This suggests that telemedicine technology can be used to supplement digital approaches for monitoring chronic health conditions [42].

Recently published literature has suggested that telemedicine approaches in general pediatric practice can be used to provide alternatives to traditional patient visits, increase people's access to health care, and reduce the number of existing disparities [50-52]. One of the goals of recent research has been to improve the standards of telemedicine services so that they can provide higher quality care with lower costs [50,51]. The management of chronic health conditions is a realm of pediatrics in which telemedicine approaches have shown promise, especially when they are used in conjunction with in-person approaches [7,53].

Health care has been rapidly evolving to adapt to the ongoing COVID-19 pandemic, and telemedicine has become an important mechanism of health care delivery [19]. A study found that telemedicine visits in urgent care and nonurgent care facilities have increased by 135% and 4345%, respectively [19]. Many pediatric patient portals have also been updated and improved to include telehealth features [54]. The use of telemedicine during the COVID-19 pandemic not only protects patients and providers from unnecessary exposure to patients with illnesses, but also conserves personal protective equipment, which should be saved for essential encounters [55]. New telemedicine technologies, such as chatbots that provide conversation-like interactions, are being used to triage patients and screen for COVID-19 symptoms [56]. However, due to the increased use of telemedicine technology in hospitals and clinics, these technologies need to be evaluated so that people can understand their effects on patients, workers, health care systems, and insurance companies [57].

The timely management of pediatric chronic illnesses, such as obesity, allergies, and genetic diseases, is paramount to providing patients and their families with the best care, especially during the COVID-19 pandemic [24,58,59]. Web-based telemedicine visits have been used to help manage chronic conditions and related medications [24,58,59]. Additionally, during the COVID-19 pandemic, glucose monitoring software has been used to regularly record type I diabetes symptoms [60]. Common symptoms, such as migraines, can worsen during times of stress, and telemedicine can aid with providing care and limiting the need to visit a hospital [61].

Telemedicine is also being used in specific pediatric subspecialty settings. In surgery, telemedicine modalities have been used to preoperatively diagnose patients, perform surgery (ie, with robotic devices), or postoperatively monitor patients [62]. Pediatric gastroenterologists have also used telemedicine to supplement in-person visits and monitor chronic conditions (eg, inflammatory bowel disease) [63]. Furthermore, due to the limited number of pediatric subspecialty physicians in certain regions, telemedicine referrals are being used to optimize the accessibility of subspecialty resources [64]. A survey study that was conducted at a pediatric headache clinic in San Francisco, California reported that all included families found telemedicine visits to be more convenient than in-person visits. These families also stated that they would choose to use a telemedicine method again [65]. Families of children with many different health conditions have shown considerable interest in telemedicine visits, and most of these families possess sufficient technology for attending these visits [52].

Pediatric patients in rural communities face distinct challenges, such as limited access to subspecialty care and long commutes to clinics. However, these challenges can be overcome with telemedicine interventions [4,66-69]. Pediatricians from rural areas of the United States have advocated for telemedicine, as it can help with maintaining patient relationships and improving the accessibility of subspecialty care [70]. Telemedicine can provide a convenient platform that patients (eg, those from rural communities) can use to obtain the health care that they need, minimize travel time, and reduce waiting times for appointments [4,66-69].

The use of telemedicine in adult medical care is similar to that in pediatric care. Web-based patient monitoring via telemedicine modalities allows intensive care unit physicians to check the status of multiple patients at any time and place [71]. In one study, neurology patients were monitored with web-based electrocardiogram and electroencephalogram machines [72]. Telemedicine technologies can also be used to improve preprocedural instructions (eg, bowel preparation instructions

for a colonoscopy) and reduce the time needed for providing adequate education [73].

Strengths and Limitations

This systematic review has multiple strengths. First, we followed recommendations for rigorous systematic review methodologies [35-37]. Second, language and country filters were not applied to the literature search. Therefore, studies from all countries and studies in any language were eligible for this review. Furthermore, these factors did not limit the scope of this review. Third, the quality of evidence from all included studies was evaluated by using the GRADE approach [38]. This increased the transparency of the quality of included studies. Fourth, although we searched for publications from the last 10 years, our earliest study was published in 2015 [39]. Therefore, it is likely that earlier studies were not missed.

The potential methodological limitations of this systematic review should also be discussed. First, this review used a single database (ie, PubMed) to conduct the literature search. However, PubMed is the most comprehensive medical database. Most studies in other databases are also likely to be found in PubMed. Therefore, it is likely that we did not miss any studies that were relevant to our review. However, the possibility of missing a study cannot be excluded. Second, even though our search criteria allowed for the inclusion of studies from all countries, all included studies were conducted in high-income countries. Telemedicine use in high-income and low-income countries may be different, and the results of this review should be viewed as results from high-income countries. Third, this review included studies with different follow-up periods and patient populations (ie, various health conditions and age groups). Therefore, there may have been several inconsistencies between the results of each study. Furthermore, these limitations did not allow us to perform a meta-analysis [74]. Fourth, to identify the strongest available evidence, we only included randomized controlled trials that were published in peer-reviewed journals. Therefore, publication bias (ie, the tendency to report positive study results) may be present in the included studies [75].

Conclusion

In recent years, telemedicine use among the pediatric population has become more common. Although a clear consensus on the benefits of telemedicine approaches in pediatrics has not been reached, recent literature has shown that telemedicine services are comparable to or better than in-person services. Patients and caregivers have also consistently reported that they are more satisfied with telemedicine visits than with in-person visits. This shows promise for telemedicine in pediatric settings, especially during times when social distancing is a requirement, such as the COVID-19 pandemic. Future studies should focus on improving telemedicine delivery services, people's access to health care, the quality of telemedicine approaches, and the integration of telemedicine into in-person physician visits. Furthermore, future studies that emphasize the cost-effectiveness of telemedicine, the use of telemedicine services in rural settings, and barriers to telemedicine technology implementation are needed to analyze the true potential of telemedicine approaches for improving children's and adolescents' health outcomes.

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Authors' Contributions

ACS conceptualized and designed the study, collected the data, analyzed the data, drafted the initial manuscript, and reviewed and revised the manuscript. SMB conceptualized and designed the study, coordinated and supervised the data collection process, and critically reviewed and revised the manuscript. Both authors reviewed and approved the final version of the manuscript.

Conflicts of Interest

None declared.

Multimedia Appendix 1
PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) checklist.
[DOC File , 65 KB - pediatrics_v4i1e22696_app1.doc]

Multimedia Appendix 2
Detailed descriptions of study outcomes.
[DOCX File , 46 KB - pediatrics_v4i1e22696_app2.docx]

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Abbreviations

ADHD: attention deficit hyperactivity disorder
GRADE: Grading of Recommendations, Assessment, Development and Evaluation
HbA_{1c}: hemoglobin A_{1c}
PRISMA: Preferred Reporting Items for Systematic Reviews and Meta-Analyses
SFD: symptom-free day

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Original Paper

Interformat Reliability of Web-Based Parent-Rated Questionnaires for Assessing Neurodevelopmental Disorders Among Preschoolers: Cross-sectional Community Study

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Abstract

Background: Early detection and intervention for neurodevelopmental disorders are effective. Several types of paper questionnaires have been developed to assess these conditions in early childhood; however, the psychometric equivalence between the web-based and the paper versions of these questionnaires is unknown.

Objective: This study examined the interformat reliability of the web-based parent-rated version of the Autism Spectrum Screening Questionnaire (ASSQ), Attention-Deficit/Hyperactivity Disorder Rating Scale (ADHD-RS), Developmental Coordination Disorder Questionnaire 2007 (DCDQ), and Strengths and Difficulties Questionnaire (SDQ) among Japanese preschoolers in a community developmental health check-up setting.

Methods: A set of paper-based questionnaires were distributed for voluntary completion to parents of children aged 5 years. The package of the paper format questionnaires included the ASSQ, ADHD-RS, DCDQ, parent-reported SDQ (P-SDQ), and several additional demographic questions. Responses were received from 508 parents of children who agreed to participate in the study. After 3 months, 300 parents, who were among the initial responders, were randomly selected and asked to complete the web-based versions of these questionnaires. A total of 140 parents replied to the web-based format and were included as a final sample in this study.

Results: We obtained the McDonald ω coefficients for both the web-based and paper formats of the ASSQ (web-based: ω=.90; paper: ω=.86), ADHD-RS total and subscales (web-based: ω=.88-.94; paper: ω=.87-.93), DCDQ total and subscales (web-based: ω=.82-.94; paper: ω=.74-.92), and P-SDQ total and subscales (web-based: ω=.55-.81; paper: ω=.52-.80). The intraclass correlation coefficients between the web-based and paper formats were all significant at the 99.9% confidence level: ASSQ (r=0.66, *P*<.001); ADHD-RS total and subscales (*r*=0.66-0.74, *P*<.001); DCDQ total and subscales (*r*=0.66-0.71, *P*<.001); P-SDQ Total Difficulties and subscales (*r*=0.55-0.73, *P*<.001). There were no significant differences between the web-based and paper formats for total mean score of the ASSQ (*P*=.76), total (*P*=.12) and subscale (*P*=.11-.47) mean scores of DCDQ, and the P-SDQ Total Difficulties mean score (*P*=.20) and mean subscale scores (*P*=.28-.79). Although significant differences were found between the web-based and paper formats for mean ADHD-RS scores (total: t_{132}=2.83, *P*=.005; Inattention subscale: t_{133}=2.15, *P*=.03; Hyperactivity/Impulsivity subscale: t_{133}=3.21, *P*=.002), the effect sizes were small (Cohen *d*=0.18-0.22).

Conclusions: These results suggest that the web-based versions of the ASSQ, ADHD-RS, DCDQ, and P-SDQ were equivalent, with the same level of internal consistency and intrarater reliability as the paper versions, indicating the applicability of the web-based versions of these questionnaires for assessing neurodevelopmental disorders.

