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Review

A Global Overview of COVID-19 Research in the Pediatric Field: Bibliometric Review

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Abstract

Background: Since the beginning of the COVID-19 pandemic, a great number of papers have been published in the pediatric field.

Objective: We aimed to assess research around the globe on COVID-19 in the pediatric field by bibliometric analysis, identifying publication trends and topic dissemination and showing the relevance of publishing authors, institutions, and countries.

Methods: The Scopus database was comprehensively searched for all indexed documents published between January 1, 2020, and June 11, 2020, dealing with COVID-19 in the pediatric population (0-18 years). A machine learning bibliometric methodology was applied to evaluate the total number of papers and citations, journal and publication types, the top productive institutions and countries and their scientific collaboration, and core keywords.

Results: A total of 2301 papers were retrieved, with an average of 4.8 citations per article. Of this, 1078 (46.9%) were research articles, 436 (18.9%) were reviews, 363 (15.8%) were letters, 186 (8.1%) were editorials, 7 (0.3%) were conference papers, and 231 (10%) were categorized as others. The studies were published in 969 different journals, headed by *The Lancet*. The retrieved papers were published by a total of 12,657 authors from 114 countries. The most productive countries were the United States, China, and Italy. The four main clusters of keywords were pathogenesis and clinical characteristics (keyword occurrences: n=2240), public health issues (n=352), mental health (n=82), and therapeutic aspects (n=70).

Conclusions: In the pediatric field, a large number of articles were published within a limited period on COVID-19, testifying to the rush to spread new findings on the topic in a timely manner. The leading authors, countries, and institutions evidently belonged to the most impacted geographical areas. A focus on the pediatric population was often included in general articles, and pediatric research about COVID-19 mainly focused on the clinical features, public health issues, and psychological impact of the disease.

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KEYWORDS

COVID-19; SARS-CoV-2; children; pediatrics; bibliometric review; publications; research; literature; review

Introduction

With its massive and rapid spread, the COVID-19 pandemic has been an unprecedented challenge for health care systems worldwide [1]. At the same rate, the research community has been extremely prolific, with a considerable amount of scientific papers published within a very short time, in an effort to address all aspects of disease management in a timely manner. Many

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journals are offering fast-track publication for COVID-19–related papers. The availability of reviewers for an expedited review process has been called for. In other words, the editorial process that usually characterizes the research on a disease has been sped up and amplified in this exceptional circumstance.

Infection by SARS-CoV-2 seems to be milder in the pediatric population than in the elderly [2], and the relative protection of children against the severe forms of COVID-19 is a major point of interest in the comprehension of the pathogenetic mechanisms of the disease [3]. Therefore, research on COVID-19 in the pediatric field has experienced a strong increase. The very high number of continuously published papers and the speed at which research on this topic has been generated makes it extremely difficult to stay updated with such a rapidly evolving knowledge base. In this regard, a bibliometric analysis of the current literature on pediatric COVID-19 can help clinicians stay updated with emerging and swiftly evolving scientific outcomes. Bibliometric analysis is the attempt to quantitatively assess the current literature on a certain topic, allowing researchers to acquire knowledge about research trends and providing insights into the contribution of a particular country or institution to that topic, as well as data on coauthorship and collaboration [4].

Therefore, we performed a bibliometric analysis of the papers on COVID-19 in the pediatric field published in the first half of 2020 to assess research on this topic on a global scale, identify publication trends, and provide some hints on the gap of knowledge to be filled by future research.

Methods

The Scopus database was comprehensively searched for all the indexed documents published between January 1, 2020, and June 11, 2020, dealing with COVID-19 in the pediatric population (0-18 years of age). The keywords used were *COVID* or *coronavirus* and *pediatric* or *child* or *children* or *adolescent*$ in the title or abstract. We used only these terms to conduct a broad search that would ensure the inclusion of relevant literature. The inclusion criteria were papers that (1) were published after the first report of COVID-19 from the Wuhan government on December 31, 2019, and (2) matched the search keywords. As COVID-19 was first reported in China and a fairly large number of research papers were written in Chinese, language was not limited during the retrieval process.

The literature retrieval group consisted of 3 trained professionals. We used the export feature of the search engine to retrieve data for further processing. All extracted literature entries were exported into Microsoft Excel (Microsoft Corp) for screening and selection. The reviewers (AM, FT, GG, SB) independently screened the titles, abstracts, and, if ambiguous, full texts for the inclusion of articles. Discrepancies were resolved through discussions among them and with a fifth reviewer (IR) in case of difficulties in reaching an agreement. The reviewers independently conducted information extraction from the included papers. Discrepancies were similarly resolved through discussion among the reviewers.

For studies that fulfilled the inclusion criteria, the following information was extracted: authors, affiliations, and country of origin (when there was more than one author, the corresponding author's information was used), journal, publication date, publication type, citation, and abstract. The abstract and title of every record retrieved were screened to determine which studies should be assessed further.

We performed explorative data analysis for:

- Total number of papers to measure global productivity;
- Total citations to assess the relevance of an author, institution, or country;
- Journal and publication types to assess topic dissemination;
- Scientific collaboration between authors, institutions, and countries to show how they related to others;
- Core keywords to show clusters of research topics.

The intracountry and intercountry collaboration indices were presented as single-country publication (SCP) and multiple-country publication (MCP), according to the country of the corresponding author, and the timing of the first COVID-19 case reported for each country was highlighted as well.

A machine learning bibliometric methodology was applied to evaluate the distribution of each factor. The *bibliometrix* R package with its Biblioshiny web interface was used [5].

No ethics approvals were considered necessary as this was a literature-based study.

Results

General Data

A total of 2301 papers were retrieved. Out of them, 1078 (46.9%) were research articles, 436 (18.9%) were reviews, 363 (15.8%) were letters, 186 (8.1%) were editorials, 7 (0.3%) were conference papers, and 231 (10%) were categorized as others.

Citation Analysis

The retrieved articles had 11,063 citations with an average of 4.8 citations per article. Of the retrieved papers, 869 (37.8%) were cited at least once. The 10 most frequently cited articles are shown in Table 1. The top-ranking paper (n=1255 citations) was published in *The New England Journal of Medicine* and was focused on the clinical characteristics of COVID-19 in China.

Table 2 shows the distribution of the prevalence of the retrieved articles according to the number of citations.

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Table 1. List of the 10 most cited articles about COVID-19 in the pediatric population.

Authors and reference	Title	Journal	Publication date	Total citations, n (%)
Guan et al [6]	Clinical characteristics of coronavirus disease 2019 in China	*The New England Journal of Medicine*	April 30, 2020	1255 (11.3)
Chan et al [7]	A familial cluster of pneumonia associated with the 2019 novel coronavirus indicating person-to-person transmission: A study of a family cluster	*Lancet*	February 15, 2020	735 (6.6)
Mehta et al [8]	COVID-19: Consider cytokine storm syndromes and immunosuppression	*Lancet*	March 13, 2020	324 (2.9)
Wu et al [9]	A new coronavirus associated with human respiratory disease in China	*Nature*	February 3, 2020	309 (2.8)
Wu et al [10]	Risk factors associated with acute respiratory distress syndrome and death in patients with coronavirus disease 2019 pneumonia in Wuhan, China	*JAMA Internal Medicine*	March 13, 2020	291 (2.6)
Chen et al [11]	Clinical characteristics and intrauterine vertical transmission potential of COVID-19 infection in nine pregnant women: A retrospective review of medical records	*Lancet*	February 12, 2020	275 (2.5)
Lai et al [12]	Severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) and coronavirus disease-2019 (COVID-19): The epidemic and the challenges	*International Journal of Antimicrobial Agents*	February 12, 2020	213 (1.9)
Xu et al [13]	Evolution of the novel coronavirus from the ongoing Wuhan outbreak and modeling of its spike protein for risk of human transmission	*Science China Life Sciences*	January 21, 2020	153 (1.4)
Dong et al [14]	Epidemiology of COVID-19 among children in China	*Pediatrics*	June 1, 2020	134 (1.2)
Lu et al [15]	SARS-CoV-2 infection in children	*The New England Journal of Medicine*	April 23, 2020	128 (1.2)

Table 2. Distribution of published papers about COVID-19 in the pediatric population according to the number of citations.

Number of citations	Articles, n (%)
>100	17 (0.74)
50-100	25 (1.09)
20-49	70 (3.04)
5-19	202 (8.78)
<5	1989 (86.44)

Journal Analysis

The studies were published in 969 different journals. *The Lancet* headed the list with a total number of 29 publications, followed by the *Journal of Medical Virology* (n=26) and *Science of the Total Environment* (n=25). In analyzing the publications' reference lists, the most cited sources were *The Lancet* (n=2336 citations), followed by *The New England Journal of Medicine* (n=1835), and the *Journal of American Medical Association* (n=1017).

Author Analysis

A total of 12,657 authors contributed to these papers, with a mean of 5.5 authors per document. Out of this total, 239 were authors of single-authored documents and 12,418 were authors of multiauthored documents.

Country Analysis

The retrieved papers were published by authors from 114 countries. The most productive country in the COVID-19 research field in pediatrics was the United States, with 178 publications, followed by China, with 138 publications, and Italy, with 87 publications. Table 3 shows the top 10 productive countries.

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Table 3. The top 10 productive countries publishing on the topic of COVID-19 in the pediatric population.

Country	Articles, n	SCP[a], n	MCP[b], n	First reported case[c]
United States	178	134	44	January 30, 2020
China	138	119	19	December 31, 2019
Italy	87	70	17	February 20, 2020
India	30	23	7	March 2, 2020
France	26	21	5	February 7, 2020
United Kingdom	23	10	13	January 28, 2020
Canada	22	10	12	January 27, 2020
Iran	19	15	4	February 19, 2020
Korea	18	14	4	January 20, 2020
Australia	17	11	6	January 25, 2020

[a]SCP: single-country publication.

[b]MCP: multiple-country publication.

[c]Source: WHO Coronavirus (COVID-19) Dashboard [16].

Institution Analysis

The published articles came from 4919 institutions, with the top 10 institutions accounting for 614 (26.7%) papers. The Huazhong University of Science and Technology was the most productive institution, with 117 documents, followed by Harvard Medical School (n=82 publications) and the University of Oxford (n=67). Table 4 displays the top 10 productive institutions.

Table 4. The top 10 productive institutions publishing on the topic of COVID-19 in the pediatric population.

Institution	Country	Articles, n (%)
Huazhong University of Science and Technology	China	117 (5.1)
Harvard Medical School	United States	82 (3.6)
University of Oxford	United Kingdom	67 (2.9)
University of Washington	United States	63 (2.7)
University of California	United States	61 (2.6)
University of Toronto	Canada	56 (2.4)
Tehran University of Medical Sciences	Iran	47 (2.0)
University College London	United Kingdom	45 (2.0)
Universal Scientific Education and Research Network	_[a]	39 (1.7)
University of Melbourne	Australia	37 (1.6)

[a]Not applicable.

Collaboration Analysis

Figure 1 shows the country collaboration analysis. The network diagram showed that China and the United States were the leaders in COVID-19 research in cooperation with other countries (each node represents a country, node size corresponds to publication number, connecting lines represents country cooperation, and line thickness indicates collaboration frequencies). Overall, developed countries had greater collaboration networks than developing territories.

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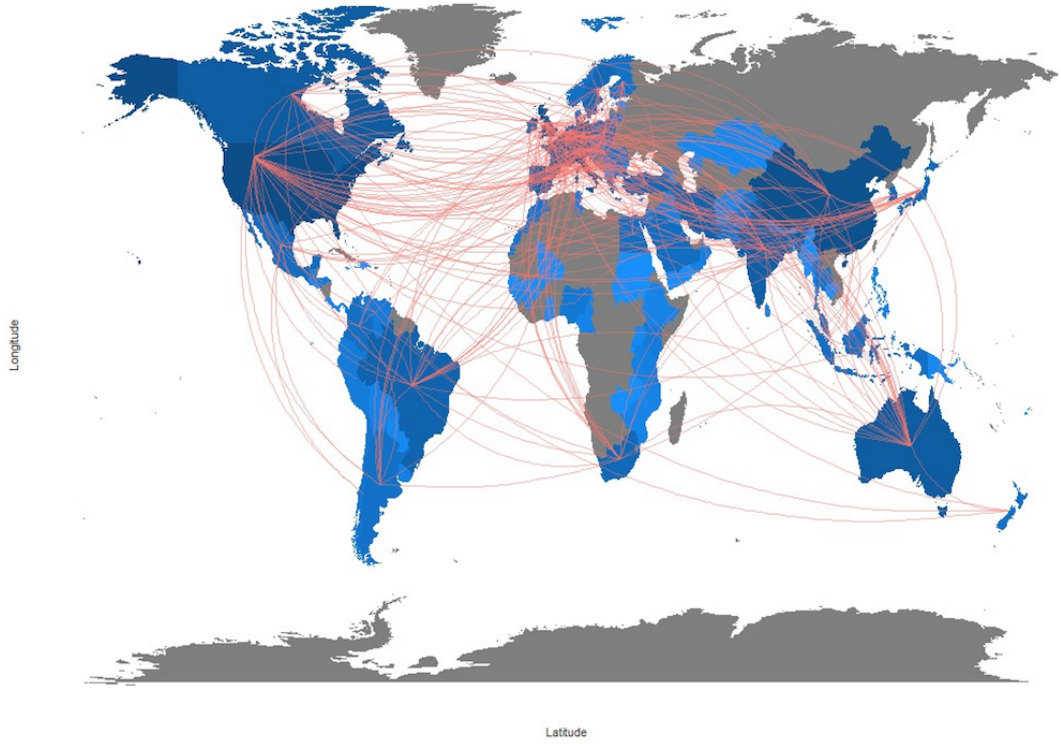
Figure 1. Collaboration map between countries in publications on COVID-19 in the pediatric population.



Keyword Analysis

Overall, the three most common keywords were *COVID-19* with 880 occurrences, *coronavirus* with 368 occurrences and *Sars-Cov2* with 294 occurrences. Multimedia Appendix 1 shows a word cloud for the most common keywords of the retrieved papers

When keywords were clustered into themes, four main clusters emerged: pathogenesis and clinical characteristics (keyword occurrence: n=2240), public health issues (n=352), mental health (n=82), and therapeutic aspects (n=70).

Discussion

Principal Findings

In this bibliometric review, we aimed to provide a comprehensive portrait of published research on COVID-19 in the pediatric population. The bibliometric approach properly fits the aim of representing all scientific publications on a certain topic in a defined time frame. Therefore, this method has been recently used by many authors to depict the state of current knowledge about COVID-19 under different perspectives [17-27], and we used it to focus on the pediatric population.

The most impressive result was the large number of articles published within a limited period on a single topic. Most were research articles, but, notably, more than 15% of the total published papers were letters, testifying to the rush to quickly spread even preliminary results and personal perspectives [28]. The low rate of conference papers was probably due to the fact that most of the national and international conferences scheduled in that period were cancelled in compliance with social distancing measures.

The citation analysis showed that only about one-third of the published papers were cited at least once, suggesting that the short time frame and the rapidly increasing amount of literature likely lowered the possibility of a paper being cited. However, the top-cited paper reached a high number of citations in a very short time.

The leading authors, countries, and institutions evidently belonged to the most impacted geographical areas, as already pointed out in previous general bibliometric analyses.

The keyword analysis, as well as the top-cited articles, revealed that a focus on the pediatric population was often included in general articles about COVID-19 as well. Analyzing the keyword clusters, it was observed that pediatric research about COVID-19 mainly focused on the clinical features of the disease and public health issues. Notably, mental health–related topics were of outstanding interest, even overcoming therapeutic aspects. Psychosocial challenges and opportunities for children with chronic health conditions during lockdown, such as digital approaches to remote pediatric health care delivery, represented a mainstream topic in the publication trends during the pandemic [29,30]. This, along with the lack of emergency preparedness–related topics among the most recurrent keywords, could suggest that COVID-19 research in pediatrics more frequently addressed the psychological impact of lockdown, per the milder clinical course of the disease among children, and unlike what emerged in a similar bibliometric analysis outside the pediatric area [17].

Taken together, our findings demonstrate that scientists around the world, facing the unprecedented emergency of the COVID-19 pandemic, felt compelled to publish their findings and opinions with the aim to contribute to the evolving knowledge base as soon as new evidence emerged. This

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occurred even in the pediatric field, which has up to now not been so seriously involved in the COVID-19 emergency.

This process was facilitated by high-impact medical journals opening special issues or columns for COVID-19 [31], some publishers waiving publication fees and providing free access to article content, and many journals opting to fast track peer review. It would be desirable that some of these positive aspects implemented in the current difficult times would become common practice. Nonetheless, in such an accelerated publishing process, it should be of great importance to keep the quality of research papers high, which is essential to spread valuable information [22,32]. In the future, it would be interesting to compare this first 6-month publication rush to later publication attitudes about the COVID-19 topic in the pediatric field.

Limitations

Our study has some limitations that need to be acknowledged, pertaining mainly to the intrinsic bias of having considered only studies indexed in Scopus within a limited time frame, knowing that results may differ had other databases been included or the time period extended. However, this bibliometric analysis of COVID-19 publications in the pediatric field offers a global overview of what has been published on the topic, which allows for the identification of possible gaps in knowledge for new lines of research. In particular, the Scopus database was chosen for its reliability, wide coverage of scientific production, and daily updates. We decided to search only one database mainly to avoid duplicate results. Moreover, as this bibliometric analysis addressed a very recent and rapidly evolving topic, we preferred to search only one reliable database to overcome gaps in the promptness of updates from different sources. Finally, we preferred Scopus as our main data source since it provides data analysis for publications and citations, and allows the results to be sorted according to the number of citations.

Another limitation consists of the limited time frame of our analysis. Using a different methodological approach, such as a living systematic review, would allow for a real-time update on the rising evidence in such a rapidly evolving field. As a result, it would be possible for authors to constantly monitor gaps in knowledge, which they could try to fill with future research.

Conclusion

In conclusion, a substantial number of papers have been published on the topic of COVID-19 in the pediatric field. It would be advisable to carry on implementing the positive changes to publication policies that emerged during the COVID-19 pandemic, which will aid in providing solid evidence to inform and support clinical and public health decision making.

Conflicts of Interest

None declared.

Multimedia Appendix 1
Word cloud of the most frequently used keywords in papers about COVID-19 in the pediatric population.
[PNG File , 926 KB - pediatrics_v4i3e24791_app1.png]

References

1. WHO Health Emergency Dashboard. URL: https://extranet.who.int/publicemergency [accessed 2020-09-15]
2. De Luca CD, Esposito E, Cristiani L, Mancino E, Nenna R, Cortis E, et al. Covid-19 in children: A brief overview after three months experience. Paediatr Respir Rev 2020 Sep;35:9-14 [FREE Full text] [doi: 10.1016/j.prrv.2020.05.006] [Medline: 32593648]
3. Cristiani L, Mancino E, Matera L, Nenna R, Pierangeli A, Scagnolari C, et al. Will children reveal their secret? The coronavirus dilemma. Eur Respir J 2020 Apr 02;55(4):2000749 [FREE Full text] [doi: 10.1183/13993003.00749-2020] [Medline: 32241833]
4. Ellegaard O, Wallin J. The bibliometric analysis of scholarly production: How great is the impact? Scientometrics 2015;105(3):1809-1831 [FREE Full text] [doi: 10.1007/s11192-015-1645-z] [Medline: 26594073]
5. Aria M, Cuccurullo C. bibliometrix : An R-tool for comprehensive science mapping analysis. Journal of Informetrics 2017 Nov;11(4):959-975. [doi: 10.1016/j.joi.2017.08.007]
6. Guan WJ, Ni ZY, Hu Y, Liang WH, Ou CQ, He JX, China Medical Treatment Expert Group for Covid-19. Clinical Characteristics of Coronavirus Disease 2019 in China. N Engl J Med 2020 Apr 30;382(18):1708-1720 [FREE Full text] [doi: 10.1056/NEJMoa2002032] [Medline: 32109013]
7. Chan J, Yuan S, Kok K, To K, Chu H, Yang J, et al. A familial cluster of pneumonia associated with the 2019 novel coronavirus indicating person-to-person transmission: a study of a family cluster. The Lancet 2020 Feb;395(10223):514-523. [doi: 10.1016/S0140-6736(20)30154-9]
8. Mehta P, McAuley D, Brown M, Sanchez E, Tattersall R, Manson J. COVID-19: consider cytokine storm syndromes and immunosuppression. The Lancet 2020 Mar 03;395(10229):1033-1034. [doi: 10.1016/S0140-6736(20)30628-0]
9. Wu C, Zhao S, Yu B, Chen Y, Wang W, Song Z, et al. A new coronavirus associated with human respiratory disease in China. Nature 2020 Feb 03;579(7798):265-269 [FREE Full text] [doi: 10.1038/s41586-020-2008-3] [Medline: 32167524]

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10. Wu C, Chen X, Cai Y, Xia J, Zhou X, Xu S, et al. Risk Factors Associated With Acute Respiratory Distress Syndrome and Death in Patients With Coronavirus Disease 2019 Pneumonia in Wuhan, China. JAMA Intern Med 2020 Jul 01;180(7):934-943 [FREE Full text] [doi: 10.1001/jamainternmed.2020.0994] [Medline: 32167524]
11. Chen H, Guo J, Wang C, Luo F, Yu X, Zhang W, et al. Clinical characteristics and intrauterine vertical transmission potential of COVID-19 infection in nine pregnant women: a retrospective review of medical records. The Lancet 2020 Mar;395(10226):809-815 [FREE Full text] [doi: 10.1016/S0140-6736(20)30360-3] [Medline: 32081636]
12. Lai CC, Shih TP, Ko WC, Tang HJ, Hsueh PR. Severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) and coronavirus disease-2019 (COVID-19): The epidemic and the challenges. Int J Antimicrob Agents 2020 Mar 21;55(3):105924-105460 [FREE Full text] [doi: 10.1016/j.ijantimicag.2020.105924] [Medline: 32081636]
13. Xu X, Chen P, Wang J, Feng J, Zhou H, Li X, et al. Evolution of the novel coronavirus from the ongoing Wuhan outbreak and modeling of its spike protein for risk of human transmission. Sci China Life Sci 2020 Mar 16;63(3):457-460 [FREE Full text] [doi: 10.1007/s11427-020-1637-5] [Medline: 32009228]
14. Dong Y, Mo X, Hu Y, Qi X, Jiang F, Jiang Z, et al. Epidemiology of COVID-19 Among Children in China. Pediatrics 2020 Jun 23;145(6):1663-1665 [FREE Full text] [doi: 10.1542/peds.2020-0702] [Medline: 32179660]
15. Lu X, Zhang L, Du H, Zhang J, Li YY, Qu J, Chinese Pediatric Novel Coronavirus Study Team. SARS-CoV-2 Infection in Children. N Engl J Med 2020 Apr 23;382(17):1663-1665 [FREE Full text] [doi: 10.1056/NEJMc2005073] [Medline: 32187458]
16. WHO Coronavirus (COVID-19) Dashboard. URL: https://covid19.who.int/ [accessed 2021-07-12]
17. Yu Y, Li Y, Zhang Z, Gu Z, Zhong H, Zha Q, et al. A bibliometric analysis using VOSviewer of publications on COVID-19. Ann Transl Med 2020 Jul;8(13):816-816 [FREE Full text] [doi: 10.21037/atm-20-4235] [Medline: 32793661]
18. Şenel E, Topal F. Holistic Analysis of Coronavirus Literature: A Scientometric Study of the Global Publications Relevant to SARS-CoV-2 (COVID-19), MERS-CoV (MERS) and SARS-CoV (SARS). Disaster Med Public Health Prep 2020 Aug 13:1-8 [FREE Full text] [doi: 10.1017/dmp.2020.300] [Medline: 32787980]
19. Odone A, Salvati S, Bellini L, Bucci D, Capraro M, Gaetti G, et al. The runaway science: a bibliometric analysis of the COVID-19 scientific literature. Acta Biomed 2020 Jul 20;91(9-S):34-39 [FREE Full text] [doi: 10.23750/abm.v91i9-S.10121] [Medline: 32701915]
20. Patil S. Indian Publications on SARS-CoV-2: A bibliometric study of WHO COVID-19 database. Diabetes Metab Syndr 2020;14(5):1171-1178 [FREE Full text] [doi: 10.1016/j.dsx.2020.07.007] [Medline: 32673837]
21. Liu N, Chee M, Niu C, Pek P, Siddiqui F, Ansah J, et al. Coronavirus disease 2019 (COVID-19): an evidence map of medical literature. BMC Med Res Methodol 2020 Jul 02;20(1):177 [FREE Full text] [doi: 10.1186/s12874-020-01059-y] [Medline: 32615936]
22. Nowakowska J, Sobocińska J, Lewicki M, Lemańska Ż, Rzymski P. When science goes viral: The research response during three months of the COVID-19 outbreak. Biomed Pharmacother 2020 Sep;129:110451 [FREE Full text] [doi: 10.1016/j.biopha.2020.110451] [Medline: 32603887]
23. De Felice F, Polimeni A. Coronavirus Disease (COVID-19): A Machine Learning Bibliometric Analysis. In Vivo 2020 Jun;34(3 Suppl):1613-1617. [doi: 10.21873/invivo.11951] [Medline: 32503819]
24. Mao X, Guo L, Fu P, Xiang C. The status and trends of coronavirus research: A global bibliometric and visualized analysis. Medicine (Baltimore) 2020 May 29;99(22):e20137 [FREE Full text] [Medline: 32481379]
25. Chahrour M, Assi S, Bejjani M, Nasrallah A, Salhab H, Fares M, et al. A Bibliometric Analysis of COVID-19 Research Activity: A Call for Increased Output. Cureus 2020 Mar 21;12(3):e7357 [FREE Full text] [doi: 10.7759/cureus.7357] [Medline: 32328369]
26. Lou J, Tian S, Niu S, Kang X, Lian H, Zhang L, et al. Coronavirus disease 2019: a bibliometric analysis and review. Eur Rev Med Pharmacol Sci 2020 Mar;24(6):3411-3421 [FREE Full text] [doi: 10.26355/eurrev_202003_20712] [Medline: 32271460]
27. Warin T. Global Research on Coronaviruses: An R Package. J Med Internet Res 2020 Aug 11;22(8):e19615 [FREE Full text] [doi: 10.2196/19615] [Medline: 32730218]
28. Fidahic M, Nujic D, Runjic R, Civljak M, Markotic F, Lovric Makaric Z, et al. Research methodology and characteristics of journal articles with original data, preprint articles and registered clinical trial protocols about COVID-19. BMC Med Res Methodol 2020 Jun 22;20(1):161 [FREE Full text] [doi: 10.1186/s12874-020-01047-2] [Medline: 32571302]
29. Serlachius A, Badawy SM, Thabrew H. Psychosocial Challenges and Opportunities for Youth With Chronic Health Conditions During the COVID-19 Pandemic. JMIR Pediatr Parent 2020 Oct 12;3(2):e23057 [FREE Full text] [doi: 10.2196/23057] [Medline: 33001834]
30. Badawy SM, Radovic A. Digital Approaches to Remote Pediatric Health Care Delivery During the COVID-19 Pandemic: Existing Evidence and a Call for Further Research. JMIR Pediatr Parent 2020 Jun 25;3(1):e20049 [FREE Full text] [doi: 10.2196/20049] [Medline: 32540841]
31. Brown A, Horton R. A planetary health perspective on COVID-19: a call for papers. The Lancet 2020 Apr;395(10230):1099. [doi: 10.1016/s0140-6736(20)30742-x]
32. Ioannidis J. Coronavirus disease 2019: The harms of exaggerated information and non-evidence-based measures. Eur J Clin Invest 2020 Apr;50(4):e13222 [FREE Full text] [doi: 10.1111/eci.13222] [Medline: 32191341]

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Abbreviations

MCP: multiple-country publication
SCP: single-country publication

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Original Paper

Experiences of Children With Osteogenesis Imperfecta in the Co-design of the Interactive Assessment and Communication Tool Sisom OI: Secondary Analysis of Qualitative Design Sessions

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Abstract

Background: Children with osteogenesis imperfecta (OI) experience a diversity of symptoms that expose them to difficult physical, mental, and social challenges. Sisom (DHealth) is an interactive assessment and communication tool designed to help children aged 6-12 years with chronic conditions express their symptoms. Recently, the co-design of the *Sisom OI* paper prototype was launched by seeking the perspectives of end users, including children with OI and their clinicians.

Objective: The aim of this study is to describe the experiences that children with OI were prompted to share with researchers during the co-design of the *Sisom OI* paper prototype.

Methods: A secondary analysis of qualitative data was conducted at a university-affiliated, pediatric, orthopedic hospital. The data sources consisted of interview transcripts, drawings, field notes, and observations derived from interviewing 12 children with OI who participated in the co-design of the *Sisom OI* paper prototype. The themes and subthemes identified from the data sources were generated using qualitative description.

Results: Three themes were identified. The first, *Relating to Others*, described the balance between feeling different versus feeling similar to other children. The subthemes were *Common OI Experience*, *Feeling Different*, and *Feeling Just Like Others*. The second, *Relating to Their Condition*, described children's positive and negative interactions with their own condition and health care. The subthemes were *Understanding Their Condition*, *Special Relationship with the Hospital*, and *Difficult Treatments and Procedures*. The third, *Reflecting on Capabilities*, described children's recognition of their strengths and limitations. The subthemes were *Perceiving Limitations*, *Overcoming Isolation*, and *Celebrating Strengths*.

Conclusions: This co-design process provided children with OI the space to not only contribute to the development of the end product but also eloquently describe their experiences. These findings, based on the descriptions given by the children themselves, offer us a unique understanding of what it means to grow up with OI.

(JMIR Pediatr Parent 2021;4(3):e22784) doi:10.2196/22784

KEYWORDS

child health; symptom assessment; communication; mobile applications; software

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Introduction

Background

Osteogenesis imperfecta (OI), otherwise known as brittle bones disease, is the most common inherited chronic bone disorder [1-4]. The prevalence of OI has been estimated at 1 in 13,500 and 1 in 9700 in 2 recent population-based studies from Scandinavia [5,6]. Its principal clinical feature is bone fragility, for which there is no cure. Thus, health services focus on the prevention and treatment of fractures to maximize mobility [7]. However, pain, fatigue, and varying degrees of physical limitations may hinder participation in daily activities and acceptance by peers and lead to feelings of fear, otherness, and isolation [4,8-13]. Relatively little attention has been directed toward understanding the day-to-day experiences of children with OI specifically and from their own perspective.

Children living with other chronic conditions have a unique understanding of their experience that cannot be conveyed through a proxy [14,15]. A growing body of knowledge demonstrates a gap between what children view as significant and what their parents perceive [15,16]. Children with chronic conditions are described as having an "intense embodied understanding of [their] disease" [17] and being "historians in their own right" [18]. As clinicians, we have a moral duty to elicit these children's voices and partner with them to truly understand their perspective. This notion has been reinforced in the United Nations' (1989) *Convention on the Rights of the Child*, which states that every child has the right to express themselves and be heard by others in matters that affect their well-being [19].

The concept of authentic listening is derived from the moral duty to treat children as competent moral agents [20]. It implies their active involvement in decision-making to help them foster a strong moral order and capacity in future decision-making. The concept of authentic listening goes beyond simply allowing a child to express himself/herself: it is about considering a child's viewpoint as unique and valuable rather than immature and underdeveloped. Research that seeks to elicit the voices of children and include them as experts in decision-making is thus of great importance.

One strategy to elicit a child's voice is to transform a child's *place* (a place for children that is run by adults and their rules) into a child's *space* (a place where children can take the lead) [21]. Efforts to create this *space* were made during the participatory co-design of a paper prototype called *Sisom OI* [13]. This was the first step our research team embarked on to address the absence of interactive computerized tools designed to assess the unique needs of children living with OI. Sisom (DHealth; Norwegian acronym derived from *Si det som det er*, meaning *Tell it as it is*) is an award-winning, rigorously tested, interactive, computerized tool that helps children aged 6-12 years with chronic conditions to express their symptoms [22]. It can be downloaded on a phone, tablet, laptop, or desktop computer. Sisom, originally designed for children with cancer, was adapted for children with congenital heart disease, and in the process of being adapted for other conditions, it also has the potential to engage children with other chronic conditions in their own care [23]. It is also considered a creative system that helps clinicians better understand children's perspectives [22]. Sisom uses spoken text, sound, and animations to depict symptoms that are each represented by an animated scene within 1 of the 5 symptom islands (Figure 1).

Figure 1. The child-created avatar has the option to travel to 1 of the 5 islands depicted in Sisom with the help of the navigator, Mary.

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After first creating an *Avatar*, one is then prompted by the Sisom Navigator *Mary*, the nurse, to indicate the presence and severity of symptoms displayed by using a 5-point Likert-type scale. Upon completion, Sisom generates a child-friendly symptom report that can be shared with the family and clinicians [22,24].

To date, the development, redesign, validation, and evaluation of Sisom demonstrate the ability of this tool to create a unique environment to elicit children's perceptions of their condition and engage them in their health care [22,23,25-28].

Objective

The aim of this study is to describe the experiences shared by children with OI when approached as experts in the co-design of *Sisom OI*, to further advance the prototype development, and to understand the children's experiences.

Methods

Design and Setting

Following the approval of the institutional review board (A06-B29-17B), a secondary analysis of qualitative data was conducted [29]. The data sources consisted of interview transcripts, drawings, field notes, and observations derived from interviewing 12 children with OI who participated in the study aimed to develop the *Sisom OI* paper prototype [13]. Both studies were conducted at a university-affiliated, nonprofit, pediatric, bilingual, orthopedic hospital in Montreal, Québec, specialized in OI care. On the basis of the average prevalence rate of 2 Scandinavian studies [5,6] of 1 in 11,600, we estimated that there were 3190 individuals with OI in Canada, of which the study site has seen 576 children or 18.06% (576/3190) of the population.

Participants

Purposive sampling was used to allow for maximum variation in age, self-identified gender, and type of OI for children. Children were eligible for the study if they (1) were diagnosed with any type of OI, (2) were aged between 6 and 12 years, (3) received health care at the study site, (4) had a consenting parent or legal guardian, and (5) spoke in either English or French. Sisom was designed with and for children aged 6-12 years to meet their varying cognitive and emotional capabilities [22]. Therefore, the age eligibility criterion was aligned with previous Sisom studies and was limited to this age group. Older children were excluded, as they think and express themselves differently than younger children [22,30]. Moreover, children aged ≥12 years, with enhanced readability, can use other tools [30,31].

The original study sample size estimate of 10-15 children and 5-10 clinicians was proposed based on leading experts [13,32,33]. These sample estimates were also aligned with previous Sisom studies with similar designs to successfully develop the prototype [22,23], translate [25], and establish the usability of the tool [27]. Although other Sisom studies have included larger samples, these studies were quantitative (N=100) [26] or a secondary analysis of 2 studies [25,34], which explicitly increased the sample size to target the multiple disease groups inherent in oncology from multiple sites (n=39) [35]. In this study, a secondary analysis of 12 interview transcripts was deemed sufficient for investigation, as the interviews were rich in children's verbal expressions of experiences. Finally, data saturation was reached, and a sense of closure was attained because the data derived from our sources yielded redundant information [36].

Recruitment

The children with OI were recruited by reaching out to clinicians who assisted by identifying, screening, and approaching families to determine if they were interested in hearing more about the study [13]. One member of the study team was charged with providing a verbal and written explanation of the study to those interested in obtaining written parental informed consent and child assent [13].

Data Collection and Procedures

Data were collected in the context of the participatory co-design of the *Sisom OI* paper prototype [13]. The early stages of the co-design of the *Sisom OI* paper prototype consisted of 3 feedback cycles with 2 to 6 semistructured, face-to-face, individual child interviews per cycle [13]. The length of the interview depended on the child's interest and varied from 20 to 60 minutes. It was the parent's or legal guardian's choice to be present during the interview. During the co-design of *Sisom OI*, the children were prompted in several ways. First, Sisom prompted the children both verbally, through the questions Mary, the nurse, was asking, and visually, through the different animations that appeared. Mary's questions prompted the children to share their experiences related to each symptom being assessed. Second, the interviewer verbally prompted the children to share and elaborate on their experiences when prompted. The interviewer would either directly ask the child a question or reflect something that the child shared in common with other participants to prompt a response. These prompts helped children to draw from their experiences to determine whether the Sisom symptoms, vignettes, or avatars were relevant or irrelevant, needed to be modified, required new additions, or remained unsure [13]. Third, if present, the parent or legal guardian also prompted their children to share their experiences (Multimedia Appendix 1). Field notes were recorded during and immediately after each audio-recorded interview, which included detailed descriptions of nonverbal data, other observations, impressions, and any drawings generated.

Data Analysis

The interviews were transcribed and read through, along with the field notes and observations taken by the interviewer and the 6 drawings created by the children [13], to gain a global picture of the data. A descriptive qualitative analysis was then applied to identify themes and patterns identified in the data sources [37]. First, open coding was performed diligently to remain close to the original data. Second, code reduction or clustering was performed to create categories and remove redundant codes [37]. Third, axial coding was performed to find the links between the main categories to generate overarching themes and subthemes. Finally, the original data were revisited to ensure that the child's experience was properly interpreted and described [37]. Through this process, themes and subthemes across the children's responses were identified. For each theme

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and subtheme generated, various examples were used to highlight particular points within the children's dialogue. Ongoing meetings with the research team were held to discuss how to answer the research question, explore how the data could be best summarized, and check whether designated quotations fit the proposed themes and subthemes. The selected French quotes were subsequently translated into English. Throughout the entire process, an audit trail, composed primarily of methodological and analytical documentation, was kept, permitting the reproducibility and transferability of the process.

Results

Sample Characteristics

A total of 12 children participated in this study (Table 1). The participation rate was 92% (12/13). No child withdrew from the study.

Table 1. Sample characteristics (n=12).

Characteristics	Values
Age (years), mean (SD; range)	9 (2; 6-12)
Self-identified gender, n (%)	
Boy	7 (58)
Girl	5 (42)
Nationality, n (%)	
Provincial (Québec)	5 (42)
National (Canada)	3 (25)
International	4 (33)
Languages spoken at home, n (%)	
English	4 (33)
French	4 (33)
Bilingual (English and French)	1 (8)
Bilingual (English and other)	3 (24)
Type of osteogenesis imperfecta, n (%)	
I	4 (33)
III	2 (17)
IV	5 (42)
VI	1 (8)
Current fracture[a], n (%)	
Yes	5 (42)
No	7 (58)
Use of mobility devices, n (%)	
Wheelchair	2 (17)
Wheelchair and walker	4 (33)
None	6 (50)
Reason for presence at the study site, n (%)	
Physiotherapy and occupational therapy	6 (50)
Regular checkup	1 (8)
Admission	4 (34)
Other	1 (8)

[a]*Current fracture* referred to whether the child was immobilized and recovering from a fracture at the time of interview.

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Child Interview Findings

Overview

In the co-design of *Sisom OI*, different prompts allowed children to open up about their experiences of life with OI. These prompts were from Sisom, the interviewer, and, if present, the parent or legal guardian. The dynamic between the child and their parent or legal guardian varied. Sometimes, the parent or legal guardian answered directly for the child. In these cases, the child would then agree, disagree, or remain silent. At other times, the parent or legal guardian gave the child space to answer on their own. The experiences shared in this analysis focused explicitly on the children's responses. Three themes and their respective subthemes were identified (Multimedia Appendix 2).

Relating to Others

Children with OI described how they saw themselves in relation to other children. They demonstrated that they were different but also very similar to any other child (Figure 2).

Figure 2. The Sisom graphic that depicts the theme "Relating to Others."

Common OI Experience

Children with OI have a distinctive experience that unites them. They all recognize their integral role within the OI community. Children used terms such as "OI kids" [P5, 10 years old, type IV OI], "We" [P5, 10 years, type IV OI], and "Us" [P7, 9 years old, type III OI], showcasing this common OI experience. One child with an extensive OI history used her experience to speak on behalf of other members of the OI community stating "Well, OI kids, we as, since I'm an OI kid, [...] since I know..." [P5, 10 years old, type IV OI]. Some children seemed to adopt advocacy roles. One child described how families with children with OI look out for one another. He explained how his mother developed a *break* kit for other OI families:

> *We had a friend that lived 45 minutes away from us that had OI and he broke and he has new parents, like he's one year old, so they don't know how to splint. So mum went over and splinted him so that's how we thought of it!* [P12, 10 years old, type IV OI]

One child answered his questions about Sisom by thinking of others:

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> *I think, we have to like, keep this because, some people, it's hard [...]...Some people are impatient.* [P4, 8 years old, type IV OI]

Another child put herself in others' shoes and remarked that older children have more difficulty than younger children in asking for help:

> *It's possible, when you're younger, that it doesn't bother you, but when you're older, it bothers you more. It embarrasses you. You want to do everything yourself...So there's a big difference.* [P11, 11 years old, type IV OI]

One child advocated for the creation of *Sisom OI* in other languages so that it could be used by more children (P12, 10 years old, type IV OI). All children showed profound empathy for others with OI in their responses. Those who were more novice asked questions to the interviewer to understand the experience of others to answer Sisom's prompts: "Probably no problem, but I never tried it though, by the way" [P6, 6 years old, type I OI]. Those who were more expert related their own experience to how others might feel:

> *Sometimes people are, are so like, you don't know what you want, but that happens to me sometimes and sometimes people will cry because of that. Like me.* [P4, 8 years old, type IV OI]

Feeling Different

Children were asked by Sisom if they felt different than other children (Figure 2). They all responded that they did. One of them stated matter-of-factly: "Ya...because other kids are different than us." [P5, 10 years, type IV OI]. They also expressed that feeling different affected their lives in several ways. First, many of the children reflected that their differences led to being bullied. One child expressed how her peers at school teased her because she was different: "They often say, let's say, that I don't run fast, and they laugh at me." [P8, 7 years, type I OI].

Second, they reflected that their differences led others to stare at them, "the gaze of others" [P3, 11 years, type IV OI], which they acutely sensed, and caused their worries about feeling different to amplify. One child described how people gawked at her when she went out to the restaurant: "It's that I often get stared at, at the restaurant, and I find that unpleasant" [P3, 11 years old, type IV OI]. Other children described strangers directly asking them questions about their bodies and having to explain their condition to others. For some, this was frustrating. In contrast, 1 participant did not let these types of questions hold her back from sharing knowledge with others about her condition:

> *I just tell them to "Be careful! Because I could get hurt more easily than you." I remember that there was someone that told me at Burger King: "Why are you more fragile than others?" I just explained to him that "Well, I have an illness, so my bones are more fragile and that if I fall it could break me..." and he was like "Ah..." Because [...] he kind of bumped into me.* [P11, 11 years old, type IV OI]

Third, they reflected that their differences led others to treat them differently. One child described how others were afraid to break her bones and that she was ostracized on a daily basis:

> *I feel very lonely. No one wants to play with me because of my illness.* [P8, 7 years old, type I OI]

Other children expressed that people's perceptions of the hardships of living with OI were overexaggerated. One child recounted the story of a stranger who wanted to give her a hug because she felt pity for her [P3, 11 years old, type IV OI]. Another child reflected on how he felt when his mother was overprotective of him:

> *That's annoying! I hate that. Be careful! You might get hurt.* [P7, 9 years old, type III OI]

However, 2 children viewed being different as positive, stating:

> *Everybody's different* [P4, 8 years old, type IV OI]
>
> *Being different from other kids is a good thing. Like, for example, I think that because I'm in a wheelchair, I can do things that other kids can't do, and that, that part's a good thing.* [P5, 10 years old, type IV OI]

Feeling Just Like Others

All children demonstrated that they were more similar to others than they were different. The children shared their desires, wishes, and preferences just as any other child would. The goal was to engage children in the co-design of the *Sisom OI* paper prototype; however, OI was sometimes left out of the conversation altogether by the participants. For instance, when 1 child was asked what makes them feel different from other children, they commented on their age. When asked if there was anything else, they stated, "That's it" [P10, 9 years old, type VI OI]. Every interview included children laughing, playing, and sharing, just as any other child would.