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KEYWORDS

neurodevelopmental disorders; web-based questionnaire; preschoolers; parents; interformat reliability

Introduction

In the American Psychiatric Association's Diagnostic and Statistical Manual for Mental Disorders, Fifth Edition (DSM-5), neurodevelopmental disorders are identified in the early developmental stages and are characterized by developmental deficits that lead to impairments in personal, social, academic, and vocational functioning [1]. Representative examples of such disorders include autism spectrum disorder, attention-deficit/hyperactivity disorder, and developmental coordination disorder. The core characteristics of autism spectrum disorder include 2 main dimensions—social communication and restricted, repetitive sensory–motor behaviors—that are irrespective of culture, race, ethnicity, or socioeconomic group [2]. Estimates of the total-population prevalence of autism spectrum disorder range from 2.2% to 3.2% [3,4]. The hallmarks of attention-deficit/hyperactivity disorder are developmentally impaired attention, motor hyperactivity, impulsivity, and the difficulties associated with them [5]. A recent meta-analysis [6] revealed that the estimated prevalence of children with attention-deficit/hyperactivity disorder is 3.4% in the general population. Developmental coordination disorder is characterized by marked impairment in the acquisition and execution of motor skills. This impairment significantly and sustainably interferes with activities of daily living, including academic achievement [1]. A recent review [7] reported that prevalence estimates for developmental coordination disorder among children range from 2% to 20%, with 5% to 6% being the most commonly reported prevalence rate.

It is known that children with these conditions not only have various secondary mental health problems [8-10] but also experience maladjustment in adulthood [11-13]. Since a number of studies [7,14,15] have reported that early detection and intervention for neurodevelopmental disorders is effective, it is necessary to develop useful screening tools for assessing these conditions in early childhood.

Several questionnaires have been developed to assess a variety of neurodevelopmental disorders. The Autism Spectrum Screening Questionnaire (ASSQ) was developed to screen for autism spectrum disorder in school-age children based on their parents' or primary caregivers' ratings [16] and has been shown to be highly accurate in screening for autism spectrum disorder [17]. Additional research has confirmed that the ASSQ has good reliability and validity and can be applied to preschool-age children, as well [18,19]. The ADHD-Rating Scale (ADHD-RS) is one of the most widely used questionnaires developed to assess ADHD symptoms in children age 5 to 18 years [20]. There are 2 versions of the ADHD-RS: the home form is rated by parents or primary caregivers of the children, and the school form is rated by teachers. It has been demonstrated that the ADHD-RS has good reliability and validity in preschool children [21]. Moreover, previous research has confirmed that the ADHD-RS shows higher sensitivity and specificity in parent ratings than those in teacher ratings among preschoolers [22]. The Developmental Coordination Disorder Questionnaire 2007 (DCDQ) was developed to identify children age 5 to 15 years who are at risk for developmental coordination disorder, based on parents' ratings [23]. It has been found that the DCDQ has good psychometric properties and has been recommended for use in clinical practice as supplemental information for the diagnosis of children with developmental coordination disorder [7]. It is known that children with neurodevelopmental disorders have behavioral and emotional difficulties. The Strengths and Difficulties Questionnaire (SDQ) is a brief behavioral screening questionnaire about externalizing and internalizing problems in children [24]. It has been reported that using parent ratings for the SDQ has satisfactory reliability and validity in a community sample of 5- to 15-year-old children [25].

These questionnaires have been developed as paper-and-pencil type questionnaires and can be useful in individual clinical settings; however, to screen large populations in a local community for early detection of neurodevelopmental disorders, web-based versions are more efficient than the paper-and-pencil version because of the significant time and effort required to distribute and collect paper-and-pencil questionnaires. Not only can costs be saved, but also there are additional advantages; for example, we can automate the process of manual data entry after completing data collection [26]. In addition, using web-based questionnaires to collect data generally improves the quality of the data because the validation checks can incorporate prompts that alert respondents if they enter incorrect or incomplete answers [27]. Furthermore, web-based instruments expand the reach of assessments, which is particularly important under pandemic conditions when it is difficult or impossible to administer in-person assessments [28,29].

On one hand, several studies [30-32] have reported psychometric equivalence between web-based and paper-and-pencil versions of the questionnaires used to assess various psychological disorders; on the other hand, some studies have revealed psychometrically significant differences between the 2 formats [26,33,34]. Therefore, it is necessary to evaluate the comparability of web-based and paper-and-pencil versions of the questionnaires [35]. In particular, high interformat reliability, meaning the level of equality between different delivery formats, indicates that the psychometric properties of the instrument are independent of the delivery format [36]. However, to our knowledge, no studies have assessed the interformat reliability of web-based questionnaires that aim to assess neurodevelopmental disorders.

We aimed to examine the interformat reliability of the web-based versions of the ASSQ, ADHD-RS, DCDQ, and SDQ. Based on previous work [36], we confirmed interformat reliability from the following 3 perspectives. First, we verified the internal consistency of the web-based and paper-and-pencil formats of each questionnaire. Second, we examined the

intraclass correlations between the 2 formats of each questionnaire to test intrarater reliability. Third, we investigated the mean score differences between the 2 formats of each questionnaire to confirm equivalence in quality.

Methods

Participants

This study was conducted as part of the Hirosaki Five-Year-Old Children Developmental Health Check-up Study (HFC Study), a large community-based cohort study initiated in 2013 that examined the impact of children's neurodevelopmental disorders and lifestyle habits on their adaptation and emotional and behavioral problems at age 5 years. Located in Aomori Prefecture in the northeastern part of Japan, Hirosaki City has approximately 175,000 residents, 1 university, and several colleges, and its main industry is agriculture.

Participants in this study were recruited in July 2018. The local government of Hirosaki City distributed a set of paper-based questionnaires for voluntary completion to the parents of 620 5-year-old children in the city via the municipal health center. The package included the ASSQ, ADHD-RS, DCDQ, parent-reported SDQ (P-SDQ), and demographic questions. Responses were received from 508 parents who agreed to participate in the study. After 3 months, 300 of the 508 respondents were randomly selected and informed of the objective of this study. The individuals who gave their written consent to participate were asked to complete web-based versions of these questionnaires. Participants were given an ID and password to complete the web-based survey on their own computers. There were no restrictions on the type of computer (eg, personal computer, tablet, or smartphone) that they could use to complete the survey. A total of 140 parents replied to the web-based format and were included in the final sample in the present study. Table 1 shows the demographic characteristics of this sample; Multimedia Appendix 1 contains the characteristics of the 368 people who did not respond to the web-based survey.

Table 1. Participants' demographic characteristics.

Characteristics	Value (N=140), n (%)
Children's gender	
Boy	78 (55.7)
Girl	62 (44.3)
Children's age (months)	
60	3 (2.1)
61	25 (17.9)
62	21 (15.0)
63	25 (17.9)
64	19 (13.6)
65	32 (22.9)
66	15 (10.7)
Respondent	
Mother	127 (90.7)
Father	13 (9.3)
Childcare during daytime	
Nursery school	115 (82.1)
Kindergarten	24 (17.1)
Mother	1 (0.7)
Household income (JPY[a])	
<2 million	10 (7.1)
2-4 million	44 (31.4)
4-7 million	56 (40.0)
7-10 million	18 (12.9)
>10 million	7 (5.0)
Don't know	5 (3.6)

[a]JPY: Japanese Yen; an approximate exchange rate of US $1= 103.80 JPY.

Measures

ASSQ

The ASSQ has 27 items that assess autistic features such as social interaction and communication problems, behaviors that are restrictive and repetitive, motor clumsiness, and other associated symptoms, including motor and vocal tics [16,17]. The items are rated on a 3-point scale ranging from 0 (not true) to 2 (true). A higher ASSQ score indicates more severe autistic problems. The total possible score of the ASSQ ranges from 0 to 54. In this study, we used the Japanese version of the ASSQ. A previous study [19] revealed that the ASSQ had good reliability (autism spectrum disorder clinical group: Cronbach α=.88; community group: Cronbach α=.87) and validity as a screening instrument for use with preschoolers in Japanese community settings.

ADHD-RS

The ADHD-RS includes 18 items to measure 2 features of attention-deficit/hyperactivity disorder: Inattention (9-item subscale) and Hyperactivity/Impulsivity (9-item subscale) [20]. It is evaluated on a 4-point scale ranging from 0 (not at all or rarely) to 3 (very often). Higher scores on the ADHD-RS indicate more severe attention-deficit/hyperactivity disorder problems, with total scores ranging from 0 to 54. This study used the Japanese version of the ADHD-RS home form [37]. A previous study [22] revealed that this version of the ADHD-RS had sufficient reliability (Inattention subscale: Cronbach α=.88, Hyperactivity/Impulsivity subscale: Cronbach α=.85) and validity to screen for children potentially living with attention-deficit/hyperactivity disorder in a community setting.

DCDQ

The DCDQ consists of 15 items organized into 3 subscales—Control During Movements (6 items), Fine Motor and Handwriting (4 items), and General Coordination (5 items) [23]. Parents or primary caregivers were asked to evaluate the degree of motor coordination in their children compared to that of other children of the same age on a 5-point scale ranging from 1 (not at all like your child) to 5 (extremely like your child). Lower scores indicate severe developmental coordination disorder symptoms. The total possible score ranges from 15 to 75. This study used the Japanese version of the DCDQ, which has sufficient criterion validity, fit indices, and internal consistency (Total: Cronbach α=.93; Control During Movements subscale: Cronbach α=.91; Fine Motor and Handwriting: Cronbach α=.91; General Coordination: Cronbach α=.81) when used with preschool- and school-age children [38].

P-SDQ

The P-SDQ includes 25 items that assess children's strengths and difficulties on 5 different subscales (each comprising 5 items): Emotional Symptoms, Conduct Problems, Hyperactivity/Inattention, Peer Relationship Problems, and Prosocial Behavior [24,25]. Parents or principal caregivers rated the items on a 3-point scale ranging from 0 (not true) to 2 (certainly true). The score for each subscale is calculated by summing the scores of 5 items, ranging from 0 to 10. The Total Difficulties score is calculated by summing the 4 difficulty subscale scores, ranging from 0 to 40. Higher scores on the 4 difficulty subscales as well as the Total Difficulties score indicate more severe emotional and behavioral deficits. Meanwhile, a higher score on the Prosocial Behavior subscale represents a more positive aspect of prosocial behavior. In this study, we used the P-SDQ, which showed favorable psychometric properties in Japanese community-based samples (Total Difficulties: Cronbach α =.77; Emotional Symptoms: Cronbach α=.61; Conduct Problems: Cronbach α=.52; Hyperactivity/Inattention: Cronbach α=.75; Peer Relationship Problems: Cronbach α=.52; Prosocial Behavior: Cronbach α=.69) [39].