They shared things about themselves that they found special, such as their likes: "We all love unicorns!" [P6, 6 years old, type I OI]; their dreams: "I want to be a vet." [P5, 10 years old, type IV OI]; and their hobbies:

> *I am a HUGE Harry Potter fan! I am at the 5th book.* [P7, 9 years old, type III OI]

One child, by having people get to know her, showed others that she was more than just an *OI kid*:

> *They just don't know me yet. So like, [...] if a friend was there, then they wouldn't look at me like that, cause they'd know that it's my, I have a condition...* [P5, 10 years old, type IV OI]

This child suggested the creation of a Sisom island called *About Me* in which children could share what was unique about them. This child also emphasized that the avatar they create of themselves within *Sisom OI* be as realistic as possible. This view was shared by all participants: "You would like [...] that it looks the most like you..." [P7, 9 years old, type III OI] so that "You could see yourself...Through a computer technological program" [P4, 8 years old, type IV OI]. Many of the children had fun creating their avatars and customizing them:

> *Ya, I like the color on that one!* [P9, 6 years old, type III OI]

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> *I'd like to put some shoes I like to wear. I got light up shoes but I'm not wearing them now...* [P10, 9 years old, type VI OI]

Many of the children reflected how they were the same as everyone else by normalizing their symptoms, stating:

> *Everyone's gonna feel like that, right?* [P7, 9 years old, type III OI]
>
> *It's pretty much [like that for] everyone.* [P3, 11 years old, type IV OI]

They often described how they felt the same way others felt and shared many of the same experiences. Similarly, when 1 child was asked about her nightmares, she answered:

> *I have more nightmares about like clowns and animals and stuff...That's what scares me. Not really surgeries.* [P11, 11 years old, type IV OI]

The same participant explained how she adapts her situation to be able to participate in events just like any other child. For instance, she takes her wheelchair out to go "trick-or-treating" to make the most of it:

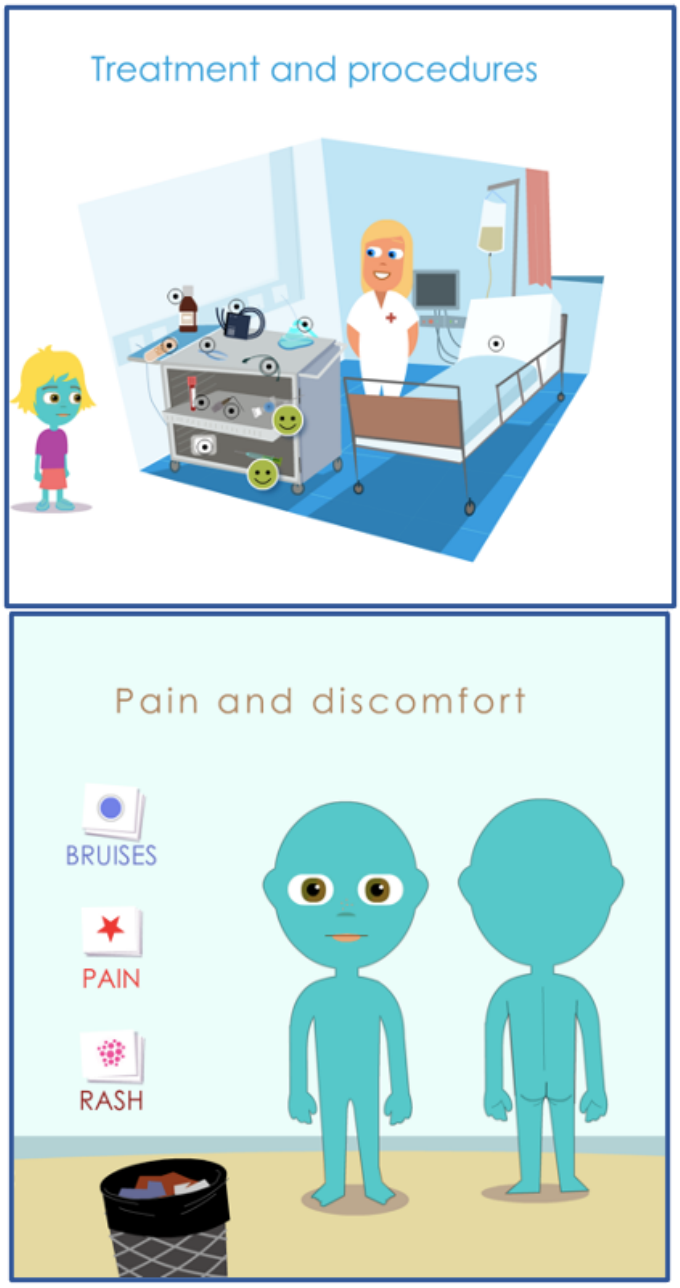
> *For example, Halloween, I would never do that with my walker! I would never be able! [...] By the time I get there wouldn't be any candy left!* [P11, 11 years old, type IV OI]

Relating to Their Condition

Overview

In *Relating to Their Condition*, children described their positive and negative interactions with their own condition and the health care system (Figure 3).

Figure 3. The Sisom graphics that depict the theme "Relating to Their Condition."



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An example of a child's quote on *Relating to Their Condition* is as follows:

> *The problem with OI is not how you look, it's like, how you feel...is there any pain? Stuff like that! Not how you look! This is actually...That's what makes sense to me.* [P4, 8 years old, type IV OI]

Understanding Their Condition

Each child interviewed had a thorough understanding of their OI. The depth of their knowledge seemed to correlate with the breadth of their experiences within the health care system. Children who were older with more severe OI had more extensive experiences with treatments, procedures, and the hospital overall. Within our sample, children ranged from knowing the general principles and basics of OI to being very informed and involved in their own health.

Some children only had rudimentary knowledge. One child was asked about her experience with OI. She simply replied:

> *No, I don't know that [OI] [...] I come here to get my bones looked at.* [P6, 6 years old, type I OI]

Similarly, another child, when asked about OI, stated:

> *What is that?...I have brittle bones like I can just hit [...] something and [...] get hurt.* [P10, 9 years old, type VI OI]

Most children understood that their condition was a hereditary genetic disorder that affected their bones and caused pain. One child described it as, "Having pain in my bones" and "For example, I could have had brothers and little sisters with this disease" [P8, 7 years old, type I OI].

In contrast, one child knew the names of many of his bones, medications, and their indications as well as the pathophysiology of OI:

> *This arm, before it was operated, it was like a banana. [...] And it was because, my muscles, they're fine. But, it's that, they pulled on my bone, since my bone is like, so much like rubber, it like, weep [sound].* [P7, 9 years old, type III OI]

One child even described her disease in a metaphor to be able to help others understand:

> *Because, the OIs, they say "bones of glass." So maybe [we could show] a glass that is broken.* [P11, 11 years old, type IV OI]

The way in which these children interpreted and related their experiences with OI to others seemed to depend on their level of involvement with their own health. However, each child chose a similar way of expressing what it meant to have OI to them by physically showing where they had pain (P6, 6 years old, type I OI), discomfort (P3, 11 years old, type IV of OI), and surgical scars (P7, 9 years old, type III OI). One child got up on a chair to show from what height she might need to fall to break a bone (P6, 6 years old, type I OI). Another child, when approached about how he felt about his appearance, stated:

> *The problem with OI is not how you look, it's like, how you feel...is there any pain? Stuff like that! Not how you look! This is actually...That's what makes sense to me.* [P4, 8 years old, type IV OI]

Special Relationship With the Hospital

Children with OI reported spending significant amounts of time in the hospital. As put positively by 1 child, "Sometimes I say the hospital and I are friends cause OI kids, we go to hospital a lot, for check-ups and stuff" [P5, 10 years old, type IV OI]. Children described many long and boring days in the hospital. One child expressed:

> *...Some patients with surgeries, they like...I don't know they like, get bored, in the, sitting in the room in a bed all day long.* [P4, 8 years old, type IV OI]

Another child described the dread of staying overnight:

> *Mostly I am scared of having pain when I turn,...I am scared of spending a bad night after my surgery [...] It's cause you never know [...] if you'll sleep, if you won't sleep...* [P11, 11 years old, type IV OI]

Children also missed their home, family, and friends. One child expressed not liking her treatments at the hospital because of "The fact that I'm far away from my sisters" [P8, 7 years old, type I OI]. Another child described missing home during a long stay after her spinal surgery:

> *We stayed there for a like a long time. Like one week, and I got sooo homesick. I missed like the city, the hustle and bustle, I missed my friends.* [P5, 10 years old, type IV OI]

It was also common for children to travel from great distances to receive treatment, as it was for one child, whose mother had to stay home, in another province:

> *Whenever I call mom I start to cry [...] She can't come here cause she has to work.* [P6, 6 years old, type I OI]

The hospital became somewhat of a home for many of these children who had formed a special relationship with the space and staff. For example, 1 child described his experience in the playroom:

> *Back when we had surgery, here, we always loved to go to the playroom [...] The volunteers, I actually like the volunteers, like I made a friend [and...] we made a made-up movie together.* [P4, 8 years old, type IV OI]

This special relationship also extended to the physicians, in whom the children placed a lot of trust to help them get better. One child described her fear of dying during surgery and that she placed trust in her physicians to help her through:

> *I trust doctors...cause they want to help me feel better. [...] Surgeons, yeah.* [P5, 10 years old, type IV OI]

Difficult Treatments and Procedures

Children described having to undergo countless unpleasant and painful procedures. When asked about medications, almost all the children expressed that they tasted horrible:

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> *...sometimes when I eat medicine, my taste buds are screaming at me like "WHY ARE YOU EATING THIS?"* [P5, 10 years old, type IV OI]

Another common and dreadful experience was having to undergo surgery. One child enumerated 3 different reasons for requiring surgery because of OI, including coxa vara repair, scoliosis repair, and rerodding. She described her experience with surgeries:

> *I have a lot of surgeries, when you have surgeries and you fall asleep for the first time, it's like very difficult, cause I see a lot of nightmares.* [P5, 10 years old, type IV OI]

It was something that she had a lot of experience with and to which she was now getting accustomed. This was similar to another child who showed off the many scars from a number of different surgeries [P7, 9 years old, type IV OI]. Another child emphasized how often he had surgeries:

> *Sometimes like, if I get hurt and I hear my leg crack [...]. They go in, and then, if I didn't have a rod in, they put a rod in, and that's how they do it...I did this MANY times.* [P10, 9 years old, type VI OI]

Children not only expressed that they felt discomfort during hospital procedures but also shared their pride in having gotten used to certain painful procedures. One child described being "Brave for everything" and doing a "Good job" by not crying during painful procedures, which she described as "another unhappy task" [P6, 6 years old, type I OI]. Another child justified his painful experiences with their purpose:

> *IVs* [intravenous accesses] are, they're good. But like, so like you could have medicine...But, then again IVs do hurt a bit. But, they're good so...[P4, 8 years old, type IV OI]

In addition, the children reflected that tolerating pain came with experience. One child reflected on how he had come so far:

> *Not a problem, for shots, for me...It's been a long...At the beginning I couldn't...* [P7, 9 years old, type III OI]

Reflecting on Capabilities

Overview

By reflecting on their capabilities, children were able to identify some of their strengths and weaknesses (Figure 4).

An example of a child's quote on *Overcoming Isolation* is as follows:

> *I do not like playing outside [alone] because everybody excludes me. [...] I like playing outside even though I have this condition, because I have many friends at school.* [P7, 9 years old, type III OI]

Figure 4. The Sisom graphics that depict the theme "Reflecting on Capabilities" and the subtheme "Overcoming Isolation."

Perceiving Limitations

All children expressed a desire to do something they were physically incapable of because of a fear of getting hurt. For some children, this activity was eating. Some described not being able to eat tasty foods that were "Like, let's say, sticky things...and things that are too hard" [P8, 7 years old, type I OI]

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and "like [...] bars, for example, I can no longer have them..." [P3, 11 years, type IV OI] because of an orthodontic mouthpiece they were required to wear for their OI. One child explained that OI prevented her from playing with her friends:

> *Like for example...there, like, in my school yard, there's like a big slide [...] where you climb up high, it's like twisty. [...] so whenever I see them, my friends, they're going down the slide, they're having so much fun, it's like, I can't do it.* [P5, 10 years old, type IV OI]

Another explained that his OI led to fractures that required him to wear a cast for healing. During this period, he was not able to play sports with his friends [P7, 9 years old, type III OI]. This sentiment was echoed by another child who drew a picture to show how it felt to be excluded because of an injury:

> *Like, if he's looking at his cast, and his friends are outside playing street hockey...He's in his room [...] It affects you to watch your friends play and you have to stay inside doing nothing.* [P11, 11 years old, type IV OI]

Children also expressed how their perceived limitations may affect their future endeavors. One child expressed her desire to be a good dancer but that this was difficult because of the way her OI affected her bones. She reflected:

> *Well, cause, I...I just wanna be a good dancer, and like, I wanna have strong bones...[P6, 6 years old, type I OI]*

Another child stated that it would be important to include the question "Are you afraid that your handicap will prevent you from doing what you really want in life?" [P11, 11 years old, type IV OI] in *Sisom OI*. She explained how a person with OI may want to become a police officer but would have to settle for working in the office answering phones instead of being out in the field.

Some children also reflected on what their lives might have been like without OI. One recounted her daydreams and how she would "Go to any place [...] hang out with [her] friends alone, [...] help [her] mom a lot, [...and] go around [...] to places with [her] dad" [P5, 10 years old, type IV OI]. She reflected on needing help for some activities for which she was dependent on others. She also asserted her independence for other tasks. Most of the other children reported a similar balance.

Overcoming Isolation

Ideas of feeling left out were expressed by all children. When prompted by Sisom "Do you ever feel left out?" children answered:

> *A lot, because, because no one, almost no one, wants to be with me because of my disease.* [P8, 7 years old, type I OI]

> *Sometimes, [...] cause sometimes kids, they don't wanna play with you, or, because of your condition they don't wanna talk to you...* [P5, 10 years old, type IV OI]

They overcame these feelings by finding independent activities and excelling in other areas, escaping through imagination, having good friends, and thinking positively.

First, the children asserted their independence in several areas such as reading and journaling and taking time for themselves to be alone: "I want to write something on here!" [P9, 6 years old, type III OI]. One child described how she turned to nature when her sister did not want to play with her: "So I just go outside and sit behind one of my trees" [P6, 6 years old, type I OI]. Another child described how while she was dependent on others in some areas, she was determined to find ways to maintain her independence:

> *When I'm at school, I have an aid, but she doesn't go in the bathroom with me, she just holds the door in case I fall [...] I do my things, and I leave. She doesn't come in with me, I do it all by myself.* [P11, 11 years old, type IV OI]

Second, building and maintaining strong friendships helped children overcome the feeling of being left out. One child referred to her pets when she reflected that "at least I have friends" [P8, 7 years old, type I OI]. Another explained that good friends made feelings of exclusion tolerable:

> *I do not like playing outside [alone] because everybody excludes me. [...] I like playing outside even though I have this condition, because I have many friends at school.* [P7, 9 years old, type III OI]

One child reflected how she did not feel bad when her friends played without her at recess but always appreciated the time that they spent with her:

> *They climb the spider webs, and all those things, and me, I couldn't do them, so I stayed sitting at the table with my friends and we talked...Sometimes they left and they came back...To have fun a bit [...] I wasn't going to prevent them from doing everything and say: "No you stay with me because I can't do it" [Laughs].* [P11, 11 years old, type IV OI]

Third, they used their imagination to escape to different worlds by making up "a game like BOOM" [P7, 9 years old, type III OI], having an "imaginary friend" [P7, 9 years old, type III OI], imagining a "candy world, [...] a land full of candy and unicorns" [P6, 6 years old, type I OI], and "daydream[ing] about [the] future [...] working with animals" [P5, 10 years old, type IV OI]. Positive thinking was implicit in the attitudes of all children. One child embraced this philosophy when she explicitly stated:

> *As a person, you always have to think positive. Cause if you think about the negative, then you'll see life in a bad way. But life is not as bad as you think.* [P5, 10 years old, type IV OI]

Celebrating Strengths

All the children experienced feelings of otherness, exclusion, and physical limitations, but they also celebrated their strengths and asserted their independence in various ways. Children were excited about their mobility improvements:

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> *I am quickly learning how to walk.* [P8, 7 years old, type I OI]
>
> *[running], that's the next step.* [P7, 9 years old, type III OI]

Some expressed confidence and pride in their current mobility status:

> *I'm an expert driver with my wheelchair!* [P4, 8 years old, type IV OI]
>
> *You know, I go really fast in my wheelchair.* [P3, 11 years old, type IV OI]

This confidence allowed them to attend special events and show off their skills. One child participated in special Paralympic tournaments: "I won [...] I got three gold medals" [P3, 11 years, type IV OI]. She also played soccer at her specialized school, where she felt comfortable being herself.

Another child also played soccer and hockey with his friends [P7, 9 years old, type III OI]. These adapted environments were important contributors in reinforcing feelings of inclusion and value.

The children also celebrated their academic accomplishments. They highlighted how well they do in school: "I'm one of the best readers in my class!" [P12, 10 years old, type IV OI]. The children described their accomplishments with pride and confidence. One child explained how she was able to excel even in an accelerated program:

> *I am in the intensive stream [...] So I do my sixth grade in six months...[Actually] five months! [...] So I have to go quickly! It's like I've become a model!* [P11, 11 years old, type IV OI]

Finally, children with OI seemed to truly look out for one another. Their involvement in this study reflected kinship. Many participants understood the research implications for other children with OI. They highlighted the importance of including "everybody's ideas, everybody's advice" [P7, 9 years old, type III OI] and hoped:

> *that kids will love it! Cause, we need to express it in a way that the public will love it [...] So we can make money and we can help this disease!* [P4, 8 years old, type IV OI]

This kinship was further exemplified by one child who was very implicated in fundraising for OI. She made it a point to encourage other children with OI to keep their heads high:

> *From my point of view, OI kids, they don't think that they're [...] awesome. [...] But you are awesome. I sometimes tell OI kids that, cause like, most of the ones that I know, they're like young, like five or six, [...] So I tell them that: "You are awesome, cause you went through so much, and you're so little!"* [P5, 10 years old, type IV OI]

Not only did she celebrate her own strengths, but she also encouraged others to find their strengths and celebrate them.

Discussion

Principal Findings

Efforts to *authentically listen* and bring to light what children with OI shared when approached as experts in their condition was made possible by conducting a secondary analysis of qualitative data sources collected for the co-design of the *Sisom OI* paper prototype [13]. This study demonstrates, as shown in other recent findings, that children who use Sisom are prompted to share their experiences [25,27]. Through the analysis of their dialogue, 3 themes were identified. The first, *Relating to Others*, described the balance between feeling different versus feeling similar to other children. The subthemes were *Common OI Experience*, *Feeling Different*, and *Feeling Just Like Others*. The second, *Relating to Their Condition*, described children's positive and negative interactions with their own condition and the health care system. The subthemes were *Understanding Their Condition*, *Special Relationship with the Hospital*, and *Difficult Treatments and Procedures*. The third, *Reflecting on Capabilities*, described children's recognition of their strengths and limitations. The subthemes were *Perceiving Limitations*, *Overcoming Isolation*, and *Celebrating Strengths*.

Children with OI had a unique way of maintaining balance in their lives. This was demonstrated through all three of the themes. First, in *Relating to Others*, children described a balance between feeling different and feeling similar to other children. They also expressed a concern about the perception of others. This has previously been described in other studies of children with OI [8,38]. An important way that children with OI overcame this feeling of otherness was through building relationships with other children and clinicians in the OI community [8,39]. Another way children navigated between feeling different and feeling similar to other children was by sharing with others the special things that defined them and by participating in activities with other children as best as they could. This helpful strategy has been described in other populations of children with chronic conditions [17,40].

Second, in *Relating to Their Condition*, children described a balance between the positive and negative aspects of living with OI. Children's depth of understanding and moral awareness can range from *rich sophistication* to *simplistic matter-of-factness* [20]. Children are capable of such awareness, and their voice should be attended to without comparison with the adult perspective [20]. Indeed, in the data, there was a wide range of depth in the understanding of OI. All children were able to share what it meant to have OI. Being in the hospital was an experience that was painful, with numerous unpleasant procedures including surgeries, needle pricks, and extended hospitalizations. The fear of needles was encountered by children living with OI in this study as well as others [8,38]. Tolerating pain and *getting used to* painful procedures was something that children with OI have expressed in other studies [38]. This idea of *getting used to* pain was expressed by the children as a proud adaptation to a difficult aspect of living with OI. Another way that children overcame the negative aspects of being hospitalized was by reflecting on the positive relationships they had developed with clinicians and volunteers,

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which other children have reported [18,41]. It has been shown previously that children with OI form special relationships with people who understand OI, for instance, their physicians [8].

Finally, in *Reflecting on Capabilities*, children described a balance between perceiving their limitations and celebrating their strengths. Feeling left out because of the fear of fractures or physical limitations was a major element of this theme and has been explored in other studies [8,38,42]. Despite the presence of this feeling, children with OI have been found to have an immensely positive and strength-based outlook on life [8,39,42]. Our study showed 1 way that these children embraced positivity was to constantly strive to improve themselves physically and academically. This concept of a strong desire to move forward was supported by other studies; however, these studies also showed that children with OI held back due to the fear of pushing too hard and fracturing [8,39]. Children's perception of these physical limitations propelled them to take on and excel in their intellectual feats [8]. Interestingly, our study showed that, given the right environment and support, children with OI are also capable of excelling in physical feats. These children are *taking life in stride* and using their positive outlook to overcome feeling different and feeling left out and making the best of every day [42].

Sisom may contribute to transforming a child's *place* into a child's *space* [13] by inviting children to participate in their care [28] and showcasing how they can serve as advisors and partners in their care [21]. Sisom allows for the promotion of discussions about what children deem the most important. This essential information may help clinicians enhance children's participation in their own care and ultimately empower them to cope with the difficult physical, mental, and social challenges they face. However, *Sisom OI* is yet to be developed, tested, or integrated into practice. Therefore, these study findings offer only preliminary insight into the types of discussions that may unfold from the future use of *Sisom OI* in practice. As OI remains a rare disease with few clinicians encountering a child with OI over the course of their career, there may be some reluctance to use Sisom. Offering training and delineating potential responses to children's expressed concerns may help integrate this novel, interactive, computerized tool into practice and contribute to the transformation of children's rightful space in their health care systems [28,35].

In this study, the partnership created between the interviewer and the child successfully elicited and highlighted the *child's perspective*, placing the child in the center of their experience. These children had many things to share about their experience, as noted by other chronically ill children [43]. Although parental involvement, at times, made it difficult for the child to take the lead, our approach revealed the child's perspective. Parents highlighted the things with which children did not necessarily agree. These differences in perspective have been reported in previous studies [15,16]. Sisom's ability to highlight such differences is considered a key feature, as it strives to promote communication between children and adults so that they may engage in shared decision-making.

Limitations

The children in this study were prompted to give their opinion on Sisom to co-design *Sisom OI*. Therefore, they were not directly prompted to share their OI experiences, unless prompted by the interviewer. The questions posed in Sisom were not tailored to specifically capture the experiences of children with OI. This may have led to significant data gaps. Future studies should be conducted to specifically aim to understand children's perspective on their OI.

Furthermore, the involvement of parents and legal guardians in the interviews had, in some cases, a muting effect on the child. Instead of giving space for their child to answer the questions independently, some took it upon themselves to answer for their child. In some cases, this prompted the child to express their feelings and share their experiences, whereas in other cases, this caused the child's voice to be lost. Further studies should consider how to highlight the child's voice by emphasizing on the child's answers and constantly reemphasizing the purpose of the study to the parents involved.

Clinical Implications

This study highlights the richness of the experiences shared by children who are approached as experts in their own condition. These children had valuable information and insights to share regarding their day-to-day lives and the factors in their lives that both hinder their development and support their endeavors. The study findings offered a glimpse of what it means to grow up living with OI but also showcased a novel approach to gain their perspective. The children's inputs will be further used to advance the development of *Sisom OI* and our overall approaches to offer care that responds directly to their needs and empower them to be active partners in their care.

Research Implications

The study findings will be also used to inform the development, verification, validation, and evaluation of *Sisom OI* to ensure that meaningful aspects of life with OI are included in the app and attended to in practice. At present, the *Sisom OI* paper prototype has been developed [13]. Although many of the existing Sisom symptoms were deemed relevant for inclusion in the *Sisom OI* paper prototype, 57 new symptoms were generated. The relevant symptoms addressed children's thoughts and feelings about hospitalization and their wishes for participation in their own care. The new symptoms addressed fractures, body image, and social isolation related to difficulties with accessibility and intimidation. Therefore, these findings, in addition to the themes identified in this study, will be used to inform the next phase of *Sisom OI* development with end user partnerships, stakeholder buy-in, and the necessary funding. We expect to build the prototype; test for usability; pilot for feasibility; and evaluate the integration of *Sisom OI* into the hospital, home, and school settings.

Conclusions

This co-design process provided children with OI the space to not only contribute to the development of the end product but also to eloquently describe their experiences. These findings, based on the description given by the children themselves, offer us a unique understanding of what it means to grow up with OI.

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Conflicts of Interest

None declared.

Multimedia Appendix 1
Child interview guide.
[DOCX File , 17 KB - pediatrics_v4i3e22784_app1.docx]

Multimedia Appendix 2
Themes and subthemes identified when children were approached as experts in the co-design of Sisom osteogenesis imperfecta.
[DOCX File , 534 KB - pediatrics_v4i3e22784_app2.docx]

References

1. Palomo T, Vilaça T, Lazaretti-Castro M. Osteogenesis imperfecta. Curr Opin Endocrinol Diabetes Obes 2017;24(6):381-388. [doi: 10.1097/med.0000000000000367]
2. Bardai G, Moffatt P, Glorieux FH, Rauch F. DNA sequence analysis in 598 individuals with a clinical diagnosis of osteogenesis imperfecta: diagnostic yield and mutation spectrum. Osteoporos Int 2016 Dec 11;27(12):3607-3613. [doi: 10.1007/s00198-016-3709-1] [Medline: 27509835]
3. Tauer JT, Robinson M, Rauch F. Osteogenesis imperfecta: new perspectives from clinical and translational research. JBMR Plus 2019 Aug 20;3(8):e10174 [FREE Full text] [doi: 10.1002/jbm4.10174] [Medline: 31485550]
4. Dahan-Oliel N, Oliel S, Tsimicalis A, Montpetit K, Rauch F, Dogba MJ. Quality of life in osteogenesis imperfecta: a mixed-methods systematic review. Am J Med Genet A 2016 Jan 14;170A(1):62-76. [doi: 10.1002/ajmg.a.37377] [Medline: 26365089]
5. Lindahl K, Åström E, Rubin C, Grigelioniene G, Malmgren B, Ljunggren Ö, et al. Genetic epidemiology, prevalence, and genotype-phenotype correlations in the Swedish population with osteogenesis imperfecta. Eur J Hum Genet 2015 Aug 06;23(8):1042-1050 [FREE Full text] [doi: 10.1038/ejhg.2015.81] [Medline: 25944380]
6. Folkestad L. Mortality and morbidity in patients with osteogenesis imperfecta in Denmark. Dan Med J 2018 Apr;65(4):B5454 [FREE Full text] [Medline: 29619932]
7. Trejo P, Rauch F. Osteogenesis imperfecta in children and adolescents-new developments in diagnosis and treatment. Osteoporos Int 2016 Dec 5;27(12):3427-3437. [doi: 10.1007/s00198-016-3723-3] [Medline: 27492436]
8. Tsimicalis A, Denis-Larocque G, Michalovic A, Lepage C, Williams K, Yao T, et al. The psychosocial experience of individuals living with osteogenesis imperfecta: a mixed-methods systematic review. Qual Life Res 2016 Aug 19;25(8):1877-1896. [doi: 10.1007/s11136-016-1247-0] [Medline: 26894269]
9. Shapiro JE, Germain-Lee EL. Osteogenesis imperfecta: effecting the transition from adolescent to adult medical care. J Musculoskelet Neuronal Interact 2012 Mar;12(1):24-27 [FREE Full text] [Medline: 22373948]
10. Fegran L, Hall EO, Uhrenfeldt L, Aagaard H, Ludvigsen MS. Adolescents' and young adults' transition experiences when transferring from paediatric to adult care: a qualitative metasynthesis. Int J Nurs Stud 2014 Jan;51(1):123-135. [doi: 10.1016/j.ijnurstu.2013.02.001] [Medline: 23490470]
11. Nghiem T, Louli J, Treherne S, Anderson C, Tsimicalis A, Lalloo C, et al. Pain experiences of children and adolescents with osteogenesis imperfecta: an integrative review. Clin J Pain 2017 Mar;33(3):271-280. [doi: 10.1097/AJP.0000000000000395] [Medline: 27258996]
12. Me, myself, and OI. The Extraordinary Adventures of a Breakable Girl. 2014. URL: https://www.youtube.com/watch?v=IAhs-piej1o [accessed 2021-08-01]
13. Siedlikowski M, Rauch F, Tsimicalis A. Giving children with osteogenesis imperfecta a voice: participatory approach for the development of the interactive assessment and communication tool Sisom OI. J Med Internet Res 2020 Sep 22;22(9):e17947 [FREE Full text] [doi: 10.2196/17947] [Medline: 32960176]
14. Ruland C, Slaughter L, Starren J, Vatne T, Moe E. Children's contributions to designing a communication tool for children with cancer. Stud Health Technol Inform 2007;129(Pt 2):977-982. [Medline: 17911861]

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15. Livesley J, Long T. Children's experiences as hospital in-patients: voice, competence and work. Messages for nursing from a critical ethnographic study. Int J Nurs Stud 2013 Oct;50(10):1292-1303. [doi: 10.1016/j.ijnurstu.2012.12.005] [Medline: 23332686]
16. Garth B, Aroni R. 'I Value What You have to Say'. Seeking the perspective of children with a disability, not just their parents. Disab Soc 2003 Aug;18(5):561-576. [doi: 10.1080/0968759032000097825]
17. Alderson P, Sutcliffe K, Curtis K. Children as partners with adults in their medical care. Arch Dis Child 2006 Apr 11;91(4):300-303 [FREE Full text] [doi: 10.1136/adc.2005.079442] [Medline: 16399782]
18. Sartain S, Clarke C, Heyman R. Hearing the voices of children with chronic illness. J Adv Nurs 2000 Oct;32(4):913-921. [Medline: 11095230]
19. United Nations. The Convention on the Rights of the Child. Geneva, Switzerland: United Nations; 1989:1-15.
20. Carnavale F. Listening authentically to youthful voices: A conception of the moral agency of children. In: Storch JL, Rodney P, Starzomski R, editors. Toward a Moral Horizon: Nursing Ethics for Leadership and Practice. Canada: Pearson Education; 2004:315-332.
21. Wyness M. Adult's involvement in children's participation: juggling children's places and spaces. Child Soc 2009;23(6):395-406. [doi: 10.1111/j.1099-0860.2008.00181.x]
22. Ruland CM, Starren J, Vatne TM. Participatory design with children in the development of a support system for patient-centered care in pediatric oncology. J Biomed Inform 2008 Aug;41(4):624-635 [FREE Full text] [doi: 10.1016/j.jbi.2007.10.004] [Medline: 18082468]
23. Arvidsson S, Gilljam B, Nygren J, Ruland CM, Nordby-Bøe T, Svedberg P. Redesign and validation of Sisom, an interactive assessment and communication tool for children with cancer. JMIR Mhealth Uhealth 2016 Jun 24;4(2):e76 [FREE Full text] [doi: 10.2196/mhealth.5715] [Medline: 27343004]
24. Sisom: Child communication. DHealth. 2021 Feb 16. URL: https://dhealthapp.no/en/products/ [accessed 2021-08-01]
25. Tsimicalis A, Le May S, Stinson J, Rennick J, Vachon M, Louli J, et al. Linguistic validation of an interactive communication tool to help French-speaking children express their cancer symptoms. J Pediatr Oncol Nurs 2017 Jul 07;34(2):98-105. [doi: 10.1177/1043454216646532] [Medline: 27226380]
26. Baggott C, Baird J, Hinds P, Ruland CM, Miaskowski C. Evaluation of Sisom: a computer-based animated tool to elicit symptoms and psychosocial concerns from children with cancer. Eur J Oncol Nurs 2015 Aug;19(4):359-369. [doi: 10.1016/j.ejon.2015.01.006] [Medline: 25691300]
27. Tsimicalis A, Stone P, Bakken S, Yoon S, Sands S, Porter R, et al. Usability testing of a computerized communication tool in a diverse urban pediatric population. Cancer Nurs 2014;37(6):25-34 [FREE Full text] [doi: 10.1097/NCC.0000000000000115] [Medline: 24457227]
28. Svedberg P, Arvidsson S, Larsson I, Carlsson I, Nygren JM. Barriers and enablers affecting successful implementation of the electronic health service sisom: multicenter study of child participation in pediatric care. J Med Internet Res 2019 Nov 15;21(11):e14271 [FREE Full text] [doi: 10.2196/14271] [Medline: 31730040]
29. Hugues KA, Tarrant A. Qualitative Secondary Analysis. Thousand Oaks, California, United States: SAGE Publications Ltd; 2019:1-240.
30. UX design for children (Ages 3-12) 4th edition. Nielsen Norman Group. URL: https://www.nngroup.com/reports/children-on-the-web/ [accessed 2021-03-10]
31. UX design for teenagers (Ages 13-17) 3rd edition. Nielsen Norman Group. URL: https://www.nngroup.com/reports/teenagers-on-the-web/ [accessed 2021-03-10]
32. Nielsen J, Landauer T. A mathematical model of the finding of usability problems. In: Proceedings of the INTERACT '93 and CHI '93 Conference on Human Factors in Computing Systems. 1993 Presented at: INTERCHI93: Conference on Human Factors in Computing; April 24 - 29, 1993; Amsterdam, The Netherlands. [doi: 10.1145/169059.169166]
33. Nielsen J. Why you only need to test with 5 users. Nielsen Norman Group. 2000. URL: https://www.nngroup.com/articles/why-you-only-need-to-test-with-5-users/ [accessed 2021-08-01]
34. Tsimicalis A, Rennick J, Stinson J, May SL, Louli J, Choquette A, et al. Usability testing of an interactive communication tool to help children express their cancer symptoms. J Pediatr Oncol Nurs 2018 May 31;35(5):320-331. [doi: 10.1177/1043454218777728] [Medline: 29848164]
35. Tsimicalis A, Rennick J, Le May S, Stinson J, Sarkis B, Séguin K, et al. “Tell it as it is”: How Sisom prompts children and parents to discuss their cancer experience. Cancer Rep 2019 Apr 23;2(3):1-15. [doi: 10.1002/cnr2.1173]
36. Polit D, Beck C. Nursing Research: Generating and Assessing Evidence for Nursing Practice. Pennsylvania: Lippincott Williams & Wilkins; 2008:1-796.
37. Sandelowski M. Whatever happened to qualitative description? Res Nurs Health 2000 Aug;23(4):334-340. [doi: 10.1002/1098-240x(200008)23:4<334::aid-nur9>3.0.co;2-g]
38. Hill CL, Baird WO, Walters SJ. Quality of life in children and adolescents with Osteogenesis Imperfecta: a qualitative interview based study. Health Qual Life Outcomes 2014 Apr 16;12(1):54 [FREE Full text] [doi: 10.1186/1477-7525-12-54] [Medline: 24742068]

XSL•FO
RenderX

39. Dogba MJ, Bedos C, Durigova M, Montpetit K, Wong T, Glorieux FH, et al. The impact of severe osteogenesis imperfecta on the lives of young patients and their parents - a qualitative analysis. BMC Pediatr 2013 Sep 30;13(153):1-8 [FREE Full text] [doi: 10.1186/1471-2431-13-153] [Medline: 24074180]
40. Kane H, Fearon D. 'It grabs me by my neck': Exploring children's experience of chronic illness in Mauritania through drawings and interviews. Childhood 2017 Nov 09;25(2):203-219. [doi: 10.1177/0907568217739354]
41. Angström-Brännström C, Norberg A, Jansson L. Narratives of children with chronic illness about being comforted. J Pediatr Nurs 2008 Aug;23(4):310-316. [doi: 10.1016/j.pedn.2007.04.006] [Medline: 18638674]
42. Wiggins S, Kreikemeier R. Bisphosphonate therapy and osteogenesis imperfecta: The lived experience of children and their mothers. J Spec Pediatr Nurs 2017 Oct 06;22(4):e12192. [doi: 10.1111/jspn.12192] [Medline: 28876506]
43. Söderbäck M, Coyne I, Harder M. The importance of including both a child perspective and the child's perspective within health care settings to provide truly child-centred care. J Child Health Care 2011 Jun;15(2):99-106. [doi: 10.1177/1367493510397624] [Medline: 21685225]

Abbreviations

OI: osteogenesis imperfecta

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Original Paper

Evaluating Digital Program Support for the Physical Activity 4 Everyone (PA4E1) School Program: Mixed Methods Study

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Abstract

Background: Effectively scaled-up physical activity interventions are urgently needed to address the high prevalence of physical inactivity. To facilitate scale-up of an efficacious school-based physical activity program (Physical Activity 4 Everyone [PA4E1]), provision of implementation support to physical education (PE) teachers was adapted from face-to-face and paper-based delivery modes to partial delivery via a website. A lack of engagement (usage and subjective experience) with digital delivery modes, including websites, may in part explain the typical reduction in effectiveness of scaled-up interventions that use digital delivery modes. A process evaluation focused on the PA4E1 website was undertaken.

Objective: The 2 objectives were to (1) describe the usage of the PA4E1 program website by in-school champions (PE teachers leading the program within their schools) and PE teachers using quantitative methods; (2) examine the usage, subjective experience, and usability of the PA4E1 program website from the perspective of in-school champions using mixed methods.

Methods: The first objective used website usage data collected across all users (n=273) throughout the 9 school terms of the PA4E1 implementation support. The 4 usage measures were sessions, page views, average session duration, and downloads. Descriptive statistics were calculated and explored across the duration of the 26-month program. The second objective used mixed methods, triangulating data from the first objective with data from a think-aloud survey and usability test completed by in-school champions (n=13) at 12 months. Qualitative data were analyzed thematically alongside descriptive statistics from the quantitative data in a triangulation matrix, generating cross-cutting themes using the "following a thread" approach.

Results: For the first objective, in-school champions averaged 48.0 sessions per user, PE teachers 5.8 sessions. PE teacher sessions were of longer duration (10.5 vs 7.6 minutes) and included more page views (5.4 vs 3.4). The results from the mixed methods analysis for the second objective found 9 themes and 2 meta-themes. The first meta-theme indicated that the website was an acceptable and appropriate delivery mode, and usability of the website was high. The second meta-theme found that the website content was acceptable and appropriate, and identified specific suggestions for improvement.

Conclusions: Digital health interventions targeting physical activity often experience issues of lack of user engagement. By contrast, the findings from both the quantitative and mixed methods analyses indicate high usage and overall acceptability and appropriateness of the PA4E1 website to school teachers. The findings support the value of the website within a multidelivery mode implementation intervention to support schools to implement physical activity promoting practices. The analysis identified suggested intervention refinements, which may be adopted for future iterations and further scale-up of the PA4E1 program.

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Trial Registration: Australian New Zealand Clinical Trials Registry ACTRN12617000681358; https://www.anzctr.org.au/Trial/Registration/TrialReview.aspx?id=372870

(JMIR Pediatr Parent 2021;4(3):e26690) doi:10.2196/26690

KEYWORDS

process evaluation; engagement; think-aloud methodology; mixed methods; physical activity; website; digital health intervention; implementation support; delivery mode; scale-up

Introduction

Background

As much as 1 in 4 (25%) adults and 4 in 5 (80%) adolescents do not meet the global recommendations for aerobic exercise and are therefore at increased risk of noncommunicable diseases and premature mortality [1,2]. Scalable programs with proven effectiveness are urgently needed to increase population physical activity, including those in the school setting, as outlined in the Global Action Plan on Physical Activity 2018-2030 [3].

Many efficacious health interventions exist [4], but scaling-up efficacious interventions with proven health benefits is important to ensure the population health benefit of such interventions can be realized [5]. Digital delivery modes are often used to facilitate scale-up of efficacious interventions, as they have the potential to achieve considerable reach at relatively low cost compared with traditional modes of program delivery [4]. A recent systematic review of scaled-up trials of obesity-prevention interventions found that the main adaptations to interventions to facilitate their scale-up were changes in the modes of delivery [4]. Specifically, websites or other digital delivery modes were commonly added or replaced face-to-face components, for example, to provide program resources and training for teachers or clinicians in program delivery [4]. Research examining digital delivery modes such as websites, however, often report poor usage and engagement with such technologies [6-8].

A lack of engagement with digital technologies [4] may, in part, explain the modest impact of physical activity interventions that have relied on such technologies to support population-wide scale-up [4,6,9]. A recent meta-analysis of digital health interventions targeting physical activity found that higher usage engagement is associated with targeted behavior changes [10]. Perski et al [9] have conceptualized engagement with digital health interventions to include both amount, duration and depth of *usage*, and user *subjective experience*, characterized by attention, interest, and affect [9]. Systematic reviews of the human–computer interaction literature suggest that good usability (ie, functionality and efficiency of the digital application) and subjective experience engagement (ie, users attention, interest, and affect) are important drivers of reducing attrition and increasing usage of digital applications [9,11].

Process evaluations are recommended as part of comprehensive evaluations of all randomized trials to aid the interpretation of trial findings and to better understand and explain why an intervention has or has not worked [12]. Process evaluations may be particularly important as part of trials of interventions that have been scaled-up, given the limited research and insight scientists currently have regarding scale-up processes. However, detailed process evaluations are seldom undertaken. For example, a recent systematic review of school-based physical activity interventions found that just 4 of the 17 trials included in the review had conducted a process evaluation [13]. Further, of those undertaken in the secondary school setting, few have used mixed methods [14-17] and many have focused on a narrow range of quantitative process outcomes (eg, fidelity, reach, dose) [18-21]. Perhaps most importantly for those interested in the application of digital technologies to support scale-up, none of the trials identified in the review [4], or in reviews of the scale-up literature generally, have undertaken a process evaluation examining the key dimensions of subjective experience and usage of digital components employed to deliver implementation support strategies.

Given the lack of process evaluations focused on the digital delivery mode of interventions utilizing multiple delivery modes, this study sought to address this evidence gap by conducting a process evaluation of the digital delivery mode used to support scale-up of an effective health intervention. This study addresses the scale-up of Physical Activity 4 Everyone (PA4E1), a secondary school–based physical activity program that included a website to support the delivery of implementation support strategies to school teachers (physical education [PE] teachers and in-school champions [PE teachers leading the program within their schools]). The objectives of this study are below, followed by an overview of the PA4E1 program and evaluation trials, which provide more detailed context for this study.

1. To describe the usage of the PA4E1 program website by in-school champions and PE teachers using quantitative methods.
2. Examine the usage, subjective experience, and usability of the PA4E1 program website from the perspective of in-school champions using mixed methods.

Physical Activity 4 Everyone (PA4E1)

School practices, programs, and policies can support adolescents to be physically active and are recommended by the World Health Organization and governments internationally through whole-school approaches [3,22-24]. One school program that has been shown to assist schools (PE teachers, principals) to support their students to become more physically active is PA4E1 [25-27].

PA4E1 Prescale: Efficacy Trial

PA4E1 was first trialed from 2012 to 2014 as a 2-year randomized controlled efficacy trial in low-socioeconomic Australian secondary schools [25]. PA4E1 had positive effects on student physical activity and unhealthy weight gain [25-31].

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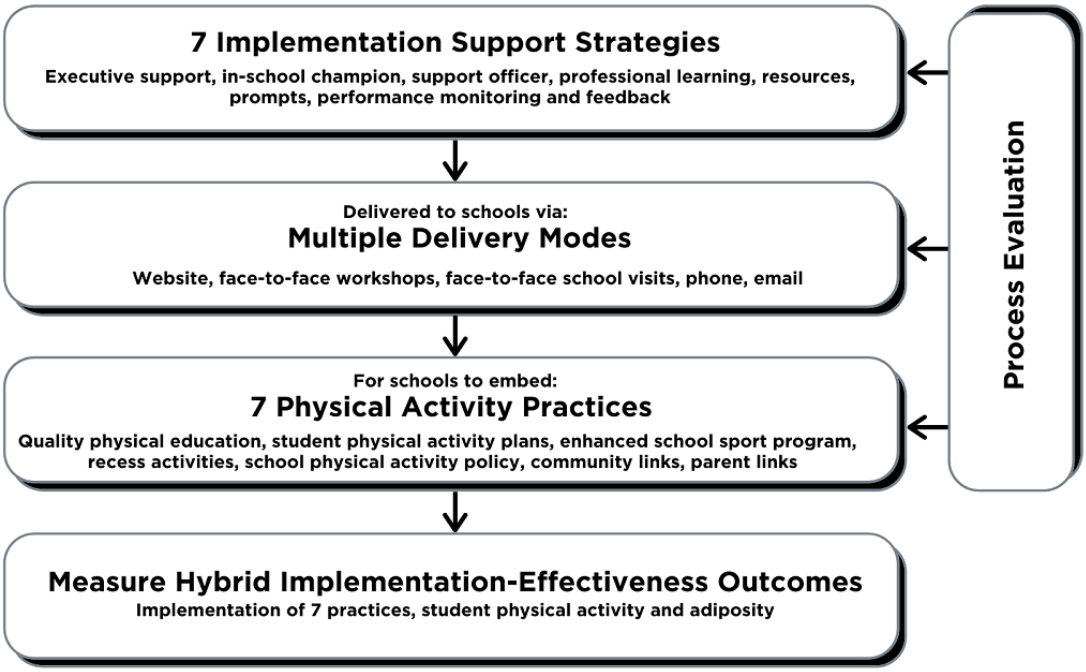
The PA4E1 program consisted of 7 physical activity practices, and 6 implementation support strategies.

PA4E1 Postscale (This Study): Type III Hybrid Implementation–Effectiveness Trial

The program was scaled-up for delivery in more schools across a larger geographic area, utilizing a website optimized for desktop, mobile, and tablet devices, to support the delivery of implementation support strategies to schools. The PA4E1 scale-up trial was a type III hybrid implementation–effectiveness cluster randomized controlled trial [29]. The trial involved 49 schools, 24 allocated to the program (intervention) group. An outline of the logic for this trial is shown in Figure 1. Program schools were offered 7 implementation support strategies to support their adoption of 7 physical activity practices, supporting school students to become more physically active. Details of the physical activity practices and implementation support strategies (as well as additional information on the timing of the implementation support strategies) are provided in Multimedia Appendix 1. Adaptations for scale-up were made to both the physical activity practices and the implementation support strategies, as detailed elsewhere [29]. The main change relevant to this study was the introduction of a digital delivery mode, a website, for the provision of program implementation support. Multimedia Appendix 1 (Table S1) shows how the website was used within the 7 implementation support strategies (n=23 substrategies). To summarize, a password-protected program website replaced face-to-face and paper-based delivery modes to part-deliver teacher professional learning to all PE teachers in participating schools; provide program resources for in-school champions and PE teachers; prompt in-school champions and PE teachers to implement PA4E1; monitor schools' performance on meeting practice implementation milestones in each school term via an in-school champion–completed termly survey; and provide feedback to school stakeholders (in-school champions, principals) based on their termly survey results [25,29].

The primary trial outcome was uptake of physical activity practices by schools at study midpoint (12 months) and 24 months. At 12 months the trial's primary outcome (proportion of schools adopting at least four of the seven physical activity practices) was significant, with more schools implementing 4 of the 7 practices in the program group (16/24, 67%) than in the control group (1/25, 4%; $P<.001$) [32]. Further process evaluation outcomes will be reported elsewhere, in line with the process evaluation protocol [28].

Figure 1. The PA4E1 scale-up trial logic model, expanded from [28]. PA4E1: Physical Activity 4 Everyone.



Methods

The methods are reported by objective.

Objective 1: Describe the Usage of the PA4E1 Program Website by In-School Champions and PE Teachers Using Quantitative Methods

Sampling

Usage data were tracked for the entire study period (October 2017 to December 2019; 9 school terms) for all users of the PA4E1 website who had a registered account with the program website, including in-school champions, PE teachers, principals, school administration staff, and nonschool staff (support officers and the PA4E1 research team). At the start of the program, all users were provided a registration link via email to register for the website. During the program, new school staff were provided a link to register for the website.

Data Collection

Usage data were collected via Google Analytics throughout the 9 terms of the program (26 months, October 9, 2017, to

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December 20, 2019). Prior to March 29, 2018 (term 3), data could not be analyzed by website user type; only overall usage could be captured. In order to further understand the different types of users, a "custom view" was applied in Google Analytics from this date to prospectively segregate different school user types (ie, in-school champions, PE teachers).

Measures

The post hoc analysis method described by the Analyzing and Measuring Usage and Engagement Data (AMUsED) framework [33] was used to guide the selection of usage variables to report (Multimedia Appendix 2). Google Analytics was used to track 4 variables for the entire study period (Textbox 1).

Textbox 1. Variables tracked using Google Analytics.

Sessions

Similar to a "login," sessions are defined as a group of user interactions (eg, page views, downloads) that occur within a single period, specifically a session ends after 30 minutes of inactivity or if users leave and revisit the website. Sessions were chosen as a better measure than "logins" for the PA4E1 website as users remained logged in for a rolling period of 30 days, whereby users were only logged out following 30 days of inactivity.

Page Views

Page view is a count of total visits to each page of the website. If a user clicks reload after reaching the page, this was counted as an additional page view. If a user navigated to a different page and then returned to the original page, a second page view was recorded.

Average Session Duration

The length of time of a session from the first click to the last, excluding the inactive period of a session immediately following the last click.

Downloads

A count of the total number of downloads of resources from the resources page of the website. Users had to click onto the particular resource to be counted as a download.

Data Analysis

Data were first downloaded from Google Analytics for the entire study period. Descriptive statistics were produced in SAS software [34] for all users for the entire study period and for each type of user from March 29, 2018. Mean and SD were calculated for each measure.

Objective 2: Examine the Usage, Subjective Experience, and Usability of the PA4E1 Program Website From the Perspective of In-School Champions Using Mixed Methods

Sampling

All in-school champions (n=24) were invited to complete a "think-aloud" survey and usability test via email after 4 school terms of website use (November 2018) [35]. Participants could complete the survey anytime through until May 2019 (Term 6). All in-school champions were emailed a study information letter and informed that completion of the survey acted as consent to the study. Completion of the survey and usability test was expected to take 25-30 minutes based on piloting. Participants were provided with an AUD $30 (US $23) e-gift card reimbursement for completing the survey.

Data Collection

The think-aloud survey and usability test was conducted remotely via in-school champions' own digital devices (ie, in-school champions' own mobile, laptop, or computers). Loop11 user-testing software was used to conduct the think-aloud survey and usability test [36]. As Figure 2 shows, Loop11 displays a set of questions and tasks imposed upon the website [36]. Throughout the think-aloud survey and usability test, participants were prompted by the Loop 11 platform to verbalize their thoughts while responding online to a series of questions (survey) and tasks (usability test) (Multimedia Appendix 3). The researchers were not present during the study, and participants were invited to complete the study in their own time using a link provided to them via email. This method was chosen to increase the real-world relevance of the findings, as the participants complete the activities in their own setting [9]. As well as the quantitative responses from the survey questions and usability tasks, a video screen capture and microphone audio data were also collected concurrently, and participants were frequently encouraged to explain their responses by "thinking aloud." A practice question was designed to give participants the opportunity to familiarize themselves with the format of the Loop11 user testing platform and to practice thinking aloud.

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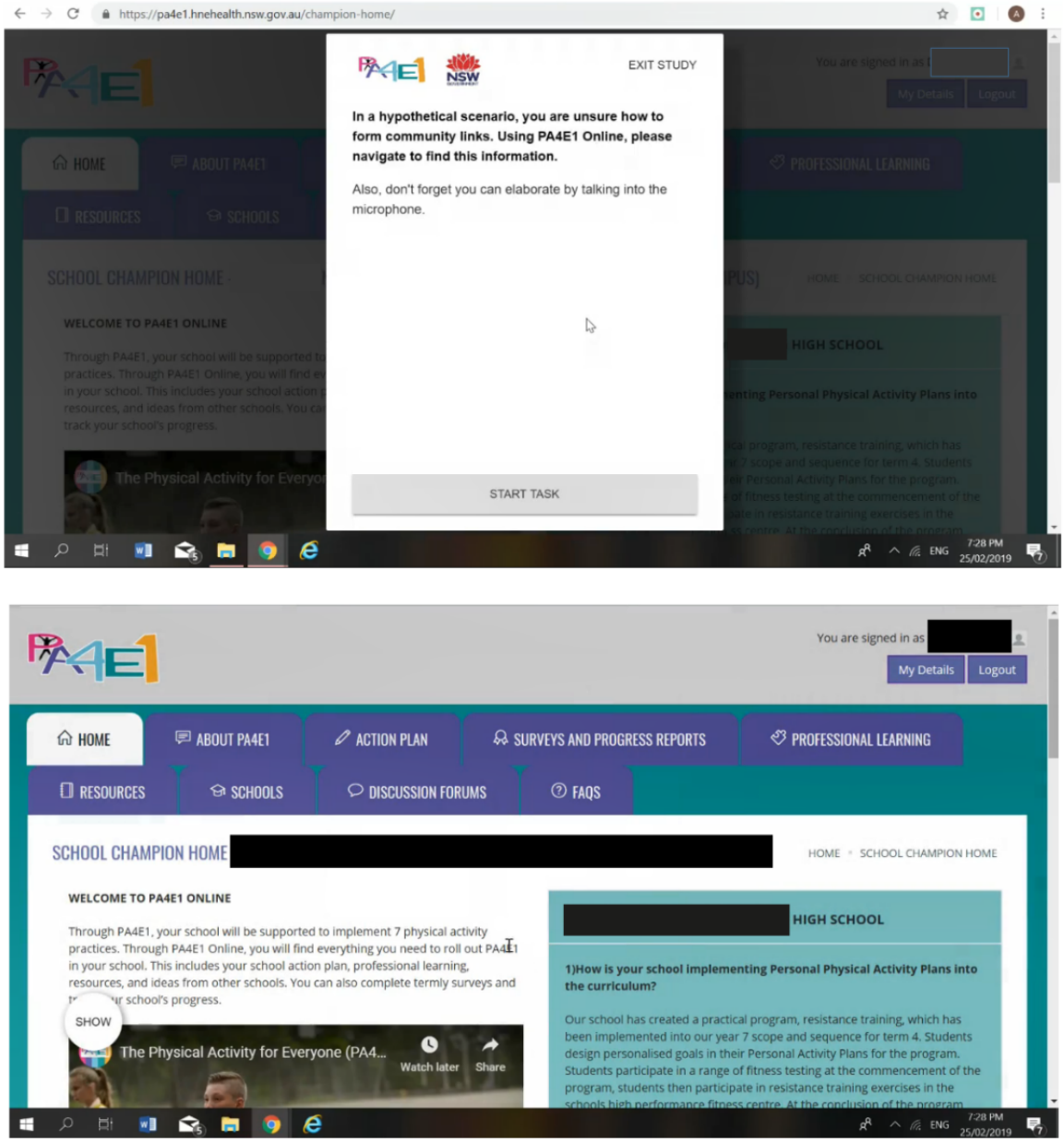
Figure 2. Two screenshots of a participant completing the think-aloud survey and usability test, with the survey imposed on the program website.



Measures

Figure 2 shows a screenshot of the think-aloud survey and usability test which is imposed upon the program website. The survey contained 4 validated tools [37-39]. All tools were on a 5-point Likert-scale (1=strongly disagree, 2=disagree, 3=neutral, 4=agree, and 5=strongly agree). The Systems Usability Scale is a 10-item scale to assess usability of the website (scores range 0-100) [37] and comparable data exist to interpret the score derived from the Systems Usability Scale [40]. The second and third tools were the Acceptability of Intervention [39] and Intervention Appropriateness Measure [39], to assess the acceptability and appropriateness of the website, respectively (scores range 0-5). The fourth tool was the long form of the User Engagement Scale [38], which is a 31-item tool split into 4 subscales (Aesthetic appeal; Focused attention; Perceived usability; and Reward; scores range 0-5 for each subscale and overall).

The usability test contained 3 tasks. The average time to task completion and success rate of task completion were recorded within Loop11. Tasks asked participants to navigate to certain sections of the website. The first task asked participants to navigate to the discussion forum, the second to find information on forming community links (see Practice 7 in Table S2 of Multimedia Appendix 1) and the final task asked participants where they would post a lesson observation (see Practice 1 in Table S2 of Multimedia Appendix 1). Participants were encouraged to verbalize their thoughts (think aloud) throughout the 3 tasks.

The survey also contained 12 prompting questions which asked participants to think aloud to verbalize their responses (see Multimedia Appendix 3 for a full list of these questions). The

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questions asked participants to explain their thoughts toward a particular hypothetical scenario or comment on a particular aspect of the website (eg, usefulness of the website if PA4E1 were to be rolled out state-wide). The questions were used to elicit additional responses from participants. Concurrently, audio data and a video of the participants screen (screen capture) were recorded.

Additional administration data were used to identify usage of other delivery modes for implementation support strategies (Figure 1), including attendance at face-to-face events and contact logs between in-school champions and support officers (data sources have been described in the process evaluation protocol [28]).

Mixed Method Data Analysis

A mixed methods data analysis was informed by Perski et al's [9] conceptualization of digital behavior change intervention engagement framework, whereby *delivery* and *content* were considered separately. *Delivery* focuses on themes that emerge relating to the aesthetics/design, challenge, complexity, control features, credibility features, ease of use, familiarity, guidance, interactivity, message tone, mode of delivery, novelty, narrative, personalization, and professional support features of the digital intervention. *Content* focuses on themes that emerge relating to the behavior change techniques (eg, feedback, goal setting, reminders, rewards, self-monitoring, social support features). Themes relating to delivery and content were extracted separately.

All quantitative data were downloaded and analyzed in MS Excel. The quantitative data from the validated tools were scored according to their instructions [37-39] and summary descriptive statistics were produced (mean score [SD]). Task completions were calculated as percentage of successful completions compared with total attempts. Time taken to complete tasks were recorded from the initial click to the final successful click by the participant. Mean time and SD were calculated across all in-school champions for each task.

Audio data were transcribed verbatim and analyzed using inductive thematic analysis [41]. Screen recording data were also downloaded (video) and analyzed concurrently with the audio data, within Nvivo 12 (QSR International) [42], to provide additional context to the audio data (eg, a participant's cursor location within website).

The quantitative and qualitative data were mixed during analysis using a triangulation matrix. Both data sets were given equal emphasis to address the objective. The descriptive statistics were produced at the same time as the initial codes of the qualitative data. As per the process evaluation protocol [28], the "following the thread" approach was employed to generate hypothesis and questions of the qualitative data from the quantitative data, and vice versa. The findings were combined and compared (triangulated) using a triangulation matrix to assess where findings from one method agreed or partially agreed (convergence), appear to contradict each other (discrepancy or dissonance), or are silent (ie, a theme arises in 1 data set but not in another). The matrix allowed the production of themes and meta-themes (higher-level themes) that combined and compared the qualitative and quantitative data sets [41]. Throughout the entire analysis, MM kept a research journal.

The iterative phases of the mixed method analysis [41] included:

- Familiarizing with the data (both audio and screen capture as well as quantitative data).
- Generating initial codes and following the thread (MM produced descriptive statistics, followed a thread, and developed initial codes of the transcriptions using Nvivo 12 [42]).
- Searching for themes (MM reviewed each code and ordered them under headings, before discussing the ordered codes with JD to produce an initial triangulation matrix).
- Reviewing themes (EC, TM, and RS reviewed the themes, participant quotes, and matrix labels—resolving disagreements through discussion with MM and JD).
- Defining and naming themes (all authors discussed and agreed upon final themes).
- Producing the report.

Ethical Approval

The trial was prospectively registered (ACTRN12617000681358) and approved by the Hunter New England Research Ethics Committee (Ref No. 11/03/16/4.05), University of Newcastle (Ref No. H-2011-0210), NSW Department of Education (SERAP 2011111), Maitland Newcastle Catholic School Diocese, Broken Bay Catholic School Diocese, Lismore Catholic School Diocese, Armidale Catholic School Diocese, and the Aboriginal Health and Medical Research Council.

Availability of Data and Materials

The full data set supporting the conclusions of this article containing data not already included within the article or its additional files is available from the corresponding author on reasonable request.

Results

The results are reported below by objective.

Objective 1: Describe the Usage of the PA4E1 Program Website by In-School Champions and PE Teachers Using Quantitative Methods

There were a total of 273 users of the PA4E1 website during the whole study period. School users of the website were in-school champions (n=30) and PE teachers (n=198). Few principals (n=7) or school administration staff (n=2) registered for the program. Additionally, there were also nonschool users (n=20; ie, support officers and the PA4E1 research team) and unidentified users (n=16) of the program website.

Table 1 provides a breakdown of website usage for in-school champions and PE teachers from March 28, 2018 (term 3) to December 20, 2019 (term 9) (when the data were available by user) and for all program users from October 9, 2017, to December 20, 2019 (entire study period). In-school champions were the most frequent users of the program, with a mean of 48.0 sessions per user compared with 5.8 sessions per user by PE teachers. They also had higher total page views (276.6) and

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downloads (33.6) per user than PE teachers. PE teachers had fewer sessions (5.8), but these were of longer duration (10.5 vs 7.6 minutes) and included more page views on average (5.4 vs 3.4).

Table 1. Summary of Physical Activity 4 Everyone (PA4E1) website usage by user type.

Usage	In-school champions (n=30)[a]	Physical education teachers (n=198)[a]	All users (n=273)[b]
Sessions per user, mean	48.0	5.8	22.8
Page views per user, mean	276.6	57.0	179.4
Page views per session, mean	3.4	5.4	7.9
Session duration (minutes), mean	7.6	10.5	8.6
Downloads per user, mean	33.6	2.3	—

[a]Data were not available for the start of the program (October 9, 2017, to March 28, 2018). Data presented are from March 29, 2018, to December 20, 2019.

[b]The "All users" group includes in-school champions (n=30), PE teachers (n=198), principals (n=7), school administration staff (n=2) as well as nonschool users (n=20) and unidentified users (n=16).

The most frequently viewed pages by in-school champions were the home page (18.4% of all views), surveys and progress reports (15%), professional learning (10.3%), and resources (7.3%). PE teachers most frequently viewed professional learning (19.7% of all views) and the home page (9.6%).

A total of 90 different resources were downloaded from the website at least once by either an in-school champion or a PE teacher, equating to a total of 1007 downloads. The top 20 most downloaded resources accounted for more than half of all total downloads (n=559). Among the top downloaded resources were those directly assisting a particular implementation support strategy (Figure 1; see Table S1 in Multimedia Appendix 1) or physical activity practice (Figure 1; see Table S1 in Multimedia Appendix 1), which included a template for lesson observation (Practice 1), a student physical activity plan template (Practice 2), newsletter snippets (Practice 6), partnership agreement (Practice 7), and a school physical activity policy template (Practice 5).

The usage over time was explored by looking at the termly number of sessions, page views per session, average session duration, and number of downloads. Figure 3 shows these data per month for both in-school champions and PE teachers.

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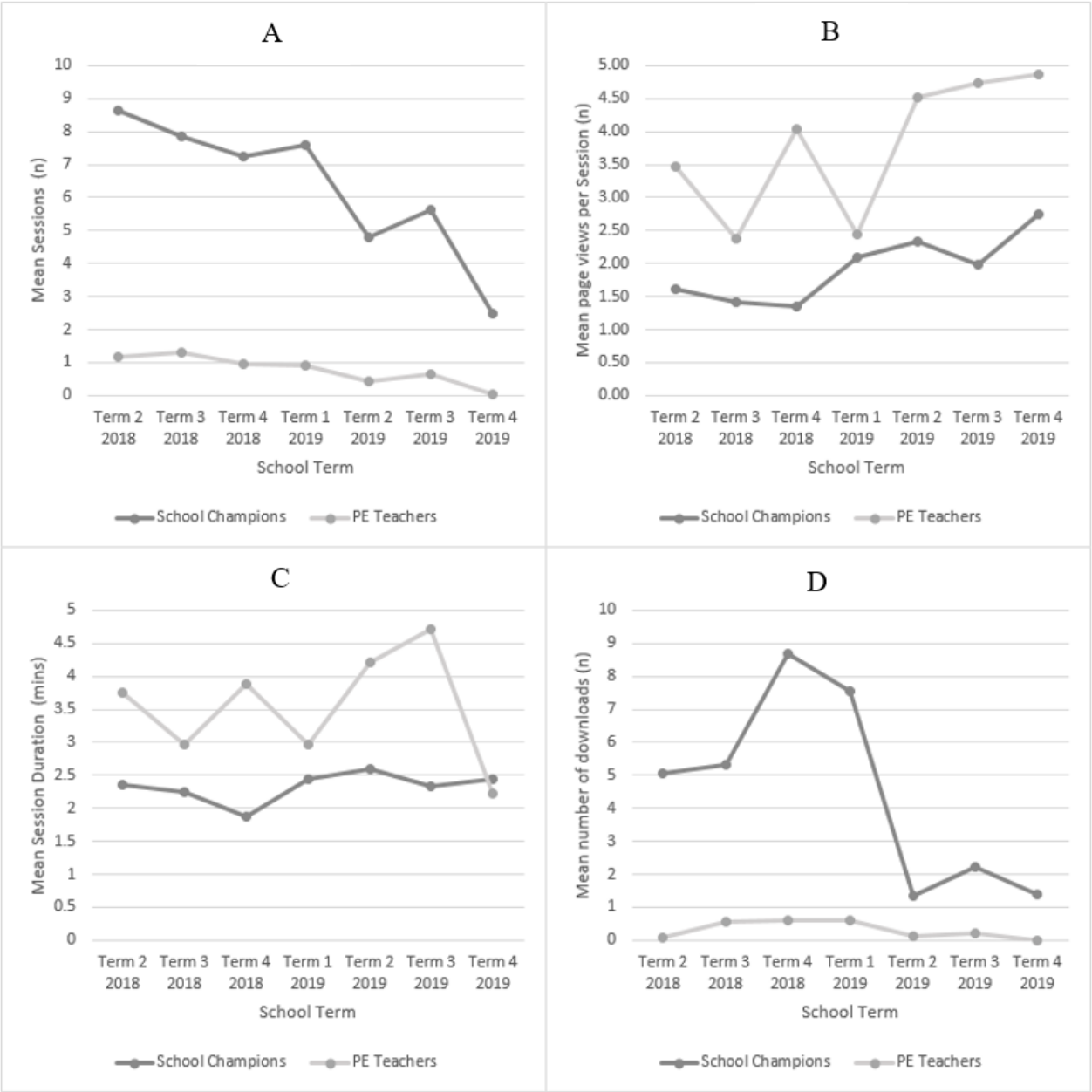
Figure 3. (A) Mean number of sessions, per term. (B) Mean number of page views per session, per term. (C) Mean session duration, per term. (D) Mean number of downloads, per term. Note that Term 4, 2017, and Term 1, 2018, data are not available (as described in the "Methods" section). PE: physical education.



Objective 2: Examine the Usage, Subjective Experience, and Usability of the PA4E1 Program Website From the Perspective of In-School Champions Using Mixed Methods

Of the 24 in-school champions invited to complete the think-aloud survey and usability test, 13 participated (54%). The average time to complete the survey and usability test was 20 minutes. In-school champions (9 females and 4 males) represented schools located in major cities (n=6), inner regional (n=6), and outer regional (n=1) regions [43]. Seven of the in-school champions also had responsibility for leading the health and PE programs in their school. All schools (9 government and 4 Catholic) were in the lower 50% of the Socio-Economic Indexes for Areas (SEIFA) of Relative Socioeconomic Disadvantage (suburb in lower 50% of NSW) [44]. School enrollments ranged from 348 to 1316 (mean 900 [SD 246]). Six schools had 10% or more Indigenous student enrollment [45]. Table 2 summarizes the meta-themes and themes for *delivery* and *content*, respectively. Multimedia Appendix 4 includes the full triangulation matrix table and convergence labels.

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Table 2. Summary of delivery and content meta-themes and themes.

Themes	Theme explained
Delivery	
Meta-theme	The website was an acceptable and appropriate delivery mode. Usability of the website was high.
Theme 1	The website complemented other delivery modes, but in-school champions preferred the other delivery modes (support officer and face-to-face).
Theme 2	The website was used as a "utility" delivery mode (ie, used only when required, not as the first delivery mode of choice).
Theme 3	If the program were delivered statewide, the website would be useful, though embedding within other systems that teachers already used may be helpful.
Theme 4	Mixed reactions toward the possible addition of a website chat feature with a support officer available to video call during business hours.
Theme 5	The discussion forum was not used, but in-school champions reported that it could be potentially useful, perhaps if delivered differently.
Content	
Meta-theme	The website content was generally acceptable and appropriate, with a few specific suggestions for improvement.
Theme 1	A lack of notifications (or prompts) to highlight new things in the website reduced return traffic to the website.
Theme 2	The lesson observation form (a resource for Practice 1) was difficult to find on the website, it was difficult to track lesson observation completion, and some usability issues with completing out of internet range.
Theme 3	Termly surveys (Support Strategy 7) were generally acceptable and completed.
Theme 4	The resources were acceptable and downloaded frequently.

Delivery

Meta-Theme: The Website Was an Acceptable and Appropriate Delivery Mode. Usability of the Website Was High (Triangulation Convergence Label: Agree)

Quantitative data indicated in-school champions agreed or strongly agreed that the PA4E1 website was acceptable and appropriate. For in-school champions (n=13), the mean acceptability of the intervention measure was 4.52 (SD 0.04) out of 5, and the mean Intervention Appropriateness Measure was 4.46 (SD 0.0) out of 5, both indicating agreement that the website is acceptable and appropriate. Additionally, the mean in-school champions' (n=13) overall User Engagement Scale score was 3.4 (SD 0.7) out of 5 [32]. The mean individual dimension scores from the User Engagement Scale were 4.2 (SD 0.1), 3.7 (SD 0.5), 3.4 (SD 0.3), and 2.4 (SD 0.3) for aesthetic appeal, reward factor, perceived usability, and focused attention, respectively.

In-school champions found the usability of the website to be "good." The overall in-school champion (n=13) Systems Usability Scale score was 77.7, which corresponds to a "good" website within the "acceptable" range of the Systems Usability Scale [31,37]. Additionally, the majority of the 3 navigation tasks were completed successfully by in-school champions (74% [29/39] of all tasks completed successfully). The average time to task completion was 71 seconds per task. The discussion forum and community links tasks had higher success rates (11/13 [85%] and 12/13 [92%], respectively) than the lesson observation task (6/13, 46%). Time to complete each task was 91, 66, and 55 seconds for discussion forum, community links, and lesson observation tasks, respectively. No tasks were abandoned.

Qualitative data also indicated good acceptability, appropriateness, and usability of the website delivery mode. Some example quotes are as follows:

> *...it's set out very clearly, easy to use, easy to access, and is updated relatively frequently.*
>
> *So, we all know that teachers are the time poor people of the world. So definitely having an online mode is very, very suitable.*

Theme 1: The Website Complemented Other Delivery Modes, But In-School Champions Preferred the Other Delivery Modes (Support Officer and Face-to-Face; Triangulation Convergence Label: Agree)

Quantitative data showed schools utilized the website (Figure 1). In-school champions also relied on the other delivery modes, for example, contact logs between support officers and in-school champions indicated that 18/24 schools received a face-to-face contact with their support officer at least once a term in the first 12 months. In the second 12 months, this dropped to 8/24 schools, though the criteria for this implementation support strategy were different for the second 12 months (Multimedia Appendix 1). However, weekly emails or phone calls between support officers and in-school champions occurred in 16/24 schools during the first 12 months, but this increased to 22/24 schools in the second 12 months. Additionally, the 2 face-to-face workshops were well attended; 23/24 in-school champions attended the first workshop at the start of the program while 22/24 attended the second workshop at the midpoint of the program.

Qualitative data indicated that in-school champions used the website to complement interactions with their support officer. The website often was not the first point of call, and it may not

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be able to fully replace face-to-face and direct contact methods from the support officers. The website may be particularly useful for after-hours support (eg, on weekends and evenings). Example quotes:

> *You've obviously got your Support Officer as well [as the website], but it's good just to have this, this portal [website].*
>
> *Yes [I like the website], but it needs to be accompanied by other things like face-to-face as well.*
>
> *...it is an invaluable experience to meet, at a common place, where you can actually roll out, the initial delivery, and training, for the first time that people are running PA4E1. However, as said before, online delivery should remain part of this program.*
>
> *...teachers are often not being able to get this extra, sort of, work, or professional learning done during their actual hours of being at school. So I often access the website from home, after hours, on the weekend.*

Theme 2: The Website Was Used as a "Utility" Delivery Mode (ie, Used Only When Required, Not as the First Delivery Mode of Choice; Triangulation Convergence Label: Agree)

Quantitative data from objective 1 indicated that in-school champions used the website on average every fortnight, corresponding to 6.3 sessions per term (SD 2.0). Qualitative data identified that the website was perhaps not used to its full potential, but instead it was used for its core functions (eg, termly surveys, professional learning), rather than to peruse additional ideas and resources. Example quotes:

> *To be honest I don't think I've used PA4E1 online [the website], probably to its full potential.*
>
> *Yeah well obviously I haven't really used this [pointing cursor at the resources section] as much as I should've. I have used some of these [resources] though...*
>
> *I'm aware of most kind of, components of it [the website], but there are things I'm not [aware of]...*

Theme 3: If the Program Were Delivered Statewide, the Website Would Be Useful, Though Embedding Within Other Systems That Teachers Already Used May Be Helpful (Triangulation Convergence Label: Slightly Agree)

All in-school champions (13/13) agreed that online delivery of the PA4E1 program should remain. The majority of in-school champions (12/13) suggested the PA4E1 website would be a suitable mode of delivery in its current form should the PA4E1 program be rolled out to schools statewide.

There were conflicting suggestions emerging from the qualitative data. In-school champions indicated that while a website would be useful for statewide delivery of the program, it could become another platform that teachers are asked to use, so it would be better embedding any roll out within existing platforms. Example quotes:

> *...it is another platform that we need to access in order to implement our teaching.*
>
> *I do think it is quite suitable, to roll out in its current form if there was to be no changes.*
>
> *The more people that are on it [the website] as well, I think the more discussion that would kind of, be generated....*

Theme 4: Mixed Reactions Toward the Possible Addition of a Website Chat Feature With a Support Officer Available to Video Call During Business Hours (Triangulation Convergence Label: Slightly Agree)

Quantitative data were generally supportive; the majority of in-school champions (11/13) liked the idea that support officers would be available during working hours for support within the PA4E1 website via video, audio, or text chat features. Qualitative data were mixed, with some supporting the feature, while some preferring traditional contact methods such as phone call and face-to-face. Some example quotes:

> *Supportive: it's [a] great [idea], because whenever you can squeeze it in in your free period there's someone there to talk to you about so you don't have to wait until the [in-School] Champion or someone in the know is available to help you…that's fantastic.*
>
> *Neutral: I mean it might ease the load on the Support Officers having to kind of travel everywhere [around the NSW state], 'cause then you have that option to sit and talk to them like you would face-to-face. But yeah I mean I like it, but I don't dislike it, but I don't know if it's hugely necessary.*
>
> *Nonsupportive: I dislike because I would prefer face-to-face. Email works just as well, and a phone call if necessary.*

Theme 5: The Discussion Forum Wasn't Used, But In-School Champions Reported It Could Be Potentially Useful, Perhaps If Delivered Differently (Triangulation Convergence Label: Agree)

Quantitative usage data showed that use of the discussion forum was very low. There were 3 discussion forum posts made by support officers to generate content; however, there were 0 discussion forum posts by in-school champions or PE teachers during the entire program. In response, the support officers created a Facebook group for in-school champions which was established on November 7, 2018. From the launch of the Facebook group until the end of the program, there were 10 posts by the PA4E1 team (support officers and staff) and 12 posts by in-school champions in the Facebook group.

Qualitative data suggested the discussion forum was not very useful, and may be better delivered within the website, or could be better delivered using Facebook. Example quotes:

> *Yep. It's [the discussion forum]...a little bit limited.*
>
> *I think this was mentioned at the training day, the suggestion that a discussion forum could pop up on the front page...so, it was the, kind of, first thing you*

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saw, and the first thing that popped up, whenever you went there.

I don't think that the discussion forum is a good mode of communication. It appears that no one's really using it. Perhaps a different form of social media such as Facebook, which is happening, that more people use.

Content

Meta-Theme: The Website Content Was Generally Acceptable and Appropriate, With a Few Specific Suggestions for Improvement (Triangulation Convergence Label: Agree)

As per quantitative data from the previous meta-theme relating to *delivery*, the majority of in-school champions agreed or strongly agreed that the website was acceptable, appropriate, and had good usability.

Qualitative data also indicated the website content was appropriate. Example quotes:

Yeah, I think the online portal in its current form is kind of suitable, I think a few minor changes and adjustments would make it better.

...the online portal is essential with implementation of the program. You couldn't do it without it. It's your go-to resource.

Theme 1: A Lack of Notifications (or Prompts) to Highlight New Things in the Website Reduced Return Traffic to the Website (Triangulation Convergence Label: Silence)

No quantitative data were available for this theme. Qualitative data highlighted that notifications would have been useful to highlight updates to the website, which may subsequently increase usage.

...sometimes new things just pop up that I didn't really realize were there

Maybe also having some form of like, notification so in the top right hand corner...so that you know, like, you can see new...things that are kind of happening...

...the portal [website] is not something I log onto everyday. As we all kind of said, like, everyone goes onto Facebook and things like that. So, that's where-and you get notifications that pops up on your phone, where often I don't know that things have been posted in the discussion forum, until I come back on, and often that's quite a while after they were posted.

Theme 2: The Lesson Observation Form (a Resource for Practice 1) Was Difficult to Find on the Website, It Was Difficult to Track Lesson Observation Completion, and Some Usability Issues With Completing Out Of Internet Range (Triangulation Convergence Label: Agree)

More than half of in-school champions (7/13) were unable to find the lesson observation section on the website. Qualitative data also indicated that in-school champions had difficulty finding the lesson observations, tracking PE teachers' completion of these observations, and also some issues with signal to the website while out of the internet range. Example quotes:

You do have to scroll down a little bit on the page [to find the Lesson Observation Form], and it is like, quite a small link, which I know some staff at our school have had trouble, kind of, finding it.

...you just kind of have to do a bit of a tally of how many observations one teacher has had.

Sometimes when we were out of mobile range or if this person didn't have a mobile, it didn't quite, ah, work out.

Theme 3: Termly Surveys (Support Strategy 7) Were Generally Acceptable and Completed (Triangulation Convergence Label: Slightly Agree)

From 0 to 12 months, 24/24 in-school champions completed all termly surveys and received the feedback reports from these surveys. From 12 to 24 months, 21/24 in-school champions completed all termly surveys and received feedback reports (3 schools missed 1 survey each). Few principals received the feedback reports, 7/24 principals had registered accounts with the program website and therefore received the feedback reports via email. The remaining principals did not directly receive the feedback reports, but may have been shown to them by their in-school champions. Qualitative data were scarce, but some support for the acceptability of the termly surveys was provided. Example quote:

...the termly surveys, I mean this was handy to see where we were at but I suppose before I did the survey I kind of had an idea of what we did and didn't do well.

Theme 4: The Resources Were Acceptable and Downloaded Frequently (Triangulation Convergence Label: Agree)

A total of 90 different resources were downloaded from the website at least once by either an in-school champion or a PE teacher, equating to a total of 1007 downloads. Example quotes:

I really love this particular resource section. Yeah, look, I just can't say enough about, you know, what I love about it.

I loved the graphics and the images. I've really, I really liked working with the Physical Activity Policy template. I thought it looked really professional, yeah, I- I did really like...those aspects.

Discussion

Principal Findings

We have previously reported that the multimode implementation support strategies used in the PA4E1 scale-up trial increased the implementation of physical activity–promoting practices in lower socioeconomic secondary schools in New South Wales, Australia [32]. The results presented in this paper expand on

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these findings with quantitative and mixed methods process evaluation data on the role of the PA4E1 website in delivery of the implementation support strategies of the PA4E1 program. To our knowledge, this is the first comprehensive process evaluation of a school-based physical activity program to focus exclusively on the digital delivery mode of a multidelivery mode implementation support strategy. The primary findings indicate that usage of the website by both the in-school champions and PE teachers was high, which aligns with the high fidelity and reach of the implementation support found at 12 months reported elsewhere [32]. As intended, in-school champions had more frequent website use than PE teachers. The results of the mixed methods analysis indicated that both the delivery and content of the website were acceptable and appropriate. A number of usability issues were identified and are included as suggested modifications for future iterations of the website.

The findings support the value of the website within a multidelivery mode implementation intervention to support schools to implement physical activity promoting practices. Although other digital health interventions targeting physical activity have often suffered with issues of engagement [6,10,46], the website delivering implementation support for the PA4E1 program does not appear to have impaired the potential of the program to have a positive impact. As discussed by Sebire et al [47], embracing technology in school physical activity interventions may be an effective way to efficiently deliver content, for example, by reducing challenges related to limited time for training.

Completing this study about website engagement as a component of our broader process evaluation has provided information not typically collected within trials of school-based physical activity programs. Previous studies have either omitted evaluation of the website delivery mode [48,49], or largely relied on quantitative website usage data to assess usage engagement [9,50-52]. These studies have reported varying levels of website usage, with studies' usage engagement varying between pilot and full trial [52,53]. Focusing only on usage data ignores the other component of engagement, subjective experience [9]. This study includes both quantitative and qualitative data exploring both usage and subjective experience engagement [9], with both data sets being triangulated to report high acceptability and appropriateness of the delivery and content of the website. Usage data showed that in-school champions and PE teachers accessed the website frequently, though this appeared to decline over time (Figure 3). As intended by the design of the PA4E1 implementation support strategies (Multimedia Appendix 1), in-school champions accessed the website more frequently than PE teachers. Additionally, compared with other websites, the PA4E1 website has a rating of "good" usability [37,40].

The study was novel in its use of "think-aloud" methodology to explore teachers' experiences of using a website to support the delivery of a health-based intervention. Think-aloud methodologies have largely been used to inform the development of websites and apps for use directly with the target user of the health intervention [54-58] (eg, an app to support weight management among adults with diabetes [54]). By contrast, we used a think-aloud methodology with teachers (in-school champions) who have been accessing a website to support the delivery of the PA4E1 program to adolescent students. Previous studies using the think-aloud methodology [54-58] have found it to be useful for identifying usability and subjective experience issues. However, the procedure used in these studies involved the presence of a researcher, which may have affected participants' reactions. Our think-aloud procedure allowed in-school champions to respond to the think-aloud survey and usability test remotely, using their own devices, in their own time. Such a method is likely to be more ecologically valid than those involving the presence of researchers, at research sites, using research devices [59].

Intervention Refinements

This process evaluation revealed several refinements and suggestions for the PA4E1 website. In-school champions suggested the website was used in conjunction with other delivery modes (face-to-face, phone, email) and that these delivery modes of the implementation support strategies were also highly valued, which was additionally supported by usage data of multiple delivery modes. In addition, while the program website was acceptable and appropriate in its current format, it was often not the "first-choice" for in-school champions; often, in-school champions would use other delivery modes first. In-school champions also suggested the website could be embedded within other systems that teachers already access, such as state education portals. Therefore, we suggest future iterations of the PA4E1 program should carefully consider the balance of delivery modes used to deliver the implementation support strategies.

In-school champions suggested that both a website chat feature (to access support officers) and a discussion forum (to chat with other in-school champions) may be useful, although this was not a unanimous suggestion. Strategies to increase engagement with such features may be required, which was further highlighted by in-school champions who suggested the addition of notifications within the website and via email to promote usage of the website and highlight new content. Other features that may improve engagement include social networking platforms [60], social support, and behavioral prompts [61]. Finally, the lesson observation form (see Practice 1 outlined in Multimedia Appendix 1, Table S2) was difficult to find on the website (task completion = 46% [6/13]) and in-school champions reported that this could be made more prominent within the website.

Future iterations of a website supporting PA4E1 delivery may benefit from user testing and refinement prior to rolling out the website for delivery [62]. Such formative evaluations are common to inform e-commerce websites and from the human–computer interaction literature [62].

Strengths and Limitations

A key strength of this study is the detailed mixed methods design, triangulating multiple data sources to provide a more coherent and actionable set of themes from the data. Another strength is the use of an ecologically valid remote think-aloud survey and usability test procedure, to collect in-school champions experiences of using the PA4E1 website.

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Limitations of this study include the low response rate to the think-aloud survey and usability test. Despite the remote nature of the survey, some respondents found it difficult to use the Loop 11 think-aloud survey and usability test software. Just over half of in-school champions (13/24, 54%) responded to the think-aloud survey and usability test. This may have introduced response bias, whereby those who responded were more likely to report high acceptability and appropriateness of the website. However, with regard to sample size, 5 participants are deemed sufficient for detecting most usability problems [63]. Another limitation was the inability to segregate user types prior to term 2, 2018. This was due to a technical difficulty in collecting the data in Google Analytics that could only be fixed prospectively from school term 2, 2018.

Conclusion

The results of this study provide context to support the primary outcome of the trial, highlighting that both in-school champions and PE teachers used the program website, and acceptability and appropriateness of the website were high among in-school champions. Our study has focused on the website delivery mode of a multidelivery mode implementation support strategy to support schools to implement physical activity promoting practices. We will publish further process evaluation results elsewhere, focusing on the remaining components of our process evaluation protocol [28].

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Authors' Contributions

MM and JD conceived the design of this study. RS, EC, JW, and LW conceived the design of Physical Activity 4 Everyone. MM and TM analyzed the quantitative data. MM and JD analyzed qualitative data and subsequently mixed the qualitative and quantitative data. MM led the writing of the manuscript. All authors contributed to multiple versions of the manuscript and gave final approval for it to be published.

Conflicts of Interest

None declared.