Statistical Analysis

To test internal consistency, we calculated McDonald ω coefficients for the total and subscale scores of each measure, based on a previous study's recommendation [40], for both web-based and paper formats. We also calculated intraclass correlation coefficients between the web-based and paper formats to evaluate the interformat reliability. Paired 2-tailed *t* tests were performed to evaluate mean score differences between the web-based and paper formats to examine equivalence in quality. A *P* value <.05 was statistically significant. Analyses were performed using SPSS software (version 25.0; IBM Corp) and R (version 4.0.3; R Foundation for Statistical Computing).

Ethics

The research was performed in accordance with the ethical guidelines of the Declaration of Helsinki. The protocol of this study was approved by the Committee on Medical Ethics of Hirosaki University (IRB 2018-168). To protect personal data, we adhered to the city's and the committee's information security policies.

Results

Internal Consistency

Table 2 shows the McDonald ω coefficients for both formats of the questionnaires.

Table 2. McDonald ω coefficients for the web-based and paper versions of the ASSQ, ADHD-RS, DCDQ, and P-SDQ (N=140).

Scale and subscales	McDonald ω	
	Web-based	Paper
Autism Spectrum Screening Questionnaire	.90	.86[a]
Attention-Deficit/Hyperactivity Disorder Rating Scale		
Total	.94	.93[b]
Inattention	.90	.88[c]
Hyperactivity/Impulsivity	.88	.87[c]
Developmental Coordination Disorder Questionnaire		
Total	.94	.92[a]
Control During Movement	.87	.86[a]
Fine Motor/Handwriting	.88	.91[d]
General Coordination	.82	.74
Parent-rated Strength and Difficulties Questionnaire		
Total Difficulties	.81	.78
Emotional Symptoms	.64	.70
Conduct Problems	.55	.50
Hyperactivity/Inattention	.78	.79
Peer Relationship Problems	.57	.52
Prosocial Behavior	.76	.80

[a]Calculated for 137 participants because of missing data.

[b]Calculated for 133 participants because of missing data.

[c]Calculated for 134 participants because of missing data.

[d]Calculated for 139 participants because of missing data.

Based on a previous study [41], an internal consistency coefficient below .70 is considered unacceptable, a coefficient from .70 to .79 is considered fair, a coefficient from .80-.89 is considered good, and a coefficient of .90 or above is considered excellent. The McDonald ω coefficients for both the web-based and paper formats of the ASSQ ranged from .86 to .90, indicating good to excellent internal consistency. The McDonald ω coefficients for both the web-based and paper formats of the overall ADHD-RS and its subscales ranged from .87 to .94, also indicating good to excellent internal consistency. Meanwhile, those for both the web-based and paper formats of the overall DCDQ and its subscales ranged from .74 to .94, indicating fair to excellent internal consistency. The McDonald ω coefficients for the web-based and paper formats of the Total Difficulties subscale and the subscales of the P-SDQ ranged from .52 to .81. Notably, the McDonald ω coefficients for both the web-based and paper versions of the Peer Relationship Problems and Conduct Problems subscales and the web-based version of the Emotional Symptoms subscale were all unacceptable [41], with coefficients ranging from .51 to .66.

Intraclass Correlation Coefficients

Table 3 presents the intraclass correlation coefficients between each format for ASSQ, ADHD-RS, DCDQ, and P-SDQ.

Table 3. Intraclass correlation coefficients between the web-based and paper formats of the ASSQ, ADHD-RS, DCDQ, and P-SDQ (N=140).

Scale and subscales	Intraclass correlation[a] (95% CI)
Autism Spectrum Screening Questionnaire	0.66[b] (0.56-0.75)
Attention-Deficit/Hyperactivity Disorder Rating Scale	
Total	0.72[c] (0.62-0.79)
Inattention	0.66[d] (0.55-0.74)
Hyperactivity/Impulsivity	0.74[d] (0.65-0.80)
Developmental Coordination Disorder Questionnaire	
Total	0.71[b] (0.61-0.78)
Control During Movement	0.71[b] (0.62-0.79)
Fine Motor/Handwriting	0.66[e] (0.55-0.74)
General Coordination	0.66 (0.56-0.75)
Parent-rated Strength and Difficulties Questionnaire	
Total Difficulties	0.73 (0.65-0.80)
Emotional Symptoms	0.59 (0.47-0.69)
Conduct Problems	0.66 (0.55-0.74)
Hyperactivity/Inattention	0.68 (0.58-0.76)
Peer Relationship Problems	0.58 (0.45-0.68)
Prosocial Behavior	0.55 (0.43-0.66)

[a]All correlations were significant at the $P<.001$ level.

[b]Calculated for 137 participants because of missing data.

[c]Calculated for 133 participants because of missing data.

[d]Calculated for 134 participants because of missing data.

[e]Calculated for 139 participants because of missing data.

Intraclass correlation coefficients between 0.50 and 0.75 are considered moderate, whereas values above 0.75 are considered high [42]. The intraclass correlation coefficient between the web-based and paper formats of ASSQ was moderate and significant ($P<.001$). There were also moderate significant ($P<.001$) intraclass correlations found between the web-based and paper formats of the overall scale and subscales of the ADHD-RS and the DCDQ, and the P-SDQ subscales.

Mean Differences Between the Web-Based and Paper-and-Pencil Formats

Table 4 shows the mean scores and standard deviations of both formats of the ASSQ, ADHD-RS, DCDQ, and P-SDQ.

Table 4. Mean scores for the web-based and paper formats of ASSQ, ADHD-RS, DCDQ, and P-SDQ (N=140).

Scale and subscales	Web-based, mean (SD)	Paper, mean (SD)	*t* test (*df*)	*P* value	Cohen *d*
Autism Spectrum Screening Questionnaire	4.10 (5.46)	3.99 (4.67)	0.31 (136)	.76	0.02
Attention-Deficit/Hyperactivity Disorder Rating Scale					
Total	4.88 (7.08)	6.14 (6.94)	2.83 (132)	.005	0.18
Inattention	2.64 (3.84)	3.22 (3.69)	2.15 (133)	.03	0.22
Hyperactivity/Impulsivity	2.22 (3.46)	2.92 (3.62)	3.22 (136)	.002	0.20
Developmental Coordination Disorder Questionnaire					
Total	58.85 (11.53)	57.74 (10.52)	1.55 (136)	.12	0.09
Control During Movement	22.69 (4.89)	22.21 (4.48)	1.57 (136)	.12	0.06
Fine Motor/Handwriting	16.54 (3.41)	16.36 (3.66)	0.73 (138)	.47	0.05
General Coordination	19.72 (4.35)	19.26 (3.95)	1.61 (139)	.11	0.11
Parent-rated Strength and Difficulties Questionnaire					
Total Difficulties	7.42 (4.83)	7.80 (4.72)	1.29 (139)	.20	0.08
Emotional Symptoms	1.74 (1.69)	1.88 (1.76)	1.08 (139)	.28	0.07
Conduct Problems	1.82 (1.50)	1.94 (1.51)	1.08 (139)	.28	0.08
Hyperactivity/Inattention	2.67 (2.16)	2.76 (2.17)	0.63 (139)	.53	0.05
Peer Relationship Problems	1.19 (1.37)	1.22 (1.36)	0.27 (139)	.79	0.02
Prosocial Behavior	7.86 (1.92)	7.77 (2.12)	0.53 (139)	.60	0.05

There was no significant difference between the web-based and paper formats for the total mean score of the ASSQ (*P*=.76). Web-based scores were significantly lower than those of the paper format for the total mean scores of the ADHD-RS (t_{132}=2.83, *P*=.005, Cohen *d*=0.18) and for its subscales (Inattention: t_{133}=2.15, *P*=.03, Cohen *d*=0.22; Hyperactivity/Impulsivity: t_{133}=3.21, *P*=.002, Cohen *d*=0.20). We found no significant differences between the web-based and paper formats for the mean scores on the DCDQ total (*P*=.12) and subscales (Control during Movement: *P*=.12; Fine Motor/Handwriting: *P*=.47; General Coordination: *P*=.11). Similarly, there were no significant differences between the web-based and paper formats for total P-SDQ scores (*P*=.20) and mean subscale scores (Emotional Symptoms: *P*=.28; Conduct Problems: *P*=.28; Hyperactivity/Inattention: *P*=.53; Peer Relationship Problems: *P*=.79; Prosocial Behavior: *P*=.60).

Discussion

Principal Findings

The purpose of this study was to examine the interformat reliability of the web-based versions of the ASSQ, ADHD-RS, DCDQ, and SDQ by comparing the internal consistency, intraclass correlation, and mean score differences of their web-based and paper formats.

For the ASSQ, ADHD-RS, and DCDQ, the McDonald ω coefficients were sufficient for both the web-based and paper versions, similar to findings in previous studies [19,22,38] that calculated Cronbach α. These results indicate that the web-based format of these questionnaires has good internal consistency. The McDonald ω coefficients for the Total Difficulties, Hyperactivity/Inattention, and Prosocial Behavior subscales of both the web-based and paper versions of the P-SDQ were also good. The McDonald ω coefficient for the paper version of the Emotional Symptoms subscale in this study was also good; however, it was unsatisfactory for the Conduct Problems and Peer Relationship Problems subscales of both the web-based and paper versions. The McDonald ω coefficient for the web-based version of the Emotional Symptoms subscale was also relatively low. These results are similar to those found in studies [25,39,43] that calculated Cronbach α for the paper format versions. Therefore, our findings suggest that, similar to the paper format, there are a few difficulties in using the web-based format of the P-SDQ for assessing externalizing and internalizing problems in children.

We found that there were significant (*P*<.001) moderate positive intraclass correlations between the web-based and paper formats of the ASSQ, ADHD-RS, DCDQ total scores, and Total Difficulties score of the P-SDQ. Similar to this study, several earlier studies [44-46] on the equivalence between web-based and paper formats of self-report questionnaires meant to assess psychiatric symptoms have reported moderate significant correlations, suggesting that web-based questionnaire administration was a reliable alternative to using the paper format. Hence, it seems that the questionnaires assessing neurodevelopmental disorders, such as the ASSQ, ADHD-RS, DCDQ, and P-SDQ, can be made available and administered in web-based situations as well. However, another study [27] had earlier pointed out that the agreement rate between the web-based and paper formats was higher for objective factual questions than for questions based on personal subjective evaluation. The questionnaires used in this study were subjective evaluations of children's developmental status by parents, which may have impacted the correlation values.

Furthermore, the analyses revealed that there were no significant differences in the ASSQ total mean scores between the web-based and paper formats (*P*=.76). This result suggests that the web-based version of the ASSQ is equal in quality to that of the paper version. However, there were significant differences in the ADHD-RS total and subscale mean scores between the web-based and paper formats (total: *P*=.005; Inattention: *P*=.03; Hyperactivity/Impulsivity: *P*=.002). Previous studies [33,36,47] have reported a significant difference in mean scores between web-based and paper formats; however, due to the small effect size (Cohen *d*=0.14-0.27), it was determined that the statistically significant difference in mean scores was not clinically meaningful in practice. In light of this finding, web-based questionnaires are a potential substitute for paper-based questionnaires. The effect sizes obtained in this study were also small (Cohen *d*=0.18-0.22). Furthermore, the web-based version McDonald ω values in this study were slightly higher than those of the paper format. These results suggest that the ADHD-RS is applicable for web-based utilization. We found that there were no significant differences in the DCDQ total and subscale mean scores between the web-based and paper formats (total: *P*=.12; Control during Movement: *P*=.12; Fine Motor/Handwriting: *P*=.47; General Coordination: *P*=.11). Additionally, we also confirmed that there were no significant differences in the P-SDQ total and subscale mean scores between the web-based and paper formats (total: *P*=.20; Emotional Symptoms: *P*=.28; Conduct Problems: *P*=.28; Hyperactivity/Inattention: *P*=.53; Peer Relationship Problems: *P*=.79; Prosocial Behavior: *P*=.60). These results suggest that both the DCDQ and P-SDQ web-based formats are equivalent in quality to those of the paper format.

Strengths and Limitations

The evidence found in this study supports the applicability of the web-based versions of the ASSQ, ADHD-RS, DCDQ, and P-SDQ. It has been pointed out that children with possible neurodevelopmental disorders may be overlooked during developmental health check-ups in Japan [48], and there is a lack of specialized organizations capable of assessing neurodevelopmental disorders. Furthermore, in the current COVID-19 pandemic, it is also difficult to conduct in-person evaluations. This study, which shows the applicability of web-based questionnaires assessing neurodevelopmental disorders, has the potential to improve early detection and intervention for these disorders in regions where specialized services are lacking and under the present pandemic conditions.

However, there are some limitations to this research. First, the discriminant validity of the web-based version of each questionnaire was not confirmed in this study. This is because this study used a cross-sectional research design as part of a community developmental health check-up. Therefore, it is unclear whether the children included in this study had been diagnosed with neurodevelopmental disorders or experienced other emotional or behavioral problems or deficits that do not meet the criteria for a clinical diagnosis. It is necessary to verify the discriminant ability of the web-based version of each questionnaire for both clinical and nonclinical groups. Second, the raters who evaluated the children's condition in this study were mostly parents. Previous research [17,49,50] conducted with paper-based questionnaires has examined the psychometric properties of the teacher-rated version of each questionnaire that was used in this study. In the future, we also need to clarify the psychometric properties of the teacher-rated web-based version of the questionnaires used in this study compared to the parent-rated web-based version. Third, we were not able to examine the impact of the order in which the questionnaires were administered. Previous studies [26,33] have confirmed that the order in which web- and paper-based questionnaires are administered has an effect on the scores of those questionnaires. In the future, it will be necessary to investigate the effect of the order of administration of the questionnaires in the research design. Fourth, the age of the children in this study was limited to 5 years. Previous studies [17,38,49,50] on the paper-based ASSQ, ADHD-RS, and SDQ have been conducted with school-age children. It is necessary to investigate the psychometric properties of the web-based version of the ASSQ, ADHD-RS, DCDQ, and P-SDQ among school-age children. Fifth, we did not control for the computer used by the participants in this study. We need to test whether the type of computer affects their responses in the future. Finally, this study was conducted in one medium-size city in Japan, thereby limiting the generalizability of its findings to other regions.

Conclusions

This study examined the interformat reliability of the web-based versions of questionnaires for assessing neurodevelopmental disorders. Our findings showed that the web-based versions of the ASSQ, ADHD-RS, DCDQ, and P-SDQ had the same level of internal consistency, intrarater reliability, and equality as their paper versions. These results indicate the web applicability of these questionnaires for assessing neurodevelopmental disorders.

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Conflicts of Interest

None declared.

Multimedia Appendix 1
Participants who took part in the paper-and-pencil survey only (N=368).
[DOCX File , 16 KB - pediatrics_v4i1e20172_app1.docx]

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Abbreviations

ADHD-RS: Attention-Deficit/Hyperactivity Disorder Rating Scale
ASSQ: Autism Spectrum Screening Questionnaire
DCDQ: Developmental Coordination Disorder Questionnaire 2007
DSM-5: Diagnostic and Statistical Manual for Mental Disorders (Fifth Edition)
P-SDQ: Parent-reported Strengths and Difficulties Questionnaire
SDQ: Strengths and Difficulties Questionnaire

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Original Paper

Monitoring Adherence Rate to Growth Hormone Therapy and Growth Outcomes in Taiwanese Children Using Easypod Connect: Observational Study

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Abstract

Background: Adherence to growth hormone therapy is difficult to detect reliably. Devices such as easypod have been developed for electronic recording of injections. The easypod connect observational study (ECOS) was an open-label, observational, multinational, phase IV study conducted in 24 countries around the world. The final results from ECOS in the Taiwanese cohort are reported in this paper.

Objective: This study aimed to evaluate the adherence and long-term outcomes of growth hormone therapy in pediatric subjects using the easypod electromechanical device.

Methods: Subjects (aged 2-18 years or >18 years without fusion of growth plates) who received Saizen (recombinant human growth hormone, somatropin) via the easypod device were enrolled in this study. The primary objective was to assess the level of adherence in subjects receiving Saizen via easypod.

Results: In Taiwan, a total of 35 and 13 children fulfilled the criteria of full analysis set and complete analysis set, respectively. The mean (SD) age of the complete analysis set was 12.08 (2.72) years. All subjects were growth hormone–naïve, with 38% (5/13) females. The mean adherence rates of 13 subjects were 87.6% at 3 months and 84.3% at 6 months, that of 8 subjects was 81.0% at 9 months, and that of 4 subjects was 91.6% at 1 year. After 1 year of treatment, subjects had a median (Q1:Q3) change in height SD score of 0.30 (0.06:0.48), median height velocity of 6.50 (4.33:8.24) cm/year, and median change in height velocity SD score of 1.81 (–0.04:3.52).

Conclusions: With the easypod device, patients with inadequate adherence and poor response to treatment can be identified. Adherence to growth hormone therapy administered via easypod was generally high in the first year of treatment but the adherence gradually decreased over time. Overall, growth outcomes after 1 year indicated a positive growth response to growth hormone treatment. Future efforts should be focused on personalized management of adherence by using the easypod system.

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KEYWORDS

growth hormone; adherence; easypod; eHealth

Introduction

Background

Human growth hormone, also known as somatotropin, is synthesized and secreted by the somatotropic cells of the anterior pituitary gland and it plays a critical role in growth and metabolism. Recombinant human growth hormone was first approved for the treatment of childhood growth hormone deficiency in 1985 [1]. Since then, synthetic human growth hormone has been widely administered for the treatment of inadequate secretion of endogenous growth hormones in children and adults. For pediatric patients, growth hormone is indicated for treating growth disorders due to a number of medical causes, including growth hormone deficiency, Turner syndrome, and children born small for gestational age. During the past several decades, growth hormone therapy has demonstrated its effects on improving growth outcomes and helping children achieve catch-up growth [2-5].

Adherence to Growth Hormone Treatment

As growth hormone therapy for children generally starts at a young age and lasts for several years, both the child and the family are involved in this long-term treatment process. For chronic non–life-threatening conditions such as growth hormone deficiency [6], adherence to treatment is relatively difficult to maintain at a high level, especially when the benefits are not immediately apparent, and regular subcutaneous injections with a frequency of up to once daily causes both physical and psychological burdens. Even though adherence can be monitored through methods such as diary cards or by comparing total expected growth hormone usage to the total amount of growth hormone prescribed, the data could easily be overestimated and become unreliable since the child or the parents may be reluctant to admit missing injections [7].

Studies have shown that growth outcomes of growth hormone therapy could be affected by multiple factors [8-10], among which, poor adherence is still a major problem in treating growth disorders for pediatric patients [11,12]. Although the results vary substantially between studies due to the methods and definitions applied, a prevalence of 5%-82% has been reported [13]. Poor adherence not only results in suboptimal growth but also increases unnecessary medical expenses [14,15].

The frequency of inadequate adherence is usually underestimated when assessed using conventional methods (eg, diary cards, questionnaires, number of returned vials) [13,16], which only give fragmentary pictures of a patient's dosing history. In addition, the aforementioned methods cannot completely reveal the patterns of nonadherence such as reduced dosage, drug holiday, or delayed initiation [16]. Thus, electronic monitoring of drug dosing histories is currently recognized as a standard for adherence quantification [16]. The electronic monitoring of injections via devices such as easypod provides information on how many doses have been taken as prescribed and about nonadministered doses, thereby reflecting the extent to which the patient is adherent to the therapy. Through adequate monitoring methods, physicians are able to promptly evaluate the adherence following an inadequate response to growth hormone therapy [17,18].