Multimedia Appendix 1
Supplementary file 1. Supplementary Table 1 and Supplementary Table 2.
[DOCX File , 30 KB - pediatrics_v4i3e26690_app1.docx]

Multimedia Appendix 2
Supplementary file 2. AMUsED framework.
[DOCX File , 46 KB - pediatrics_v4i3e26690_app2.docx]

Multimedia Appendix 3
Supplementary file 3. Think-aloud survey and usability test - questions and tasks.
[DOCX File , 23 KB - pediatrics_v4i3e26690_app3.docx]

Multimedia Appendix 4
Supplementary file 4. Triangulation matrix.
[DOCX File , 34 KB - pediatrics_v4i3e26690_app4.docx]

References

1. Guthold R, Stevens G, Riley L, Bull F. Worldwide trends in insufficient physical activity from 2001 to 2016: a pooled analysis of 358 population-based surveys with 1·9 million participants. Lancet Glob Health 2018 Oct;6(10):e1077-e1086 [FREE Full text] [doi: 10.1016/S2214-109X(18)30357-7] [Medline: 30193830]

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2. Guthold R, Stevens GA, Riley LM, Bull FC. Global trends in insufficient physical activity among adolescents: a pooled analysis of 298 population-based surveys with 1·6 million participants. Lancet Child Adolesc Health 2020 Jan;4(1):23-35 [FREE Full text] [doi: 10.1016/S2352-4642(19)30323-2] [Medline: 31761562]
3. Global Action Plan on Physical Activity 2018-2030: More Active People for a Healthier World. Geneva: World Health Organization; 2018. URL: https://apps.who.int/iris/bitstream/handle/10665/272722/9789241514187-eng.pdf [accessed 2021-07-20]
4. McCrabb S, Lane C, Hall A, Milat A, Bauman A, Sutherland R, et al. Scaling-up evidence-based obesity interventions: A systematic review assessing intervention adaptations and effectiveness and quantifying the scale-up penalty. Obes Rev 2019 Jul 13;20(7):964-982. [doi: 10.1111/obr.12845] [Medline: 30868745]
5. Katz D. School-based interventions for health promotion and weight control: not just waiting on the world to change. Annu Rev Public Health 2009 Apr;30(1):253-272. [doi: 10.1146/annurev.publhealth.031308.100307] [Medline: 19705560]
6. Davies CA, Spence JC, Vandelanotte C, Caperchione CM, Mummery W. Meta-analysis of internet-delivered interventions to increase physical activity levels. Int J Behav Nutr Phys Act 2012 Apr 30;9(1):52 [FREE Full text] [doi: 10.1186/1479-5868-9-52] [Medline: 22546283]
7. Romeo A, Edney S, Plotnikoff R, Curtis R, Ryan J, Sanders I, et al. Can Smartphone Apps Increase Physical Activity? Systematic Review and Meta-Analysis. J Med Internet Res 2019 Mar 19;21(3):e12053 [FREE Full text] [doi: 10.2196/12053] [Medline: 30888321]
8. Bort-Roig J, Gilson ND, Puig-Ribera A, Contreras RS, Trost SG. Measuring and influencing physical activity with smartphone technology: a systematic review. Sports Med 2014 May 5;44(5):671-686. [doi: 10.1007/s40279-014-0142-5] [Medline: 24497157]
9. Perski O, Blandford A, West R, Michie S. Conceptualising engagement with digital behaviour change interventions: a systematic review using principles from critical interpretive synthesis. Transl Behav Med 2017 Jun 13;7(2):254-267 [FREE Full text] [doi: 10.1007/s13142-016-0453-1] [Medline: 27966189]
10. Mclaughlin M, Delaney T, Hall A, Byaruhanga J, Mackie P, Grady A, et al. Associations Between Digital Health Intervention Engagement, Physical Activity, and Sedentary Behavior: Systematic Review and Meta-analysis. J Med Internet Res 2021 Feb 19;23(2):e23180 [FREE Full text] [doi: 10.2196/23180] [Medline: 33605897]
11. Maia CLB, Furtado ES. A systematic review about user experience evaluation. In: Marcus A, editor. Design, User Experience, and Usability: Design Thinking and Methods. DUXU 2016. Lecture Notes in Computer Science, vol 9746. Berlin, Germany: Springer; 2016:445-455.
12. Moore GF, Audrey S, Barker M, Bond L, Bonell C, Hardeman W, et al. Process evaluation of complex interventions: Medical Research Council guidance. BMJ 2015 Mar 19;350(mar19 6):h1258-h1258 [FREE Full text] [doi: 10.1136/bmj.h1258] [Medline: 25791983]
13. Love R, Adams J, van Sluijs EMF. Are school-based physical activity interventions effective and equitable? A meta-analysis of cluster randomized controlled trials with accelerometer-assessed activity. Obes Rev 2019 Jun 09;20(6):859-870 [FREE Full text] [doi: 10.1111/obr.12823] [Medline: 30628172]
14. Jong ST, Croxson CH, Guell C, Lawlor ER, Foubister C, Brown HE, et al. Adolescents' perspectives on a school-based physical activity intervention: A mixed method study. J Sport Health Sci 2020 Jan;9(1):28-40 [FREE Full text] [doi: 10.1016/j.jshs.2019.06.007] [Medline: 31921478]
15. Cosme Chavez R, Nam EW. Process Evaluation of a School-Based Program Aimed at Preventing Obesity in Adolescents from Lima and Callao, Peru. Int J Environ Res Public Health 2020 Jul 03;17(13):4804 [FREE Full text] [doi: 10.3390/ijerph17134804] [Medline: 32635324]
16. Sebire SJ, Banfield K, Jago R, Edwards MJ, Campbell R, Kipping R, et al. A process evaluation of the PLAN-A intervention (Peer-Led physical Activity iNtervention for Adolescent girls). BMC Public Health 2019 Sep 02;19(1):1203 [FREE Full text] [doi: 10.1186/s12889-019-7545-z] [Medline: 31477088]
17. Gorely T, Harrington DM, Bodicoat DH, Davies MJ, Khunti K, Sherar LB, et al. Process evaluation of the school-based Girls Active programme. BMC Public Health 2019 Aug 29;19(1):1187 [FREE Full text] [doi: 10.1186/s12889-019-7493-7] [Medline: 31464592]
18. Proctor E, Silmere H, Raghavan R, Hovmand P, Aarons G, Bunger A, et al. Outcomes for implementation research: conceptual distinctions, measurement challenges, and research agenda. Adm Policy Ment Health 2011 Mar 19;38(2):65-76 [FREE Full text] [doi: 10.1007/s10488-010-0319-7] [Medline: 20957426]
19. Gebremariam MK, Arah OA, Bergh IH, Andersen LF, Bjelland M, Grydeland M, et al. Factors affecting the dose of intervention received and the participant satisfaction in a school-based obesity prevention intervention. Prev Med Rep 2019 Sep;15:100906 [FREE Full text] [doi: 10.1016/j.pmedr.2019.100906] [Medline: 31194160]
20. Parrish A, Trost SG, Howard SJ, Batterham M, Cliff D, Salmon J, et al. Evaluation of an intervention to reduce adolescent sitting time during the school day: The 'Stand Up for Health' randomised controlled trial. J Sci Med Sport 2018 Dec;21(12):1244-1249. [doi: 10.1016/j.jsams.2018.05.020] [Medline: 29891406]
21. Bonde AH, Stjernqvist NW, Sabinsky MS, Maindal HT. Process evaluation of implementation fidelity in a Danish health-promoting school intervention. BMC Public Health 2018 Dec 27;18(1):1-10. [doi: 10.1186/s12889-018-6289-5]

XSL•FO
RenderX

22. Bellew B, Nau T, Smith B, Bauman A, editors. Getting Australia Active III: A systems approach to physical activity for policy makers. Sydney, Australia: The Australian Prevention Partnership Centre and The University of Sydney; 2020.
23. What is a Health Promoting School? World Health Organization. 2021. URL: https://www.who.int/health-topics/health-promoting-schools#tab=tab_1 [accessed 2021-07-19]
24. Milton K, Cavill N, Chalkley A, Foster C, Gomersall S, Hagstromer M, et al. Eight Investments That Work for Physical Activity. J Phys Act Health 2021 May 14;18(6):1-6. [doi: 10.1123/jpah.2021-0112] [Medline: 33984836]
25. Sutherland R, Campbell E, Lubans DR, Morgan PJ, Okely AD, Nathan N, et al. A cluster randomised trial of a school-based intervention to prevent decline in adolescent physical activity levels: study protocol for the 'Physical Activity 4 Everyone' trial. BMC Public Health 2013 Jan 22;13(57):1-10. [doi: 10.1186/1471-2458-13-57]
26. Sutherland R, Campbell E, Lubans DR, Morgan PJ, Okely AD, Nathan N, et al. 'Physical Activity 4 Everyone' school-based intervention to prevent decline in adolescent physical activity levels: 12 month (mid-intervention) report on a cluster randomised trial. Br J Sports Med 2016 Apr;50(8):488-495 [FREE Full text] [doi: 10.1136/bjsports-2014-094523] [Medline: 26359346]
27. Sutherland RL, Campbell EM, Lubans DR, Morgan PJ, Nathan NK, Wolfenden L, et al. The Physical Activity 4 Everyone Cluster Randomized Trial: 2-Year Outcomes of a School Physical Activity Intervention Among Adolescents. Am J Prev Med 2016 Aug;51(2):195-205 [FREE Full text] [doi: 10.1016/j.amepre.2016.02.020] [Medline: 27103495]
28. Mclaughlin M, Duff J, Sutherland R, Campbell E, Wolfenden L, Wiggers J. Protocol for a mixed methods process evaluation of a hybrid implementation-effectiveness trial of a scaled-up whole-school physical activity program for adolescents: Physical Activity 4 Everyone (PA4E1). Trials 2020 Mar 17;21(268):1-11. [doi: 10.1186/s13063-020-4187-5]
29. Sutherland R, Campbell E, Nathan N, Wolfenden L, Lubans DR, Morgan PJ, et al. A cluster randomised trial of an intervention to increase the implementation of physical activity practices in secondary schools: study protocol for scaling up the Physical Activity 4 Everyone (PA4E1) program. BMC Public Health 2019 Jul 04;19(1):883 [FREE Full text] [doi: 10.1186/s12889-019-6965-0] [Medline: 31272421]
30. Sutherland R, Reeves P, Campbell E, Lubans DR, Morgan PJ, Nathan N, et al. Cost effectiveness of a multi-component school-based physical activity intervention targeting adolescents: the 'Physical Activity 4 Everyone' cluster randomized trial. Int J Behav Nutr Phys Act 2016 Aug 22;13:94 [FREE Full text] [doi: 10.1186/s12966-016-0418-2] [Medline: 27549382]
31. Hollis JL, Sutherland R, Campbell L, Morgan PJ, Lubans DR, Nathan N, et al. Effects of a 'school-based' physical activity intervention on adiposity in adolescents from economically disadvantaged communities: secondary outcomes of the 'Physical Activity 4 Everyone' RCT. Int J Obes (Lond) 2016 Oct;40(10):1486-1493 [FREE Full text] [doi: 10.1038/ijo.2016.107] [Medline: 27430652]
32. Sutherland R, Campbell E, McLaughlin M, Nathan N, Wolfenden L, Lubans DR, et al. Scale-up of the Physical Activity 4 Everyone (PA4E1) intervention in secondary schools: 12-month implementation outcomes from a cluster randomized controlled trial. Int J Behav Nutr Phys Act 2020 Aug 08;17(1):100 [FREE Full text] [doi: 10.1186/s12966-020-01000-y] [Medline: 32771011]
33. Miller S, Ainsworth B, Yardley L, Milton A, Weal M, Smith P, et al. A Framework for Analyzing and Measuring Usage and Engagement Data (AMUsED) in Digital Interventions: Viewpoint. J Med Internet Res 2019 Feb 15;21(2):e10966 [FREE Full text] [doi: 10.2196/10966] [Medline: 30767905]
34. SAS. Enterprise Guide 6. SAS. 2014. URL: https://www.sas.com/en_au/home.html [accessed 2021-07-19]
35. Charters E. The Use of Think-aloud Methods in Qualitative Research An Introduction to Think-aloud Methods. Brock Education Journal 2003 Jul 01;12(2):68-82. [doi: 10.26522/brocked.v12i2.38]
36. Loop11. URL: https://www.loop11.com/ [accessed 2021-07-19]
37. Brooke J. SUS: A quick and dirty usability scale. In: Systems Usability Scale. Reading: Redhatch Consulting; 1996.
38. O'Brien HL, Cairns P, Hall M. A practical approach to measuring user engagement with the refined user engagement scale (UES) and new UES short form. International Journal of Human-Computer Studies 2018 Apr;112:28-39. [doi: 10.1016/j.ijhcs.2018.01.004]
39. Weiner BJ, Lewis CC, Stanick C, Powell BJ, Dorsey CN, Clary AS, et al. Psychometric assessment of three newly developed implementation outcome measures. Implementation Sci 2017 Aug 29;12(1):1-12. [doi: 10.1186/s13012-017-0635-3]
40. Bangor A, Kortum P, Miller J. Determining what individual SUS scores mean: adding an adjective rating scale. J Usability Studies 2009;4(3):114-123 [FREE Full text]
41. Braun V, Clarke V. Using thematic analysis in psychology. Qualitative Research in Psychology 2006 Jan;3(2):77-101. [doi: 10.1191/1478088706qp063oa]
42. NVivo. 2018. URL: https://www.qsrinternational.com/nvivo-qualitative-data-analysis-software/about/nvivo [accessed 2021-07-20]
43. Australian Bureau of Statistics. Statistical Geography Volume 1- Australian Standard Geogrphical Classification (ASGC). Canberra: Commonwealth of Australia; 2011.
44. Australian Bureau of Statistics. 2033. In: 0.55.001 001 - Socio-economic Indexes for Areas (SEIFA) - State Suburb (SSC) Index of Relative Socio-economic Disadvantage. Canberra: Commonwealth of Australia; 2011.
45. My School is a resource for parents, educators and the community to find information about each of Australia's schools. In: Australian Curriculum, Assessment and Reporting Authority. Canberra: Commonwealth of Australia; 2019.

XSL•FO
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46. Vandelanotte C, Müller AM, Short CE, Hingle M, Nathan N, Williams SL, et al. Past, Present, and Future of eHealth and mHealth Research to Improve Physical Activity and Dietary Behaviors. J Nutr Educ Behav 2016 Mar;48(3):219-228.e1. [doi: 10.1016/j.jneb.2015.12.006] [Medline: 26965100]
47. Sebire SJ, Kesten JM, Edwards MJ, May T, Banfield K, Tomkinson K, et al. Using self-determination theory to promote adolescent girls' physical activity: Exploring the theoretical fidelity of the Bristol Girls Dance Project. Psychol Sport Exerc 2016 May;24:100-110 [FREE Full text] [doi: 10.1016/j.psychsport.2016.01.009] [Medline: 27175102]
48. Smedegaard S, Brondeel R, Christiansen LB, Skovgaard T. What happened in the 'Move for Well-being in School': a process evaluation of a cluster randomized physical activity intervention using the RE-AIM framework. Int J Behav Nutr Phys Act 2017 Nov 16;14(1):159 [FREE Full text] [doi: 10.1186/s12966-017-0614-8] [Medline: 29145868]
49. Kostamo K, Jallinoja P, Vesala KM, Araújo-Soares V, Sniehotta FF, Hankonen N. Using the critical incident technique for qualitative process evaluation of interventions: The example of the "Let's Move It" trial. Soc Sci Med 2019 Jul;232:389-397 [FREE Full text] [doi: 10.1016/j.socscimed.2019.05.014] [Medline: 31146148]
50. Lubans DR, Smith JJ, Skinner G, Morgan PJ. Development and implementation of a smartphone application to promote physical activity and reduce screen-time in adolescent boys. Front Public Health 2014;2:42 [FREE Full text] [doi: 10.3389/fpubh.2014.00042] [Medline: 24904909]
51. Jong ST, Croxson CHD, Foubister C, Brown HE, Guell C, Lawlor ER, et al. Reach, Recruitment, Dose, and Intervention Fidelity of the GoActive School-Based Physical Activity Intervention in the UK: A Mixed-Methods Process Evaluation. Children 2020 Nov 17;7(11):231. [doi: 10.3390/children7110231]
52. Corder K, Sharp SJ, Jong ST, Foubister C, Brown HE, Wells EK, et al. Effectiveness and cost-effectiveness of the GoActive intervention to increase physical activity among UK adolescents: A cluster randomised controlled trial. PLoS Med 2020 Jul;17(7):e1003210 [FREE Full text] [doi: 10.1371/journal.pmed.1003210] [Medline: 32701954]
53. Corder K, Brown HE, Schiff A, van Sluijs EMF. Feasibility study and pilot cluster-randomised controlled trial of the GoActive intervention aiming to promote physical activity among adolescents: outcomes and lessons learnt. BMJ Open 2016 Nov 11;6(11):e012335. [doi: 10.1136/bmjopen-2016-012335]
54. Simpson SA, Matthews L, Pugmire J, McConnachie A, McIntosh E, Coulman E, et al. An app-, web- and social support-based weight loss intervention for adults with obesity: the HelpMeDoIt! feasibility RCT. Public Health Res 2020 Mar;8(3):1-270. [doi: 10.3310/phr08030] [Medline: 32186839]
55. Nurmi J, Knittle K, Ginchev T, Khattak F, Helf C, Zwickl P, et al. Engaging Users in the Behavior Change Process With Digitalized Motivational Interviewing and Gamification: Development and Feasibility Testing of the Precious App. JMIR Mhealth Uhealth 2020 Jan 30;8(1):e12884 [FREE Full text] [doi: 10.2196/12884] [Medline: 32003750]
56. Castle EM, Greenwood J, Chilcot J, Greenwood SA. Usability and experience testing to refine an online intervention to prevent weight gain in new kidney transplant recipients. Br J Health Psychol 2021 Feb;26(1):232-255. [doi: 10.1111/bjhp.12471] [Medline: 32931645]
57. Simons D, De Bourdeaudhuij I, Clarys P, De Cocker K, Vandelanotte C, Deforche B. A Smartphone App to Promote an Active Lifestyle in Lower-Educated Working Young Adults: Development, Usability, Acceptability, and Feasibility Study. JMIR Mhealth Uhealth 2018 Feb 20;6(2):e44 [FREE Full text] [doi: 10.2196/mhealth.8287] [Medline: 29463491]
58. Poppe L, Van der Mispel C, De Bourdeaudhuij I, Verloigne M, Shadid S, Crombez G. Users' thoughts and opinions about a self-regulation-based eHealth intervention targeting physical activity and the intake of fruit and vegetables: A qualitative study. PLoS One 2017;12(12):e0190020 [FREE Full text] [doi: 10.1371/journal.pone.0190020] [Medline: 29267396]
59. Shiffman S, Stone AA, Hufford MR. Ecological momentary assessment. Annu Rev Clin Psychol 2008;4:1-32. [doi: 10.1146/annurev.clinpsy.3.022806.091415] [Medline: 18509902]
60. Petersen JM, Prichard I, Kemps E. A Comparison of Physical Activity Mobile Apps With and Without Existing Web-Based Social Networking Platforms: Systematic Review. J Med Internet Res 2019 Aug 16;21(8):e12687 [FREE Full text] [doi: 10.2196/12687] [Medline: 31420956]
61. Bardus M, van Beurden SB, Smith JR, Abraham C. A review and content analysis of engagement, functionality, aesthetics, information quality, and change techniques in the most popular commercial apps for weight management. Int J Behav Nutr Phys Act 2016 Mar 10;13:35 [FREE Full text] [doi: 10.1186/s12966-016-0359-9] [Medline: 26964880]
62. Blandford A, Gibbs J, Newhouse N, Perski O, Singh A, Murray E. Seven lessons for interdisciplinary research on interactive digital health interventions. Digit Health 2018;4:2055207618770325 [FREE Full text] [doi: 10.1177/2055207618770325] [Medline: 29942629]
63. Faulkner L. Beyond the five-user assumption: benefits of increased sample sizes in usability testing. Behav Res Methods Instrum Comput 2003 Aug;35(3):379-383. [doi: 10.3758/bf03195514] [Medline: 14587545]

Abbreviations

AMUsED: Analyzing and Measuring Usage and Engagement Data
PA4E1: Physical Activity 4 Everyone
PE: physical education
SEIFA: Socio-Economic Indexes for Areas

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Original Paper

Associations Between Psychosocial Measures and Digital Media Use Among Transgender Youth: Cross-sectional Study

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Abstract

Background: Transgender, nonbinary, and gender-diverse (TNG) youth encounter barriers to psychosocial wellness and also describe exploring identities and communities on the web. Studies of cisgender youth connect increased digital technology use with lower well-being, parent relationships, and body image scores as well as increased loneliness and fear of missing out (FOMO). However, little is known about the psychosocial factors associated with digital technology use among TNG compared with cisgender youth.

Objective: This study aims to examine the associations between psychosocial measures and digital technology use and its importance for cisgender and TNG youth.

Methods: We surveyed a nationally representative sample of adolescents (aged 13-18 years) about psychosocial wellness and digital technology use. Psychosocial measures included assessment of well-being, parental relationships, body image, loneliness, and FOMO. Digital media use assessments included the short Problematic and Risky Internet Use Screening Scale-3 and the Adolescent Digital Technology Interactions and Importance (ADTI) scale and subscales. We compared psychosocial measures between gender identity groups. We also compared stratified correlations for psychosocial measures (well-being, parent relationships, body image, loneliness, and FOMO) with ADTI and Problematic and Risky Internet Use Screening Scale-3 scores between gender identity groups. All comparisons were adjusted for age, race, and ethnicity.

Results: Among 4575 adolescents, 53 (1.16%) self-identified as TNG youth. TNG youth had lower scores for well-being (23.76 vs 26.47; $P<.001$), parent relationships (19.29 vs 23.32; $P<.001$), and body image (13.50 vs 17.12; $P<.001$), and higher scores for loneliness (9.28 vs 6.55; $P<.001$) and FOMO (27.93 vs 23.89; $P=.004$), compared with cisgender peers. In a pattern different from that of their cisgender peers, better well-being scores and body image for TNG youth predicted higher problematic internet use (PIU) scores (correlation coefficients of 0.32 vs −0.07; $P=.004$ and 0.26 vs −0.21; $P=.002$, respectively). FOMO was a stronger positive predictor of higher ADTI total and subscale scores for cisgender youth compared with TNG youth.

Conclusions: Overall, this study supports previously demonstrated disparities in the psychosocial wellness of TNG youth and adds that these disparities include loneliness and FOMO. This study shows prediction of PIU by both higher well-being and better body image, indicating that PIU may not be unilaterally driven by problematic factors among TNG youth. We suggest that this may be because of the specific digital media functions that TNG youth engage with as a disenfranchised population.

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KEYWORDS

transgender person; internet; sex and gender minorities; well-being; adolescent; mobile phone

Introduction

Background

Transgender, nonbinary, and gender-diverse (TNG) youth are a marginalized population that experiences multiple barriers to psychosocial wellness. Experiences of discrimination and oppression are thought to lead to minority stress, which can lead to an increased risk of various negative health effects such as psychological distress, eating disorders, and suicidality [1-3]. Despite being a varied population, it is well recognized that TNG adolescents have disparities in different aspects of psychosocial wellness, including happiness [4] and parent support [5]. Although TNG youth may also experience lower body image and overall quality of life, these can improve with gender-affirming therapies [6,7].

Social support, community connectedness, and coping strategies may protect the psychosocial wellness of TNG youth [1]. However, this population also identifies barriers to support and to identity exploration and expression [8,9]. TNG youth may, therefore, compensate for the risk of poor social support in their communities by using the internet and social media to connect with others to access social and informational support they may not receive elsewhere [10]. How factors in psychosocial wellness are related to TNG youth's digital media use is not well understood.

In general youth populations, several negative psychosocial measures have been associated with high levels of digital media use. Lower well-being and higher rates of suicidal thoughts are associated with high levels of internet use among adolescents [11]. In addition, depression and higher perceived stress scores are associated with problematic internet use (PIU) [12-14], which is defined as use that is "risky, excessive, or impulsive" and leads to "physical, emotional, functional, or social impairment" [15].

Other areas of disparity in psychosocial wellness for TNG youth, including low parent support, body image, and well-being, have also been tied to media use in general adolescent populations. For example, increased parental control, restrictive mediation, and parental neglect predict smartphone addiction [16]. Good parent-child communication and web-based parent support, such as being friends with one's parents on Facebook, are associated with decreased PIU [17,18]. The use of social media is also associated with an increase in concern about body image [19], with increased use associated with increased body dissatisfaction across genders [20]. The barriers that TNG youth experience in these psychosocial domains may influence how and the degree to which they use digital media.

Two additional factors connected to social media use that may be areas of vulnerability for TNG youth are loneliness and fear of missing out (FOMO), the tendency to feel anxious over missing out on rewarding experiences of others. These factors have not been studied in TNG youth but have been associated with digital media use in general adolescent populations [21,22], including frequent social media checks [23]. As gender identity minorities, TNG youth may be at risk of increased loneliness and FOMO, which may impact digital media use in this group. Notably, belonging, the opposite of loneliness, has been identified as a mediator of positive outcomes for TNG people. Community belonging fully mediates the relationship between transgender identity and well-being in TNG adults [24]. In TNG youth, school belonging is associated with decreased drug use among TNG youth [25] and better mental health, and it also mediates the relationship between peer victimization and mental health concerns [26]. Understanding disparities in loneliness and FOMO in TNG youth and the intersection of those factors with digital media may help identify opportunities to facilitate belonging in web-based spaces using evidence-based interventions [27].

Although multiple negative psychosocial wellness factors have been associated with digital media use or PIU, more typical patterns of internet use have also been associated with well-being or life satisfaction in some studies. Better overall well-being is associated with the use of social media to connect with others [28]. Similarly, a study in which participants had to decrease their social media use resulted in a decrease in self-reported life satisfaction [29]. Lesbian, gay, bisexual, transgender, queer, and questioning (LGBTQ+) youth, in particular, are more likely to identify web-based friends and to describe them as more supportive than in-person friends [30]. During the COVID-19 pandemic, LGBTQ+ youth identify the specific importance of web-based spaces and support during a time when they may be stuck at home with unsupportive family members [31]. Thus, previous research suggests potential benefits and risks associated with digital media use, especially for LGBTQ+ youth.

Objectives

Given that TNG youth identify the importance of web-based spaces while simultaneously experiencing risks to psychosocial wellness, an understanding of the relationship between psychosocial measures and digital technology use is critical for cultivating positive digital experiences for this at-risk population. The aim of this study is to examine the associations between psychosocial measures and digital technology use and its importance for cisgender and TNG youth. We hypothesized that TNG youth would show disparities in measures of psychosocial wellness and that PIU and digital media importance would be tied to negative psychosocial measures in both TNG and cisgender youth.

Methods

Setting

We conducted a secondary analysis of a cross-sectional survey administered between February and March 2019. This survey was conducted using Qualtrics, a web-based survey platform. The study was approved by the University of

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Wisconsin-Madison Education and Social Behavioral Sciences Institutional Review Board.

Participants

We used a survey panel approach to study a representative sample of adolescents in the United States. Panels facilitate increased speed of data collection with a wide geographic reach compared with traditional approaches for survey distribution [32]. Due to this increased efficiency and ability to provide demographic samples within 10% of their reciprocal US population values, Qualtrics was selected as the survey platform [33]. Recruited participants had previously signed up to get survey invitations through Qualtrics. Upon joining Qualtrics, panelists were asked to complete demographic assessments, which allow relevant survey invitations to be targeted to potentially eligible sample populations. Participants received invitations by email, and the survey remained open until the requested number of participants had completed the survey.

Adult parents who were US residents who spoke English, had adolescent children (aged 13-18 years), and had signed up for survey panel participation were recruited by a Qualtrics survey manager. Information was provided to parents who had potentially eligible children and were interested in the study. Consent was obtained from parents of children aged between 13 and 17 years at the beginning of the survey; adolescents who were aged 18 years gave their own consent before beginning the survey. Participants aged 13-17 years also provided web-based assent before survey initiation. Youth participants had the option to discontinue the survey at any time without loss of benefits. Participants completed demographic information as part of this survey (refer to the full survey in Multimedia Appendix 1) and were included in the study if they answered the question that asked them to describe their gender identity. Youth were excluded if they selected *Preferred not to answer* in response to the question about their gender identity. Although some youth that choose *Preferred not to answer* may be gender-questioning, we deemed it inappropriate to make assumptions about whether this group was primarily cisgender, gender-questioning, or TNG.

Measures

The full text for all survey measures and scoring summaries can be found in Multimedia Appendix 1.

Digital Media Use Measures

Problematic and Risky Internet Use

The validated short version of the Problematic and Risky Internet Use Screening Scale-3 (PRIUSS-3) was used to measure problematic and risky internet use [34]. This scale includes three questions that evaluate anxiety when away from the internet, loss of motivation when on the internet, and feelings of withdrawal when away from the internet. A Likert scale was used to measure how often participants experienced these items (0=never, 1=rarely, 2=sometimes, 3=often, and 4=very often). A summed score of three or more on PRIUSS-3 indicates that the participant is at risk for PIU. The α coefficient for this scale was .87.

Adolescent Digital Technology Interactions and Importance

Perceived motivations for adolescents' technology interactions were measured using the validated Adolescent Digital Technology Interactions and Importance (ADTI) scale [35]. There are three ADTI subscales: factor 1, technology to bridge web-based and offline experiences and preferences; factor 2, technology to go outside one's identity or offline environment; and factor 3, technology for social connection. Each of the 18 items included in the ADTI is associated with one of the subscales. For example, participants were asked how important it was to use social media platforms to "Provide an important accomplishment or update on your life using social media" (factor 1), "video chat" (factor 3), "manage my mood" (factor 2), and "create a profile with a different identity" (factor 2). A five-point Likert scale was used to score perceived importance levels (1=not at all important and 5=extremely important). A higher perceived importance of technology use was indicated by a higher summation score. The α coefficient was .95 for the total ADTI scale. The α coefficients for the three subscales were .90 (factor 1), .92 (factor 2), and .89 (factor 3).

Psychosocial Wellness Measures

Overall Well-being

Well-being was measured using the validated Short Warwick-Edinburg Mental Well-being Scale [36]. This seven-item scale asks participants to specify how often in the past 2 weeks they experienced the following feelings or experiences: "I've been feeling useful," "I've been dealing with problems well," and "I've been able to make up my own mind about things." Participants responded based on a five-point Likert scale (1=none of the time and 5=all of the time). A summary score was calculated by adding the individual responses for each item. A higher summary score indicated higher levels of well-being. The α coefficient for this measure was .91 in the general population [36].

Parental Relationships

To assess parental relationships, we used the eight-question Parent-Adolescent Relationship Scale [37]. This scale consisted of three statements about the adolescent's identification with the parent, such as "I think highly of him/her," assessed with a Likert scale of 0=strongly disagree to 4=strongly agree. In addition, this scale includes questions about parent-adolescent relationships, such as "How often does she/he praise you for doing well?" or "How often does she/he blame you for her/his problems?" These were scored with responses ranging from 0 to 4 (0=never and 4=always). Three questions (Multimedia Appendix 1) were reverse scored, as they were framed with negative connotations. A summary score was calculated, with a higher numeric output indicating better parental relationships. The α coefficient for this scale was .68.

Body Image

Body image was assessed using the previously validated Body Image Scale, which consists of four items: "I would like to change a good deal about my body," "I am satisfied with my looks," "I would like to change a good deal about my looks," and "I am satisfied with my body" [38]. Each is answered based on a six-point Likert scale, ranging from 1 for "does not apply

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at all" and 6 for "applies exactly." A summary score was generated by summing all items; items 1 and 3 were reverse scored because of their negative framing. A higher summary score indicates a more positive body image. The α for this four-item scale was .82 [38].

Loneliness

Loneliness was measured using subscale questions from the validated Comprehensive Inventory of Thriving [39]. Participants were asked to agree or disagree with the following statements: "I feel lonely," "I often feel left out," and "there is no one I feel close to," using a five-point Likert scale (1=strongly disagree and 5=strongly agree). A summary score for each participant was calculated, with a higher score indicating increased loneliness. The range of α coefficients for this subscale previously published is .79 to .87 [39] and the α coefficient in our study was .90.

Fear of Missing Out

FOMO was measured via a 10-item scale that has been used in previous studies [40-42], which includes statements such as "I get worried when I find out my friends are having fun without me" and "It bothers me when I miss an opportunity to meet up with friends" [43]. Response options were based on a Likert scale (1=not at all true of me and 5=extremely true of me). A summary score was computed for each participant by averaging the responses of all 10 statements. The α coefficient for this scale was .87 [43].

Demographics

Demographic questions assessed age, gender, race, and ethnicity. Participants were asked to identify their age by selecting a whole number (12-18) in response to the question, "What is your age in years?." Gender was assessed by asking, "Which response best describes your gender?," with response options of "Female," "Male," "Non-binary gender," "Female to male transgender," "Male to female transgender," "Other," or "Prefer not to answer." Participants were considered cisgender if they answered "Female" or "Male" and TNG if they answered "Non-binary gender," "Female to male transgender," "Male to female transgender," or "Other." To assess ethnicity and race, respectively, participants were asked, "Are you of Hispanic, Latino, or Spanish origin or descent?" and "What would you consider your race?" (refer to Multimedia Appendix 1 for the full text). Due to limited power in the TNG group, racial groups were dichotomized into White or Caucasian people and people of color and ethnicity into non-Hispanic, Latinx, or Spanish people and Hispanic, Latinx, or Spanish people when used as controls.

Analysis

Demographic information was compared between gender groups using the Fisher exact test for categorical analysis, which included comparison by age group (13-14 years vs 15-18 years to demonstrate representation of middle school– and high school–aged youth), race, and ethnicity between gender identity groups. A two-tailed *t* test was used to compare age as a continuous variable. Psychosocial outcomes were compared between gender groups while adjusting for age, race, and ethnicity using analysis of covariance in PROC GLM procedure in SAS. Stratified correlations for psychosocial measures (well-being, parent relationships, body image, loneliness, and FOMO) with ADTI and PRIUSS-3 scores were compared between gender identity groups using PROC NLMIXED procedure in SAS while adjusting for age, race, and ethnicity. We compared the regression coefficients of standardized values between the gender groups; in this case, the slopes were equal to the correlation coefficients. All reported *P* values were two-sided, and $P<.05$ was used to define statistical significance. Statistical analyses were performed using SAS software (version 9.4; SAS Institute).

Results

Demographics

Among 4575 adolescent participants, there were 53 (1.16%) TNG youth. Mean age (cisgender youth: 14.62 years, SD 1.68; TNG youth: 14.57 years, SD 1.66; $P=.82$) and age distribution did not vary between the two gender identity groups. Compared with cisgender peers, TNG youth were less likely to identify their race as White people (26/53, 49% vs 3041/4522, 67.25%; $P<.001$) and more likely to identify their ethnicity as Hispanic (31/52, 60% vs 786/4469, 17.59%, $P<.001$). Few TNG youth identified with transfeminine identities (5/4575, 0.11% of the total study population) compared with youth with nonbinary identities (23/4575, 0.5%) and transmasculine identities (25/4575, 0.55%). Refer to Table 1 for demographic information.

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Table 1. Demographics of cisgender and transgender, nonbinary, and gender-diverse youth participants (N=4575).

Demographic measure	Cisgender		TNG[a]		*P* value
	Total	Participant, n (%)	Total	Participant, n (%)	
Gender identity[b]					
Female or feminine identity	4522	2130 (47.1)	53	5 (9.4)	__[c]
Male or masculine identity	4522	2392 (52.9)	53	25 (47.2)	—
Nonbinary identity	N/A[d]	N/A	53	23 (43.4)	—
Total	4575	4522 (98.84)	4575	53 (1.16)	—
Age[e] (years), n (%)					
13-14	4506	2160 (47.94)	53	29 (54.72)	.34
15-18	4506	2346 (52.06)	53	24 (45.28)	.34
Ethnicity, n (%)					<.001
Non-Hispanic	4469	3683 (82.41)	52	21 (40.38)	
Hispanic	4469	786 (17.59)	52	31 (59.62)	
Race, n (%)					<.001
White	4521	3041 (67.25)	53	26 (49.06)	
Black	4521	692 (15.3)	53	5 (9.43)	
Native	4521	139 (3.07)	53	12 (22.64)	
Asian	4521	227 (5.02)	53	8 (15.09)	
Multiracial	4521	219 (4.84)	53	2 (3.77)	
Other	4521	204 (4.51)	53	0 (0)	

[a]TNG: transgender, nonbinary, and gender-diverse.

[b]No youth selected “Other” in identifying their gender identity.

[c]Unable to perform comparison and derive *P* value given that there are different numbers of gender subcategories for cisgender and transgender, nonbinary, and gender-diverse youth.

[d]N/A: not applicable.

[e]Cisgender: mean age 14.62 years (SD 1.68); transgender, nonbinary, and gender-diverse: mean age 14.57 years (SD 1.66); *P*=.82.

Psychosocial Outcomes

TNG youth had lower scores for well-being (23.76 vs 26.47; *P*<.001), parent relationship scores (19.29 vs 23.32; *P*<.001), and body image (13.5 vs 17.12; *P*<.001) and had higher scores for loneliness scores (9.28 vs 6.55; *P*<.001) and FOMO (27.93 vs 23.89; *P*=.004) compared with cisgender youth. Across these categories, this remained significant when TNG youth were compared with cisgender females, cisgender males, and a combined group of cisgender males and females (Table 2).

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Table 2. Comparison of mean scores of psychosocial measures between cisgender and transgender, nonbinary, and gender-diverse youth[a].

Psychosocial outcome measure	Cisgender female and TNG[b] youth			Cisgender male and TNG youth			Cisgender and TNG youth		
	Cisgender female participants, mean score (SE)	TNG participants, mean score (SE)	*P* value	Cisgender male participants, mean score (SE)	TNG participants, mean score (SE)	*P* value	Cisgender male and female participants, mean score (SE)	TNG participants, mean score (SE)	*P* value
Well-being	26.16 (0.13)	23.76 (0.69)	<.001	26.76 (0.12)	23.76 (0.69)	<.001	26.47 (0.10)	23.76 (0.70)	<.001
Parent relationship	23.46 (0.11)	19.29 (0.62)	<.001	23.18 (0.11)	19.29 (0.62)	<.001	23.32 (0.09)	19.29 (0.62)	<.001
Body image	16.96 (0.11)	13.50 (0.61)	<.001	17.27 (0.11)	13.50 (0.61)	<.001	17.12 (0.09)	13.50 (0.61)	<.001
Loneliness	6.44 (0.09)	9.28 (0.50)	<.001	6.65 (0.09)	9.28 (0.50)	<.001	6.55 (0.07)	9.28 (0.50)	<.001
FOMO[c]	23.88 (0.25)	27.93 (1.37)	.004	23.89 (0.24)	27.93 (1.37)	.004	23.89 (0.20)	27.93 (1.37)	.004

[a]Adjusted for age, race, and ethnicity; all the values are significant compared with *P*=.05.

[b]TNG: transgender, nonbinary, and gender-diverse.

[c]FOMO: fear of missing out.

Digital Media Use Measures and Psychosocial Outcomes

When correlations between PRIUSS-3 scores and psychosocial measures were assessed, TNG youth showed patterns that differed from their cisgender peers (Figure 1). Positive body image and higher well-being positively predicted PIU scores for TNG youth (body image: 0.26 and well-being: 0.32), whereas a negative correlation was seen for cisgender youth for both categories (body image: −0.21; *P*=.002 and well-being: −0.07; *P*=.004). TNG and cisgender youth showed similar patterns of PIU correlating negatively with parental relationship scores and positively with loneliness and FOMO. Figure 1 shows the associations between psychosocial outcomes and PRIUSS-3 scores.

There were also some differences in the correlation patterns for cisgender and transgender youth when examining associations between ADTI scores and wellness measures. Better parent relationships predicted higher total ADTI scores in TNG youth (correlation coefficient 0.14), whereas higher parent relationship scores predicted lower total ADTI scores in cisgender males (−0.19; *P*=.04). A significant difference in correlation was not found between TNG youth and cisgender females. Better body image scores also positively predicted higher ADTI-3 scores for TNG youth (correlation coefficient 0.43) compared with slight negative correlation for cisgender youth (−0.01; *P*=.004). Increased FOMO was a positive predictor of ADTI total and subscale scores for youth of all genders, although it was a stronger predictor for cisgender compared with TNG youth. Table 3 shows the associations between the ADTI scores and psychosocial outcomes.

Figure 1. Comparison of correlation coefficients of well-being, parent relationship, body image, loneliness, and fear of missing out versus Problematic and Risky Internet Use Screening Scale-3 for cisgender and transgender, nonbinary, and gender diverse youth. **P*<.01. TNG: transgender, nonbinary, and gender-diverse.

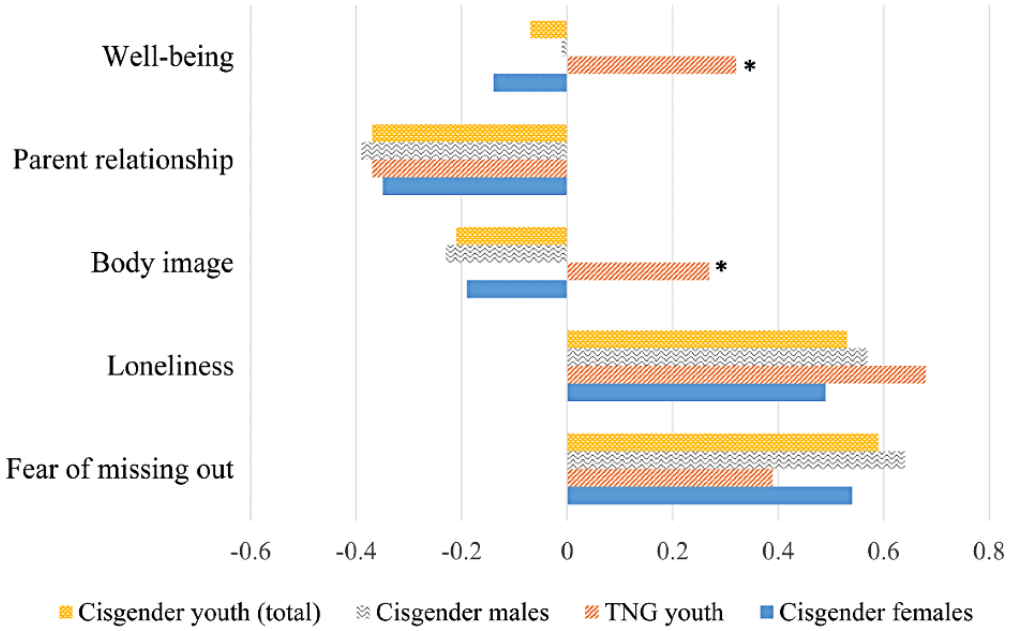

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Table 3. Comparison of correlation coefficients of well-being, parent relationships, body image, loneliness, and fear of missing out versus digital technology interactions and problematic internet use for transgender, nonbinary, and gender-diverse and cisgender youth[a].

Outcome and predictor	Cisgender female and TNG[b] youth			Cisgender male and TNG youth			Cisgender and TNG youth		
	Cisgender females	TNG youth	*P* value	Cisgender males	TNG youth	*P* value	Cisgender males and females	TNG youth	*P* value
PRIUSS-3[c] (problematic internet use)									
Well-being	*−0.14*[d]	*0.32*	*<.001*	*−0.01*	*0.32*	*.02*	*−0.07*	*0.32*	*.004*
Parent relationship	−0.35	−0.37	.90	−0.39	−0.37	.84	−0.37	−0.37	.96
Body image	*−0.19*	*0.27*	*.003*	*−0.23*	*0.27*	*.001*	*−0.21*	*0.26*	*.002*
Loneliness	0.49	0.68	.10	0.57	0.68	.38	0.53	0.67	.22
FOMO[e]	0.54	0.39	.22	0.64	0.39	.05	0.59	0.39	.11
ADTI[f] total									
Well-being	0.10	0.40	.02	0.23	0.40	.17	0.17	0.40	.06
Parent relationship	−0.16	0.14	.06	*−0.19*	*0.14*	*.04*	−0.18	0.13	.05
Body image	−0.20	−0.05	.28	−0.29	−0.05	.08	−0.25	−0.05	.15
Loneliness	0.28	0.33	.71	0.41	0.33	.53	0.35	0.33	.85
FOMO	0.52	0.27	.05	0.67	0.27	.002	0.60	0.27	.01
ADTI factor 1 (technology to bridge web-based and offline experiences or preferences)									
Well-being	0.11	0.34	.10	0.23	0.34	.45	0.18	0.34	.23
Parent relationship	−0.14	−0.07	.57	−0.23	−0.07	.23	−0.19	−0.07	.36
Body image	−0.07	0.15	.17	−0.14	0.15	.06	−0.11	0.14	.10
Loneliness	0.19	0.16	.82	0.34	0.16	.23	0.27	0.16	.45
FOMO	0.45	0.19	.05	0.61	0.19	.002	0.54	0.19	.01
ADTI factor 2 (technology to go outside one's identity or offline environment)									
Well-being	0.06	0.29	.07	0.18	0.29	.39	0.13	0.29	.20
Parent relationship	−0.21	−0.04	.16	−0.34	−0.04	.02	−0.28	−0.04	.05
Body image	−0.30	−0.13	.20	−0.30	−0.13	.20	−0.31	−0.13	.19
Loneliness	0.32	0.26	.65	0.47	0.26	.11	0.40	0.26	.27
FOMO	0.45	0.24	.09	0.65	0.24	<.001	0.56	0.24	.009
ADTI factor 3 (technology for social connection)									
Well-being	0.07	0.34	.05	0.21	0.34	.35	0.15	0.34	.15
Parent relationship	−0.15	−0.03	.37	−0.19	−0.03	.24	−0.17	−0.03	.29
Body image	0.01	0.43	.006	*−0.02*	*0.43*	*.003*	*−0.01*	*0.43*	*.004*
Loneliness	0.18	0.35	.22	0.29	0.35	.66	0.23	0.35	.42
FOMO	0.44	0.16	.04	0.54	0.16	.006	0.49	0.16	.02

[a]All comparisons were adjusted for age, race, and ethnicity.

[b]TNG: transgender, nonbinary, and gender-diverse.

[c]PRIUSS-3: Problematic and Risky Internet Use Screening Scale-3.

[d]Values in italics denote statistically significant difference and different patterns of correlation (±) between cisgender and transgender, nonbinary, and gender-diverse youth.

[e]FOMO: fear of missing out.

[f]ADTI: Adolescent Digital Technology Interactions and Importance.

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Discussion

Principal Findings

This cross-sectional study is the first to explore factors in psychosocial wellness as predictors of PIU and the importance of digital media use among TNG youth. Our findings show continued barriers to psychosocial wellness in TNG youth compared with cisgender youth. In addition, our study shows that the pattern of prediction of psychosocial risk factors with PIU differs in TNG youth, with some positive factors predicting higher PIU scores, suggesting that digital engagement may function differently for this group. Finally, we found that FOMO was a stronger predictor of digital technology importance for cisgender youth compared with TNG youth across ADTI scales and subscales.

The comparison of psychosocial measures for TNG versus cisgender youth is consistent with previous studies that show significant threats to psychosocial wellness in this population [4,5]. Loneliness and FOMO, which have not previously been well studied in TNG youth, were also increased, which may speak to the social isolation that can occur as a gender minority. This is particularly notable given that loneliness and FOMO are constructs inverse to belonging, which has been shown to have a protective mediating effect in TNG youth and adults [24-26]. Future research to better understand the role and development of FOMO in TNG youth may offer insights into and avenues for interventions to facilitate belonging.

Although disparities in psychosocial wellness in TNG youth were consistent across categories, our findings show that positive PRIUSS-3 screens for PIU were predicted by some negative psychosocial experiences in this group (low parent relationship score, loneliness, and FOMO) and also predicted by positive attributes in a pattern different from cisgender peers. Well-being and body image scores predicted positive screens for PRIUSS-3 among TNG youth. In contrast, body image and well-being scores were negatively correlated with positive PRIUSS-3 screens in cisgender peers. This correlation of well-being with the outcome of PIU may complicate the very definition of PIU in this population, as PIU by definition interferes with functions that may be central to well-being. In TNG youth, this finding may represent the complexity of PIU. Although internet and digital media use may be a site of increased web-based bullying for TNG youth [30] and may interfere with day-to-day activities that are considered standard, appropriate activities for youth (such as school), web-based experiences may be varied enough to also support wellness as an alternative to environments where TNG youth may encounter in-person bullying [44] and other forms of harm (again, school).

The prediction of PIU by positive body image for TNG youth may also relate to a function of digital media more specific to this population: the importance of being read as the gender of their identification. TNG youth sometimes experience gender dysphoria ("a marked difference between the individual's expressed/experienced gender and the gender others would assign him or her" [45]), which may relate to a person's experience in their body and how their physical appearance is read by others as a certain gender. In general adolescent populations, social media use has been associated with eating disorder behaviors [46]. Limited research suggests that TNG young adults are at higher risk of such behaviors [3], which makes the connection between PIU and positive body image seen here more surprising. This relationship between PIU and body image in TNG youth may be mediated by the digital media function of being able to present and be recognized as their identified gender. Literature from adult transgender populations shows that being able to present (and be *read* by others) as one's identified gender is related to improved body image [47]. Digital media offers the opportunity to represent oneself using chosen names, pronouns, and selected photographs that may simplify this process compared with offline communities, with different platforms facilitating this in different ways. In addition, disclosure of identity on social media by TNG adults is followed by increased positive sentiment in subsequent posts [48], such that there may be a positive snowball effect from web-based engagement.

This complexity in the relationship of TNG youth with internet use is also evident in the relationship between FOMO and digital media importance. For cisgender youth compared with TNG youth, FOMO is a stronger predictor of higher levels of digital media importance across ADTI and ADTI subscales, measuring overall importance, technology to bridge web-based and offline experiences, technology to go outside one's identity or offline environment, and technology for social connection. It is notable that, despite increased FOMO in TNG youth, FOMO was both a less powerful predictor of the *importance* of digital media and did not differ as a predictor of PIU for TNG compared with cisgender youth. Although a predominantly cisgender culture in real life reflects cisgender identities as the norm, TNG youth may have increased FOMO as they see only that cisgender narrative in their offline experience, but our results suggest that this feeling of missing out may not be a powerful driver of why this population turns to digital media. This is in line with qualitative descriptions of digital media use, in which TNG youth describe seeking out digital media as a positive resource where they find validation of their identities in their web-based interactions and classify space on the web as a source of informational and emotional support [10,49]. This may show a pivot to digital media as an example of positive action and resilience in TNG youth, as they go on the web to access reflections of their experiences in that of others, a step identified by TNG youth in identity formation [50]. Haimson [51] posits that social media is a *social transition machinery* that facilitates these rites of passage during the process of gender transition. This understanding of web-based interactions driven by motivation for positive interactions (rather than escapism) may help to reframe approaches to building web-based spaces for TNG youth, with increased focus on resilience, belonging through connection, celebration of TNG identities, and community.

Limitations

Although this study included a large adolescent sample with a prevalence of TNG identity similar to the general US population of approximately 0.5%-3% [52-54], this secondary analysis was not targeted to maximize recruitment of TNG youth, and the absolute number of TNG youth in our study was limited

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(53/4575, 1.16%). This also limited additional subgroup analyses (eg, by specific gender identity or race and ethnicity). Furthermore, although the ethnic and racial breakdown of participants in this sample mirrors that of the US population [55], a response rate is not available because of the recruitment methods used by Qualtrics. Although these factors may limit generalizability, this study represents an important opportunity to examine the experiences of TNG youth in a large national sample. The TNG youth in this study are more likely to identify as people of color and as Hispanic compared with cisgender youth participants; as such, we controlled for race and ethnicity in our analysis.

In addition, this study did not assess whether TNG youth had accessed gender care. Gender care has been associated with improved body image and quality of life among young TNG people [7], and there is a strong web-based community around gender transition. Indeed, there are a number of YouTube channels, Tumblr blogs, and vlogs [51,56] documenting steps in gender transition, as adolescents and young adults engage in hormone therapy or move through the process of accessing different surgeries. It may be that TNG youth with access to gender care may have improved well-being and body image as well as more web-based engagement in these digital spaces with associated increased measures of PIU. Understanding access to gender care, gender transition, web-based engagement, and PIU would be a rich area for future research.