Objectives

The easypod connect observational study (ECOS) was an open-label, observational, longitudinal study conducted in 24 countries (Argentina, Australia, Austria, Canada, China, Colombia, Czech Republic, Finland, France, Greece, Hungary, Indonesia, Italy, Kingdom of Saudi Arabia, Korea, Mexico, Norway, Singapore, Slovakia, Spain, Sweden, Taiwan, United Arab Emirates, and the United Kingdom) with a total of 1203 subjects included for analyses. It aimed to evaluate the adherence and long-term outcomes of therapy in pediatric subjects using the easypod electromechanical device for growth hormone treatment and to undertake population-based analyses to generate hypotheses relating to drivers of individual adherence [19]. The results of the ECOS have been published by Koledova et al [19]. Among the countries involved, the results of Spain [20], Italy [21], and Mexico [22] have been published. However, there is no related publication in the Asia-Pacific region. The culture and living habits might possibly influence medication adherence. Clinically, some children in Taiwan go to bed late, resulting in late administration of the growth hormone, which may indirectly affect adherence. Therefore, in this study, we present the results of Taiwanese pediatric subjects.

Methods

Study Design

ECOS was a multinational, multicenter, observational, longitudinal, open-label, phase IV study conducted between November 2010 and February 2016. The study was conducted in accordance with principles of the Declaration of Helsinki and the protocol, as well as the good clinical practice (ICH-GCP E6) and the applicable national legal and regulatory requirements. The study protocol was approved by the institutional review board at each study site, and written informed consent or assent was obtained from all subjects' parents or legal guardians before enrolment.

Patients

Subjects (aged 2-18 years or >18 years without fusion of growth plates) who received Saizen (Merck KGaA) via the easypod electromechanical device were enrolled. Subjects who were receiving growth hormones in whom growth plates had fused (ie, for taking growth hormones for its metabolic effects), subjects with contraindications to Saizen as per locally approved prescribing information, subjects using an investigational drug, or subjects participating in an interventional clinical study were excluded from the study. The duration of follow-up for growth hormone treatment was planned to be at least 6 months and up to 5 years. There was 1 baseline visit followed by 1-4 subsequent visits per year as per routine practice. All assessments were performed during the visits. As an observational study, growth hormone treatment and other aspects of patient management were entirely at the discretion of the physician and his or her patient, following a standard clinical practice.

Data Collection and Study Endpoints

The primary endpoint was treatment adherence rate (percentage of prescribed injections that were administered) over time. Data

on injection time, date, dose, planned frequency were uploaded to a secure web-based database via a specific connection kit and the physician's computer. For subjects who had consented to participate in the observational study, deidentified data were then uploaded to the web-based registry/observational study. While adherence data from the enrolled subjects were primarily derived from the easypod device, other information such as demographics, relevant medical and treatment history, and auxological data (eg, height, growth velocity, and bone age) were entered by the physician into the electronic case report form. Data were collected at every visit, as available per routine practice.

Statistical Analysis

Analysis sets included a full analysis set and a complete analysis set. The full analysis set consisted of all the subjects included in the study, whereas the complete analysis set consisted of all the subjects of the full analysis set without missing the treatment start date on the electronic case report form, without gap in the injection information of more than a week after the start of the treatment, and with height measurement closest to the treatment start date not missing using a window of 3 months (91 days). All statistical analyses on adherence rates were performed on the complete analysis set and were performed in a descriptive way for the endpoints, considering this was a single-arm, noninterventional study. Continuous variables were described with the number of subjects, number of subjects with missing data, mean (SD), median, first and third quartiles (Q1, Q3), and minimum and maximum values. For categorical variables, summary statistics were the number and percentage of subjects in each category. To calculate height standard deviation score (SDS) and height velocity (HV) SDS, the reference median growth parameter and the SD of the reference growth parameter were applied. The World Health Organization reference growth table [23] and the Tanner and Whitehouse reference growth table [24] were used for height SDS and HV SDS derivation, respectively.

Results

Patient Characteristics

The ECOS was conducted in 3 medical centers in Taiwan. A total of 35 children had sufficient data and were included in the full analysis set, of which 13 subjects fulfilled the criteria of the complete analysis set. Among the 35 subjects of the full analysis set, 32 had growth hormone deficiency, 2 were born small for gestational age, and 1 had Turner syndrome. The average age was 12.26 years. More than half of the subjects were male (19/35, 54%). At baseline, all subjects were growth hormone–naïve, with a mean height of 137.06 cm and a mean growth velocity of 4.14 cm/year. The baseline demographic characteristics and auxological data are shown in Table 1.

Table 1. Demographic and auxological data of the subjects at baseline.

Demographic and auxological data	Full analysis set (N=35)	Complete analysis set (n=13)
Age (years) (min, max)	12.26 (7.0, 16.0)	12.08 (6.0, 16.0)
Sex, n (%)		
Female	16 (46)	5 (39)
Male	19 (54)	8 (61)
Asian ethnicity, n (%)	35 (100)	13 (100)
Pubertal stage, n (missing)	21 (14)	5 (8)
Tanner 1, n (%)	4 (19)	0
Tanner >1, n (%)	17 (81)	5 (100)
IGF-1 status, n (missing)	10 (25)	2 (11)
Abnormally low, n (%)	0	0
Normal, n (%)	8 (80)	2 (100)
Abnormally high, n (%)	2 (20)	0
Bone age, n (missing)	15 (20)	9 (4)
Greulich and Pyle assessment (years), (min, max)	11.84 (3.0, 15.7)	10.72 (3.0, 15.0)
Growth velocity (cm/year), (min, max)	4.14 (0.0, 9.4)	3.05 (0.0, 4.9)
Height (cm), (min, max)	137.06 (103.0, 161.0)	139.26 (103.0, 161.0)
Indication for growth hormone treatment, n (%)		
Growth hormone deficiency	32 (91)	11 (84)
Small for gestational age	2 (6)	1 (8)
Turner syndrome	1 (3)	1 (8)
Other	0	0
Adjusted mid-parent's height (cm), (min, max)	162.79 (151.0, 179.0)	164.55 (153.0, 179.0)

Adherence Rates of the Subjects

The primary endpoint was the adherence rate of subjects receiving Saizen via easypod over a period of time. Among the 13 subjects in the complete analysis set, the longest follow-up period was approximately 1.5 years, with a mean (SD) treatment duration of 332 (113.1) days, and the proportions of subjects with adherence data available for 3, 6, 9 months, and 1 year were 100% (13/13), 100% (13/13), 62% (8/13), and 31% (4/13), respectively. The median (IQR) of adherence rates over increasing periods of follow-up are presented in Figure 1. The mean adherence rate was 87.6% at 3 months, 84.3% at 6 months, and 81.0% at 9 months, indicating a slight decrease in adherence rate over time. The mean adherence rate was calculated by averaging all patients' adherence rates during a period of time. The majority of the complete analysis set subjects maintained an adherence rate of ≥80%, and these percentages remained steady at 3, 6, and 9 months (Figure 2). Subgroup analysis by sex revealed that the median adherence rates were similar between the female and male subjects (Figure 3).

Figure 1. Treatment adherence rates over time (complete analysis set). Boxes show Q1 and Q3, with median as white line and mean as red squares.

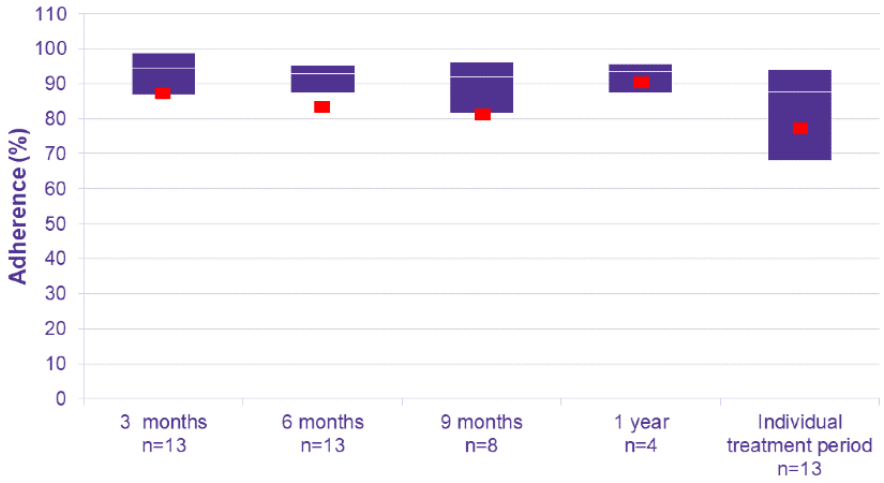


Figure 2. The proportion of patients treated with growth hormone using easypod with adherence rates of at least 80% over time and for all patients at any time within the study period.

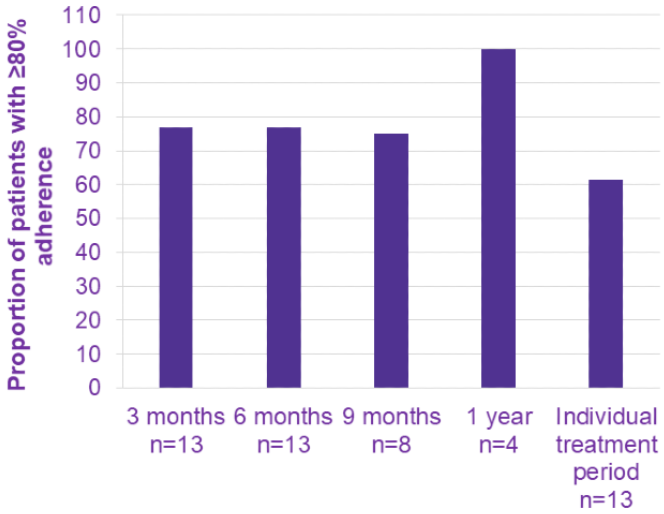
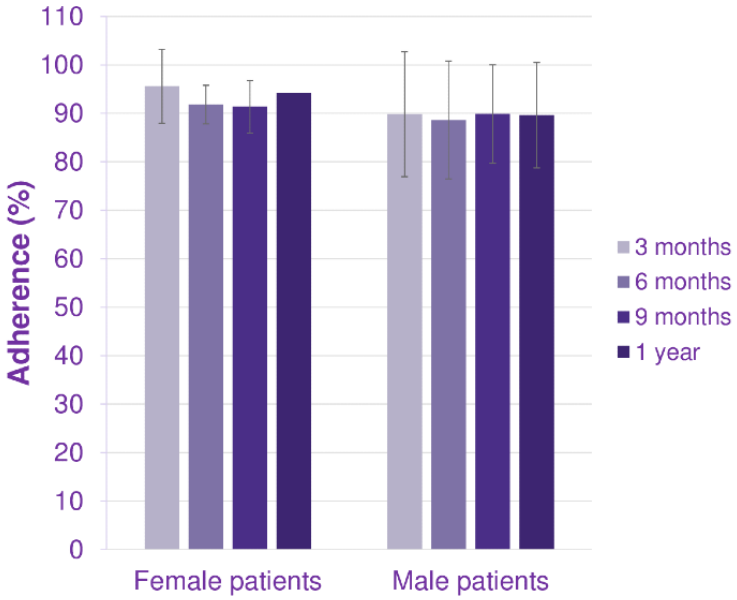


Figure 3. Treatment adherence rates over time by gender (complete analysis set).



Growth Outcomes of the Subjects

After 1 year of treatment, subjects had a mean (SD) change in height of 6.25 (3.07) cm, height SDS of +0.27 (0.30), mean (SD) HV of 6.49 (2.95) cm/year, and mean (SD) change in HV SDS of 1.51 (2.22). The growth outcomes and changes from baseline after 1 year of growth hormone treatment are summarized in Table 2. Spearman product-moment correlations between these outcomes and adherence rates were assessed to further investigate the impact of adherence on growth outcomes. Nevertheless, limited by the number of subjects with available data (n=4), no significant and consistent correlation was identified in the complete analysis set (data not shown). Overall, growth outcomes after 1 year indicated a positive growth response to growth hormone treatment.

Table 2. Growth outcomes and changes from baseline of the subjects after 1 year of growth hormone treatment using easypod (complete analysis set).

Growth outcome	Subjects with growth hormone deficiency (n=11)	Subject who was small for gestational age (n=1)	Subject with Turner syndrome (n=1)	Overall (n=13)
Baseline height (cm), mean (SD)	141.95 (14.32)	103.00	146.00	139.26 (17.05)
Change in height (cm), mean (SD)	6.20 (3.24)	8.50	4.50	6.25 (3.07)
Baseline height, SDS[a] (SD)	–1.74 (0.94)	–2.68	–2.38	–1.86 (0.91)
Change in height SDS at 1 year, mean (SD)	0.22 (0.29)	0.59	0.56	0.27 (0.30)
Baseline height velocity (cm/year), mean (SD)	3.30 (1.17)	3.92	0	3.05 (1.47)
1-year height velocity (cm/year), mean (SD)	6.56 (3.12)	7.92	4.33	6.49 (2.95)
1-year height velocity SDS, mean (SD)	1.45 (2.31)	2.23	—[b]	1.51 (2.22)

[a]SDS: standard deviation score.

[b]Not available because of missing data.

Discussion

The ECOS assessed the adherence to recombinant human growth hormone treatment as well as growth outcomes in pediatric patients with growth disorders. The results of the European and American countries involved in this open-label, observational, longitudinal study have been published [20-22]. To our knowledge, there is no literature exploring the adherence to growth hormone treatment in Taiwanese or Chinese pediatric patients in a real-life setting, especially by using an electronic monitoring method. In Taiwan, physicians' clinical experience has shown that some children have relatively low adherence owing to late administration of growth hormones with late bedtime. The culture and living habits of Asians such as children's daily routines, the time of going to and coming from school, and the activities after school are quite different from others in the world. The medication adherence might be possibly influenced by these differences. In this study, the Taiwanese cohort of the ECOS is reported. The mean adherence rate was generally high in the first year of the treatment, with the majority of the complete analysis set subjects maintaining an adherence rate of greater than 80%. Although the adherence gradually decreased with a longer duration of follow-up, it is in line with the global ECOS results [19] as well as with that of previous studies showing that the adherence rate diminished over time [15,25].

Reduced adherence to growth hormone therapy is detrimental to therapeutic outcomes [11,14,26] and is considered one of the major causes of suboptimal growth [27]. To maximize the effect of growth hormone therapy, it is necessary to maintain good adherence throughout the entire treatment course. Nonadherence not only represents an obstacle to effective treatment, but it also leads to an increase in the medication costs from direct and indirect aspects [13]. Previous literature has shown that adherence could be negatively or positively associated with a variety of factors such as reduced HV [13], comprehensive medical education/training [28], duration of treatment [10,26,29], and choice of injection device [29]. Through real-time monitoring, timely interventions can be prompted in response to nonadherence, rather than signaled by suboptimal growth at a later stage. Moreover, with reliable information regarding adherence at hand, physicians are able to tell whether suboptimal growth arises from nonadherence or other possible causes.

The prevalence and the level of adherence rate vary considerably among studies, which is partly attributable to the methods applied as well as inconsistent definitions used across the studies [13]. While it has been reported that 39%-66% of the patients missed more than 1 injection per week [10,14], another study showed that the median adherence rate might be up to 95% [30]. Most previous studies investigating adherence were cross-sectional, and the adherence was assessed with a questionnaire-based survey [28,29,31]. As an electronic monitoring device is currently recognized as a standard for quantifying adherence [16], it provides information of the precise time and doses of injections, which allows further analyses for nonadherence patterns [16]. A concordance of 84.3% between adherence reported by patients and recorded using easypod has been demonstrated in a study, and the authors found that there was a trend toward self-reported adherence being higher than the recorded adherence [7]. Nevertheless, it is not known whether the difference resulted from forgetfulness, fear of disappointing practitioners, or a combination of factors [32], and no data are currently available to assess this supposition.

In this study, one of the study objectives was to describe the impact of adherence on clinical outcomes for subjects receiving Saizen via easypod. In fact, with 1190 evaluable subjects, the ECOS global results revealed that statistically significant correlations of 0.13 and 0.08 were observed between adherence rate and change in height SDS and between adherence rate and HV SDS, respectively, indicating a positive correlation between adherence rate and growth outcomes. Unfortunately, the number of subjects was not sufficient to support such analyses for the subgroup of Taiwanese patients, since only 4 patients were administered Saizen for more than 1 year. To consolidate the correlation between adherence and growth outcomes, larger sample sizes are required for future studies.

Suboptimal adherence is a common problem in growth hormone treatment. Since adherence to growth hormone therapy is critical for the optimization of treatment outcomes, it has to be taken into account while evaluating the therapeutic effects for treatment modulation in routine clinical practice. Detection of nonadherence can be difficult using pre-electronic monitoring methods because the patient may be reluctant to admit such behavior [7]. The electronic monitoring of injections via devices such as easypod provides reliable and objective information on how many doses have been taken as prescribed and about the nonadministered doses, which reflect the extent to which the patient is adherent to the therapy. The electronic monitoring device, as distinct from conventional monitoring methods, is less labor-intensive and enables physicians to review the timing, date, and dosage of recombinant human growth hormone delivered in a real-time manner. It may help promote adherence and prompt disease management for routine practice.

This study was restricted by its observational nature as there was a considerable level of missing data and intersubject variability. In addition, as mentioned above, the number of subjects included in the complete analysis set was also limited, and most patients had a treatment duration of less than 1 year. Nevertheless, this paper shows the adherence patterns of pediatric patients using an electronic monitoring device, which have not been previously reported in a Taiwanese patient population. To the best of our knowledge, this is the first study providing insight into the adherence rate and characteristics of Taiwanese pediatric patients who require growth hormone treatment. Of note, future studies are warranted to confirm the results and to further explore the effects of individual variables such as bone age at baseline, socioeconomic statuses, and parental, marital, or employment status.

This was a phase IV, open study, and its conditions were different from the phase II or phase III randomized controlled trial. As a phase IV postmarketing study, it generally aims to explore treatment effectiveness and long-term safety. Compared with a randomized controlled trial, observational trials usually reflect the actual clinical treatment effectiveness because the trial design does not have as many limitations in the inclusion/exclusion conditions as a randomized controlled trial. This study did not specify the length of time of patients receiving easypod treatment. Many patients were treated for less than 1 year. In addition to being limited by the number of results, the length of the treatment period is also one of the possible reasons.

Collectively, this study unveils the adherence over time among Taiwanese pediatric patients receiving growth hormone treatment via the easypod electronic monitoring device. The growth outcomes and changes after 1 year of treatment are also presented, although the associations between adherence rate and growth outcome as well as factors affecting adherence to growth hormone therapy in Taiwanese patients were limited by the sample size. The electronic monitoring/injection device serves as a useful tool for both patients and physicians to help disease management and provide direct information regarding adherence to growth hormone therapy.

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Conflicts of Interest

PHS, CY, and MCC declare no conflicts of interest. CLC is an employee of Merck Ltd.

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Abbreviations

ECOS: easypod connect observational study
HV: height velocity
SDS: standard deviation score

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Original Paper

Patient-Generated Health Data in Pediatric Asthma: Exploratory Study of Providers' Information Needs

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Abstract

Background: Adolescents are using mobile health apps as a form of self-management to collect data on symptoms, medication adherence, and activity. Adding functionality to an electronic health record (EHR) to accommodate disease-specific patient-generated health data (PGHD) may support clinical care. However, little is known on how to incorporate PGHD in a way that informs care for patients. Pediatric asthma, a prevalent health issue in the United States with 6 million children diagnosed, serves as an exemplar condition to examine information needs related to PGHD.

Objective: In this study we aimed to identify and prioritize asthma care tasks and decisions based on pediatric asthma guidelines and identify types of PGHD that might support the activities associated with the decisions. The purpose of this work is to provide guidance to mobile health app developers and EHR integration.