Conclusions

Overall, this study is the first to examine the relationship between psychosocial wellness and digital media use as well as the first to show disparities in loneliness and FOMO in TNG compared with cisgender youth. These findings support the importance of a nuanced approach to the interpretation of positive PIU screens in TNG youth. The prediction of PIU by both well-being and improved body image shows that PIU may not be unilaterally problematic among TNG youth, and the definition of and screening tools for PIU may need to be further explored in this population. The pattern of these results may support a picture in which digital media use offers critical functions that may engage and reinforce TNG youth with some strengths in certain areas of psychosocial wellness, including overall well-being and body image. In addition, this highlights the limited role of FOMO in digital media importance in this group compared with cisgender youth, which may offer opportunities to better understand and facilitate resilience and belonging in web-based spaces. In clinical settings, a nuanced, harm reduction approach may assist with counseling and creating a realistic media plan to reduce screen time while honoring that a young TNG person may benefit from specific functions of digital media.

Whether this more complicated picture of PIU applies to other minority populations besides TNG youth will be an important area for future research. A better understanding of positive PRIUSS-3 screens, PIU, and reasons for and predictors of digital media use, particularly in disenfranchised or oppressed populations (such as Black and indigenous youth and other youth of color; lesbian, gay, bisexual, and questioning youth; and disabled youth), will better inform opportunities for intervention and support.

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Conflicts of Interest

None declared.

Multimedia Appendix 1
Survey details, with keys to scoring and interpretation.
[PDF File (Adobe PDF File), 247 KB - pediatrics_v4i3e25801_app1.pdf]

References

1. Valentine SE, Shipherd JC. A systematic review of social stress and mental health among transgender and gender non-conforming people in the United States. Clin Psychol Rev 2018 Dec;66:24-38 [FREE Full text] [doi: 10.1016/j.cpr.2018.03.003] [Medline: 29627104]
2. James S, Herman J, Rankin S, Keisling M, Mottet L, Anafi M. The Report of the 2015 U.S. Transgender Survey. National Center for Transgender Equality, Washington, DC. 2016. URL: https://transequality.org/sites/default/files/docs/usts/USTS-Full-Report-Dec17.pdf [accessed 2021-07-14]
3. Diemer EW, Grant JD, Munn-Chernoff MA, Patterson DA, Duncan AE. Gender identity, sexual orientation, and eating-related pathology in a national sample of college students. J Adolesc Health 2015 Aug;57(2):144-149 [FREE Full text] [doi: 10.1016/j.jadohealth.2015.03.003] [Medline: 25937471]

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4. Aparicio-García ME, Díaz-Ramiro EM, Rubio-Valdehita S, López-Núñez MI, García-Nieto I. Health and well-being of cisgender, transgender and non-binary young people. Int J Environ Res Public Health 2018 Sep 28;15(10):2133 [FREE Full text] [doi: 10.3390/ijerph15102133] [Medline: 30274141]
5. Clark TC, Lucassen MF, Bullen P, Denny SJ, Fleming TM, Robinson EM, et al. The health and well-being of transgender high school students: results from the New Zealand adolescent health survey (Youth'12). J Adolesc Health 2014 Jul;55(1):93-99 [FREE Full text] [doi: 10.1016/j.jadohealth.2013.11.008] [Medline: 24438852]
6. Kuper LE, Stewart S, Preston S, Lau M, Lopez X. Body Dissatisfaction and Mental Health Outcomes of Youth on Gender-Affirming Hormone Therapy. Pediatrics 2020 Apr 27;145(4):e20193006 [FREE Full text] [doi: 10.1542/peds.2019-3006] [Medline: 32220906]
7. de Vries AL, McGuire JK, Steensma TD, Wagenaar EC, Doreleijers TA, Cohen-Kettenis PT. Young adult psychological outcome after puberty suppression and gender reassignment. Pediatrics 2014 Oct 08;134(4):696-704. [doi: 10.1542/peds.2013-2958] [Medline: 25201798]
8. Kosciw J, Greytak E, Giga N, Villenas C, Danischewski D. The 2015 National School Climate Survey: The experiences of lesbian, gay, bisexual, transgender, and queer youth in our nation's schools. New York: GLSEN; 2016:1-174.
9. Botsford J, Allen B, Andert B, Budge S, Rehm J. Meeting the healthcare needs of transgender, nonbinary, and gender expansive/nonconforming youth in Wisconsin: a report of the 2017 Wisconsin Transgender Youth Community Needs Assessment. 2018. URL: https://www.med.wisc.edu/media/medwiscedu/documents/about-us/CH-174891-18-TNG-Youth-Report-Full.pdf [accessed 2021-07-14]
10. Selkie E, Adkins V, Masters E, Bajpai A, Shumer D. Transgender adolescents' uses of social media for social support. J Adolesc Health 2020 Mar;66(3):275-280. [doi: 10.1016/j.jadohealth.2019.08.011] [Medline: 31690534]
11. Twenge JM, Campbell WK. Media use is linked to lower psychological well-being: evidence from three datasets. Psychiatr Q 2019 Jun 11;90(2):311-331. [doi: 10.1007/s11126-019-09630-7] [Medline: 30859387]
12. Ko C, Yen J, Chen C, Yeh Y, Yen C. Predictive values of psychiatric symptoms for internet addiction in adolescents: a 2-year prospective study. Arch Pediatr Adolesc Med 2009 Oct 05;163(10):937-943. [doi: 10.1001/archpediatrics.2009.159] [Medline: 19805713]
13. Yen J, Ko C, Yen C, Wu H, Yang M. The comorbid psychiatric symptoms of internet addiction: attention deficit and hyperactivity disorder (ADHD), depression, social phobia, and hostility. J Adolesc Health 2007 Jul;41(1):93-98. [doi: 10.1016/j.jadohealth.2007.02.002] [Medline: 17577539]
14. Derbyshire KL, Lust KA, Schreiber LR, Odlaug BL, Christenson GA, Golden DJ, et al. Problematic internet use and associated risks in a college sample. Compr Psychiatry 2013 Jul;54(5):415-422. [doi: 10.1016/j.comppsych.2012.11.003] [Medline: 23312879]
15. Moreno MA, Jelenchick LA, Christakis DA. Problematic internet use among older adolescents: a conceptual framework. Comput Human Behav 2013 Jul;29(4):1879-1887. [doi: 10.1016/j.chb.2013.01.053]
16. Fischer-Grote L, Kothgassner OD, Felnhofer A. Risk factors for problematic smartphone use in children and adolescents: a review of existing literature. Neuropsychiatr 2019 Dec 06;33(4):179-190 [FREE Full text] [doi: 10.1007/s40211-019-00319-8] [Medline: 31493233]
17. Anderson EL, Steen E, Stavropoulos V. Internet use and problematic internet use: a systematic review of longitudinal research trends in adolescence and emergent adulthood. Int J Adolesc Youth 2016 Oct 10;22(4):430-454. [doi: 10.1080/02673843.2016.1227716]
18. Kerr B, D'Angelo JD, Diaz-Caballero A, Moreno MA. College student problematic internet use and digital communication medium used with parents: cross-sectional study. JMIR Pediatr Parent 2020 Apr 23;3(1):e17165 [FREE Full text] [doi: 10.2196/17165] [Medline: 32324140]
19. Richards D, Caldwell PH, Go H. Impact of social media on the health of children and young people. J Paediatr Child Health 2015 Dec 26;51(12):1152-1157. [doi: 10.1111/jpc.13023] [Medline: 26607861]
20. Holland G, Tiggemann M. A systematic review of the impact of the use of social networking sites on body image and disordered eating outcomes. Body Image 2016 Jun;17:100-110. [doi: 10.1016/j.bodyim.2016.02.008] [Medline: 26995158]
21. Berryman C, Ferguson CJ, Negy C. Social media use and mental health among young adults. Psychiatr Q 2018 Jun 1;89(2):307-314. [doi: 10.1007/s11126-017-9535-6] [Medline: 29090428]
22. Subrahmanyam K, Lin G. Adolescents on the net: internet use and well-being. Adolescence 2007;42(168):659-677. [Medline: 18229503]
23. Barry CT, Sidoti CL, Briggs SM, Reiter SR, Lindsey RA. Adolescent social media use and mental health from adolescent and parent perspectives. J Adolesc 2017 Dec;61:1-11. [doi: 10.1016/j.adolescence.2017.08.005] [Medline: 28886571]
24. Barr SM, Budge SL, Adelson JL. Transgender community belongingness as a mediator between strength of transgender identity and well-being. J Couns Psychol 2016 Jan;63(1):87-97. [doi: 10.1037/cou0000127] [Medline: 26751157]
25. Hatchel T, Marx R. Understanding intersectionality and resiliency among transgender adolescents: exploring pathways among peer victimization, school belonging, and drug use. Int J Environ Res Public Health 2018 Jun 19;15(6):1289 [FREE Full text] [doi: 10.3390/ijerph15061289] [Medline: 29921806]

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26. Hatchel T, Valido A, De Pedro KT, Huang Y, Espelage DL. Minority stress among transgender adolescents: the role of peer victimization, school belonging, and ethnicity. J Child Fam Stud 2018 Jun 25;28(9):2467-2476. [doi: 10.1007/s10826-018-1168-3]
27. Eccles AM, Qualter P. Review: Alleviating loneliness in young people - a meta-analysis of interventions. Child Adolesc Ment Health 2021 Feb 13;26(1):17-33. [doi: 10.1111/camh.12389] [Medline: 32406165]
28. Liu D, Baumeister R, Yang CB, Hu B. Digital communication media use and psychological well-being: a meta-analysis. J Comput Mediat Commun 2019 Sep;24(5):259-274. [doi: 10.1093/jcmc/zmz013]
29. Vally Z, D'Souza CG. Abstinence from social media use, subjective well-being, stress, and loneliness. Perspect Psychiatr Care 2019 Oct 12;55(4):752-759. [doi: 10.1111/ppc.12431] [Medline: 31402459]
30. Ybarra ML, Mitchell KJ, Palmer NA, Reisner SL. Online social support as a buffer against online and offline peer and sexual victimization among U.S. LGBT and non-LGBT youth. Child Abuse Negl 2015 Jan;39:123-136 [FREE Full text] [doi: 10.1016/j.chiabu.2014.08.006] [Medline: 25192961]
31. Fish JN, McInroy LB, Paceley MS, Williams ND, Henderson S, Levine DS, et al. "I'm Kinda Stuck at Home With Unsupportive Parents Right Now": LGBTQ Youths' experiences with COVID-19 and the importance of online support. J Adolesc Health 2020 Sep;67(3):450-452 [FREE Full text] [doi: 10.1016/j.jadohealth.2020.06.002] [Medline: 32591304]
32. Dillman D, Smyth J, Christian L. Internet, Phone, Mail, and Mixed-Mode Surveys: The Tailored Design Method. Hoboken, New Jersey, United States: John Wiley & Sons; 2008:1-528.
33. Heen M, Lieberman J, Miethe T. A comparison of different online sampling approaches for generating national samples. Centre for Crime and Justice Policy, University of Neveda, Las Vegas. 2014. URL: https://www.unlv.edu/sites/default/files/page_files/27/ComparisonDifferentOnlineSampling.pdf [accessed 2021-07-14]
34. Moreno MA, Arseniev-Koehler A, Selkie E. Development and testing of a 3-item screening tool for problematic internet use. J Pediatr 2016 Sep;176:167-172 [FREE Full text] [doi: 10.1016/j.jpeds.2016.05.067] [Medline: 27395768]
35. Moreno MA, Binger K, Zhao Q, Eickhoff J. Measuring interests not minutes: development and validation of the adolescents' digital technology interactions and importance scale (ADTI). J Med Internet Res 2020 Feb 12;22(2):e16736 [FREE Full text] [doi: 10.2196/16736] [Medline: 32049068]
36. Tennant R, Hiller L, Fishwick R, Platt S, Joseph S, Weich S, et al. The Warwick-Edinburgh Mental Well-being Scale (WEMWBS): development and UK validation. Health Qual Life Outcomes 2007 Nov 27;5(1):63 [FREE Full text] [doi: 10.1186/1477-7525-5-63] [Medline: 18042300]
37. Hair E, Moore K, Garrett S, Kinukawa A, Lippman L, Michelson E. The parent-adolescent relationship scale. In: Moore K, Lippman L, editors. What Do Children Need to Flourish?. Boston: Springer; 2005:183-202.
38. Alsaker FD. Pubertal timing, overweight, and psychological adjustment. J Early Adolesc 1992;12(4):396-419. [doi: 10.1177/0272431692012004004]
39. Su R, Tay L, Diener E. The development and validation of the Comprehensive Inventory of Thriving (CIT) and the Brief Inventory of Thriving (BIT). Appl Psychol Health Well Being 2014 Nov 12;6(3):251-279. [doi: 10.1111/aphw.12027] [Medline: 24919454]
40. Elmer T, Mepham K, Stadtfeld C. Students under lockdown: Comparisons of students' social networks and mental health before and during the COVID-19 crisis in Switzerland. PLoS One 2020 Jul 23;15(7):e0236337 [FREE Full text] [doi: 10.1371/journal.pone.0236337] [Medline: 32702065]
41. Fuster H, Chamarro A, Oberst U. Fear of missing out, online social networking and mobile phone addiction: a latent profile approach. Aloma 2017 Oct 13;35(1):22-30. [doi: 10.51698/aloma.2017.35.1.22-30]
42. Franchina V, Abeele MV, van Rooij A, Lo Coco G, De Marez L. Fear of missing out as a predictor of problematic social media use and phubbing behavior among flemish adolescents. Int J Environ Res Public Health 2018 Oct 22;15(10):2319 [FREE Full text] [doi: 10.3390/ijerph15102319] [Medline: 30360407]
43. Przybylski AK, Murayama K, DeHaan CR, Gladwell V. Motivational, emotional, and behavioral correlates of fear of missing out. Comput Hum Behav 2013 Jul;29(4):1841-1848. [doi: 10.1016/j.chb.2013.02.014]
44. Reisner SL, Greytak EA, Parsons JT, Ybarra ML. Gender minority social stress in adolescence: disparities in adolescent bullying and substance use by gender identity. J Sex Res 2015 Apr 17;52(3):243-256 [FREE Full text] [doi: 10.1080/00224499.2014.886321] [Medline: 24742006]
45. Gender dysphoria. American Psychiatric Association. 2013. URL: https://www.psychiatry.org/File%20Library/Psychiatrists/Practice/DSM/APA_DSM-5-Gender-Dysphoria.pdf [accessed 2019-01-07]
46. Saul JS, Rodgers RF. Adolescent eating disorder risk and the online world. Child Adolesc Psychiatr Clin N Am 2018 Apr;27(2):221-228. [doi: 10.1016/j.chc.2017.11.011] [Medline: 29502748]
47. van de Grift T, Cohen-Kettenis P, Elaut E, De Cuypere G, Richter-Appelt H, Haraldsen I, et al. A network analysis of body satisfaction of people with gender dysphoria. Body Image 2016 Jun;17:184-190. [doi: 10.1016/j.bodyim.2016.04.002] [Medline: 27137814]
48. Haimson O. Mapping gender transition sentiment patterns via social media data: toward decreasing transgender mental health disparities. J Am Med Inform Assoc 2019 Aug 01;26(8-9):749-758 [FREE Full text] [doi: 10.1093/jamia/ocz056] [Medline: 31120498]

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49. Evans YN, Gridley SJ, Crouch J, Wang A, Moreno MA, Ahrens K, et al. Understanding online resource use by transgender youth and caregivers: a qualitative study. Transgend Health 2017 Dec;2(1):129-139 [FREE Full text] [doi: 10.1089/trgh.2017.0011] [Medline: 29082333]
50. Jackson SJ, Bailey M, Foucault Welles B. #GirlsLikeUs: Trans advocacy and community building online. New Media Soc 2017 Jun 09;20(5):1868-1888. [doi: 10.1177/1461444817709276]
51. Haimson O. Social media as social transition machinery. Proc ACM Hum-Comput Interact 2018 Nov;2(CSCW):1-21. [doi: 10.1145/3274332]
52. 2018 Dane County Youth Assessment: Dane County High Schools Combined Report. Dane County Youth Commission: Madison, WI. 2018. URL: https://www.dcdhs.com/About-Us/Commissions-Boards-and-Committees/Youth-Commission/Youth-Assessment [accessed 2021-07-14]
53. Rider GN, McMorris BJ, Gower AL, Coleman E, Eisenberg ME. Health and care utilization of transgender and gender nonconforming youth: a population-based study. Pediatrics 2018 Mar 05;141(3):e20171683 [FREE Full text] [doi: 10.1542/peds.2017-1683] [Medline: 29437861]
54. Herman J, Flores A, Brown T, Wilson B, Conron K. Age of individuals who identify as transgender in the United States. The Williams Institute: UCLA School of Law. 2017. URL: https://williamsinstitute.law.ucla.edu/publications/age-trans-individuals-us/ [accessed 2021-07-14]
55. United States Census Bureau. URL: https://www.census.gov/data.html [accessed 2021-03-15]
56. Raun T. Video blogging as a vehicle of transformation: Exploring the intersection between trans identity and information technology. Int J Cult Stud 2014 Jan 16;18(3):365-378. [doi: 10.1177/1367877913513696]

Abbreviations

ADTI: Adolescent Digital Technology Interactions and Importance
FOMO: fear of missing out
LGBTQ+: lesbian, gay, bisexual, transgender, queer, and questioning
PIU: problematic internet use
PRIUSS-3: Problematic and Risky Internet Use Screening Scale-3
TNG: transgender, nonbinary, and gender-diverse

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Original Paper

Development of a Mobile App to Improve Numeracy Skills of Children With Autism Spectrum Disorder: Participatory Design and Usability Study

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Abstract

Background: The use of information and communication technologies is transforming the lives of millions of people including children with autism spectrum disorder (ASD). However, the process of developing a user-friendly and effective mobile app needs to follow a complex standard protocol and culture-sensitive customization, and involves multiple sectors. This complex work becomes even more challenging when considering children with ASD in low- and middle-income countries as the users.

Objective: This study aimed to design and develop a more intuitive mobile app to improve numeracy skills of children with ASD in Rwanda and evaluate the usability of the app.

Methods: A participatory design approach was utilized in this study in which 40 children with ASD, 5 teachers, and 10 parents of children with ASD participated in focus group discussions (FGDs) and usability testing. A narrative literature review was performed to explore existing mobile apps and compare previous studies to design the questions for FGD and facilitate a framework for designing the app. The agile methodology was used to develop the mobile app, and the heuristics evaluation method was used to test and evaluate the usability of the initial version of the app to improve its functionalities. The interviews were recorded, transcribed, and analyzed following the guidelines of the qualitative narrative analysis (QNA) method.

Results: During the FGDs the respondents shared their need for a mobile app in teaching and learning numeracy for children with ASD and pointed to possibilities of integrating the mobile app into existing curriculum. Ten themes emerged from the FGDs and exercise of developing the mobile app. The themes were related to (1) teaching and learning numeracy for children with ASD, (2) planning and development of a mobile app for a person with ASD, (3) testing a mobile app, (4) strength of the developed app against the existing ones, (5) behavioral maintenance and relapse prevention, (6) possibilities to integrate the mobile app into the existing curriculum, (7) data protection for users, (8) social implications, (9) challenges in Rwanda, and (10) focus on future.

Conclusions: The community plays an important role in the planning, development, and evaluation of a mobile app for children with ASD. In this study, inputs from teachers and parents resulted in an optimally designed mobile app that can improve numeracy skills in children diagnosed with ASD to support the implementation of competency-based curriculum in Rwanda.

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KEYWORDS

autism spectrum disorder; mobile app; learning; information and communication technologies; education; numeracy; mathematics

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Introduction

Autism spectrum disorder (ASD) is a neuro-developmental disorder with persistent deficit in (1) social communication and social interaction and (2) restricted or repetitive pattern of behavior, interest, or activities from the developmental period [1]. The prevalence of autism among children is increasing as a result of different biopsychosocial and environmental factors as well as with the change in diagnostic criteria. Initially, it was considered that globally around 1% of children have autism; however, recent data from the Centers for Disease Control and Prevention (CDC) indicated that 1 in 54 children have autism in the United States. As the number of children with autism is increasing, their need for health care and education is also accelerating. Studies have consistently documented that children with autism can learn and acquire different life and living skills with appropriate training and support. The study by Kamaruzaman and Azahari [2] documented the potential of these children to learn and develop their skills in various domains such as mathematics. The use of systematic instruction, such as task-analytic instruction, proved the possibility of children with significant cognitive disabilities learning basic mathematics [3]. However, only few teachers are interested about the nature and extent of ASD symptoms that cause challenges in social communication for these children; besides, their unique behavioral patterns and need for intensive support are less recognized [4-6].

Computer-based interventions available on mobile devices improve the learning of children with ASD even when they are used with minimal supervision [7]. Different studies have suggested that electronic devices such as iPads and smartphones installed with assistive apps are effective in teaching children with ASD when additional features such as audio and video are included in the provided content [8]. Previous studies have found that children can learn numeracy skills through digital devices with touchscreen technology [9,10]. Many children with ASD are drawn toward computers as they tend to be visual learners. For this reason, e-learning systems are a natural choice for them as both static and dynamic images as well as videos can be included as part of the learning process [11].

Rwanda is promoting the use of information and communication technologies (ICTs) at all levels through multiple initiatives that include the One Laptop Per Child Project (for those at basic education levels) and loan schemes for students (for those in higher learning institutions) [12]. These interventions have increased the number of computer users [13] and reduced the access gap in ICT in both urban and rural populations. Rwanda's competence-based curriculum divides numeracy into different categories from level 1 primary (P1) to level 3 primary (P3) [14]. However, there are some children with ASD who have never attended the formal education system due to the stigma against them [15,16].

Previous studies by Tanner et al [17], Fairus et al [18], and Soomro and Soomro [19] revealed that mobile app technologies are crucial to improve the learning ability of individuals with ASD. The study by Rajagopal and Ying [20] revealed that mobile apps with repetitive instruction can improve learning numeracy skills for children with ASD. According to Díaz and Barco [21] and Alcantud et al [22], the development of assistive technologies for a person with ASD has increased in the last decade. However, few studies have taken place in sub-Saharan Africa and other low-income countries such as Rwanda [23]. According to De Macedo and Ulbricht [24], children with ASD show different behaviors, and hence parents and educators need to contextualize the content of the technological apps. Our previous work [5] revealed how worthy the development and tailor-made designs of mobile apps are and that these have immense potentiality to help improve the learning competencies of children with ASD in Rwandan schools. This study has been prompted by previous studies that revealed how mobile technologies enhance academic skills in different ways such as word matching or picture perception [25,26]. The introduction of mobile apps is the one of most attempted solutions to include persons with ASD in society [27], as the use of multimedia technologies can improve their writing skills and social communication.

The focus of children with ASD is compromised by the sensory onslaught in the learning environment which prevents them from paying attention when studying [28]. Assistive technologies such as virtual environments, augmented reality, and smart glasses [29,30] have been developed in recent years to improve the collaborative interactive environment in order to help children with ASD stay focused. The development of mobile apps to support individuals with ASD has been recommended by many researchers including Fraccaro et al [31] and Read et al [32]. However, mobile apps should be culture and context specific [12,33]. This can be achieved by including Indigenous features and examples of motivations available in their local context.

Different mobile apps such as iConverse [34] and SPEAK all! [35] have successfully improved the communication ability of children with ASD [36]. Studies by Kamaruzaman and Azahari [2] and Tashnim et al [37] introduced numeracy and calculation as a psychoanalysis to improve the lives of children with ASD. However, mobile apps designed for persons with ASD will be successful only when it is designed in response to zones of instructional opportunity, which include central coherence, a theory of mind, and executive function [38]. These zones of instruction outline a clear and validated structure of the specific instructional needs of the user, considering that the deficits and impairment associated with ASD impact the user's learning style and also reduce the number of new skills memorized, so as to allow them to refer materials later to make the appropriate decision [39,40]. This information is significant in the argument for using mobile apps to teach and instruct children with ASD because it helps them to focus their attention on a particular topic/skill [38]. Khorrami et al [41] reported that there is a tendency among children with ASD to read by decoding before establishing an understanding of comprehension. Many studies have sought to teach simple mathematical skills (eg, number matching, counting) [42] by addressing the deficit in communication as a common impairment in children with ASD [43]. Rwanda is implementing the competency-based curriculum to develop skills for all children regardless of their disabilities from preprimary to upper secondary education [44]. Children

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with ASD are interested in using digital tools and their teachers and parents believe that the use of ICTs might improve their learning when the tools are developed by considering their sociocultural context [45,46]. However, how such a local culture–sensitive digital tool can be developed is not yet documented in Rwanda. This study aimed to fill this gap in knowledge by developing a user-friendly mobile app to improve numeracy skills in children with ASD level 3 in Rwanda.

Methods

Design

This participatory design and usability study was performed from November 2019 to June 2020 and intended to design a mobile app to improve numeracy skills in children with ASD. Mixed methodologies were employed in this study: (1) a participatory design approach [47] to improve the outcomes due to the context-sensitive need and a future-oriented approach to the design of technological solutions by involving workers and professionals from the field of education of children with ASD in the design; (2) a narrative literature review [48] to compare our app with existing mobile apps and previous studies. For this purpose, the agile [49] methodology, which considers customers, developers, stakeholders, and end users, was used to inspect the app elements at every stage of the development process and make adjustments according to the requirement; (3) a heuristics evaluation that used focus group and observation was utilized to test and evaluate the initial version of the app to improve its functionalities [50,51].

Participants

The local participants were selected based on data from our previous study [5]. The international participants were randomly selected based on their responses to 5 questions posted on the Quora platform [52], which is an open public forum to exchange ideas.

The participants of this study were children with ASD, teachers, parents, and the international community. A total of 40 children with ASD (32 boys and 8 girls) participated in testing the developed app. Among these, 5 children (4 boys and 1 girl) were not enrolled in schools and thus were visited at their home with their parents. The remaining 35 children (30 boys and 5 girls) were enrolled in Autisme Rwanda [53], which is the center established in 2014 in Rwanda that cares for children with ASD. Five teachers (2 males and 3 females) were recruited from Autisme Rwanda. Parents (n=10) of children with ASD who participated in the design and evaluation of the app were recruited from the Rwanda Parent's Initiative on Autism (RPIA) [54], which is an association of parents having children with ASD in their families in Rwanda. Within the parents' group, there were 4 fathers and 6 mothers.

Interview Guide and App Development

A series of open questions were used as the interview guide during focus group discussions (FGDs), which is a way to gather people from similar backgrounds or experiences to discuss a specific topic of interest (Multimedia Appendix 1). Some of these questions were posted on the Quora forum [52] (Multimedia Appendix 2) to collect data from the international community.

The design of the mobile app was guided by the Autism Spectrum Disorder Inclusion Collaboration Model [55]. The user interface is designed by following the principles of the Universal Mobile Application Accessibility and Inclusion [56] and included the following: (1) perceivable (to ensure that content is discernible by all users), (2) operable (ie, all features should be fully employable by everyone, regardless of the limitations of the user), (3) understandable (relates to the cognitive ability of the user to comprehend the meaning of the presented information), and (4) robust (ie, its content is flexible so it can be easily interpreted by an array of users).

The development of the app was accomplished in 2 steps: (1) user requirement analysis and (2) design process, as recommended by Soomro and Soomro [19].

User Requirement Analysis

The app that will be designed for the education of children with ASD needed a set of requirements to improve the probability of successful learning [56]. We used detailed methods and developed a list of possible requirements from existing studies and participants [52,53,57]. The FGDs were conducted by involving teachers of special education and practitioners in the fields as participants while the children with ASD observed their behavior inside and outside the class.

Information on the software interaction design was gathered from experts around the world who had experience in treating and dealing with children with ASD as well as individuals with ASD who were successful in their academic careers. This involved posting questions on the Quora Digest platform [52] by one of the authors and analyzing their replies. We chose to use Quora as our data source because (1) Google Trends data show that it is increasing in popularity against other platforms available globally [58]; (2) it tries to match questions with experts and most questions are answered by users with authority on the content [59]; and (3) it also offers users the ability to edit the way a question is asked, and thus allows users to connect people with questions they feel the user could answer [60]. The information collected from this platform served as complimentary to data collected from teachers and parents. The profiles and responses from the Quora platform are shared as web links in Multimedia Appendix 1.

Design Process

To understand the strength of ICT devices and examine the effectiveness of available apps for students with ASD, we next designed the mobile app interface that can respond to the needs of children with ASD in the current context. From the requirements analysis, we identified a set of characteristics needed for the software interaction, as explained by Mejía et al [25], and using the following equations:

$$U=\{Sn,Cg,Mp\} \quad \textbf{(1)}$$

$$Sn=\{Sn_1, ..., Sn_n\} \quad \textbf{(2)}$$

$$Cg=\{Cg_1, ..., Cg_n\} \quad \textbf{(3)}$$

$$Mp = \{Mp_1, ..., Mp_n\} \quad \textbf{(4)}$$

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where *U* represents a user (Equation 1), *Sn* represents the set of the senses (Equation 2), such as eyesight, hearing, and touch; *Cg* represents cognitive functions, such as memory and attention (Equation 3); and *Mp* represents a motor function for different parts of the body (Equation 4).

Designing a task-centered user interface for the app involved multiple processes, according to the established guidelines and as recommended by Punchoojit and Hongwarittorrn [55].

The steps in the task-centered design process were adapted and modified as necessary during the aforesaid process. We also referred to the universal mobile app accessibility and inclusion guidelines proposed by Ballantyne et al [56]. While designing the mobile app, 9 steps were followed: (1) task analysis, (2) choose the representative tasks, (3) find existing interfaces, (4) rough out the design, (5) analyze the user interaction, (6) create a prototype, (7) test design with users, (8) iterate, and (9) build the design.

Task Analysis

To design this app, we first agreed on the target audience, as the background of users (ie, children with ASD) will help the designer to include elements in the design considering their ability.

In this process, we referred to the previous work by Díaz and Barco [21], where a software (*S*) is defined a set of functionalities (*F*) operated by the user, which also includes a user interface (*I*):

$$S=\{\{F_1, \ldots, F_n\}I\} \quad \textbf{(5)}$$

In this mobile app, functionalities are defined as tasks (T) that can be done.

$$S=\{\{T_1, \ldots, T_n\}I\} \quad \textbf{(6)}$$

Children with ASD level 3 [1] are the only actors in the system.

The mobile app includes a graphical user interface (*I*) that allows users to interact with the system. From Equation 6, the interface is

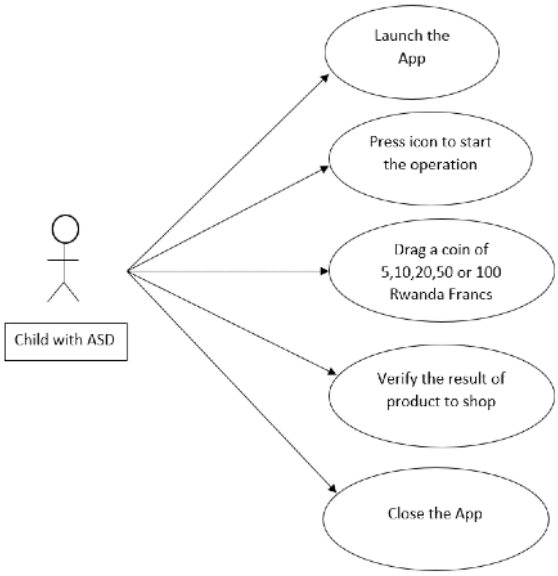
$$I=\{Uip_1, \ldots, Uip_n\} \quad \textbf{(7)}$$

$$Uip=\{\{Cp\},\{C\}\} \quad \textbf{(8)}$$

Equations 7 and 8 show that an interface (*I*) is a set of user interface patterns (*Uip*) which is a group of graphic user interface–style *Cp* to solve a particular problem. It is also associated with specific characteristics of users (*C*).

The use case diagram (Figure 1) illustrates the different tasks to be performed by a user.

Figure 1. Use case diagram of the system (app). ASD: autism spectrum disorder.



From Figure 1, it can be seen that each task is associated with a user interface pattern *Uip*.

$$\forall T{:}Uip \quad \textbf{(9)}$$

where ∀T represents the universal quantifier, meaning that for all cases, *T* (task) is associated with a user interface.

From Equation 9, the task includes all actions executed by the user.

$$T=\{Ac_1, \ldots, Ac_n\} \quad \textbf{(10)}$$

In this app, actions are press, drag, and drop coins into a specific area.

$$T=\{\text{Press, Drag, Drop}\} \quad \textbf{(11)}$$

A child with ASD drags at least one coin of the Rwandan currency system and drops it into a provided space to get the result. Specific feedback (eg, candy, banana, strawberries, and donuts) comes immediately after a successful operation. For all users with the definite ability (A), these gifts exist as user interface patterns.

$$(\forall U \boxed{\times} A) \boxed{\times} Uip \quad \textbf{(12)}$$

The user interface pattern has coins that the user can drag and drop into the specified place, which are added up before getting a reward.

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Choose Representative Tasks

In this step, the mobile app designer analyzed a syllabus of skip counting in the competence-based curriculum [44]. Participants suggested numbers and coins to represent tasks in this app. The flowchart (Figure 2) illustrates the process of learning skip counting through the developed app.

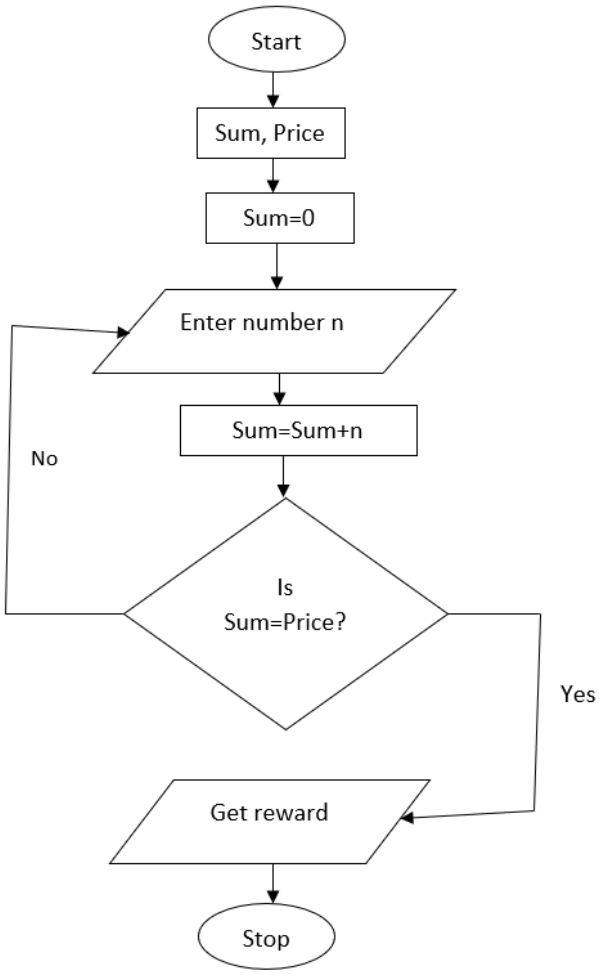
From the flowchart (Figure 2), the proposed number is given as follows:

$$N = \Sigma(V_0, ..., V_n) \quad \textbf{(13)}$$

where N is the sum of the expected value of coins and $\Sigma(V_0, ..., V_n)$ is the sum of the current value of coins in the target space plus the value of the dropped coin V_n.

$$\Sigma(V_0, ..., V_n) = V_0 + V_1, \cdots, + V_n \quad \textbf{(14)}$$

Figure 2. Flowchart of the developed app.



Find Existing Interfaces

In this step, we identified existing interfaces such as iConverse [35], SPEAK all! [36], and 123 number [61] to build ideas into the system as much as practically possible prior to making it as a reference to build the actual system. These apps were installed on smartphones and tablets and given to the children to observe their ability to use these interfaces. The results from this observation were closer to those from our previous study, in which children with ASD were able to use digital gadgets when the apps installed are easy for them to use [5]. The same methodology of observation and FGDs was used in data collection of that study.

This step helped researchers to design a good user interface which depends on how often the users will be using the system compared with how often they will be using systems that they already know [58].

Rough Out the Design

First, the preliminary (rough) description of the design is penciled on a paper. In this stage, the researchers and education practitioners had in-depth discussions on the features the system should have before building a prototype so that it can be tested out with the end users. Figure 3 presents an example of the design that simulates the intended interface.

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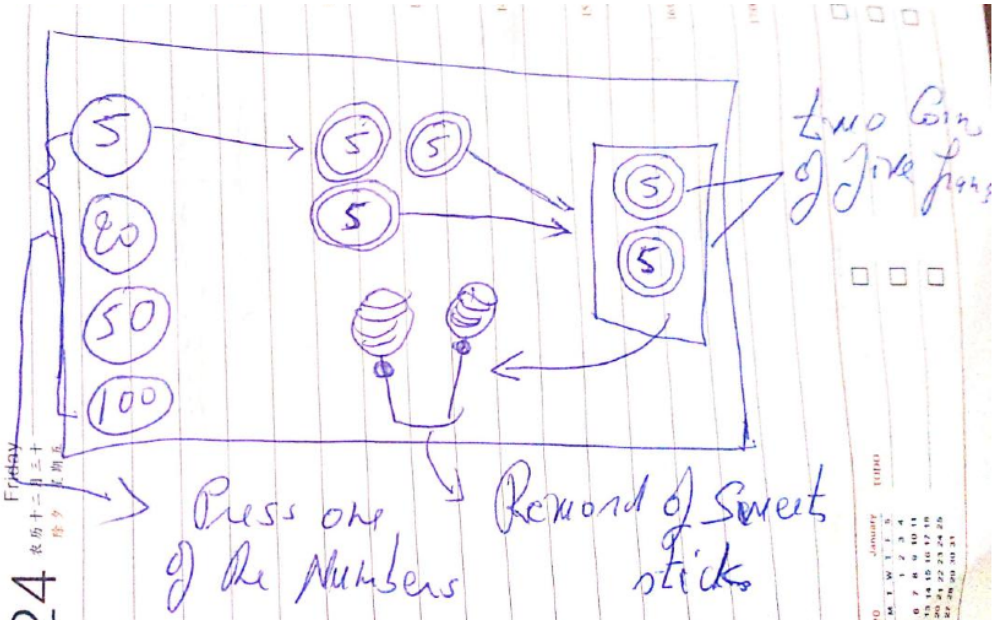
Figure 3. Rough out the design.



As can be seen in the figure, a child with ASD presses and drags 1 image of a coin and drops it into the designed space at the right-hand side. The system will count the number of currencies the child puts into the space to present the reward. The child can repeat the same action as much as s/he can.

Analyze User Interaction

Before designing the mock-up or prototype, we first analyzed how the users can interact with the interface that has been roughed out while performing specific tasks. At this stage, researchers identified areas where the users might make mistakes, such as pressing the wrong icon of coins or trying to go back. The analysis of the temporary interface was also supported by results from previous steps and experiences of study participants.

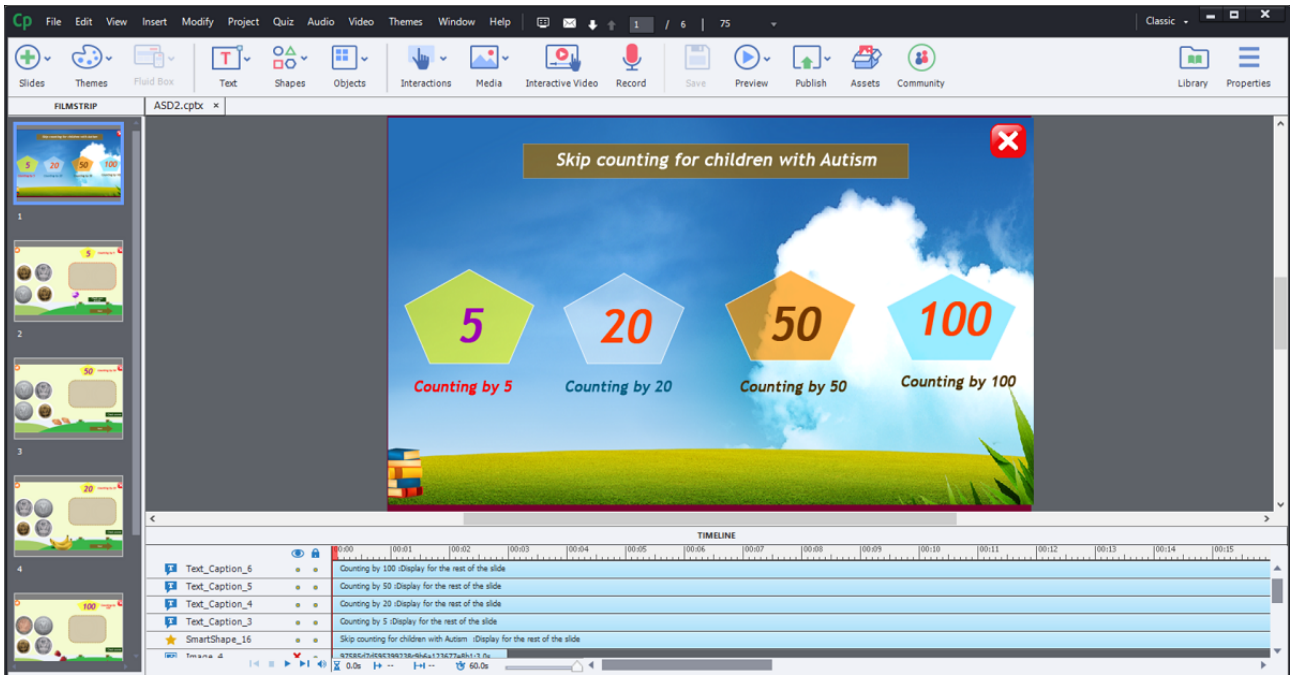
To use the developed app, children with ASD level 3 will get support from their teachers, parents, or family members who live with them.

Create a Prototype

The prototype of this app was designed using Adobe Captivate version 19.0 [62], which has features to design a responsive interface and is compatible with both Android [63], the common platform installed in smartphones, and iOS, installed on Apple devices [64].

This prototype was made only to find flaws in the system and ways to improve them in terms of functionality and usability. The Adobe Captivate interface (Figure 4) shows the template used to design the user interface of the mobile app.

Figure 4. Adobe Captivate interface.



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Test Design With Users

Afterward, the prototype built was tested by chosen users at Autisme Rwanda to bridge the gap that might still exist. This stage helped researchers to further improve the system to suit children with ASD in Rwanda. The researchers were then able to analyze the list of features that may need to be improved. After testing the interface with the children, FGD was completed with educators at the Autisme Rwanda Center to get more information on the interface design.

The children were identified randomly from the class, regardless of their subject and the economic status of their parents. The gender did not influence children's interaction during the test.

Both teachers and parents helped researchers in the recruitment of children that tested the app and continuously supported us in monitoring the changes in the behavior of these children. Parents of children who are not in school supported us by monitoring their children when they used the app at their homes during the test period. Every child that participated in this study was given a tablet on which the app was installed. The children used the app for a 30-day period.

Iterate

Based on the test phase results, the participants (teachers and parents) reported problems that need to be accounted for, such as changes in the background colors of the app, images, and feedback messages. The researchers re-designed the app to make the recommended changes and reverified it with the participants. The design of the interface was revised 2 times.

Build the Design

The final step in the development of this app before it may be commercialized is building the product. We built the app after solving the problems identified in the previous steps and FGDs. Continuous testing was performed to modify the interface according to the needs of children with ASD whose behavior may vary from one another.

The developed app was installed on smartphones and tablets that will be used in the next study to evaluate the impact of this app on children who will be using it.

Ethics

With respect of research ethics, a letter of consent (Multimedia Appendix 3) was signed by each teacher and parent of children with ASD who participated in the study. Participation in the study was entirely voluntary and no special reward was given to the participants at the end of sessions as an acknowledgment of their time. Ethical clearance (Multimedia Appendix 4) was obtained from the Collegial Research Ethical Committee of the University of Rwanda – College of Education. The collaboration letter between the principal investigator (TN) and Autisme Rwanda was signed before the start of the study (Multimedia Appendix 5). The collected data are stored in a confidential and secure place on a physical memory drive.

Results

Identified Themes

A total of 10 themes emerged while developing this mobile app that responds to the needs of children with ASD in order to improve their numeracy skills (basic mathematics). The themes are related to (1) teaching and learning numeracy for children with ASD, (2) planning and development of a mobile app for a person with ASD, (3) testing a mobile app, (4) strength of the developed app against the existing ones, (5) behavioral maintenance and relapse prevention, (6) possibilities to integrate the mobile app into existing curriculum, (7) data protection for users, (8) social implications, (9) challenges in Rwanda, and (10) focus on future.

Teaching and Learning Numeracy for Children With ASD

This theme elucidates models and techniques used to support learning numeracy for children with ASD. Teachers understood the application of the applied behavior analysis model in a classroom environment and were trying to implement some of its strategies to motivate children with ASD to stay focused. This was supported by a common statement from teachers which says:

> *Learners with Autism need rewards like giving them a pen or a toy to encourage them to study. We use available tools in the school like pens or rulers to motivate them to learn.*

This succeeded when the teachers considered the needs of each child, as they are different across the spectrum. This is supported by an international expert who replied to the question posted on a public forum:

> *You must consider the needs of each child, as everybody on the spectrum is different. Some people on the Autism Spectrum might find it difficult even to do simple math problems, while others may find even complicated math problems too easy. Some may need more help with the X and Y values, while others may need more help with counting money.*

This statement suggests that individuals with ASD do not learn the same way and thus the methods for teaching them need to be different. In the public forum, a person with Asperger syndrome said

> *Well, the first flaw in your question is that you assume we all learn the same. We don't. However, if you take the approach that all individuals have different learning styles, and you try to incorporate that into your app, you might find a modicum of success.*

Changes in methodologies and integration of digital tools that respond to the individual needs of a child with ASD can help teachers teach numeracy skills for these children.

Planning and Development of a Mobile App for a Person With ASD

This theme explains the approach to follow when planning and developing a mobile app for children with ASD. It is important

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to familiarize the environment in which children with ASD live. This is confirmed by the period for which a researcher spent time in the school caring for children with ASD and the collaboration letter signed by both the manager of the center and the principal investigator (TN) of this research. While planning the development of an app that helps children with ASD learn, the involved collaborators sat together and examined the behavior of children with ASD under different conditions.

Equations 1-4 show that to design an interface for children with ASD, the designer considered (1) the senses of the children such as vision, hearing, and tactile; (2) cognitive level of a child to perform a given task; and (3) the ability to exercise motor functions to perform a task.

A list of user's requirements (Table 1) was collected from participants when planning the development of the mobile app.

Table 1. List of user's requirements.

Requirements/Difficulties	Requirements	Methods for gathering requirements
• Senses (Sn)	• For each action, there must be a voice associated with it. • Use clear images of coins from the Rwandan currency system. • Number of images of coins should be ≤5. • The size of icons/coins must be big enough to enable the precisions of children when dragging them to the right place. • Ignore the background sounds.	• Group interviews, observation, literature review, and online open discussion.
• Cognitive functions (Cg)	• Use of a soft sound that can attract children to use the mobile app. • Facilitate direct feedback using an image of the product that is liked by children, such as doughnuts, bananas, and candy. • Reduce background colors that can distract children. Better to use 1 background image whose existence is known to children. • Use a clear image that tells the user to return to the home screen or to close the app.	• Group interviews, observation, literature review, and online open discussion.
• Motor function (Mp)	• The user interface needs to be responsive to devices with high resolutions such as tablets. • Allow the user to repeat actions until the dragged object reaches the target. • Choose the app to be in a landscape position.	• Group interviews, observation, and literature review.

Equations 1, 2, 3, and 5 were also supported by a common response from the global practitioners who replied to the questions posted on the forum that the app design should take advantage of the interaction strengths by making the accessibility limited to skill instruction and improve information processing by being both engaging and enjoyable.

The participants suggested 3 points to consider when designing the interface for children with ASD: (1) graphic design guidelines, (2) user interface design, and (3) success recognition and messages.

Graphic Design Guidelines

To design the graphical interface, we followed the recommendations by Hussain et al [65] who suggested the interface, number of pictures, size of the screen, icons, colors, and content to ensure the comfort of children with ASD when they use mobile apps. The graphic interface is made up of 2 main menus: (1) home and (2) exit.

The design flow (Figure 5) illustrates the app's structure that facilitates users to navigate.

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Figure 5. Design flow of the app.

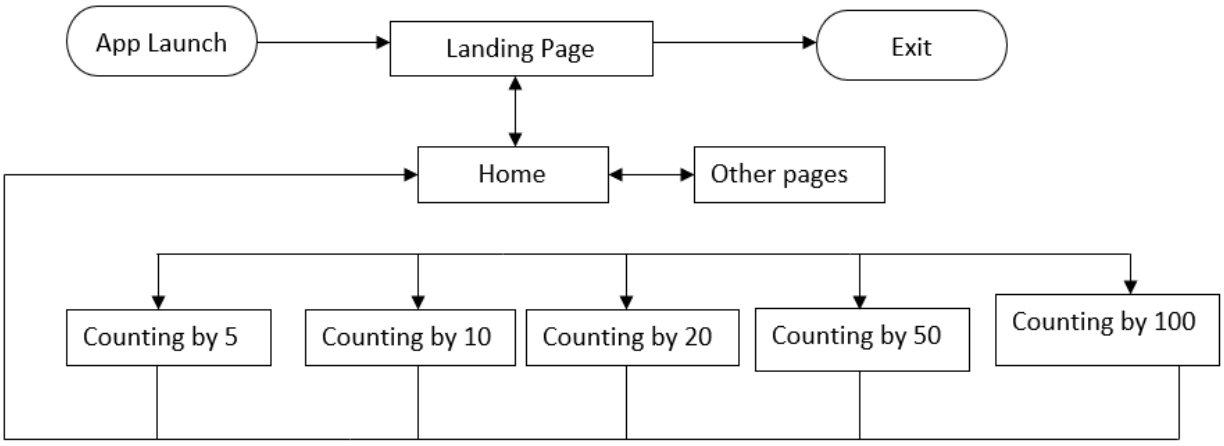

Figure 5 shows that the home menu is the starting point when the app is opened while the exit icon is used only to close it. Equation 5 defined a software app as a set of functions that are associated with user interface. Home menu, exit, and images that illustrate numbers to count are the elements of the developed mobile app's interface. Equation 6 substituted the functionalities with tasks that users are expected to do within a software app via an interface. In this app, a child with ASD presses one of the figures to start counting numbers.

Equation 7 shows that an interface (*I*) is a set of user interface patterns (*Uip*) that facilitate users to interact with the system. From the home menu, a user can navigate through the system by touching 1 out of 5 icons: (1) counting by 5, (2) counting by 10, (3) counting by 20, (4) counting by 50, and (5) counting by 100. Equation 8 shows that a child with ASD who presents different characteristics presses one of these icons to start counting to achieve a certain goal.

Equation 9 demonstrates that under each submenu, a user counts Rwandan francs coins [66] that correspond to the number he or she heard from the voice (Equations 1-4).

To perform a task presented in Equations 10 and 11, a child with ASD drags at least one coin to a provided space to get the feedback (eg, candy, banana, strawberries, and others). The feedback for each action is the interfaces proposed in Equation 12.

User Interface Design

The user interface is mainly divided into 2 categories based on menus specified in Figure 5: (1) landing page and (2) other pages.

Landing Page

After launching the app, the landing page shows all categories of skip counting numbers. The images of currency were not put on the home page to prevent the loss of concentration of children as recommended in Equation 3. Figure 6 shows the design of the landing page of the app, which presents 4 options, among which a user can press one to initiate the operation (Equations 5 and 6).

Figure 6. The landing page of the app.

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Other Pages

Other pages become viewable after pressing an icon from the landing page as indicated in Equation 9. Each task is associated with a specific use interface (Equation 6). The interface contains a set of coins that are used in the Rwandan currency system, namely, 5 francs, 10 francs, 20 francs, 50 francs, and 100 francs [65]. The child is asked to drag coins one by one to the destination place to get a reward. Figures 7 and 8 show examples of the pages of counting by 5 before and after an operation, respectively. A child identifies coins of 5 francs and drags them into the box at the right-hand side and then after a successful operation s/he is given a reward of candy (Equations 10 and 11).

A child is asked to drag and drop coins of 5 francs to get a candy (Figure 7). Equation 13 shows that the current value is set to 0 initially, with values added after dropping each coin, as presented in Equation 14.

Using an example of counting by 5, the value of Equation 14 is as follows:

$$N=V_0+V_1+\cdots+V_n$$

$$V_0=0,\ V_1=5,\ V_2=5$$

$$N=0+5+5$$

$$N=10$$

Thus, the price of 1 candy is 10 Rwandan francs. Figure 8 shows the gift of candy offered to a child after a successful operation. In each step, the child with ASD has an interface containing the image, text, and audio (Equation 9).

Figure 7. Counting by 5 interface.

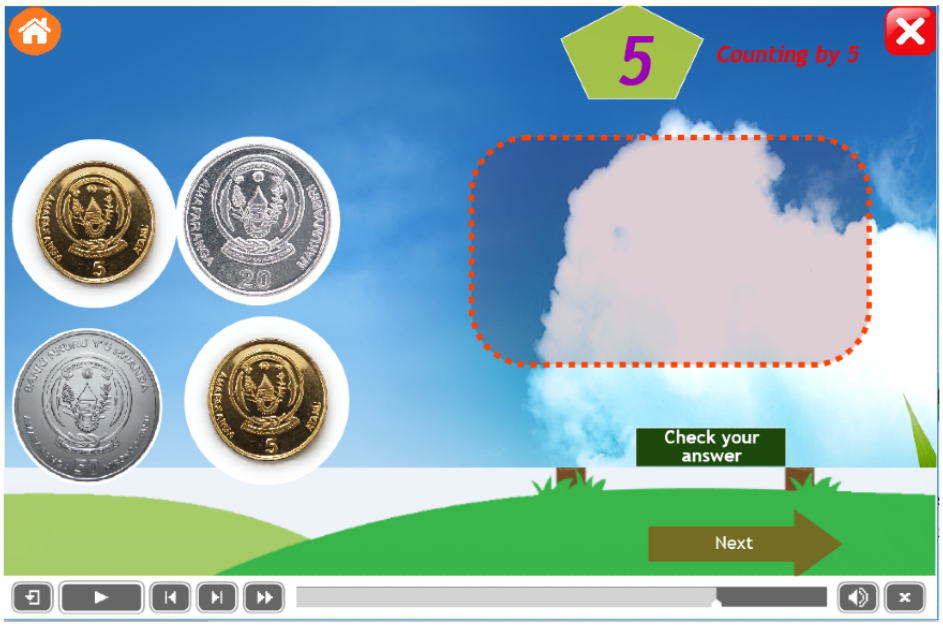
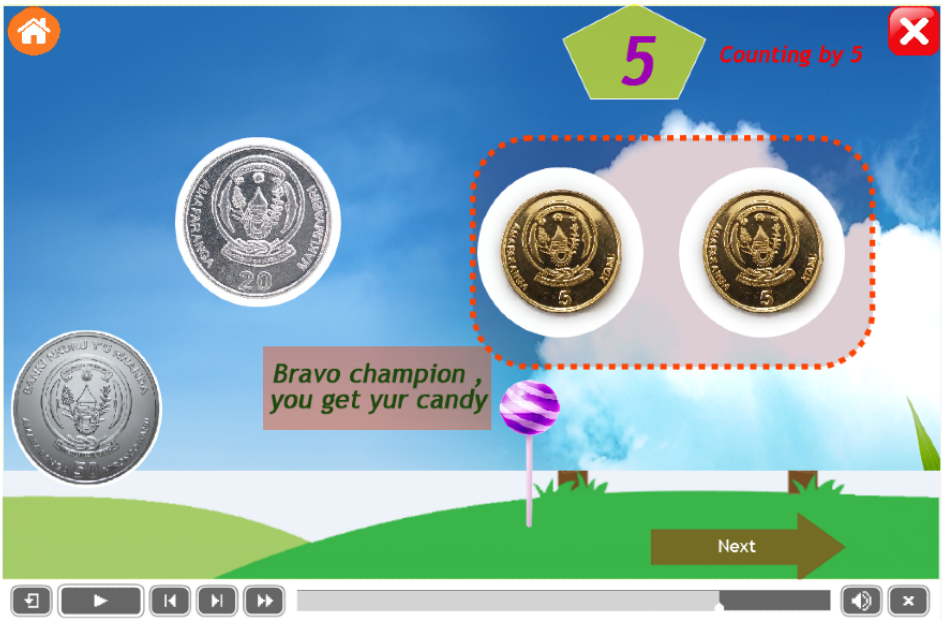


Figure 8. The interface of counting by 5 after dragging 2 coins of 5 francs.



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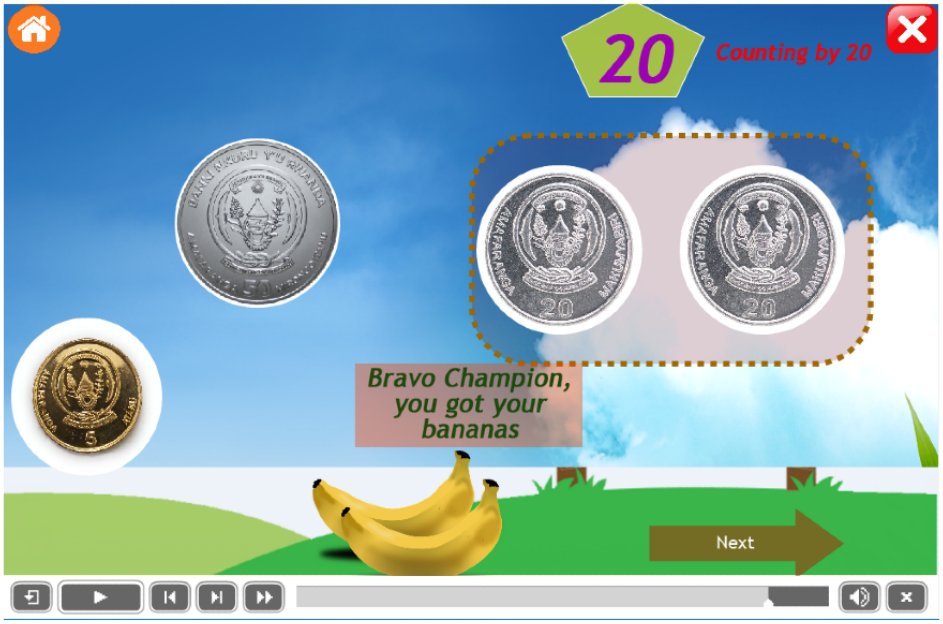
Success Recognition and Messages

To encourage children with ASD, it was recommended to award a special gift at every step of the operation. This mobile app provides different rewards that are common in their surrounding environment, such as doughnuts, bananas, and candies. Some example are as follows: the reward of bananas after dragging 2 coins of 20 francs (Figure 9), the reward of doughnuts after dragging 2 coins of 50 francs (Figure 10), and the reward of strawberries after dragging 2 coins of 100 francs (Figure 11).

Figure 9. The reward of bananas after dragging 2 coins of 20 francs.



Children with ASD were interested in the recognition message and image of the product which was offered to them after successfully completing an operation. This was supported by a teacher who said:

> *Children are most excited with the sound of congratulations after they succeed to count coins. As an educator, I found that this application can help much in my teaching activities when it will be improved to cover the whole curriculum.*

The integration of visual and audio features in the ICT-enabled technologies can improve the attention of children with ASD when they are learning. In this app, every action is associated with a sound that informs the child what s/he is expected to do to get a reward.

Figure 10 shows a reward of 2 doughnuts after a child successfully counted 2 coins of 50 Rwandan francs. Counting 2 coins makes the child happy because of the reward—2 doughnuts. Continuing the exercises in this app stimulates children to be engaged in using the local currency to buy basic needs.

The changes in the product of reward encourage children to continue using the mobile app to find the next message and product. Figure 11 shows that the child succeeded in dragging 1 coin of 100 Rwandan francs and got a reward of 2 strawberries.

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Figure 10. The reward of doughnuts after dragging 2 coins of 50 francs.

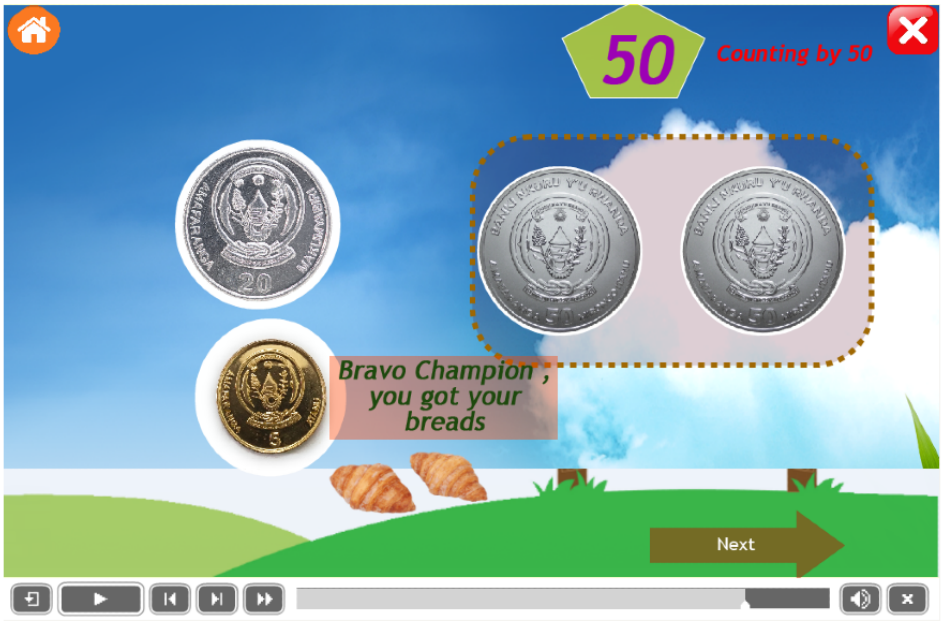

Figure 11. The reward of strawberries after dragging 2 coins of 100 francs.

The children who use the developed mobile app get a reward after successfully counting the number of coins that are equal to the cost of the product (reward) set in the system. The children get a message to try again when s/he dragged the wrong coin.

Testing a Mobile App

This theme demonstrates the result of testing the developed app. A total of 35 children in the Autisme Rwanda tested the app, which was monitored by the teachers in the center. Five children who are not in the center were visited at their homes. Table 2 presents an overview of staff and children at the center.

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Table 2. Status of Autisme Rwanda in 2020.

Information on children and staff	Value
Children, n	
Male	38
Female	7
Total	45
Children age (years), range	2-12
Staff, n	
Teachers	12
Teachers' assistants	8
Technicians and hygienic personnel	16
Total	36

An observation of the children using the developed app demonstrated its success, as it answered to their individual needs.

Findings from the test showed that it is crucial to customize the app in different forms, but with the same goal, so as to cater to each child's behavior.

This was supported by a comment by a participant on the Quora platform who responded to the question regarding the background color to be used in the app:

> *That's totally up to the child. Many of them don't show any special indication of liking certain colors. When my autistic son was young, he liked orange, but after about the age of seven, he wore nothing but black. He's 36 years old and still wears nothing but black. Occasionally we can talk him into charcoal, but it doesn't last long.*

The development of apps for children with ASD is a long process to accompany an individual in his/her life. It is therefore crucial to always change the content and format of the app to respond to the behavior of each child with ASD. These changes are also supported by another parent from the Quora forum who said

> *My son loves very intense, bright colors. But that doesn't mean everyone on the spectrum would. They are all individuals.*

While suggestions were reported by some users, many found the app promising to improve the learning of children with ASD in Rwanda. This was supported by a parent who said,

> *What I can see is that by using this application, my kid will gradually increase the knowledge in counting money and I hope it will help him to use that knowledge in the society even though he is not enrolled in the school.*

Using ICT tools, children with ASD learn more skills, which they can employ in their daily lives without being at school.

Children using the app showed more interest in continuing to use with the app, as they are familiar with the images of coins used in the app, which were similar to the ones they use in their families. This was supported by one parent who said,

> *My son was very interested in using this application because he always sees the coins we use at home. I believe within the time; my child will be able to count money.*

Children with ASD can learn counting from the electronic device when the app is contextualized with the environment.

Table 3 shows the phases in which 55 participants participated during the design, development, test, and evaluation of the mobile app.

Table 3. The number of participants.

Participants	Design and evaluation, n	Development, n	Test, n	Total, n
Teacher	5	0	0	5
Parents	10	0	0	10
Children with ASD enrolled in a school	0	0	35	35
Children with ASD not enrolled in school	0	0	5	5
Total	15	0	40	55

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Strength of the Developed App Against the Existing Apps

This theme confirmed the positive effect of the app on children with ASD in their learning of numeracy skills. However, the result from the observation of children with ASD using SPEAK all! [36] and 123 number [61] indicated that it was difficult to adapt the existing interface to the needs of children with ASD when they were developed in a context different from what they are currently living in.

This was supported by all teachers who participated in this study. One of the teachers said,

> *Normally these children face the problem of communication in general. It is very difficult for them to learn new content which is in the foreign language and the content is not adapted to the local environment.*

Many teachers and parents suggested re-creation of content adapted to the existing environment to enable children with ASD to learn from the objects they are familiar with.

The participants found the developed mobile app to have more impact, compared with other apps, on the learning of children with ASD in Rwanda.

This was supported by a common statement from parents:

> *This application will have a positive impact on the learning of our children because the coins used in the application are those we use in our daily life.*

Despite this positive statement, participants suggested the need to have the app in the local language. Children with ASD have difficulties in verbal communication. Thus, it is crucial to use their mother tongue to enable them to learn new skills.

Behavioral Maintenance and Relapse Prevention

Children with ASD tend to lose focus when communicating with others. This mostly happens due to distraction, which is commonly noted in these children in an inclusive environment. The instant motivational feedback when a child completes the operation makes him/her focus on using the app.

This is supported by an autism consultant who replied to a question posted on the Quora forum:

> *Just because they might not look at you does not mean that they are not paying attention or focusing. Is their ability to focus compromised by the sensory onslaught that is going on in the room?*

The participants also suggested the consideration of different factors such as level of noise in the room, smells, and movement that can make children with ASD lose their focus. These factors can be also associated with the health conditions as reported by the same consultant.

> *Some autistic people have difficulty with interception, understanding messages from our body. The child might have a headache, earache, stomachache, etc. but not be able to let you know. Or, this might be a constant state for them, again, making it harder to focus.*

The parents who participated in the study suggested giving time to the children when they are not following the instructions.

> *The children with ASD generally do not follow all the instructions we are giving them, but we are surprised to see them practicing what we were expected from them to do when they follow the instructions, and we hope with ICT can help better to improve their level of skills.* [A common statement from parents]

The provision of educational mobile apps that provide instant feedback at every step can help children with ASD stay focused and gradually improve their level of skills in different subjects.

Possibilities to Integrate the Mobile App Into Existing Curriculum

Teachers who participated in this study said that children with ASD enjoyed using digital devices that have audio and visual features. Thus, it is recommended to provide digital devices in the class with content related to the intended learning outcomes.

> *ICT can bring a positive impact when they are used in class, but it is better to select applications that can respond to the planned objective.* [A common statement from teachers]

The results from the test phase of the developed app were promising for its use in one section of the mathematics syllabus in a competency-based curriculum.

> *I found that the level of children with ASD in my class has been improved after using this application. However, it takes much time to learn from it compared to normal students.*

Learning a new subject for children with ASD takes longer than that for normal students. Therefore, the teachers suggested additional time to the time planned in the curriculum for normal students.

Response from parents proved that it is possible to learn basic mathematics outside the classroom by using ICT-enabled assistive technologies.

> *my kid is now trying to count money after using this mobile application. But it took time to learn it, it requires more time to expose the child to the application and play with it.* [Mother of a child with ASD]

Changes in the structure of the curriculum and integration of ICT in every section of the syllabus are recommended to include children with ASD in the normal educational system. This succeeds when the changes in methods of teaching children are also adopted.

> *An autistic child can learn anything that a neurotypical child can. The challenge is in the different approach to teaching (with careful consideration to the different way their brain/mind works) (e.g. using pictures rather than words to explain concepts, or using tactile feeling to aid memory/understanding), and the learning environment is accepting of and accommodating their*

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> *needs.* [Response from Adult diagnosed on the autism spectrum from Quora forum]

The adaptation of content to children with ASD is possible when the use of text in teaching is reduced and the ICT is empowered with audio–visual features.

Data Protection for Users

Privacy of information about users of mobile apps remains critical among participants.

The participants suggested not to expose any information about children with ASD. This information includes their images and information about their families. This policy was also supported by the school management which prohibited the use of children's photograph within the app.

Parents suggested disabling the camera when children are using the mobile app and preventing the app from taking any other information from children when using it.

> *Usually in the community, there is a misunderstanding of the cause of the ASD. We accept our children, but we don't want to share their images outside unless they are at school.* [A common statement from parents]

This is also supported by a teacher who said,

> *I remember when I was doing my academic research, one parent told me that many people are coming to visit him to do marketing of his child for business purposes. Now I cannot not allow anyone to take a picture of my child.*

Although the use of ICT suggests the possibility of improving the learning ability of children with ASD, some features such as pervasive monitoring, facial recognition, biometrics, and blanket data retention practices are not recommended to be used in the educational app for children with ASD.

Social Implications

This theme illustrates the social benefits and challenges associated with the use of the mobile app. Children with ASD have limited social communication skills, which is a major barrier to their learning. Thus, the participants expected the mobile app to improve the social relationship among children with ASD and the community.

> *I believe that the continuous use of mobile applications that are designed by considering this environment can also make children cope with the environment like going to shop, bringing some products and others.* [A father of a child with ASD]

Children with ASD can benefit from the practical knowledge of using money, thereby improving their relationship and communication with economic dealers.

However, some participants also reported a negative effect of the use of a mobile app for children with ASD that can affect their lives. They suggested time limit to restrict the continuous use of the app by children, which will also allow them to learn from other normal persons while not solely depending on the app for learning.

> *Using the mobile application is helpful to our children, however allowing them to learn only from the application can make them addicted with technology.* [A common statement from teachers]

Thus, mixing the technology with traditional educational interventions for children with ASD can bring about a positive impact on the life of these children and improve their social living with the community.

Challenges in Rwanda

Changes in behavior and feelings among children with ASD may be a barrier to the development of ICT tools enabled with assistive technologies in Rwanda. The response from the Quora forum about the colors the children with ASD like indicates that their color preference changes with their age.

> *That's totally up to the child. Many of them don't show any special indication of liking certain colors. When my autistic son was young, he liked orange, but after about the age of seven he wore nothing but black. He's 36 years old and still wears nothing but black. Occasionally we can talk him into charcoal, but it doesn't last long.*

Teachers who participated in this study suggested a continuous change of features in app depending on the behavior of a child.

> *The developed application can be helpful when children have a common preference. As we see it will succeed if the designer of the system can continually update it depending on the changes of behavior of our children.* [A common statement from teachers]

The development of a mobile app for children with ASD requires continuous iteration in the design process. However, the insufficient number of ICT technicians and qualified teachers available to participate in the design and development of a mobile app that responds to the needs of children with ASD remains a challenge in Rwanda.

Participants also reported difficulties in accessing the app because it can only be used on tablets or smartphones. This makes the mobile app inaccessible for children from families with a less privileged background. The provision of low-cost devices to families with low income is recommended by the participants.

Focus on Future

The last theme is to identify the views and recommendations from participants to be considered in the future when planning and developing mobile apps for children with ASD. The findings of this study suggested the importance of developing a family-centered process that requires parents to support their children. In this study, most teachers suggested a collaboration between software developers to develop apps that respond to the needs of the children.

> *As we see, the design of the application for these children is a long process. We suggest building a good collaboration with application developers to make sure that the application can respond to the needs of the children.* [A common statement form teachers]

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Developing a software app involves collaboration with the stakeholders in every step of development. However, if the users are persons with mental disorders such as ASD, this requires strong participation of persons who live with them. The development of a mobile app for the whole competency-based curriculum is also recommended by teachers.

The parents reported the use of the mobile app to support the transition of children from childhood to adult life if it is continuously updated according to the level of children and the changes in their behavior.

> *The behavior of our children is changed within time. We suggest a continuous change in application to accompany the children in their development till adulthood.* [A common statement from parents]

Using digital technologies supports social inclusion of children with ASD when these tools are systematically updated and meet the environmental context. Succeeding in the development and use of mobile apps for children with ASD requires a strong collaboration between educators, parents, and digital technology industries.

Discussion

Principal Findings

There has been continuous effort to increase community engagement in the development of learning tools for children with ASD. This study suggested continuous changes in the methodology used to teach children with ASD, and a systematic update of features in the developed app by considering the social norms, behavior, and privacy of these children. The responses collected from Quora [52] also suggested continuous changes in the features of the app so as to adapt it to the behavior of children with ASD during their different levels of development. This was supported by the study by Buteau-poulin et al [67], in which the authors recommended the regular update of information related to special education of children with disabilities that is available online. Participants from the Quora platform said that children can learn anything from digital devices even in cases where they tend not to look at the instructor during their class hours. This is possible only when the content meets the needs of the child and is structured around many different learning activities intended for real-world use [6,68].

We used coins and voices to keep the children focused. This was used as a reinforcement, as positive reinforcement has been utilized to keep children motivated during learning [7,69]. A study by Tzanakaki et al [70] reported that succeeding in teaching numeracy to children with ASD requires the use of various models that help students to stay focused. Moreover, children with ASD learn more easily from visual cues and videos promoted to enhance teaching and learning in an inclusive classroom [71,72]. Conoyer et al [73] evaluated different numeracy domains in the early grades such as oral counting, number recognition, touch count, missing number, and number relations. Hussain et al [65] recommended avoiding destructive voices and images that can make children lose focus. The interface designed in this study used 4 images of coins and each action is associated with a sound. Moreover, the success of ICT tools for education depends on the full participation of parents and educators of children with ASD [74]. Gamification of content [33] when teaching children with ASD is useful, as this made it possible to keep the children stay focused in this study; in this regard, having an interface that is more friendly and enjoyable is also important [56]. Many mobile apps were developed to support children with ASD [19], but some of them failed when used in different geographical contexts. To avoid this mistake, the interface designer in this study followed the guidelines prescribed previously [75] and the curriculum of basic education in Rwanda [44].

The inclusive curriculum, which aims at improving access and successful participation in the education of a person with disabilities, can bring a positive impact when ICT is fully integrated within the teaching and learning practices [76]. Results of this study show that by using ICT tools, children with ASD learn more skills commonly practiced in their daily lives without attending a school. However, it is difficult to follow the formal curriculum design when teaching children with ASD [15]. This is supported by different strategies [77], such as the development of competencies of teachers in teaching children with ASD before enrolling them into mainstream schools. The improvement of infrastructure, such as assistive technologies enabled with ICT, is reported to have a positive impact on facilitating the learning of children with ASD [78].

The study by Diener et al [79] reported that assistive technologies can help improve the inner skills hidden in children with ASD. The exposure of children with ASD to digital tools can bring about positive effects [80]. However, children with ASD are engaged only when more time is given to them to play with digital devices [5,81]. This approach is also supported by Song et al [82] who reported success in assisting persons with communication difficulties through ICT tools. Our results are no different. To sum up, children with ASD can benefit from app knowledge and utilize the knowledge gained to improve their communication in day-to-day economic activities.

Despite the advantages of using ICT tools in the education of children with ASD in Rwanda, there remain challenges of social implication when children with ASD are exposed to the digital tools for a long period [33,83]. However, parents and teachers are encouraged to supervise children with ASD to be able to manage challenging behavior(s) that may occur upon removal of the digital device [7]. Although the use of ICT tools in education continues to be encouraged to support learning of children with ASD, there is an increasing need for educational ICT tools such as mobile apps that support inclusion of children with ASD in the general education system [84]. By engaging the stakeholders to drive the development of the mobile app, we are addressing the needs of families and educators to improve the lives of children with ASD in the society.

Strengths and Limitations

The researchers of this study intended to find an ICT solution that can support the current competency-based curriculum of basic education in the Rwandan education system.

This study has the following strengths:

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- It reflects the experiences of teachers and parents who are in service to support children with ASD.
- It was conducted by experienced researchers in the field of education, information technology, special education, and ASD.
- The test was performed by children with ASD themselves while parents and teachers contributed to the improvement of the developed app.

This study followed the practical recommendations of Gowen et al [75], which helped researchers to familiarize with the autism community. These recommendations are (1) prestudy considerations, (2) recruitment of participants, (3) study visit considerations, and (4) poststudy considerations.

The rationale was that teachers can adapt to different new methods and tools that are innovative to improve the teaching and learning process. The researchers in this study selected skip counting as a learning method that can help children with ASD to improve their mathematical skills that are used in real life [73].

The study has the following limitations: (1) it explored only 1 topic among the 36 topics (3%) of planned contents of the syllabus that is supposed to help children learn basic numeracy at an early age and (2) there may be some long-term disadvantages experienced by the teachers due to constant changes and updates to the app (ie, technical and pedagogy challenges).

In future studies, all content corresponding to the existing syllabus should be explored and allow more time for participants to test the interface and take into consideration much of their views. A study on the impact of the developed app may help uncover both the long-term advantages and disadvantages that teachers may experience during the implementation of new curriculum. Another potential solution that should be further explored is the development of personalized ICT solutions for each subject planned in the competency-based curriculum that can respond to the needs of children with ASD.

Conclusions

The ICT has promisingly paved its way into the field of teaching and treating children with ASD. Rapid advancement has been achieved by developing different ICT tools to improve the quality of life for children with ASD. However, the community plays a robust role in the planning, development, and evaluation of a mobile app for children with ASD. In this study, we focused on developing a user-friendly effective tool (a mobile app) that responds to the needs of children with ASD and eliminates the barriers of learning basic mathematical skills. The community and end users should play the pivotal role in the planning, development, and evaluation of any digital tools and techniques to ensure their acceptability and successful integration. This study may be the starting point for future studies that facilitate the successful integration of individuals with autism into the society of Rwanda by utilizing different digital tools in order to ensure their participation in different learning and economic activities.

Conflicts of Interest

None declared.

Multimedia Appendix 1
Questions posed on Quora.
[DOCX File , 13 KB - pediatrics_v4i3e21471_app1.docx]

Multimedia Appendix 2
Interview guide.
[DOCX File , 14 KB - pediatrics_v4i3e21471_app2.docx]

Multimedia Appendix 3
Letter of consent/Participant consent form.
[DOCX File , 16 KB - pediatrics_v4i3e21471_app3.docx]

Multimedia Appendix 4
Ethical clearance.
[PNG File , 2501 KB - pediatrics_v4i3e21471_app4.png]

Multimedia Appendix 5
Letter of collaboration.
[PDF File (Adobe PDF File), 151 KB - pediatrics_v4i3e21471_app5.pdf]

References

1. American Psychiatric Association. Diagnostic and Statistical Manual of Mental Disorders (5th ed.). Washington, DC: American Psychiatric Association; 2013.

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2. Kamaruzaman MF, Azahari MH. Form design development study on autistic counting skill learning application. 2014 Sep Presented at: 2014 International Conference on Computer, Communications, and Control Technology (I4CT); September 2014; Langkawi, Kedah, Malaysia p. 1-5. [doi: 10.1109/I4CT.2014.6914148]
3. Hudson ME, Rivera CJ, Grady MM. Research on Mathematics Instruction with Students with Significant Cognitive Disabilities: Has Anything Changed? Research and Practice for Persons with Severe Disabilities 2018 Jan 28;43(1):38-53. [doi: 10.1177/1540796918756601]
4. Rwanda National Council of Persons with Disabilities. Disability Mainstreaming Guidelines. 2014. URL: http://www.ncpd.gov.rw/cetest-firstpage/reports [accessed 2020-12-02]
5. Ntalindwa T, Soron TR, Nduwingoma M, Karangwa E, White R. The Use of Information Communication Technologies Among Children With Autism Spectrum Disorders: Descriptive Qualitative Study. JMIR Pediatr Parent 2019 Sep 27;2(2):e12176 [FREE Full text] [doi: 10.2196/12176] [Medline: 31573940]
6. Ruparelia K, Abubakar A, Badoe E, Bakare M, Visser K, Chugani DC, et al. Autism Spectrum Disorders in Africa: Current Challenges in Identification, Assessment, and Treatment: A Report on the International Child Neurology Association Meeting on ASD in Africa, Ghana, April 3-5, 2014. J Child Neurol 2016 Jul 15;31(8):1018-1026 [FREE Full text] [doi: 10.1177/0883073816635748] [Medline: 26979098]
7. Novack MN, Hong E, Dixon DR, Granpeesheh D. An Evaluation of a Mobile Application Designed to Teach Receptive Language Skills to Children with Autism Spectrum Disorder. Behav Anal Pract 2019 Mar 9;12(1):66-77 [FREE Full text] [doi: 10.1007/s40617-018-00312-7] [Medline: 30918771]
8. Kagohara DM, van der Meer L, Ramdoss S, O'Reilly MF, Lancioni GE, Davis TN, et al. Using iPods(®) and iPads(®) in teaching programs for individuals with developmental disabilities: a systematic review. Res Dev Disabil 2013 Jan;34(1):147-156. [doi: 10.1016/j.ridd.2012.07.027] [Medline: 22940168]
9. Kamaruzaman MF, Nor HM, Azahari MHH. Using touchscreen technology to support basic numeracy learning process for high functioning children with autism. Turkish Online J. Educ. Technol 2016;2016(July):632-639 [FREE Full text]
10. Kamaruzaman MF, Md Noor H, Azahari MH. Modelling Basic Numeracy Learning Application for Children with Autism: A Pilot Study. Soc. & Manage. Res. J 2017 Dec 31;14(2):125. [doi: 10.24191/smrj.v14i2.5496]
11. da Silva M, Gonçalves D, Guerreiro T, Silva H. A Web-based Application to Address Individual Interests of Children with Autism Spectrum Disorders. Procedia Computer Science 2012;14:20-27. [doi: 10.1016/j.procs.2012.10.003]
12. Rwanda Ministry of Education. ICT in Education Policy. 2016. URL: http://mineduc.gov.rw [accessed 2020-11-07]
13. Rwanda Ministry of Education. Statistics of Education. 2019. URL: http://mineduc.gov.rw [accessed 2020-11-13]
14. Rwanda Education Board. Mathematics Subject Syllabus for Lower Primary Education. 2015. URL: https://tinyurl.com/3cjj2989 [accessed 2020-10-21]
15. Virginia Department of Education, Office of Special Education and Student Services. Models of Best Practice in the Education of Students with Autism Spectrum Disorders. Richmond, VA: Virginia Department of Education, Office of Special Education and Student Services; 2011 May. URL: https://www.doe.virginia.gov/special_ed/disabilities/autism/technical_asst_documents/autism_models_of_best_practice.pdf [accessed 2021-08-10]
16. Aboagye F. Autism in an Inclusive Class: A Case Study of How Teachers Manage to Include Pupils With Autism. Oslo, Norway: University of Oslo; 2017. URL: https://www.duo.uio.no/bitstream/handle/10852/57472/can-16.pdf?sequence=5 [accessed 2021-08-10]
17. Tanner K, Verenikina I, Tanner K. The digital technology in the learning of students with autism spectrum disorders (ASD) in applied classroom settings. 2010 Presented at: Proceedings of World Conference on Educational Multimedia, Hypermedia and Telecommunications; June 26 to August 1, 2010; Chesapeake, VA p. 2586-2591. [doi: 10.1007/978-0-387-71799-9_36]
18. Kamaruzaman MF, Rani NM, Nor HM, Azahari MHH. Developing User Interface Design Application for Children with Autism. Procedia - Social and Behavioral Sciences 2016 Feb;217:887-894. [doi: 10.1016/j.sbspro.2016.02.022]
19. Soomro N, Soomro S. Autism Children's App using PECS. AETiC 2018 Jan 1;2(1):7-16. [doi: 10.33166/AETiC.2018.01.002]
20. Rajagopal K, Ying GS. The effects of mobile flashcard application on two special need children's numeracy skills. 2015 Presented at: SAHConFed 2018; November 2018; Kedah, Malaysia URL: https://www.researchgate.net/publication/329167001_THE_EFFECTS_OF_MOBILE_FLASHCARD_APPLICATION_ON_TWO_SPECIAL_NEED_CHILDREN'S_NUMERACY_SKILLS
21. Díaz M, Barco A. Robot assisted play with a mobile robot in a training group of children with autism. 2012 Presented at: International Workshop on Human-Agent Interaction/IEEE/RSJ International Conference on Intelligent Robots and Systems (IROS 2012); October 21-24, 2012; Vilamoura, Algarve, Portugal URL: https://www.academia.edu/13972315/Robot_Assisted_Play_with_a_Mobile_Robot_in_a_Training_Group_of_Children_with_Autism
22. Alcantud F, Alonso Y, Coret J, Jiménez E. Assistive technologies and autism spectrum disorders. In: Disability Informatics and Web Accessibility for Motor Limitations. Hershey, PA: IGI Global; 2014:263-293.
23. Abubakar A, Ssewanyana D, de Vries PJ, Newton CR. Autism spectrum disorders in sub-Saharan Africa. The Lancet Psychiatry 2016 Sep;3(9):800-802. [doi: 10.1016/s2215-0366(16)30138-9]
24. de Macedo CMS, Ulbricht VR. Accessibility Guidelines for the Development of Learning Objects. Procedia Computer Science 2012;14:155-162. [doi: 10.1016/j.procs.2012.10.018]

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25. Mejía A, Juárez-Ramírez R, Inzunza S, Valenzuela R. Implementing adaptive interfaces: A user model for the development of usability in interactive systems. New York, NY: ACM; 2012 Sep Presented at: CUBE '12: Proceedings of the CUBE International Information Technology Conference; September 3-5, 2012; Pune, India p. 598-604. [doi: 10.1145/2381716.2381831]
26. Allen ML, Hartley C, Cain K. iPads and the Use of "Apps" by Children with Autism Spectrum Disorder: Do They Promote Learning? Front Psychol 2016;7:1305 [FREE Full text] [doi: 10.3389/fpsyg.2016.01305] [Medline: 27625621]
27. Xin JF, Leonard DA. Using iPads to Teach Communication Skills of Students with Autism. J Autism Dev Disord 2015 Dec;45(12):4154-4164. [doi: 10.1007/s10803-014-2266-8] [Medline: 25294648]
28. Omar S, Bidin A. The Impact of Multimedia Graphic and Text with Autistic Learners in Reading. ujer 2015 Dec;3(12):989-996. [doi: 10.13189/ujer.2015.031206]
29. Kirk HE, Gray K, Riby DM, Taffe J, Cornish KM. Visual attention and academic performance in children with developmental disabilities and behavioural attention deficits. Dev Sci 2017 Nov;20(6):1-6. [doi: 10.1111/desc.12468] [Medline: 27649816]
30. Vahabzadeh A, Keshav N, Abdus-Sabur R, Huey K, Liu R, Sahin N. Improved Socio-Emotional and Behavioral Functioning in Students with Autism Following School-Based Smartglasses Intervention: Multi-Stage Feasibility and Controlled Efficacy Study. Behav Sci (Basel) 2018 Sep 20;8(10):85 [FREE Full text] [doi: 10.3390/bs8100085] [Medline: 30241313]
31. Fraccaro P, Arguello Casteleiro M, Ainsworth J, Buchan I. Adoption of clinical decision support in multimorbidity: a systematic review. JMIR Med Inform 2015 Jan 07;3(1):e4 [FREE Full text] [doi: 10.2196/medinform.3503] [Medline: 25785897]
32. Read T, Bárcena E, Kukulska-Hulme A. Exploring the application of a conceptual framework in a social MALL app. In: Pareja-Lora A, Calle-Martínez C, Rodríguez-Arancón P, editors. New Perspectives on Teaching and Working With Languages in the Digital Era. Dublin, Ireland: Research-publishing.net; 2016:223-232.
33. Jordine T, Liang Y, Ihler E. A new game-based learning approach for learning Java on mobile devices. In: Connolly T, Boyle L, editors. Proceedings of the 10th European Conference on Games Based Learning. Reading, UK: Academic Conferences and Publishing International Limited; Oct 6, 2016.
34. Spikins P, Wright B, Hodgson D. Are there alternative adaptive strategies to human pro-sociality? The role of collaborative morality in the emergence of personality variation and autistic traits. Time and Mind 2016 Nov 15;9(4):289-313. [doi: 10.1080/1751696x.2016.1244949]
35. Disabled World. iConverse App - Augmentative Alternative Communication Tool. 2011. URL: https://www.disabled-world.com/assistivedevices/apps/iconverse.php [accessed 2020-12-20]
36. Purdue University. SPEAK all! - Application for Developing Language Skills. 2013. URL: https://www.purdue.edu/ [accessed 2021-01-14]
37. Tashnim A, Nowshin S, Akter F, Das AK. Interactive Interface Design for Learning Numeracy and Calculation for Children with Autism. New York, NY: IEEE; 2017 Presented at: 2017 9th International Conference on Information Technology and Electrical Engineering (ICITEE); October 12-13, 2017; Phuket, Thailand p. 1-6. [doi: 10.1109/ICITEED.2017.825050]
38. Nancy R, Pourcho J, Wei S, Qian CZ, Chen VY. Literacy LABELS: Emergent literacy application design for children with autism. New York, NY: ACM; 2013 Jul Presented at: SIGGRAPH '13: Special Interest Group on Computer Graphics and Interactive Techniques Conference; July 21-25, 2013; Anaheim, CA. [doi: 10.1145/2503385.2503411]
39. Qian N, Lipkin RM. A learning-style theory for understanding autistic behaviors. Front Hum Neurosci 2011;5:77 [FREE Full text] [doi: 10.3389/fnhum.2011.00077] [Medline: 21886617]
40. Saad Yahya Athbah B. Parents' Attitudes Toward the Use of Technology and Portable Devices With Children With Autism Spectrum Disorder (ASD) in Saudi Arabia. 2015 Aug. URL: http://hdl.handle.net/2376/6161 [accessed 2021-01-04]
41. Khorrami A, Tehrani-Doost M, Esteky H. Comparison between Face and Object Processing in Youths with Autism Spectrum Disorder: An event related potentials study. Iran J Psychiatry 2013 Oct;8(4):179-187 [FREE Full text] [Medline: 25628712]
42. Dixon MR, Belisle J, Stanley CR, Daar JH, Williams LA. Derived Equivalence Relations of Geometry Skills in Students with Autism: an Application of the PEAK-E Curriculum. Anal Verbal Behav 2016 Jun;32(1):38-45 [FREE Full text] [doi: 10.1007/s40616-016-0051-9] [Medline: 27606220]
43. Johnson CP, Myers SM, American Academy of Pediatrics Council on Children With Disabilities. Identification and evaluation of children with autism spectrum disorders. Pediatrics 2007 Nov;120(5):1183-1215. [doi: 10.1542/peds.2007-2361] [Medline: 17967920]
44. Curriculum Framework Pre-Primary to Upper Secondary. 2015. URL: https://www.reb.gov.rw/publications [accessed 2021-01-11]
45. Centers for Disease Control and Prevention. Autism Spectrum Disorders. Atlanta, GA: Centers for Disease Control and Prevention; 2020. URL: https://www.cdc.gov/ncbddd/autism/facts.html [accessed 2021-01-05]
46. Chown N. More on the ontological status of autism and double empathy. Disability & Society 2014 Sep 10;29(10):1672-1676. [doi: 10.1080/09687599.2014.949625]
47. Schuler D, Namioka A. Participatory Design: Principles and Practices. London, UK: Taylor and Francis; 1993.
48. Baker JD. The Purpose, Process, and Methods of Writing a Literature Review. AORN J 2016 Mar;103(3):265-269. [doi: 10.1016/j.aorn.2016.01.016] [Medline: 26924364]