Methods: We searched the literature for exemplar asthma mobile apps and examined the types of PGHD collected. We identified the information needs associated with each decision in accordance with consensus-based guidelines, assessed the suitability of PGHD to meet those needs, and validated our findings with expert asthma providers.

Results: We mapped guideline-derived information needs to potential PGHD types and found PGHD that may be useful in meeting information needs. Information needs included types of symptoms, symptom triggers, medication adherence, and inhaler technique. Examples of suitable types of PGHD were Asthma Control Test calculations, exposures, and inhaler use. Providers suggested uncontrolled asthma as a place to focus PGHD efforts, indicating that they preferred to review PGHD at the time of the visit.

Conclusions: We identified a manageable list of information requirements derived from clinical guidelines that can be used to guide the design and integration of PGHD into EHRs to support pediatric asthma management and advance mobile health app development. Mobile health app developers should examine PGHD information needs to inform EHR integration efforts.

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KEYWORDS

information needs; asthma; symptom management; mobile health; patient-generated health data; pediatrics; adolescents

Introduction

Background

Poorly controlled pediatric asthma continues to be a challenge. Pediatric asthma, the leading chronic disease among children, remains prevalent, and improvements in outcomes have stalled [1]. It is estimated that 6 million children under the age of 18 years in the United States have the chronic airway disease [2]. Despite evidence-based clinical guidelines, suboptimal treatment continues to contribute to a lack of asthma control [3]. Pediatric asthma can be managed with medications and trigger avoidance but requires continuous monitoring to assess control and detect triggers [1,4].

Understanding the complete picture of triggers and symptoms is essential for management, requiring health care providers to perform periodic assessments, adjust treatment plans, and personalize care [5]. However, a lack of objective data from patients means that a provider must depend on patient self-report, known to have reliability challenges, to make clinical decisions [6-8]. A potential solution may be the presentation of relevant patient-generated health data (PGHD) directly in the clinical documentation used by providers as they make decisions. PGHD such as biometric and physical activity, surveys, and health history are data captured electronically by patients outside of the clinic or hospital.

Mobile health (mHealth) technologies offer feasible opportunities to engage adolescents, persons between ages 10 to 19 years, in collecting PGHD [9]. Younger generations in every country are more likely than others to own a phone and are likely to use new technologies [8,10]. Moreover, adolescents engage with their mobile devices even while sick or hospitalized [11]. For pediatric asthma patients, mHealth apps support self-management, and wearable sensors provide ongoing monitoring capabilities [12,13]. The types of data collected from patients using smartphone asthma apps include symptoms, medication adherence, night awakenings, physical activity, and peak-flow expiratory rates [4,5,12,14]. Authors of two studies suggested that collecting the patient's local environmental data, such as pollen counts, ambient temperature, and humidity, should also be considered [3,4]. When shared during clinical encounters, PGHD have the potential to facilitate assessment, diagnosis, and ongoing patient monitoring [15]. Presenting PGHD within the electronic health record (EHR) is envisioned as an optimal approach so that providers do not need to interrupt their cognitive processes and workflows to navigate between different systems.

Not much is known about which PGHD are of value or how to present PGHD to the providers in the EHR. A recent scoping review showed that EHR integration of PGHD is at an emergent phase; another identified only three asthma apps with the ability to share data with other apps [16,17]. Although many asthma apps exist, only a few of the mHealth technologies developed for childhood asthma have elicited feedback from clinicians [8], and even highly rated apps have not reported integration into clinical workflows [5]. The need for EHR data sharing has been recognized [15], and one study concluded that the introduction of smart-inhaler monitoring data into the EHR might support the development of individualized asthma treatment plans [7].

Despite the potential benefits, clinicians have expressed concerns that incorporating PGHD into the EHR will further contribute to information overload [13,18]. Additionally, studies reported issues with embedding mHealth technologies into clinical workflows and identified uncertainties about organizational readiness to integrate other data sources [13]. To ensure the clinical utility of PGHD, it is vital to understand the clinical workflows in which to integrate PGHD, as well as the specific tasks and decisions that PGHD must support and the relevant information needs of providers. Moreover, the discovery of information needs is necessary to inform future mHealth app implementations.

Purpose

The purpose of this exploratory study was to identify and characterize a discrete set of tasks, decisions, and information needs of providers caring for patients with pediatric asthma and assess whether PGHD might provide useful information. We used outpatient care of patients with pediatric asthma as an exemplar clinical encounter where PGHD might have clinical value. By understanding these needs, we will be able to design interfaces and displays that optimally support the integration of PGHD into EHRs for the management of pediatric asthma.

Methods

Framework and Recruitment

We applied qualitative, descriptive methods to gain insights into key provider tasks, decisions, and information needs regarding PGHD and pediatric asthma. The procedures included analyzing published clinical guidelines to identify relevant decisions and consulting with providers treating patients with pediatric asthma to validate a discrete set of tasks and elicit their perspectives and priorities regarding the decisions, information needs, and potentially relevant PGHD.

We referred to the 3-phase model of needs assessment described by Altschuld and Kumar [19] as a guide. This model or framework proposes a practical process to assess needs that can be molded for a specific situation or setting. In the first phase of the model, preassessment, the goal is to determine what is already known regarding clinician needs and PGHD. We considered the preliminary examination of existing clinical guidelines as the activity to satisfy the preassessment phase or part 1 of this study. For the second phase or part 2 of the study, the assessment consisted of needs assessment procedures and data collection and validation with experts to move toward a full understanding. The third phase of the model, postassessment, involves the identification of strategies or development of solutions to meet the needs that were found during the assessment phase. We will consider the activities related to the postassessment phase in future research.

We recruited a convenience sample of three subject-matter experts (SMEs), domain specialists in pediatric asthma. Although no empirical evidence exists for the most appropriate number of experts for guideline review, similar studies that explored knowledge elicitation for consensus-based guidelines

used at least three task experts [20,21]. We solicited SMEs by reputation using local clinical contacts. Inclusion criteria were the ability to read, understand, and use English as a primary language; self-reported expertise in pediatric asthma; and a history of medical practice in the United States. We excluded providers with adult-only asthma experience, and we did not compensate participants for their time. We received consent from all participants and obtained ethical approval for this study from the institutional review board of the University of Utah.

Identify Tasks, Decisions, and Patient-Generated Health Data

We began with a review of the evidence-based pediatric asthma guidelines. We used the two main authoritative sources in pediatric asthma management from the National Institutes of Health National Heart, Lung, and Blood Institute (NHLBI) Expert Panel Report 3: Guidelines for the Diagnosis and Management of Asthma [22] and the Global Initiative for Asthma (GINA) Pocket Guide for Health Professionals [23]. The NHLBI asthma guidelines, in place for more than 25 years, focus on treatment protocols and monitoring for quality asthma care [3]. The GINA report serves as a practical tool to support asthma care and provides the basis for ongoing guideline revisions [24]. The development of these guidelines consisted of formal consensus methods commonly used for clinical guidelines.

According to the preassessment phase of the model, we conducted the needs assessment procedures and derived tasks, decisions, and information needs directly from the guidelines. In this context, a task is a professional duty or clinical responsibility related to patient care [25]. In the development of valid clinical guidelines, tasks are recommended to satisfy the goals of the guideline. Each of the tasks is linked to decisions: cognitive activities involving choices between alternatives or choices about what to believe or what to do [26]. In order to support the appropriate decision, information must be acquired from a person or an external system (an information need). One informatics expert on the research team extracted a list of high-level tasks from the GINA report and the primary task components from the NHLBI guidelines. Most of these were readily identified within each of the guideline documents, with tasks and decisions explicitly identified as such. Then, using the guidelines, the high-level decisions supported by each of the tasks were identified and listed alongside the information collected from the patient that assists with, or could assist with, making the decision. Once the extraction of tasks, decisions, and information needs was competed, the list was discussed with two other clinical informatics experts from the research team for agreement.

To further explain the extraction process, we used the assessment and monitoring task identified in the NHLBI guidelines as an example [22]. The assessment and monitoring task section of the guidelines identified two major decisions: assess the severity of the child's asthma and decide the level of asthma control. The guidelines listed several information needs related to the decision for severity and control, such as frequency and intensity of the symptoms, functional limitations, exacerbations, lung function, and adverse effects from medication. The information needs were not labeled as such but were obvious from the text of the guidelines. Once we completed this exercise for all tasks from each of the guidelines, we synthesized the findings from both sources to create a single integrated set.

After we assembled the set of tasks, decisions, and information needs, we searched the literature for exemplar asthma mobile apps to assess whether PGHD might provide useful information. In January 2020, we searched PubMed using the terms *asthma mobile health applications* for studies that described asthma PGHD collection features. Given the small number of publications, we did not limit the search to pediatric-specific asthma apps. We examined the types of PGHD collected by each asthma mHealth app [7,14,17]. We inferred the ability of the discovered PGHD types to meet specific information needs by referring to the literature and using our clinical knowledge. Continuing from our previous example, one of the decisions for the assessment and monitoring task is to evaluate the level of asthma control. One asthma app collects the answers from the patient or caregiver and calculates an Asthma Control Test (ACT) score. The ACT is a well-validated, symptom-based tool used to assess symptom control that correlates clinically with specialist ratings and lung function [27]. The ACT is widely used and commonly part of strategies to stratify patients as having poorly controlled or well-controlled asthma [28].

We matched the discovered PGHD types to the corresponding information need in the integrated set. We continued with this process until we had a full set of mapped decisions, information needs, and PGHD for each major task category. All three clinical informatics experts from the research team reviewed the final set of tasks, decisions, information needs, and PGHD types and achieved consensus through discussion.

Clinician Perspectives

We scheduled a 30-minute, in-person meeting with each SME independently to review the integrated set of tasks, decisions, information needs, and PGHD types. We also solicited general perceptions of the use of PGHD for adolescent asthma management. In a systematic fashion, we presented the SMEs with the mapped list and asked if it was the right list, if the items were in the order of importance for asthma treatment, and their general thoughts on using PGHD in practice.