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49. Vaishnavi P, Panicker S, Maitreyi K. Use of Agile Methodology for Mobile Applications. Int. J. Latest Technol. Eng. Manag. Appl. Sci 2016;5(10):73-77.
50. Borys M, Laskowski M. Expert vs novice evaluators: Comparison of heuristic evaluation assessment. In: Proceedings of the 16th International Conference on Enterprise Information Systems. 2014 Jan Presented at: ICEIS 2014; April 27-30, 2014; Lisbon, Portugal p. 144-149 URL: https://www.researchgate.net/publication/287048010_Expert_vs_novice_evaluators_Comparison_of_heuristic_evaluation_assessment [doi: 10.5220/0004970901440149]
51. Heo J, Ham D, Park S, Song C, Yoon WC. A framework for evaluating the usability of mobile phones based on multi-level, hierarchical model of usability factors. Interacting with Computers 2009 Aug;21(4):263-275. [doi: 10.1016/j.intcom.2009.05.006]
52. Quora Digest. URL: https://www.quora.com/ [accessed 2021-02-01]
53. Specialist autism education and support. Autism Rwanda. URL: http://www.autismerwanda.org/ [accessed 2020-11-27]
54. Rwanda Parent's Initiative on Autism. 2020. URL: http://www.rpia.rw [accessed 2021-01-13]
55. Punchoojit L, Hongwarittorrn N. Usability Studies on Mobile User Interface Design Patterns: A Systematic Literature Review. Advances in Human-Computer Interaction 2017;2017:1-22. [doi: 10.1155/2017/6787504]
56. Ballantyne M, Jha A, Jacobsen A, Scott Hawker J, El-Glaly YN. Study of accessibility guidelines of mobile applications. New York, NY: ACM; 2018 Presented at: MUM 2018: Proceedings of the 17th International Conference on Mobile and Ubiquitous Multimedia; November 25-28, 2018; Cairo, Egypt.
57. Fletcher-Watson S, Pain H, Hammond S, Humphry A, McConachie H. Designing for young children with autism spectrum disorder: A case study of an iPad app. International Journal of Child-Computer Interaction 2016 Jan;7:1-14. [doi: 10.1016/j.ijcci.2016.03.002]
58. Google Trends: Quora. URL: https://trends.google.com/ [accessed 2022-12-01]
59. Paul SA, Hong L, Chi EH. Who is authoritative? Understanding reputation mechanisms in Quora. Proceedings, CI2012. 2010. URL: https://arxiv.org/ftp/arxiv/papers/1204/1204.3724.pdf [accessed 2021-08-10]
60. Kumar S, Parveen N, Goel N, Sanwal K. Opinion Extraction From Quora Using User-Biased Sentiment Analysis. Singapore: Springer; 2018.
61. Apple Inc. 123 Numbers - Count & Tracing. 2019. URL: https://apps.apple.com/us/app/123-numbers-count-tracing/id1210356444 [accessed 2021-02-12]
62. Adobe. Adobe Captivate. 2019. URL: https://www.adobe.com/hk_en/products/captivate.html [accessed 2020-11-17]
63. Google. Android Platform. 2020. URL: https://www.android.com/ [accessed 2021-01-02]
64. Apple Inc. iOS. URL: https://www.apple.com/ios/ios-13/ [accessed 2021-01-11]
65. Hussain A, Abdullah A, Husni H. The design principles of edutainment system for autistic children with communication difficulties. AIP Conference Proceedings 2016;1761:020047. [doi: 10.1063/1.4960887]
66. National Bank of Rwanda. Rwandan Currency. 2020. URL: https://bnr.rw/ [accessed 2020-11-27]
67. Buteau-Poulin A, Gosselin C, Bergeron-Ouellet A, Kiss J, Lamontagne M, Maltais D, et al. Availability and Quality of Web Resources for Parents of Children With Disability: Content Analysis and Usability Study. JMIR Pediatr Parent 2020 Nov 10;3(2):e19669 [FREE Full text] [doi: 10.2196/19669] [Medline: 33170127]
68. Bernardini S, Porayska-Pomsta K, Smith TJ. ECHOES: An intelligent serious game for fostering social communication in children with autism. Information Sciences 2014 Apr;264:41-60. [doi: 10.1016/j.ins.2013.10.027]
69. Loiacono V, Valenti V. General education teachers need to be prepared to co-teach the increasing number of children with autism in inclusive settings,. International Journal of Special Education 2010 Jan;25(3):24-32.
70. Tzanakaki P, Grindle CF, Saville M, Hastings RP, Hughes JC, Huxley K. An individualised curriculum to teach numeracy skills to children with autism: programme description and pilot data. Support for Learning 2015 Jan 10;29(4):319-338. [doi: 10.1111/1467-9604.12069]
71. Ibrahim N, Wan Ahmad WF, Shafie A. Multimedia Mobile Learning Application for Children's Education: The Development of MFolktales. ASS 2015 Aug 18;11(24):1-13. [doi: 10.5539/ass.v11n24p203]
72. Brame CJ. Effective Educational Videos: Principles and Guidelines for Maximizing Student Learning from Video Content. CBE Life Sci Educ 2016;15(4):1-6 [FREE Full text] [doi: 10.1187/cbe.16-03-0125] [Medline: 27789532]
73. Conoyer SJ, Foegen A, Lembke ES. Early Numeracy Indicators. Remedial and Special Education 2016 Feb 09;37(3):159-171. [doi: 10.1177/0741932515619758]
74. Sevastopoulou S. Collaboration towards the inclusion of children with autistic spectrum disorders: the views of the general and special education teachers in Greek primary schools. 2016. URL: https://www.duo.uio.no/handle/10852/54037 [accessed 2021-01-13]
75. Gowen E, Taylor R, Bleazard T, Greenstein A, Baimbridge P, Poole D. Guidelines for conducting research studies with the autism community. Autism Policy Pract 2019 Sep 09;2(1 A new beginning):29-45 [FREE Full text] [Medline: 32226635]
76. Guenaga M, Mechaca I, Romero S, Eguíluz A. A Tool to Evaluate the Level of Inclusion of Digital Learning Objects. Procedia Computer Science 2012;14:148-154. [doi: 10.1016/j.procs.2012.10.017]
77. MacLeod K, Causton JN, Radel M, Radel P. Rethinking the Individualized Education Plan process: voices from the other side of the table. Disability & Society 2017 Mar 08;32(3):381-400. [doi: 10.1080/09687599.2017.1294048]

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78. Fage C, Consel CY, Balland E, Etchegoyhen K, Amestoy A, Bouvard M, et al. Tablet Apps to Support First School Inclusion of Children With Autism Spectrum Disorders (ASD) in Mainstream Classrooms: A Pilot Study. Front Psychol 2018;9:2020 [FREE Full text] [doi: 10.3389/fpsyg.2018.02020] [Medline: 30405498]
79. Diener ML, Wright CA, Smith KN, Wright SD. Assessing Visual-Spatial Creativity in Youth on the Autism Spectrum. Creativity Research Journal 2014 Aug 08;26(3):328-337. [doi: 10.1080/10400419.2014.929421]
80. Lozano-Martínez J, Ballesta-Pagán FJ, Alcaraz-García S. Software for Teaching Emotions to Students with Autism Spectrum Disorder. Comunicar: Revista Científica de Comunicación y Educación 2011 Mar 01;18(36):139-148. [doi: 10.3916/c36-2011-03-05]
81. Morrison BB, DiSalvo B. Khan Academy gamifies computer science. 2014 Mar Presented at: Proceedings of the 45th ACM technical symposium on Computer Science Education; March 5-8, 2014; Atlanta, GA URL: https://www.researchgate.net/publication/261961212_Khan_Academy_gamifies_computer_science [doi: 10.1145/2538862.2538946]
82. Song HSY, Yusof AM. Current Review of the Use of Mobile Technology to Enhance Learning and Communication Among Children With Developmental Disabilities. 2010 Jul Presented at: EDULEARN10: Proceedings 2nd International Conference on Education and New Learning Technologies; July 5-7, 2010; Barcelona, Spain p. 5990-5598 URL: https://tinyurl.com/faskk2mt
83. Aresti-Bartolome N, Garcia-Zapirain B. Technologies as support tools for persons with autistic spectrum disorder: a systematic review. Int J Environ Res Public Health 2014 Aug 04;11(8):7767-7802 [FREE Full text] [doi: 10.3390/ijerph110807767] [Medline: 25093654]
84. Competence based Curriculum. 2015. URL: https://www.reb.gov.rw/curriculum-teaching-learning-resources-department [accessed 2021-01-02]

Abbreviations

ASD: autism spectrum disorder
CDC: Centers for Disease Control and Prevention
FGD: focus group discussion
ICT: information and communication technology
QNA: qualitative narrative analysis
RPIA: Rwanda Parent's Initiative on Autism

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Original Paper

Mobile Technology Access and Use Among Adolescent Mothers in Lima, Peru: Mixed Methods Study

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Abstract

Background: Research shows promise for the use of mobile health interventions to improve access to care for mothers and infants. Although adolescent mothers in particular are comfortable with technology and often face barriers to accessing care, data on the use of digital interventions with young mothers are limited.

Objective: This study aims to examine technology access and use behavior among adolescent mothers in Lima, Peru, to inform the development of technology-mediated perinatal interventions for high-risk mothers and infants in low- and middle-income countries and other areas with limited access to care.

Methods: This mixed methods study consisted of a phone survey about technology access (N=29), focus group discussions with clinicians (N=25), and semistructured in-depth interviews with adolescent mothers (N=10) and their family members (N=8) in Lima.

Results: All adolescent mothers surveyed had access to a smartphone, and nearly half had access to a computer or tablet. However, participants reported a number of obstacles to consistent smartphone access related to the financial precarity of their situations. Examples of this included difficulty affording phone services, using shared plans, and losing smartphones because of theft.

Conclusions: These findings indicate that adolescent mothers are connected to technology, highlighting the potential scalability of technology-based health interventions for adolescent mothers in low- and middle-income countries while identifying barriers that need to be addressed.

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KEYWORDS

access to care; adolescent motherhood; LMICs; mobile phone; perinatal mental health; telehealth

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Introduction

Background

According to the International Telecommunication Union's 2016 report, 95% of the world lives in an area covered by a mobile cellular network and 85% is covered by cell phone signals [1,2]. Over the past few years, there has been significant growth in the use of mobile health (mHealth) technologies in low- and middle-income countries (LMICs), with infectious diseases and maternal health being the most frequent targets for these interventions [3,4]. mHealth interventions can be categorized by their purpose, including prevention and education, information sharing, and communication [5]. mHealth technologies hold promise for addressing challenges in health care access, delivery, and knowledge by better reaching individuals who may have difficulty accessing services [6].

Studies have identified maternal and child health as areas in which mHealth and other digital interventions hold significant promise for improving health outcomes in LMICs. A systematic review of the use of mHealth interventions in LMICs to address maternal health challenges found that mobile technologies had been used successfully in several areas, including data collection, decision support, and appointment reminders. Although these interventions showed promise for improving maternal health in LMICs, most of the studies were of low to moderate quality, indicating a need for more rigorous methods in this area [7]. Mothers in LMICs often do not receive the recommended number of antenatal or postnatal care contacts and experience gaps in knowledge and self-efficacy to provide care for their children at home [8]. mHealth interventions during this time can help improve access to information and care [8]. A recent systematic review focused on the use of mHealth educational interventions during the perinatal period and found that mothers who received these interventions had a significantly greater number of antenatal and postnatal care contacts [9]. A particularly successful example of an mHealth intervention that was scaled up in South Africa was the Mobile Alliance for Maternal Action project. This project connected mothers to the health care system by SMS text messaging, encouraged them to attend antenatal visits, and gave them information about pregnancy and childcare [10].

Maternal mental health is closely linked to maternal and child health outcomes [11-13]. For this reason, digital psychological interventions also hold promise for improving maternal and child health in resource-limited settings. For example, a digital intervention developed for low-income mothers living in rural areas of the United States was associated with increased maternal sensitivity and reduced maternal depression [14]. Despite the promise of technology to decrease disparities in access to care, research on digital interventions in LMICs has lagged behind high-income countries (HICs). However, in the past decade, researchers have begun to explore the use of technology-mediated mental health interventions in LMIC settings. In a systematic review of 19 telemental health intervention studies, the findings varied markedly based on how the intervention was delivered and the control condition used [15]. A recent systematic review and meta-analysis of 22 studies of digital psychological interventions in LMICs found that the interventions were moderately effective compared with usual care and concluded that digital interventions should be used in areas where there is inadequate access to in-person treatment [16]. Overall, this research indicates a rising trend in the use of mHealth and other digital interventions in LMICs, as well as a need for more empirical studies in this area [15].

LMICs account for 95% of adolescent births globally [17]. The adolescent pregnancy rate in LMICs is 48 births per 1000 girls (aged 15-19 years) per year, more than triple the rate in HICs (15 births per 1000 girls) [18]. Although mHealth interventions, specifically for adolescent mothers, are largely absent from the literature, the success of digital psychological interventions with adolescents suggests that it could be a useful tool to close the gap in access to care in this population. Digital psychological interventions have been demonstrated to be acceptable and feasible for adolescents [19,20]. The anonymity that internet-based interventions provide is reassuring to adolescents who are particularly sensitive to social stigma and have greater comfort with technology [21]. One systematic review of 22 randomized controlled trials of technology-mediated interventions for adolescents with depression or anxiety found improvements in depression and anxiety symptoms at the end of the intervention, but few studies conducted long-term follow-up [20].

Finally, mobile technology may also hold promise for measuring outcomes for parenting interventions, although there is limited research in this area in LMICs. Traditional methods of measuring parenting behavior, including dyad observation and retrospective questionnaires, can be resource intensive. To address these challenges, Fatori et al [22] tested the use of an electronic daily diary to measure the effects of a nursing home visiting program for adolescent mothers in Brazil. They found both high compliance to the electronic daily diary (84%) and a positive impact of the intervention on parental well-being and maternal parenting [22].

Although mHealth interventions hold potential to improve maternal health and adolescent parenting, LMIC settings face unique barriers to successful implementation. For the Mobile Alliance for Maternal Action project, described earlier, facilitators for this intervention's scale-up included political will, stakeholders' engagement, and adaptation to the South African context, whereas cost and financial sustainability were identified as barriers to scale-up [10]. Additional barriers identified to the scale-up and use of mHealth interventions in LMICs include lack of infrastructure or equipment, limited economic resources and literacy of users, privacy and confidentiality concerns, data costs, and network coverage and speed; only 7% of broadband subscriptions in LMICs have speeds of 10 megabits per second or higher [1,3].

Objectives

In summary, research shows promise for the use of mHealth interventions to improve maternal and child health by increasing affordable access to care. However, although adolescent mothers, in particular, are more comfortable with technology and can have difficulty accessing hospital- or clinic-based care, data for the use of mHealth interventions with adolescents and

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in LMIC settings are limited. To address some of these gaps in the literature, this study examined technology access and use behavior among adolescent mothers in Lima, Peru, to inform the development of technology-mediated perinatal interventions for high-risk mothers and infants in LMICs and other areas with limited access to care. Peru is a middle-income country, with an adolescent fertility rate of 55 births per 1000 girls aged 15-19 years.

Methods

Study Setting

This study was conducted in Lima, Peru. Participants were recruited from 2 different clinical sites in Lima. Instituto Nacional Materno Perinatal (INMP) is the primary reference establishment for maternal and perinatal care operated by the Ministry of Health of the Peruvian Government. It serves low-income women who are publicly insured. Taller de Niños (TANI) is a nongovernmental organization that has been operating in Peru for more than 40 years, providing medical care and other services to infants and their families residing in the San Juan de Lurigancho District of Lima. With more than one million people, this district is Lima's most populous district. Many of its residents have recently migrated from rural province areas.

Participants and Procedures

Overview

This mixed methods study comprising quantitative phone surveys, qualitative focus groups, and in-depth individual interviews. The qualitative data were collected as part of a broader assessment of the needs and preferences of adolescent mothers and their families to inform the development of a perinatal intervention. This study focuses specifically on the findings related to the use of technology.

Quantitative

Quantitative data collection consisted of a 47-item phone survey that included questions about basic demographic information and technology access and use. The survey took approximately 15 minutes to administer. Pregnant and postpartum adolescents were recruited from patient records at INMP. Women between the ages of 14 and 19 who had given birth between May 1, 2019, and May 1, 2020, or were due to give birth before September 1, 2020, were eligible to participate. The goal was to survey 25 to 40 mothers to obtain a representative sample. Potential participants were contacted in May 2020. A total of 57 adolescents were contacted by phone and invited to participate in the survey; of these, 28 (49%) were unreachable by phone, whereas the remaining 29 (51%) were reached and agreed to participate.

Qualitative

Qualitative data collection consisted of 4 clinician focus groups (n=25), 10 in-depth interviews with adolescent mothers (n=10), and 8 in-depth interviews with other caregivers (n=8). The focus groups and interviewers were coconducted by 2 Spanish-speaking members of the research team. All focus groups and interviewers were audio recorded. One member of the research team asked questions, whereas the other took notes. Three focus groups were composed of clinicians from INMP, and one focus group was conducted with clinicians from TANI. After each focus group and interview, a member of the research team listened to the recording to review what had been discussed and consider how to explore topics of interest in greater depth until theoretical saturation was reached [23].

Participants in the clinician focus groups were recruited through the heads of the clinical services, who disseminated information about the study and worked with study staff to organize and schedule the focus groups. All clinicians who cared for adolescents during the perinatal period were eligible to participate in the study. The focus groups were conducted from July 1 to July 5, 2019. Each focus group was conducted in a private room at the clinical site and lasted for 90-120 minutes. The focus group discussion guide was designed to elicit clinicians' perspectives on the needs of pregnant adolescents, adolescent mothers, and their families. This included educational, medical, physical, and emotional needs and ideas about how these needs could best be addressed. Focus group discussions were conducted before individual interviews, and major themes were identified and further explored in individual interviews.

Adolescent mothers who participated in the in-depth interviews were identified from birth records at INMP. Adolescents between the ages of 14 and 19 years who had given birth at INMP within the previous 15 months were eligible to participate. Potential participants were excluded if their infant had died, if they were not living in the Lima area, or if they did not speak Spanish. Interviews began on July 8, 2019, and were completed on February 22, 2020. Each interview was conducted in a private space at the participant's home. The interviewer spent 4 hours at each home. Interviews were conducted in Spanish by a trained research assistant and lasted about 1.5 hours, and the rest of the time was spent conducting a more informal ethnographic observation of the living situation of the mother and infant. Similar to the focus groups, the recordings were reviewed after each interview to determine which topics needed to be explored in greater depth and identify when theoretical saturation was reached [23].

Participants were asked whether there was anyone helping them care for their infants who were available to be interviewed. This way, 8 other caregivers were identified—3 were fathers of the infants, 2 were mothers of the adolescent mothers, 1 was a mother-in-law, 1 was a grandmother, and 1 was a grandfather. In some cases, the adolescent mother and the other caregiver were interviewed sequentially during a single visit; in other cases, a separate in-person interview was scheduled.

The interview guide for adolescent mothers was designed and implemented by the researchers to understand the experiences of adolescent pregnancy and motherhood, identify their unmet needs, potential intervention targets, and potential barriers to and facilitators of acceptance of a perinatal intervention in this population. The interview guide for other caregivers was designed to elicit caregivers' perceptions of the mother's experience, as well as the caregivers' own experiences and their relationships with their mothers. The research team made

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revisions to the interview guide after each interview to refine and deepen the questions. All participants provided informed consent. The institutional review boards of the INMP, Lima, Peru, and the Harvard T H Chan School of Public Health, Office of Human Research Administration, Boston approved all the procedures used in this study.

Analysis

Quantitative Analysis

The survey data were stored and analyzed in Microsoft Excel [24]. Mean and SD statistics were calculated using this software.

Qualitative Analysis

Focus groups and interviews were transcribed verbatim by a Peruvian member of the research team and were then translated into English by a bilingual team member. The codebooks were developed in Spanish, and the transcripts were coded in Spanish. Once the coding was completed, the same coding was applied to the English translations so that the findings could be disseminated in English. A directed content analysis was used based on the goals of informing intervention adaptation [25]. Coders worked in 3 teams of 2, with one team coding focus groups, another team coding mother interviews, and a third team coding caregiver interviews. First, coders read the transcripts and developed initial codebooks over a series of meetings. Once coding began, the teams met after coding each transcript to discuss and revise the codebooks. The transcripts were then recoded using the revised codebook. Memoing was used throughout the process to generate new codes, refine definitions, and relate codes to each other [23]. Intercoder reliability was measured using the κ statistic [26].

Both inductive and deductive approaches were used to analyze data [23]. First, the manifest content was grouped thematically. Thematic groupings were then labeled, and these group labels were used to generate broad themes. These broad, overarching themes were divided into subthemes. Within each theme and subtheme, the researchers drew comparisons, looking for overlap and differences and newly emerging topics and patterns. The themes identified included technology access, barriers to access, attitudes toward technology use, problems associated with technology use, and expectations and desires related to technology. Responses were reviewed to identify each theme and subtheme. NVivo (QSR International) was used for data management [27]. Intercoder reliability was substantial (κ>0.6) [28,29]. For the focus groups, the intercoder reliability was κ=0.70. For adolescent mother interviews, the intercoder reliability was κ=0.67. For other caregiver interviews, the intercoder reliability was κ=0.79.

Results

Descriptive Data

A total of 29 participants completed the phone surveys (57 were contacted; 28 were unreachable). They were all between the ages of 14 and 19 years, with an average age of 17.8 years (SD 1.5). A total of 23 participants were pregnant, and 6 had given birth within the past year (Table 1).

Table 1. Demographic characteristics of phone survey participants (N=29).

Characteristic	Value
Age (years), mean (SD)	17.8 (1.5)
Pregnant, n (%)	23 (79)
Postpartum, n (%)	6 (21)

Participant demographics for the qualitative data are summarized in Tables 2-4. A total of 25 clinicians participated in the focus group. They were between the ages of 24 and 66 years, with an average age of 47.7 (SD 14.5) years. The clinical disciplines represented were physicians (obstetrician/gynecologists), nurses, psychologists, midwives, social workers, and community health workers. A majority of the clinicians were female; there were 3 male physicians and 1 male psychologist. The duration of their clinical experience ranged from less than a year to 43 years, with an average of 16.6 (SD 13.2) years (Table 2). Participants recruited from TANI were typically younger and had less clinical experience than those recruited from the INMP.

In-depth interviews were conducted with 10 adolescent mothers and 8 other caregivers. Adolescent mothers were an average age of 16.6 years (SD 1.6); the fathers of their infants were an average age of 21.2 years (SD 3.9). The age range of the infants was 2-14 months, with an average age of 6.8 (SD 4.2) months (Table 3). Of the 8 other caregivers interviewed, 3 (38%) were infants' fathers, 2 (25%) were maternal grandmothers (mothers of the adolescents), 1 (13%) was a paternal grandmother, 1 (13%) was a maternal great-grandmother, and 1 (13%) was a maternal great-grandfather (Table 4).

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Table 2. Demographic characteristics of focus group participants (N=25).

Characteristic	Value
Age (years), mean (SD)	47.7 (14.5)
Female, n (%)	21 (84)
Male, n (%)	4 (16)
Physicians, n (%)	5 (20)
Nurses, n (%)	5 (20)
Psychologists, n (%)	5 (20)
Midwives, n (%)	4 (16)
Social workers, n (%)	4 (16)
Community health workers, n (%)	2 (8)
Clinical experience (years), mean (SD)	16.6 (13.2)

Table 3. Demographic characteristics of adolescent mothers (N=10).

Characteristic	Value
Age of mothers (years), mean (SD)	16.6 (1.6)
Age of fathers (years), mean (SD)	21.2 (3.9)
Age of infants (months), mean (SD)	6.8 (4.2)
Living with family, n (%)	6 (60)
Living with partner, n (%)	4 (40)
Graduated high school, n (%)	3 (30)
Enrolled in high school, n (%)	3 (30)
Did not graduate and not currently enrolled in high school, n (%)	4 (40)

Table 4. Relationship of other caregivers to infants (N=8).

Relationship category	Participants, n (%)
Father	3 (38)
Maternal grandmother	2 (25)
Paternal grandmother	1 (13)
Maternal great-grandmother	1 (13)
Maternal great-grandfather	1 (13)

Access to Technology

A phone survey of 29 pregnant and postpartum adolescents explored their access to technology (Table 5). All had access to a smartphone, and nearly half had access to a computer or tablet. However, there were a number of obstacles to consistent smartphone access. A total of 8 participants did not have their own phone but had only shared access to someone else's phone and 9 had been without phone access at some time in the past 12 months for various reasons: 3 had their phones stolen, 3 had their phones break, and 3 lost their phones. A total of 21 participants used a prepaid phone plan, which was the most common way to access the internet, and 15 participants had wireless internet in their home. Difficulty in paying for phone or internet services was reported by 11 participants, and interruptions to electricity services were reported by 8 participants.

All participants reported using social media at least once per week, with 46% (13/28) reporting daily use, 36% (10/28) using it every other day, 14% (4/28) using it every three days, and 4% (1/28) reporting weekly use. Participants reported using the following 3 social media platforms: WhatsApp (28/28, 100%), Facebook (23/28, 82%), and Instagram (17/28, 61%). A total of 26 participants made video calls using their smartphones. Video streaming on their phones was done by 28 participants, but 54% (15/28) reported poor video quality.

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Table 5. Mobile technology access among adolescent mothers (N=29).

Access type	Participants, n (%)
Computer or tablet	14 (48)
Smartphone	29 (100)
Shared smartphone	8 (28)
Lost smartphone access in previous 12 months	9 (31)
Prepaid phone plan	21 (72)
Wireless internet	15 (52)
Difficulty affording phone or internet service	11 (38)
Any social media use	29 (100)
Ability to make video calls	26 (90)

Summary of Qualitative Findings

In the focus groups and in-depth interviews, participants spoke about access to technology, barriers or limitations to the use of technology, and how technology is used by adolescent mothers and their families, particularly as it relates to caring for themselves and their infants. Although many ideas were shared across participants, there were some key differences between the perspectives of clinicians and adolescent mothers regarding how adolescent mothers use technology. The other caregivers were heterogeneous—3 of them were fathers, and their experience with technology was similar to that of adolescent mothers. The remaining caregivers were older. As a group, they were less comfortable with technology, and some did not have smartphones.

Barriers

A number of barriers were identified that limited access to technology, including theft, changing carriers, running out of data, and inability to afford any plan. Clinicians explained that many adolescents lived in dangerous areas where theft was common. For this reason, clinicians who did home visits were reluctant to bring their phones with them. They viewed this as a missed opportunity to use web-based tools to educate patients about pregnancy and childcare during their home visits. Phone theft also meant that adolescents were frequently without phones or using a different number. They also changed numbers when a particular carrier offered a special rate, or when they lost access to a shared phone. In addition, adolescent mothers reported that their access was limited because they were on a shared data plan, and they might run out of data, or the data could be cut off for periods of time if their family could not afford it. When this happened, they shifted to texting and other forms of communication that did not require data use. Alternatively, if they had no phone minutes and only internet access, they used apps such as WhatsApp, which uses internet rather than phone minutes. This was often the reason that they began using social media platforms to communicate. As one adolescent mother shared:

> *I only use WhatsApp because I don't have a phone plan [now]. I talk to my dad and my grandmother, video calling with them.*

As this example illustrates, adolescents with limited access to technology tended to limit communication to family and have less contact with those outside their immediate circle, including friends. In Peru and many LMICs, users are changed for placing calls and texting but not for receiving them. Thus, when they ran out of minutes, adolescents could receive calls, but they could not place them. For these reasons, it was difficult for clinicians to have reliable contact with their patients by phone.

Adolescents also preferred newer and more powerful technology; if they did not like their phones, they were less inclined to use them. As one adolescent mother explained, she did not use her phone as much since her previous phone was stolen because her new phone was smaller and less enjoyable to use: "I don't feel like using this phone because it's little, as opposed to mine, which was big." Technology became a venue for comparison, including comparing devices and access. Adolescents who felt diminished by this comparison engaged less with technology.

Technology for Education and Support

Clinicians described how technology can support adolescent mothers and improve their access to care. Some clinicians used texting to confirm appointments or alert patients that test results had become available. Community health workers said they gave their phone numbers to patients, and occasionally, they would use it to ask for help.

Some clinicians said that adolescents did not use technology for educational purposes, for example, to access information about their infants' health or their own. However, a number of adolescent mothers and fathers reported that they used web-based searches to access information. One mother used web-based searches to learn about childbirth when she was pregnant. Others used the internet to learn more about introducing solid foods and how to manage common childhood ailments like diaper rash, as in the case of this father: "Most of all it was with the diaper rash and some test results that were sent to us online [that we used our smartphones]."

Adolescent mothers also used technology to help manage their daily tasks. This included coordinating with other members of their household and searching for answers to any number of practical questions related to cooking or home repair. They also used their smartphones to show their infants cartoons to quiet

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them or entertain them when they had to do household chores. They believed it was positive and stimulating for their infants: "That is what he likes because they do these mimics, thankfully, [he likes] the talking animals."

Social Connection

Adolescent mothers and clinicians both reported that adolescents used social media, but their attitudes toward this differed. Clinicians felt that social media use was not productive or useful in the service of the tasks that adolescent mothers needed to care for themselves and their infants. However, the accounts of adolescents painted a more complex picture. Social media did make it easier to stay connected with friends while being home with their babies, but the quality of the connection varied. Adolescent mothers reported that they were less socially engaged after having their babies. They did not go out and meet their friends regularly. They also reported that their social media use decreased but that social media still made it possible to stay connected in a more limited way. For example, they could post photos on social media and receive supportive comments from their friends. One adolescent found some classmates from childhood on Facebook and saw that some were also mothers. This was comforting to her as none of her friends had babies. However, she did not reach out to reconnect with these classmates:

> *There are several classmates who have had babies, but it's not that we talk; just that I see on Facebook that they do have babies. But they don't talk to me, we weren't very close, we were just acquaintances from class.*

This participant found some comfort in finding others in similar situations on social media, but this did not necessarily lead to connection with them.

Those who were feeling alone because of estrangement from their families or significant conflict with them looked to social media for a sense of connection, but they found more disconnection and conflict. One adolescent described viewing pictures her friends posted and feeling like she was missing out on the social life that she had before becoming a mother. Another adolescent mother learned that her baby's father had a new partner from pictures he posted on social media. For participants managing conflictual relationships, social media was a more acceptable way to engage because it felt more impersonal and, therefore, safer. For this reason, it could also be perceived as distancing and offending the recipient, as was the case with the mother of an adolescent, who responded to a Facebook message she received from her adolescent daughter:

> *How cruel that you mark your indifference against me, that you do not greet me, that you do as if you do not see me, you do not look up, you do not speak to me, you do not visit me, you do not know or ask how I am or anything.*

Facebook greeting was experienced as slight when an in-person greeting was expected. In these instances, social media became another space in which the participants experienced relationship difficulties.

Adolescent participants also reported that they had difficulty setting boundaries around their social media communications. In the case of one participant who was estranged from her child's father, the father discovered that she was in labor through Facebook and sent her a message. This was experienced as both an intrusion and a reminder of his absence. She was angry at him because of the way their relationship had ended, and contact from him at the moment she was in labor was overwhelming:

> *He's the one that came here asking to meet his son because he found out that I had been at [the hospital]. The day that I gave birth he sent me a message on Facebook, but I didn't know because I was giving birth, right? When they finally gave me my cellphone at midnight I saw his messages, and he wanted to come all the way from [neighboring country] that same day that my son was born. But I didn't write back at all because I didn't want him to meet my son. But he did meet him recently.*

Although this participant ultimately allowed her child's father to meet his son, her experience demonstrates that social media can make it easy to initiate contact and difficult to manage incoming contact. Another participant was awaiting a child support payment from her child's father and received this message from the father's new partner through social media:

> *Don't worry, he'll send you the monthly payment. Don't get upset; don't try to control him.*

The participant experienced this as an unwanted intrusion. It was a communication about the missing payment, but it did not address the issue. Some adolescents found social media distracting when they were supposed to be doing other things, such as attending school. One participant avoided having data minutes on her phone so she could only use it when she was connected to internet at home:

> *No, if I reload it [with data minutes], it will distract me when I'm at school. Better not, no.*

Adolescents found it difficult to restrict unwanted contacts on social media, and they also found it difficult to restrict their own use.

Discussion

This study examined technology access, use patterns, and barriers among adolescent mothers in Peru. To the best of our knowledge, this is the first study to investigate the access and use of technology by adolescent mothers in LMICs. Despite the potential benefits of technology in promoting and improving access to care, the use of technology to support perinatal health among adolescent mothers remains relatively unexplored.

Overall, we found that adolescent mothers are connected to technology; all had smartphone access, and nearly half also had access to a laptop or tablet. These findings match the available data on adolescent technology access in HICs [30,31] and highlight the potential scalability of technology-based health interventions for adolescent mothers in LMICs. In addition, we found that all participants used social media at least once per week (13/28, 46% connected daily), and they had a range of

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experiences with it. Adolescent mothers reported difficulty in setting boundaries around their social media communications. These findings are consistent with past research, which has described the adverse impact of excessive social media use on adolescent mental health, well-being, and productivity [32-35]. Importantly, evidence indicates that these negative effects are stronger in girls than in boys [36]. Adolescent mothers and clinicians had different perspectives on adolescent technology use. They agreed that social media use could be problematic, but some of the helpful functions of technology were not visible to clinicians. Adolescents reported finding smart devices useful in searching for web-based health information, improving their health literacy, performing daily tasks, and supporting their care.

Similarly, some clinicians found these tools beneficial for disseminating test results, enhancing patient communication, and ensuring adequate follow-up care. However, they did not believe that adolescents used technology to attend to their own or their infant's health; they saw it as purely a distraction. The clinician's perspective may reflect a bias among clinicians to problematize patient behavior and focus on what they perceive patients to be doing wrong [37]. This may also indicate the need for guidance in adolescent technology use. Aschbrenner reported that adolescents preferred digital health interventions that included a professional moderator to manage peer-to-peer interactions [30].

Quantitative surveys were conducted during the pandemic, and qualitative interviews were conducted before the pandemic. We did not obtain qualitative data about the experience of the pandemic for adolescent mothers, but data on the impact of the pandemic on other vulnerable youth are relevant to this question [38]. School enrollment declined in many parts of the world [39], whereas violence and criminal behavior increased. However, in areas with adequate technology penetration, the increased availability of digital services improved access for some groups, including youth with chronic illnesses [40]. Adolescent mothers find it more difficult to leave their homes compared with other adolescents, and the expansion of services such as mHealth and remote learning may be particularly beneficial for them, which warrants further study.

Although a majority of participants had consistent access to technology, 3 major barriers were identified: (1) cost, (2) phone theft and loss, and (3) internet speed and signal strength. The cost of phones and data plans was a barrier; many adolescent mothers had difficulty affording smartphone services and thus relied on shared plans and sometimes shared devices. If they could no longer afford their plan, they would deactivate their device and use someone else's. Although this allows them to maintain access, it means that they frequently change phone numbers and cannot be reached consistently by health care providers. This finding highlights the fact that adolescents can use mobile technology to access resources, but the health care system may have difficulty maintaining contact with them. This could limit the effectiveness of interventions that rely on consistent contact between adolescents and care providers, as adolescents in precarious circumstances may not be able to bear the responsibility for maintaining contact. They may be ambivalent about contact with the health care system, particularly if they feel criticized by providers. They may also be unable to manage and organize the information required to maintain contact. In addition, most phone plans in Peru are organized such that charges apply to calls and texts made but not to those received. Thus, each time adolescents change numbers, they must bear the expense of reconnecting.

Limited financial resources also meant that many adolescent mothers lived in neighbors with high rates of poverty where crime was endemic, and they experienced frequent device theft. This added to the need for shared devices, phone number changes, and other barriers to consistent contact. Owing to the crime, clinicians also feared bringing their devices on home visits to patients, limiting their ability to use technology as an educational tool.

Participants also reported that slow internet speed could interfere with the use of certain apps, including video streaming and videoconferencing, and rolling blackouts limited consistent access. Internet speed is typically slower and signal strength is weaker in LMICs, where 3G access exceeds 4G [41]. Areas where poverty is greatest and access to in-person care is most limited also face the greatest internet coverage gaps.

This study has several limitations. First, the sample size was small, with 29 adolescent mothers surveyed. Although the sample size for both the focus group discussions and the interviews was small, data analysis indicated that theoretical saturation was reached. In both phases of data collection, only Spanish speakers were recruited, which could limit generalizability and specifically applicability to indigenous communities in Peru. Second, participants were recruited from a single tertiary care women's hospital in Lima, Peru. Adolescent mothers in other parts of Peru, or, for that matter, those receiving care at other clinical sites, were not included, and the data may not represent their experience. Third, there is the possibility of selection bias, as adolescents who chose to participate may be different from those who did not.

Our results indicate that adolescent mothers in Peru have access to digital technology, and some already use technology to help them care for their infants. These findings highlight the promise and potential of using scalable digital health interventions in LMIC settings to expand access to care and improve perinatal health outcomes. Of note, the pandemic is making it increasingly clear about the importance of telehealth in addressing health needs. However, barriers remain in the implementation of telehealth solutions in LMICs. A coordinated global approach centered around equity is needed to reduce these barriers by increasing personal device access, addressing limitations to maintaining access related to poverty, and growing internet connectivity. Subsidies are needed to increase technology access at the local level, and a coordinated global effort is needed to increase internet access as an important step toward reducing global health inequity.

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Conflicts of Interest

None declared.

References

1. Wallis L, Blessing P, Dalwai M, Shin SD. Integrating mHealth at point of care in low- and middle-income settings: the system perspective. Glob Health Action 2017 Jun 25;10(sup3):1327686 [FREE Full text] [doi: 10.1080/16549716.2017.1327686] [Medline: 28838302]
2. ICT facts and figures 2016. International Telecommunication Union. 2016. URL: https://www.itu.int/en/ITU-D/Statistics/Documents/facts/ICTFactsFigures2016.pdf [accessed 2021-03-31]
3. Kruse C, Betancourt J, Ortiz S, Valdes Luna SM, Bamrah IK, Segovia N. Barriers to the use of mobile health in improving health outcomes in developing countries: systematic review. J Med Internet Res 2019 Oct 09;21(10):e13263 [FREE Full text] [doi: 10.2196/13263] [Medline: 31593543]
4. Poorman E, Gazmararian J, Parker RM, Yang B, Elon L. Use of Text Messaging for Maternal and Infant Health: A Systematic Review of the Literature. Matern Child Health J 2014 Aug 1;19(5):969-989. [doi: 10.1007/s10995-014-1595-8]
5. Lee SH, Nurmatov UB, Nwaru BI, Mukherjee M, Grant L, Pagliari C. Effectiveness of mHealth interventions for maternal, newborn and child health in low- and middle-income countries: Systematic review and meta-analysis. J Glob Health 2016 Jun;6(1):010401 [FREE Full text] [doi: 10.7189/jogh.06.010401] [Medline: 26649177]
6. Mayberry LS, Jaser SS. Should there be an app for that? The case for text messaging in mHealth interventions. J Intern Med 2018 Feb 25;283(2):212-213 [FREE Full text] [doi: 10.1111/joim.12687] [Medline: 28881402]
7. Colaci D, Chaudhri S, Vasan A. Mhealth interventions in low-income countries to address maternal health: a systematic review. Ann Glob Health 2016;82(5):922-935 [FREE Full text] [doi: 10.1016/j.aogh.2016.09.001] [Medline: 28283147]
8. Langlois EV, Miszkurka M, Zunzunegui MV, Ghaffar A, Ziegler D, Karp I. Inequities in postnatal care in low- and middle-income countries: a systematic review and meta-analysis. Bull World Health Organ 2015 Apr 01;93(4):259-270. [doi: 10.2471/blt.14.140996]
9. Dol J, Campbell-Yeo M, Tomblin Murphy G, Aston M, McMillan D, Richardson B. Impact of mobile health interventions during the perinatal period for mothers in low- and middle-income countries. JBI Database System Rev Implement Rep 2019;17(2):137-146. [doi: 10.11124/jbisrir-2017-003801]
10. Ezezika O, Varatharajan C, Racine S. Scaling mhealth in Africa: Lessons from the implementation of the momconnect program. Res Square 2020 Nov 01:1-30 (forthcoming). [doi: 10.21203/rs.3.rs-103769/v1]
11. Harpham T, Huttly S, De Silva MJ, Abramsky T. Maternal mental health and child nutritional status in four developing countries. J Epidemiol Community Health 2005 Dec 01;59(12):1060-1064 [FREE Full text] [doi: 10.1136/jech.2005.039180] [Medline: 16286495]
12. Atkinson L, Paglia AC, Coolbear J, Niccols A, Parker KC, Guger S. Attachment security: a meta-analysis of maternal mental health correlates. Clin Psychol Rev 2000;20(8):1019-1040. [doi: 10.1016/s0272-7358(99)00023-9]
13. Engle PL. Maternal mental health: program and policy implications. Am J Clin Nutr 2009;89(3):963-966. [doi: 10.3945/ajcn.2008.26692g]
14. Baggett K, Davis B, Feil E, Sheeber L, Landry S, Leve C, et al. A randomized controlled trial examination of a remote parenting intervention: engagement and effects on parenting behavior and child abuse potential. Child Maltreat 2017 Nov 07;22(4):315-323 [FREE Full text] [doi: 10.1177/1077559517712000] [Medline: 28587520]
15. Acharibasam JW, Wynn R. Telemental health in low- and middle-income countries: a systematic review. Int J Telemed Appl 2018 Nov 01;2018:9602821 [FREE Full text] [doi: 10.1155/2018/9602821] [Medline: 30519259]
16. Fu Z, Burger H, Arjadi R, Bockting CL. Effectiveness of digital psychological interventions for mental health problems in low-income and middle-income countries: a systematic review and meta-analysis. Lancet Psychiatry 2020 Oct;7(10):851-864. [doi: 10.1016/s2215-0366(20)30256-x]
17. WHO Guidelines on preventing early pregnancy and poor reproductive outcomes among adolescents in developing countries. World Health Organization. 2011. URL: http://apps.who.int/iris/bitstream/handle/10665/44691/9789241502214_eng.pdf;jsessionid=8BF30DBDACD7E43D8611DC645C9BFEA3?sequence=1 [accessed 2021-02-22]
18. Adolescent Fertility Rates. World Bank. 2019. URL: https://data.worldbank.org/indicator/SP.ADO.TFRT [accessed 2021-03-05]

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19. Sansom-Daly UM, Wakefield CE, Bryant RA, Patterson P, Anazodo A, Butow P, Recapture Life Working Party. Feasibility, acceptability, and safety of the Recapture Life videoconferencing intervention for adolescent and young adult cancer survivors. Psychooncology 2019 Feb 28;28(2):284-292. [doi: 10.1002/pon.4938] [Medline: 30414219]
20. Välimäki M, Anttila K, Anttila M, Lahti M. Web-based interventions supporting adolescents and young people with depressive symptoms: systematic review and meta-analysis. JMIR Mhealth Uhealth 2017 Dec 08;5(12):e180. [doi: 10.2196/mhealth.8624]
21. Havas J, de Nooijer J, Crutzen R, Feron F. Adolescents' views about an internet platform for adolescents with mental health problems. Health Edu 2011 Apr 19;111(3):164-176. [doi: 10.1108/09654281111123466]
22. Fatori D, Argeu A, Brentani H, Chiesa A, Fracolli L, Matijasevich A, et al. Maternal parenting electronic diary in the context of a home visit intervention for adolescent mothers in an urban deprived area of São Paulo, Brazil: randomized controlled trial. JMIR Mhealth Uhealth 2020 Jul 28;8(7):e13686 [FREE Full text] [doi: 10.2196/13686] [Medline: 32720906]
23. Hennink M, Hutter I, Bailey A. Qualitative Research Methods. Los Angeles: SAGE Publications; 2011:1-328.
24. Cobb D, Mynhier J, Cobb S, McGuffey A. Complete Guide to Microsoft EXCEL on the Apple Macintosh. Seattle: Microsoft Press; 1990:1-750.
25. Hsieh H, Shannon SE. Three approaches to qualitative content analysis. Qual Health Res 2005 Nov 01;15(9):1277-1288. [doi: 10.1177/1049732305276687] [Medline: 16204405]
26. Burla L, Knierim B, Barth J, Liewald K, Duetz M, Abel T. From text to codings: intercoder reliability assessment in qualitative content analysis. Nurs Res 2008;57(2):113-117. [doi: 10.1097/01.nnr.0000313482.33917.7d]
27. QSR. NVivo qualitative data analysis software. QSR International Pty Ltd. 2015. URL: https://www.qsrinternational.com/nvivo-qualitative-data-analysis-software/home [accessed 2021-09-03]
28. Cohen J. A coefficient of agreement for nominal scales. Edu Psychol Measure 2016 Jul 02;20(1):37-46. [doi: 10.1177/001316446002000104]
29. McHugh ML. Interrater reliability: the kappa statistic. Biochem Med 2012:276-282. [doi: 10.11613/bm.2012.031]
30. Aschbrenner KA, Naslund JA, Tomlinson EF, Kinney A, Pratt SI, Brunette MF. Adolescents' use of digital technologies and preferences for mobile health coaching in public mental health settings. Front Public Health 2019 Jul 2;7:178 [FREE Full text] [doi: 10.3389/fpubh.2019.00178] [Medline: 31312629]
31. Hausmann JS, Touloumtzis C, White MT, Colbert JA, Gooding HC. Adolescent and young adult use of social media for health and its implications. J Adolesc Health 2017 Jun;60(6):714-719 [FREE Full text] [doi: 10.1016/j.jadohealth.2016.12.025] [Medline: 28259620]
32. Cho YG. Excessive and problematic smartphone use and poor mental health in adolescents. Korean J Fam Med 2020 Mar;41(2):73-74. [doi: 10.4082/kjfm.41.2e]
33. Kelly Y, Zilanawala A, Booker C, Sacker A. Social media use and adolescent mental health: findings from the UK millennium cohort study. EClinicalMedicine 2018 Dec;6:59-68 [FREE Full text] [doi: 10.1016/j.eclinm.2018.12.005] [Medline: 31193561]
34. O'Reilly M. Social media and adolescent mental health: the good, the bad and the ugly. J Ment Health 2020 Apr 28;29(2):200-206. [doi: 10.1080/09638237.2020.1714007] [Medline: 31989847]
35. Woods HC, Scott H. #Sleepyteens: Social media use in adolescence is associated with poor sleep quality, anxiety, depression and low self-esteem. J Adolesc 2016 Aug;51:41-49. [doi: 10.1016/j.adolescence.2016.05.008] [Medline: 27294324]
36. Abi-Jaoude E, Naylor KT, Pignatiello A. Smartphones, social media use and youth mental health. Can Med Asso J 2020 Feb 10;192(6):136-141 [FREE Full text] [doi: 10.1503/cmaj.190434] [Medline: 32041697]
37. Shoveller JA, Johnson JL. Risky groups, risky behaviour, and risky persons: Dominating discourses on youth sexual health. Critical Public Health 2006 Mar;16(1):47-60. [doi: 10.1080/09581590600680621]
38. Badawy SM, Radovic A. Digital approaches to remote pediatric health care delivery during the COVID-19 pandemic: existing evidence and a call for further research. JMIR Pediatr Parent 2020 Jun 25;3(1):e20049 [FREE Full text] [doi: 10.2196/20049] [Medline: 32540841]
39. Gettleman J, Raj S. As Ccovid-19 closes schools, the world's children go to work. New York Times. 2020 Sep 27. URL: https://www.nytimes.com/2020/09/27/world/asia/covid-19-india-children-school-education-labor.html?action=click&module=Top%20Stories&pgtype=Homepage [accessed 2021-09-03]
40. Serlachius A, Badawy SM, Thabrew H. Psychosocial challenges and opportunities for youth with chronic health conditions during the COVID-19 pandemic. JMIR Pediatr Parent 2020 Oct 12;3(2):e23057 [FREE Full text] [doi: 10.2196/23057] [Medline: 33001834]
41. Bahia K, Delaporte A. The state of mobile internet connectivity 2020. Connected Society. 2020. URL: https://www.gsma.com/r/wp-content/uploads/2020/09/GSMA-State-of-Mobile-Internet-Connectivity-Report-2020.pdf [accessed 2021-03-05]

Abbreviations

HIC: high-income country
INMP: Instituto Nacional Materno Perinatal

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LMIC: low- and middle-income country
mHealth: mobile health
TANI: Taller de Niños

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Original Paper

Perceptions of Social Media Use to Augment Health Care Among Adolescents and Young Adults With Cystic Fibrosis: Survey Study

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Abstract

Background: For individuals with cystic fibrosis (CF), adolescence and young adulthood are times of significant vulnerability and have been associated with clinical and psychosocial challenges. Social media may offer innovative care delivery solutions to address these challenges.