We assessed the suitability of PGHD to support their information needs and generated field notes throughout the interview process. Based on the expert feedback, we created a final prioritized list of decisions, information needs, and PGHD types. We recorded participant responses as notes, examined the field notes for themes, and summarized responses. All three clinical informaticists reviewed the findings.

Results

Information Needs and Patient-Generated Health Data Types

Our analysis of the GINA report and NHLBI guidelines identified 4 high-level tasks:

- Assessment and monitoring

- Education for self-management in partnership with the patient and family
- Control of environmental factors and comorbid conditions
- Clinical management and pharmacotherapy

We found that many decisions corresponded to each of the tasks and that some decisions had multiple information needs. This analysis identified several key decisions needed to accomplish the 4 guideline-derived tasks. In our examination of exemplar mobile apps for asthma, we found 9 mHealth apps and 15 different PGHD types (Table 1). We matched the types of PGHD to the information needs derived from the guidelines (Table 2). However, we found that not all types of PGHD configured in the mHealth apps correspond directly with a guideline-derived information need.

Table 1. Asthma mobile health apps and patient-generated health data types.

App	Asthma action plan	ACT[a]	Journal	Activity level	Symptoms	Triggers	Medication reminders	Peak expiratory flow	Inhaler use	Medication use	Environmental factors	Survey data	Location	Mood
Asthma MD[b]	x		x		x	x	x	x						
Asthma Health App[c]				x	x	x				x		x	x	
Asthma Storylines[b]					x			x		x				x
Hailie[b]									x					
Kagen Air[b]	x				x	x	x			x	x		x	
Kiss My Asthma[b]	x				x									
My Asthma Pal[b]	x	x			x		x			x				
Smart Track[d]									x					
Propeller Health[b]									x	x	x			

[a]ACT: Asthma Control Test.
[b]Kagan and Garland [17].
[c]Genes et al [14].
[d]Chan et al [7].

Table 2. Types of asthma-relevant patient-generated health data.

Patient-generated health data	Information generated
Asthma Control Test	Symptom trajectory from the last 4 weeks
Exposures	Symptom triggers such as allergens, smoking, and perfume
Activity level	Level of physical activity
Symptoms	Type of symptoms and if daytime or nighttime
Peak-flow meter	Measurement of peak expiratory flow rates
Inhaler use	Medication adherence, last dose, missed doses
Asthma action plan	Progression toward goals and attitudes
Environmental factors	Pollen count and air quality
Concerns and/or questions	Ability to recognize worsening symptoms

Perceptions About Patient-Generated Health Data and Pediatric Asthma Management

In August 2019, three primary care providers—two physicians and a nurse practitioner—participated in part 2. Based on their input, including their suggestions on importance to asthma treatment, we modified the initial guideline-derived list; the final list of high-priority information elements is provided in Table 3. The SMEs indicated that there were additional information needs related to triggers of asthma symptoms such as an insufficient level of dustproofing, pets, inadequate pest control measures, cleaning fluids, and other allergens; all of which may not be captured by PGHD. There was a specific interest in pollen, grass, pollution, and other environmental

factors, and we added these triggers to the information needs of the decision point on determining exposure to risk factors. Although we identified decisions related to diagnosing in our initial integrated set, we excluded diagnostic decisions from our reviews with the providers based on our assumption that asthma-specific PGHD would be most useful for, and most likely collected by, children already diagnosed with asthma.

Table 3. Guideline-derived decisions and information needs with types of patient-generated health data.

Decision	Information needs	PGHD[a]
Determine level of symptom control	Symptom trajectory, types of symptoms, medication adherence, last dose, missed doses	Symptoms, ACT[b], inhaler use
Determine exposure to risk factors	Symptom triggers such as allergens, smoking, pollen, poor air quality, perfume, inadequate dustproofing, pets, inadequate pest control measures, and cleaning fluids	Exposures, symptoms, environmental factors
Determine adjustments to medication regimen	Symptom trajectory, medication adherence, last dose, missed doses	Symptoms, inhaler use
Determine adjustments to action plan	Progression toward goals, attitudes, child's and family's ability to recognize worsening symptoms, level of physical activity	Asthma action plan, activity level
Determine ability to take medication	Observation of inhaler technique, medication adherence, last dose, missed doses	Inhaler use
Determine lung function	Lung function assessment, peak flow expiratory rates	Peak flow meter
Determine educational needs	Subjective questions or concerns from child or family	Concerns and/or questions

[a]PGHD: patient-generated health data.

[b]ACT: Asthma Control Test.

A few common perspectives resulted from the input of the SMEs. Each placed primary focus on uncontrolled asthma and indicated that PGHD would be most useful for adolescents, a subset of pediatric patients, who have trouble controlling their asthma. Perspectives concerning the timing for viewing PGHD were also prevalent. The SMEs expressed a desire to see the PGHD in the EHR at the time of the visit. They thought that it would be unusual to view the PGHD before a patient visit or between visits without an alert in the EHR or communication from the patient. Last, there was an interest in observing the inhaler technique and knowing whether the patients use spacers with their inhalers. The SMEs viewed proper inhaler technique as a critical component of medication adherence and pediatric asthma self-management. They thought that although patients might perform the proper inhaler technique during clinic visits, the technique might be inadequate outside of the visits.

Discussion

Principal Findings

A complete understanding of the information needs supported by PGHD is essential for the seamless integration of PGHD into workflows [13]. Identification of provider information needs is the first step in supporting the integration of PGHD into EHRs. In this study, we identified a list of high-priority decisions, information needs, and potential PGHD sources that can address the information needs of providers treating patients with pediatric asthma. We believe our findings demonstrate the suitability of PGHD to support clinical decisions for pediatric asthma. This work serves as a foundation to support future postassessment work such as evaluating the use of PGHD from mHealth apps and the integration of PGHD to EHRs.

In addition to the main findings, we uncovered aspects of PGHD use in pediatric asthma that may inform future research questions. Providers treating patients with pediatric asthma considered the use of PGHD to determine asthma triggers to be an essential part of treatment. Similar to other studies, providers expressed a need to know about general environmental factors, including air pollution and pollen levels [29,30]. We found that providers also wanted to learn about pest control (eg, roaches or rodents) in addition to contact with pets and other animals. Exposure products such as cleaning fluids, detergents, and perfumes were also of interest. Although it may prove difficult to capture all triggers using mobile technologies or sensors, the majority of asthma mHealth apps have the potential to include local air quality [17]. Further exploration is needed to fully understand the clinical utility of the inclusion of triggers in mHealth asthma apps.

The providers commented that inhaler use was an essential gap in their knowledge of patient behaviors, and in our review of the guidelines we found inhaler use to be a clear information need. Previous research reported that many patients use their inhaler poorly or share inhalers with friends or family members [1,8]. In our limited search, we found a lack of smart-inhaler mHealth apps that can capture and transmit data on inhaler technique or the use of spacers to providers. Technologies such as Respiro and Capmedic provide technique-related feedback to the user, but it is unclear whether providers can access technique assessment data [31,32]. As technologies advance, evidence related to the features of audio and video capture of inhaler use may be beneficial.

We found that providers were most interested in PGHD collected by patients whose asthma was severely uncontrolled. According to pediatric asthma guidelines, asthma severity is classified as

mild, moderate, or severe, with severe asthma requiring the highest level of treatment [33]. Given the potential long-term repercussions, it is vital to treat children adequately in order to establish control early in life [34]. Although an emphasis on the PGHD of patients with severe asthma is reasonable, mHealth technologies identified gaps by noncompliant patients [8]. However, a study by Chan et al [7] reported that the use of mobile technologies for severe asthma might be most promising. As the number of mHealth apps increases, it may be worthwhile to collect additional evidence on the PGHD use—or lack of use—of pediatric mHealth apps for all types of asthma severity before focusing solely on uncontrolled cases.

Although particular care models determine the point in the care process when providers should review PGHD, we determined that in the context of outpatient pediatric asthma care, providers preferred to see the PGHD at the time of the visit. Other researchers have described programs that use nurse care coordinators or community health workers to review PGHD on a more ongoing basis [35,36]. Given that many patients with asthma have visits at the time of an exacerbation [1], it is reasonable that specialists in pediatric asthma see the most value for PGHD as part of their during-visit workflows [17].

In this study, providers expressed a keen interest in viewing PGHD directly from the EHR and not from another app. However, there is a risk that the potential richness of PGHD may get lost once it is added to an already complex and sometimes unsearchable EHR. Because of these comments, it would be worthwhile to investigate the requirements and provider preferences for the display of PGHD alongside EHR data and the locations in the EHR that may be most beneficial.

If the future of health care is personalization and individualized approaches to care, new strategies to harvest data from mobile technologies are needed [1]. As a first step, information technology specialists and health care providers should work together to determine clinical information needs for available PGHD and to update needs as new PGHD sources become available. There is great potential for PGHD to support the longitudinal care of patients with chronic disease. An understanding of the PGHD needs for pediatric asthma provides the opportunity to similarly explore the PGHD needs of other chronic diseases. The next step in the needs assessment framework (phase 3) indicates that actions must resolve the needs-based priorities [19]. We suggest a strategy for future research that examines the PGHD visualization and display preferences of providers to support the design of EHR integration.

Limitations

Although this work was grounded in widely accepted guidelines, there may be nuances that were not accounted for, and all providers may not agree with or use the guidelines in practice. In addition, we validated our findings with primary care providers. It may be helpful to explore information needs with providers in other settings as needed.

Conclusion

To optimally inform implementation approaches that integrate PGHD, the identification of provider information needs is essential. We extracted a set of tasks, decisions, and information needs derived from clinical guidelines and aligned them with PGHD types that may be collected by patients. By reviewing with providers caring for pediatric asthma patients in the outpatient setting, we validated the information needs and found that they align with some types of PGHD currently collected. This preliminary work serves to support the future design and development of mHealth apps and methods to integrate PGHD into EHRs that are in alignment with clinical information needs for chronic disease management.

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Conflicts of Interest

None declared.

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Abbreviations

ACT: Asthma Control Test
EHR: electronic health record
GINA: Global Initiative for Asthma
mHealth: mobile health
NHLBI: National Heart, Lung, and Blood Institute
PGHD: patient-generated health data
SME: subject-matter expert

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