Objective: This study explored motivations and attitudes regarding current social media use and preferences for a social media platform in a sample of adolescents and young adults (AYA) with CF.

Methods: A cross-sectional survey was administered to 50 AYA with CF followed at a large pediatric-adult CF center. The survey included questions regarding social media platform utilization, attitudes toward general and CF-specific online activities, and preferences for a CF-specific care delivery platform.

Results: YouTube, Snapchat, and Instagram were the most commonly used social media platforms. AYA with CF do not report routinely using social media for health-related information acquisition, social support, or help with adherence. However, their perceptions of social media utilization and preferences for platform development suggest interest in doing so in the future.

Conclusions: AYA with CF use social media and expressed interest in the development of a social media platform. Platform development will allow for gaps in health care delivery to be addressed by improving social support and adherence while augmenting current methods of health information acquisition.

(JMIR Pediatr Parent 2021;4(3):e25014) doi:10.2196/25014

KEYWORDS

cystic fibrosis; social media; mobile health; adherence; adolescents; young adults

Introduction

Background

Cystic fibrosis (CF) is the most common life-shortening genetic disease in White people in the United States [1,2]. The CF care model, which focuses on coordinated, multidisciplinary care delivery in specialized CF care centers, has contributed to improvements in clinical outcomes. CF was previously a disease with nearly uniform death in childhood but improved care has resulted in a median predicted survival age of patients of 46.2 years [1]. Current clinical guidelines for patient care recommend patients be evaluated quarterly and more frequently during the first year of life or with the illness [3-5].

Despite care delivery improvements, adolescents and young adults (AYA) with CF still face significant vulnerabilities. Studies in CF adolescents have revealed reductions in medication adherence [6], reductions in lung function despite more aggressive management and implementation of new therapies [7], increased symptom burden [8], and development or acceleration of CF-related complications [8]. Individuals

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with CF often face significant feelings of isolation [6,9] and are unable to congregate in typical support group structures [10] due to infection control guidelines [11]. These issues remain largely unaddressed by the current care delivery paradigm. Novel approaches to health care delivery are needed during this vulnerable time to support AYA with CF.

Prior Work

Electronic health applications such as telehealth, mobile health (mHealth), and social media may be beneficial in addressing gaps in the current care delivery paradigm. Previously, mHealth and social media have been proposed as opportunities to engage AYA and augment care delivery [12,13]. These platforms may allow for improvements in social support, provide innovative networking opportunities for patients and their care team, and provide novel methods to improve disease self-management and adherence.

Previous mHealth studies have been carried out across diseases and were found to improve clinical outcomes, medication adherence, and self-monitoring in solid organ transplantation [14-17], essential hypertension [18,19], congestive heart failure [20], type 2 diabetes [21], and coronary artery disease [22]. However, not all have shown a clinically significant impact or improvements in adherence [23]. Within CF, a focus group study suggested implementation of a web-enabled cellphone would improve knowledge, provide social support, and result in improved adherence [24]. Another study exploring mHealth preferences noted the importance of automated assistance with disease management, improving communication with the multidisciplinary care team, and facilitating socialization with others with CF [25]. Although mHealth applications have been shown to have high acceptability [26], their long-term uptake remains problematic [13].

Social media has been used in other pulmonary diseases to understand patient experience [27], to characterize patient preferences for information acquisition [28], and to improve self-management and social support [29]. Social media has been leveraged to develop a disease-specific social network for chronic obstructive pulmonary disease to address loneliness and improve social support [30,31]. However, studies investigating social media usage in AYA with CF are lacking. Facebook has been described as a method for information dissemination by a large adult CF center in the United Kingdom [32]. Facebook, Facetime, and Instagram were described in a small qualitative study of 9 Canadian AYA with CF as a method to reduce social isolation and improve support [33]. A more recent study assessed 66 adults with CF or direct connection to CF (caregiver, significant other, immediate family member) regarding social media usage. The study found 98% of respondents used social media, with 96% of respondents using an online forum (Facebook, cysticfibrosis.com, healingwell.com). The study however had significant limitations in that it only captured adults, surveyed predominantly those without CF (39% had CF), and suffered from ascertainment bias as recruitment was performed online via email, on social media websites, and using CF dedicated message boards [34]. Larger survey studies to assess preferences for a CF-related social media platform, CF-related social media usage, perceptions regarding online CF health activities, and current social media platform usage among AYA with CF are lacking. It is crucial that patient motivations for social media usage and preferences for such platforms are explored to inform care delivery.

Goal of This Study

The goal of this study was to characterize current social media utilization patterns and attitudes, motivations for social media utilization, and preferences for social media platform development in AYA with CF.

Methods

Recruitment

We performed a cross-sectional survey of AYA with CF receiving care at a large pediatric-adult CF center. Participants were eligible for enrollment if they were aged between 13-30 years and diagnosed with CF. Patients were only excluded if they had previously received a solid organ transplantation. Participants were recruited from the ambulatory CF clinic and during hospital admissions on the inpatient pulmonary service from October-December 2018. Informed consent/assent was obtained electronically, and the study was approved by the institutional review board at Boston Children's Hospital (IRB-P0025949). The survey was administered by iPad using an online link to the survey.

Clinical Measurements

Clinical parameters including percent predicted forced expiratory volume in 1 second (ppFEV1), hospitalizations over the last year, BMI, cystic fibrosis–related diabetes status, age, and gender were abstracted from the medical record.

Survey Battery

The survey included 60 multiple choice questions (Multimedia Appendix 1) and explored general health/demographics (11 questions), potential components to include in a social media platform (12 questions), CF-related social media usage (11 questions), perceptions regarding online CF health activities (16 questions), and current social media platform utilization (10 questions). The topics included in the instrument were developed on the basis of previous research and scholarship surrounding social media utilization in CF [35]. The survey was piloted with 3 patients. Responses were reviewed to ensure appropriate question branching logic based on patient responses. Study data were collected and managed using Research Electronic Data Capture (REDCap) tools [36,37].

Data Analysis

Descriptive statistics were used to analyze respondent demographics, social media utilization, attitudes/perceptions/motivations during utilization, and preferred components of a social platform. Respondent Likert scale ratings for social media platforms and attitudes during social media utilization were operationalized in the following way: "1=never" and "2=rarely" were categorized together and "3=sometimes" and "4=often" were categorized together. Additional questions for perceptions/motivations during social

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media utilization were operationalized as follows: "1=strongly disagree" and "2=disagree" were categorized together and "3=agree" and "4=strongly agree" were categorized together. Finally, Likert scale ratings for components of social media platforms were operationalized in the following way: "1=not at all important" and "2=somewhat unimportant" were categorized together and "3=somewhat important" and "4=very important" were categorized together. The associations of respondents' social media utilization, attitudes/perceptions/motivations of current social media utilization, and component preferences for a social media platform with clinical parameters were assessed via chi-square test, paired *t* test, or analysis of variance, as appropriate. All analyses were conducted using STATA (version 15.1; StataCorp LLC).

Results

During the study period, 147 patients met the eligibility requirements for inclusion in the study. Of these, 80 patients were not approached as they cancelled their appointment, were felt to be inappropriate for research by their primary pulmonologist, or finished their clinic visit before the research team could approach them. Of the 67 remaining patients, 17 declined to participate in the study. A total of 50 AYA completed the survey (Table 1).

Table 1. Cohort characteristics.

Characteristics of participants	Values
Gender, n (%)	
Entire cohort	50 (100)
Males	25 (50)
Females	25 (50)
Age (n=50), mean/range (SD)	19.8/13-30 (5.2)
Racial/ethnic background,[a] n (%)	
White	49 (98)
Latino	2 (4)
Black	1 (2)
Other	1 (2)
Highest education level achieved, n (%)	
Some high school or less	22 (44)
High school or General Educational Development (GED)	4 (8)
Some college	6 (12)
College/vocational degree	13 (26)
Professional/graduate degree	5 (10)
Current health status, n (%)	
Excellent	9 (18)
Very good/good	32 (64)
Fair/poor	9 (18)
Cystic fibrosis severity, n (%)	
Mild	24 (48)
Moderate	23 (46)
Severe	3 (6)
Cystic fibrosis–related diabetes[b] (n=50), n (%)	9 (18)
BMI[b] (n=50), mean/range (SD)	21.3/15.4-35.9 (3.6)
Percent predicted forced expiratory volume in 1 second[b] (n=50), mean/range (SD)	77.2/26-126 (23.5)
Hospitalizations[b] (n=50), mean/range (SD)	1.4/0-8 (1.9)

[a]Respondents instructed to select all racial/ethnic backgrounds with which they identify.

[b]Abstracted from the electronic medical record.

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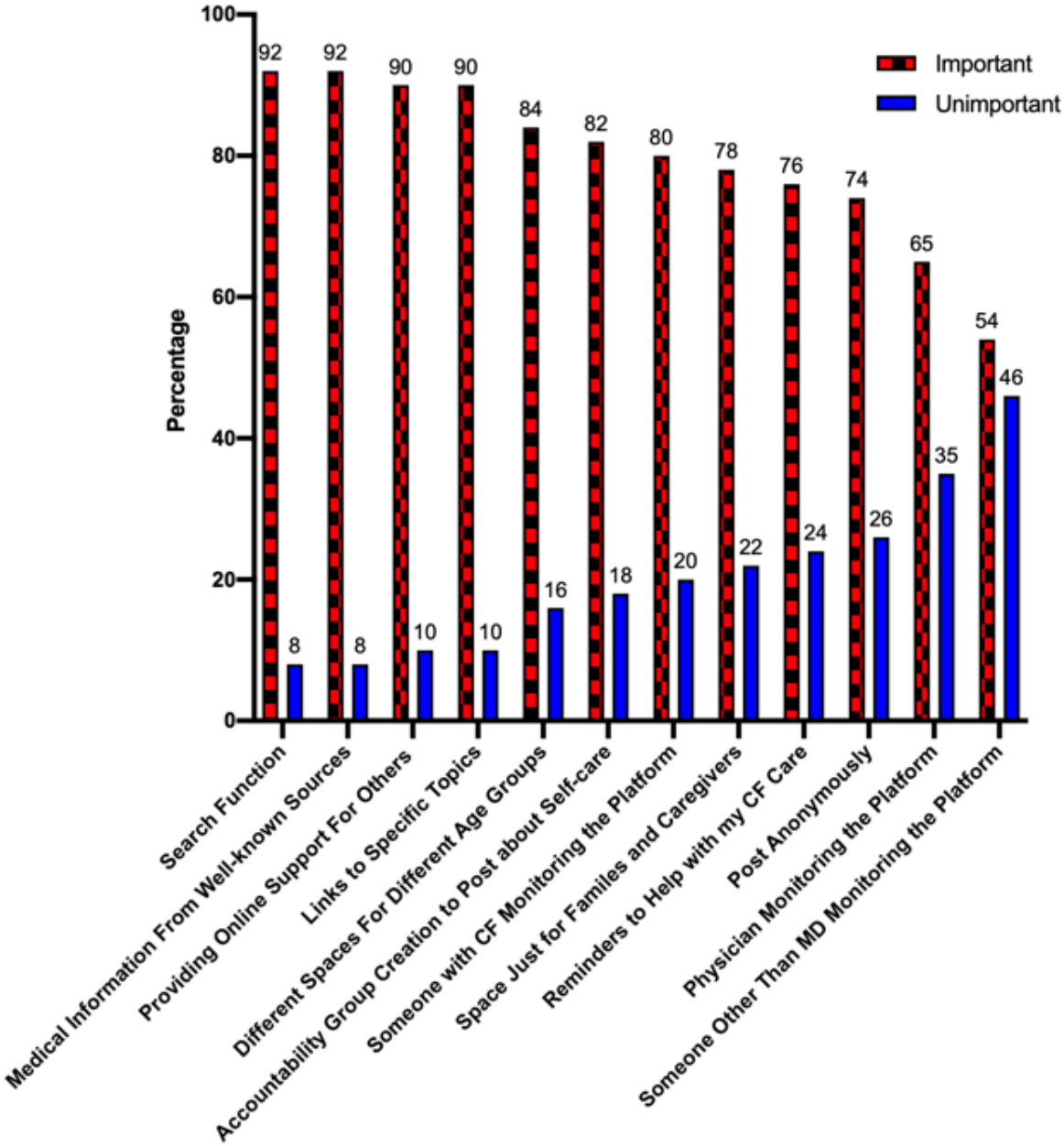
Components to include in a social platform for AYA with CF are noted in Figure 1. Respondents conveyed that a platform should include elements to help with care delivery. Overall, 44 of 49 (90%) endorsed that it was important to include a forum to provide online support for people with CF, 82% (41/50) requested accountability group creation where participants can post about self-care, and 76% (38/50) desired reminders to help with CF self-care. In addition, respondents also expressed strong support for including elements for acquisition of accurate health information. For example, 92% (45/49) wanted medical information available that came from well-known sources such as the CF Foundation and 90% (45/50) felt it was important to include links to specific CF topics.

Figure 1. Potential components to include in a CF social media platform.



Current CF-related social media usage is shown in Figure 2. When evaluating current online health-related activities, 50% (25/50) endorsed reading about CF-related information online "sometimes" or "often." These respondents were older (mean age of readers 22 years versus 17.6 years for nonreaders; *P*=.002) and were more likely to be female (64% female readers versus 36% male readers; *P*=.05). In addition, 42% (21/50) of respondents learned about other people's experiences with CF online. Only 24% of respondents (12/50) endorsed interacting with other patients with CF online and even fewer (7/50, 14%) sought support from other people with CF online.

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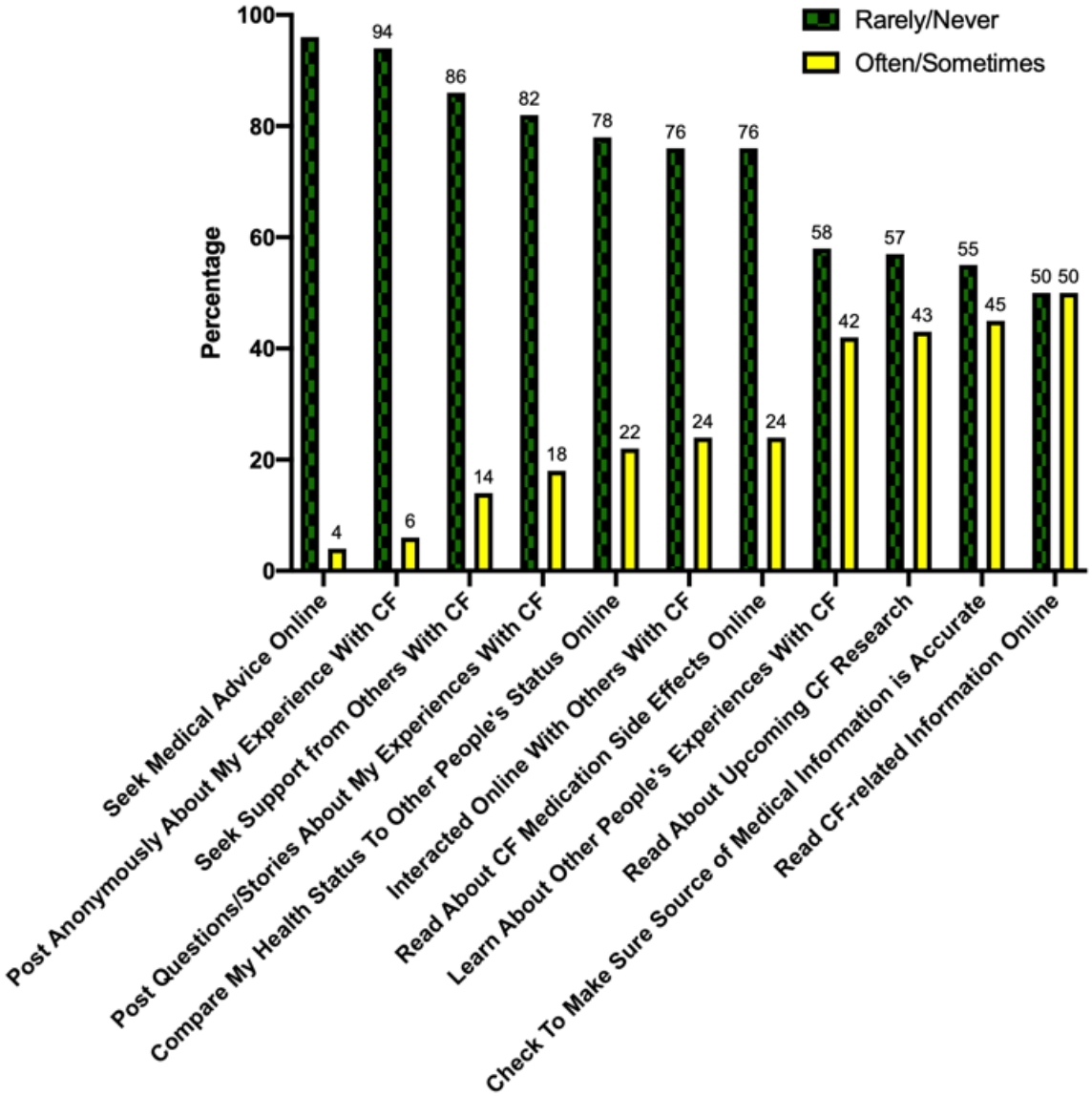
Figure 2. Cystic fibrosis–related social media use.



Although only a minority of respondents reported current social media use for online CF-related health activities, their perceptions regarding the potential uses for online CF-related health activities were different (Table 2). For example, only 42% (21/50) endorsed learning about other people's experiences with CF online. However, 77% (37/48) reported they "agree" or "strongly agree" that they feel inspired by stories of other people with CF online (37/48) and 65% (32/49) noted that they wanted to motivate and inspire others with CF.

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Table 2. Perception regarding online cystic fibrosis health activities.

Perceptions	Strongly agree/agree, n (%)	Strongly disagree/disagree, n (%)
Medical information online should come from a source like the CF Foundation (n=49)	45 (92)	4 (8)
Medical information should be from a source like a doctor (n=50)	45 (90)	5 (10)
I feel inspired by stories of other people with CF online (n=48)	37 (77)	11 (23)
It is important to me to keep my privacy online (n=50)	35 (70)	15 (30)
I want to inspire and motivate others with CF online (n=49)	32 (65)	17 (35)
Online bullying or trolling is a serious risk (n=50)	32 (64)	18 (36)
I feel motivated to perform my own self-care when I see other people with CF online (n=49)	31 (63)	18 (37)
I feel less alone when I read stories about other people's struggles with CF (n=48)	27 (56)	21 (44)
Medical information should come from a source like pharmaceutical companies (n=50)	27 (54)	23 (46)
I feel supported by other people with CF online (n=48)	23 (48)	25 (52)
I wish I could meet other people with CF in real life after meeting them online (n=48)	22 (46)	26 (54)
I feel sad or scared when I learn about other people's CF stories online (n=49)	13 (27)	36 (73)
I feel uncomfortable when comparing my health status to those of others with CF online (n=49)	12 (24)	37 (76)
I avoid other people with CF online (n=49)	7 (14)	42 (86)
I have met other people with CF in real life after meeting them online (n=48)	6 (13)	42 (87)
I feel less motivated to perform my own self-care when I interact with others with CF online (n=48)	2 (4)	46 (96)

Nearly two-thirds (31/49, 63%) endorsed feeling more motivated to perform their own self-care when interacting with others with CF online. In addition to the positive impact on motivation and inspiration, 56% (27/48) noted feeling less alone when they read stories about other people's struggles with CF. Nearly half (23/48, 48%) of respondents noted feeling supported by other people with CF online. Only 14% (7/49) reported that they actively avoid other people with CF online. When considering the potential negative ramifications of online interaction, 27% (13/49) endorsed feeling sad or scared when learning about other people's CF stories online, 24% (12/49) felt uncomfortable when comparing their health status to those of others with CF online, and 4% (2/48) felt less motivated to perform self-care when interacting with others with CF online. Regarding these perceptions, women were more likely to feel motivated to perform self-care (83% of females [20/24] versus 44% of males [11/25]; P=.004), feel less alone (71% of females [17/24] versus 42% of males [10/24]; P=.04), and endorse feeling supported by others with CF online (63% of females [15/24] versus 33% of males [8/24]; P=.04).

Social media platform utilization among AYA with CF is shown in Figure 3. Respondents who endorsed YouTube usage were younger (mean age 19.1 years in users versus 23.8 years in nonusers; P=.02). In contrast, Facebook usage was higher among older respondents (mean age 21.2 years in users versus 18.1 years in nonusers; P=.03). Female respondents had higher usage of Instagram compared to males (92% of females [23/25] versus 60% of males [15/25]; P=.008).

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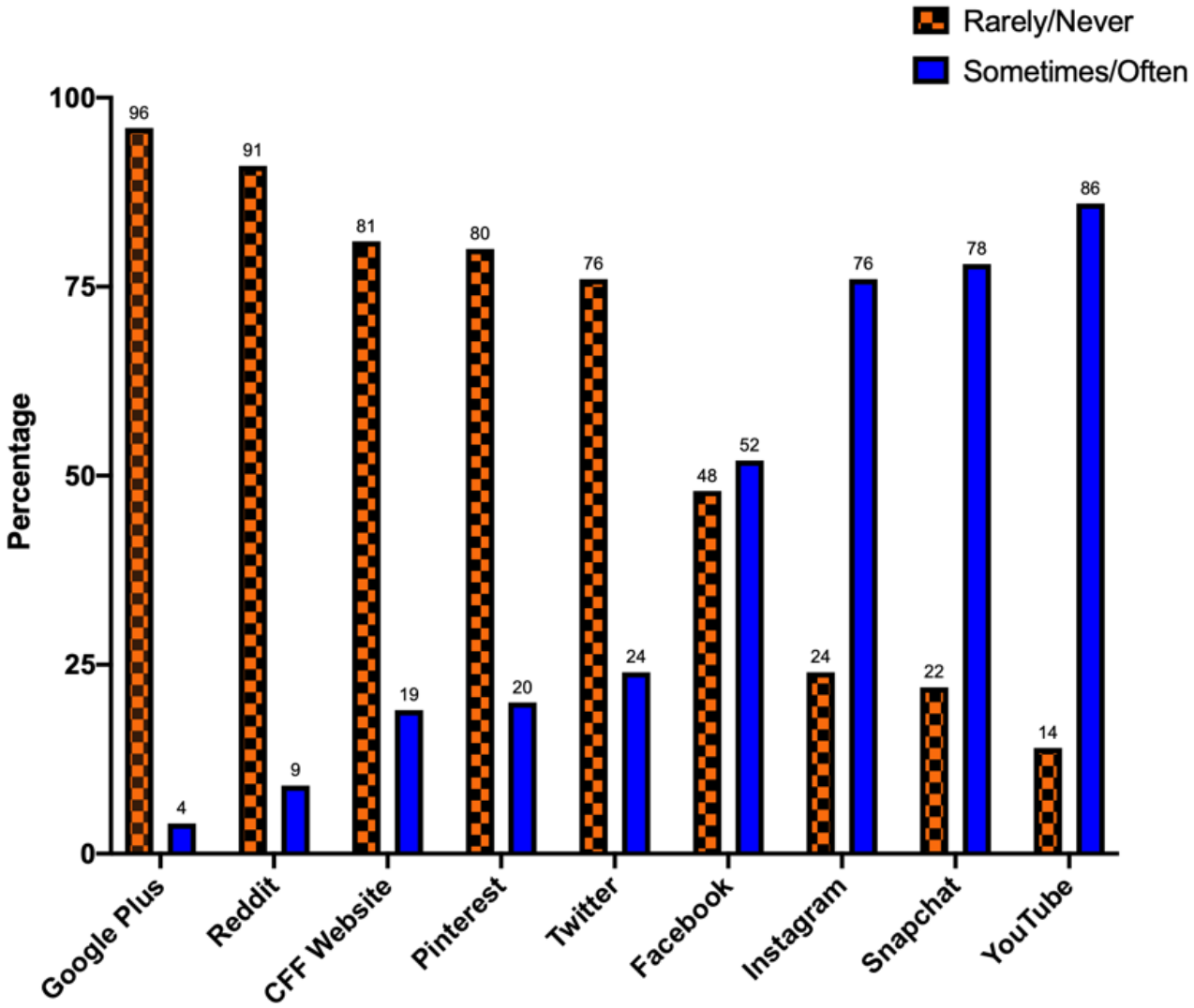
Figure 3. Social media platform use among adolescents and young adults with CF. CFF: Cystic Fibrosis Foundation.



Discussion

Principal Findings

This study highlights several potential avenues for CF social media platforms to improve care delivery for AYA with CF. Social support was a theme routinely identified by study participants as a potential area for exploration. Our study indicated that AYA with CF do not currently use social media for social interaction or support. This may be due to lacking a dedicated platform for interaction, not knowing others with CF, concerns about privacy of identity or medical-related information, or concerns for potential negative emotional consequences that may develop from interactions. Despite not currently using social media platforms for interaction or support, respondents generally expressed encouraging perceptions (feel more supported, feel less alone, feel inspired) regarding their attitudes after interacting with others with CF. This theme was also supported by respondents' preferences for components in a CF social media platform. Their desire for the creation of online forums to provide support to others and the creation of accountability groups suggest participants' perceived importance of connectedness and communication with one another. It should be noted that nearly one-fourth of respondents did experience feelings of sadness, being scared, or discomfort as a result of social interactions online. This suggests that a CF-centered platform may not be an appropriate social support structure for all, particularly those with higher risk of mental health concerns. One previous mHealth study did anticipate the need for mental health resources, medical supervision, and online moderators to provide guidance and support to address this concern [24].

There is growing evidence demonstrating the importance of social support among patients with CF and their health-related outcomes. A longitudinal survey study of 250 adults with CF explored this relationship and found fewer self-reported mental and physical health symptoms, digestive symptoms, and eating disturbances. Social support was associated with improved emotional, social, and role functioning, vitality, and body image. Those who reported more social support perceived less treatment burden and experienced better overall perceptions of their health [38]. Another cross-sectional study of 233 adults with CF highlighted the association of gender on perceived social support in adults with CF, with females perceiving greater levels of support [39]. Likewise, our study also revealed a gender disparity, with females perceiving a greater level of support, increased motivation to perform self-care, and feeling less isolated after interacting with others compared to males in our

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study. These studies highlight the potential role for a CF social media health platform in improving care delivery through augmenting social support and improving health outcomes.

Respondents to the survey also felt a CF social platform could be used to improve motivations for adherence. Participants generally endorsed increased motivation to perform self-care after interacting with others with CF. It is plausible that increased emotional support could lead to improved motivation among respondents and ultimately to an increase in adherence with performing self-care. Increased social interaction did not appear to have a negative reported effect on adherence. Respondents' desires for potential components of an electronic platform (developing accountability groups, reminders to help with self-care) also suggest their interest in improving care delivery through virtual community building while also allowing for individualizing adherence assessments. The respondents' motivations and desires are congruent with interventions proposed to improve adherence through the implementation of patient-centered treatment plans, harnessing technology and application development to increase patient motivation and virtual support, and improving the CF health service model to address individual barriers to adherence [2]. These motivations and desires suggest the possible positive implications that a CF social media health platform can have in improving care delivery through improved patient adherence.

This study indicated that AYA with CF do commonly use social media. Despite this, the majority of respondents do not currently use social media platforms for health-related information acquisition (read about upcoming research, read CF-related information online, read about medication side effects). Their perceptions about what components are important to include in a social media platform suggest their interest in health-related information acquisition online as evidenced by the perceived importance of including a search function, medical information coming from well-known sources, and links to CF-specific topics. It is unclear if participants do not currently perform these actions because they are unaware of where to acquire this information or if they do not trust social media platforms as sources of information. The latter is consistent with a cross-sectional survey of 204 AYA patients without CF in which only 25% felt social media provided useful health information [40]. The Cystic Fibrosis Foundation website provides robust information about CF, medications, and upcoming research that is evidence- and consensus-based, although our study indicates that AYA rarely use this resource.

Limitations of Study

This study has several limitations. First, participants in our study were varied in age, with the majority of respondents under the age of 21. This age distribution may skew the cohort sample and make results less generalizable to adults with CF. However, all of the respondents would be potential users of a future social media platform. Second, though small, our study represents one of the largest samplings to date of AYA patients with CF to investigate social media. Our participants were actively recruited from a CF clinic and during inpatient admissions, which resulted in a study population more representative of the CF community at our large, urban CF center. However, these findings may not be representative of all AYA with CF.

Future Directions

Our study suggests that although AYA with CF do not routinely use existing social media platforms for health-related behaviors, they do express interest in harnessing social media platforms for improving care delivery, accessing social support, and improving therapy adherence. The COVID-19 pandemic created psychosocial challenges for youth with chronic disease, which have implications for mental health and social supports [41]. Likewise, the pandemic also allowed for rapid-cycle digital advances in health care delivery [42] (eg, the popularization of the use of video-based applications for routine clinical communication). Many social media applications currently integrate video-based functions for users. This may allow for synchronous communication between people/care teams using videoconferencing rooms on Facebook or messaging/video calling features on WhatsApp, Facebook Messenger, or Google Duo. In addition, asynchronous communication may also have utility by posting recordings using Facebook Live or Instagram Live, or posting video stories (a feature of many social media platforms) for others as a form of encouragement or as a reminder. These features may have utility as a means for providing group-based support, facilitating accountability groups, or fostering socialization between AYA with CF and warrant further investigation. Additional considerations should be given to exploring the perceptions surrounding social media platform integration with the CF multidisciplinary team and routine clinical care, privacy concerns, and the implications of social determinants of health on social media use.

Finally, additional insight is needed to characterize baseline social isolation in this population and the desired modalities for support (individual video chats, messaging functionality, group video meetings, etc). It would be interesting to assess the use of social media and patients' reported sense of social isolation. For example, do AYA with CF who use social media have a lower sense of social isolation compared to those who do not use social media? Are there differences based on the type of social media modality used? Does gender have an impact on the sense of social isolation? Addressing social isolation is important as a possible means to improve self-care, adherence, and health outcomes. The implications are likely relevant for other chronic illnesses.

In conclusion, our study highlights the possible utility of social media platforms as an innovative intervention for improving health care delivery, social support, and treatment adherence. Overall, gaining insight from AYA with CF during any kind of intervention development will be critical to ensure effectiveness and improving value.

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Authors' Contributions

RCP coordinated and supervised data collection, carried out statistical analysis, and drafted the initial manuscript. RG and KR collected data and reviewed/revised the manuscript. LB conceptualized the study, designed the data collection instrument, and reviewed/revised the manuscript. GSS conceptualized the study, supervised the design of the data collection instrument, assisted with statistical analysis, and reviewed/revised the manuscript. All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

Conflicts of Interest

None declared.

Multimedia Appendix 1
Social media survey battery.
[DOCX File , 24 KB - pediatrics_v4i3e25014_app1.docx]

References

1. Cystic Fibrosis Foundation Patient Registry 2019 Annual Data Report. Cystic Fibrosis Foundation. URL: https://www.cff.org/Research/Researcher-Resources/Patient-Registry/2019-Patient-Registry-Annual-Data-Report.pdf [accessed 2020-02-20]
2. Bishay LC, Sawicki GS. Strategies to optimize treatment adherence in adolescent patients with cystic fibrosis. Adolesc Health Med Ther 2016;7:117-124 [FREE Full text] [doi: 10.2147/AHMT.S95637] [Medline: 27799838]
3. Cystic Fibrosis Foundation, Borowitz D, Robinson KA, Rosenfeld M, Davis SD, Sabadosa KA, et al. Cystic Fibrosis Foundation evidence-based guidelines for management of infants with cystic fibrosis. J Pediatr 2009 Dec;155(6 Suppl):S73-S93 [FREE Full text] [doi: 10.1016/j.jpeds.2009.09.001] [Medline: 19914445]
4. Lahiri T, Hempstead SE, Brady C, Cannon CL, Clark K, Condren ME, et al. Clinical Practice Guidelines From the Cystic Fibrosis Foundation for Preschoolers With Cystic Fibrosis. Pediatrics 2016 Apr;137(4):1-26. [doi: 10.1542/peds.2015-1784] [Medline: 27009033]
5. Yankaskas JR, Marshall BC, Sufian B, Simon RH, Rodman D. Cystic fibrosis adult care: consensus conference report. Chest 2004 Jan;125(1 Suppl):1S-39S. [doi: 10.1378/chest.125.1_suppl.1s] [Medline: 14734689]
6. George M, Rand-Giovannetti D, Eakin MN, Borrelli B, Zettler M, Riekert KA. Perceptions of barriers and facilitators: self-management decisions by older adolescents and adults with CF. J Cyst Fibros 2010 Dec;9(6):425-432 [FREE Full text] [doi: 10.1016/j.jcf.2010.08.016] [Medline: 20846910]
7. Sawicki GS, Heller KS, Demars N, Robinson WM. Motivating adherence among adolescents with cystic fibrosis: youth and parent perspectives. Pediatr Pulmonol 2015 Feb;50(2):127-136 [FREE Full text] [doi: 10.1002/ppul.23017] [Medline: 24616259]
8. Sawicki GS, Ostrenga J, Petren K, Fink AK, D'Agostino E, Strassle C, et al. Risk Factors for Gaps in Care during Transfer from Pediatric to Adult Cystic Fibrosis Programs in the United States. Ann Am Thorac Soc 2018 Feb;15(2):234-240. [doi: 10.1513/AnnalsATS.201705-357OC] [Medline: 29220199]
9. Roehrer E, Cummings E, Beggs S, Turner P, Hauser J, Micallef N, et al. Pilot evaluation of web enabled symptom monitoring in cystic fibrosis. Inform Health Soc Care 2013 Dec;38(4):354-365. [doi: 10.3109/17538157.2013.812646] [Medline: 23957685]
10. Govan JR, Brown PH, Maddison J, Doherty CJ, Nelson JW, Dodd M, et al. Evidence for transmission of Pseudomonas cepacia by social contact in cystic fibrosis. Lancet 1993 Jul 03;342(8862):15-19. [doi: 10.1016/0140-6736(93)91881-l] [Medline: 7686239]
11. Saiman L, Siegel JD, LiPuma JJ, Brown RF, Bryson EA, Chambers MJ, Cystic Fibrous Foundation, Society for Healthcare Epidemiology of America. Infection prevention and control guideline for cystic fibrosis: 2013 update. Infect Control Hosp Epidemiol 2014 Aug;35(S1):S1-S67. [doi: 10.1086/676882] [Medline: 25025126]
12. Wong CA, Merchant RM, Moreno MA. Using social media to engage adolescents and young adults with their health. Healthc (Amst) 2014 Dec;2(4):220-224 [FREE Full text] [doi: 10.1016/j.hjdsi.2014.10.005] [Medline: 25984444]
13. Rudolf I, Pieper K, Nolte H, Junge S, Dopfer C, Sauer-Heilborn A, et al. Assessment of a Mobile App by Adolescents and Young Adults With Cystic Fibrosis: Pilot Evaluation. JMIR mHealth uHealth 2019 Nov 21;7(11):e12442. [doi: 10.2196/12442]
14. Fleming JN, Taber DJ, McElligott J, McGillicuddy JW, Treiber F. Mobile Health in Solid Organ Transplant: The Time Is Now. Am J Transplant 2017 Sep;17(9):2263-2276. [doi: 10.1111/ajt.14225] [Medline: 28188681]

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15. McKenzie RB, Berquist WE, Foley MA, Park KT, Windsheimer JE, Litt IF. Text Messaging Improves Participation in Laboratory Testing in Adolescent Liver Transplant Patients. J Particip Med 2015;7:1-15 [FREE Full text] [Medline: 26213633]
16. McGillicuddy JW, Gregoski MJ, Weiland AK, Rock RA, Brunner-Jackson BM, Patel SK, et al. Mobile Health Medication Adherence and Blood Pressure Control in Renal Transplant Recipients: A Proof-of-Concept Randomized Controlled Trial. JMIR Res Protoc 2013;2(2):e32 [FREE Full text] [doi: 10.2196/resprot.2633] [Medline: 24004517]
17. DeVito Dabbs A, Song MK, Myers BA, Li R, Hawkins RP, Pilewski JM, et al. A Randomized Controlled Trial of a Mobile Health Intervention to Promote Self-Management After Lung Transplantation. Am J Transplant 2016 Jul;16(7):2172-2180 [FREE Full text] [doi: 10.1111/ajt.13701] [Medline: 26729617]
18. Magid DJ, Olson KL, Billups SJ, Wagner NM, Lyons EE, Kroner BA. A pharmacist-led, American Heart Association Heart360 Web-enabled home blood pressure monitoring program. Circ Cardiovasc Qual Outcomes 2013 Mar 1;6(2):157-163 [FREE Full text] [doi: 10.1161/CIRCOUTCOMES.112.968172] [Medline: 23463811]
19. Margolis KL, Asche SE, Bergdall AR, Dehmer SP, Groen SE, Kadrmas HM, et al. Effect of home blood pressure telemonitoring and pharmacist management on blood pressure control: a cluster randomized clinical trial. JAMA 2013 Jul 3;310(1):46-56 [FREE Full text] [doi: 10.1001/jama.2013.6549] [Medline: 23821088]
20. Weintraub A, Gregory D, Patel AR, Levine D, Venesy D, Perry K, et al. A multicenter randomized controlled evaluation of automated home monitoring and telephonic disease management in patients recently hospitalized for congestive heart failure: the SPAN-CHF II trial. J Card Fail 2010 Apr;16(4):285-292. [doi: 10.1016/j.cardfail.2009.12.012] [Medline: 20350694]
21. Quinn CC, Clough SS, Minor JM, Lender D, Okafor MC, Gruber-Baldini A. WellDoc mobile diabetes management randomized controlled trial: change in clinical and behavioral outcomes and patient and physician satisfaction. Diabetes Technol Ther 2008 Jun;10(3):160-168. [doi: 10.1089/dia.2008.0283] [Medline: 18473689]
22. Chow CK, Redfern J, Hillis GS, Thakkar J, Santo K, Hackett ML, et al. Effect of Lifestyle-Focused Text Messaging on Risk Factor Modification in Patients With Coronary Heart Disease: A Randomized Clinical Trial. JAMA 2015;314(12):1255-1263. [doi: 10.1001/jama.2015.10945] [Medline: 26393848]
23. Bhavnani SP, Narula J, Sengupta PP. Mobile technology and the digitization of healthcare. Eur Heart J 2016 Dec 07;37(18):1428-1438 [FREE Full text] [doi: 10.1093/eurheartj/ehv770] [Medline: 26873093]
24. Marciel KK, Saiman L, Quittell LM, Dawkins K, Quittner AL. Cell phone intervention to improve adherence: cystic fibrosis care team, patient, and parent perspectives. Pediatr Pulmonol 2010 Feb;45(2):157-164 [FREE Full text] [doi: 10.1002/ppul.21164] [Medline: 20054860]
25. Hilliard ME, Hahn A, Ridge AK, Eakin MN, Riekert KA. User Preferences and Design Recommendations for an mHealth App to Promote Cystic Fibrosis Self-Management. JMIR mHealth uHealth 2014;2(4):e44 [FREE Full text] [doi: 10.2196/mhealth.3599] [Medline: 25344616]
26. Francis J, Cross D, Schultz A, Armstrong D, Nguyen R, Branch-Smith C. Developing a smartphone application to support social connectedness and wellbeing in young people with cystic fibrosis. J Cyst Fibros 2020 Mar;19(2):277-283. [doi: 10.1016/j.jcf.2019.12.011] [Medline: 31917112]
27. Cook NS, Kostikas K, Gruenberger J, Shah B, Pathak P, Kaur VP, et al. Patients' perspectives on COPD: findings from a social media listening study. ERJ Open Res 2019 Feb;5(1):1-10. [doi: 10.1183/23120541.00128-2018] [Medline: 30775374]
28. Cherrez OI, Baptist A, Calderon J. Preferences of using social media among COPD patients in Latinoamerica. European Respiratory Society 2016;48(Suppl 60):12. [doi: 10.1183/13993003.congress-2016.pa2894]
29. Apperson A, Stellefson M, Paige SR, Chaney BH, Chaney JD, Wang MQ, et al. Facebook Groups on Chronic Obstructive Pulmonary Disease: Social Media Content Analysis. Int J Environ Res Public Health 2019 Oct 09;16(20):1-17 [FREE Full text] [doi: 10.3390/ijerph16203789] [Medline: 31600907]
30. Stellefson M, Paige SR, Alber JM, Stewart M. COPD360social Online Community: A Social Media Review. Health Promotion Practice 2018 Jun 08;19(4):489-491. [doi: 10.1177/1524839918779567]
31. Welch L, Orlando R, Lin SX, Vassilev I, Rogers A. Findings from a pilot randomised trial of a social network self-management intervention in COPD. BMC Pulm Med 2020 Jun 08;20(1):162 [FREE Full text] [doi: 10.1186/s12890-020-1130-1] [Medline: 32513163]
32. Dale C, Reid N, Cox K, Jones A, Williams H, Shawcross A, et al. Using social media to improve communication with people with cystic fibrosis. ERJ Open Res 2016 Jan;2(1):1-3 [FREE Full text] [doi: 10.1183/23120541.00015-2016] [Medline: 27730164]
33. Moola F. The complexities of contagion: The experience of social isolation among children and youth living with cystic fibrosis in Canada. J Child Health Care 2018 Dec;22(4):631-645. [doi: 10.1177/1367493518767784] [Medline: 29614879]
34. Shuman DM. An Assessment of the Cystic Fibrosis Community's Use of Social Media as a Community and Medical Resource. eScholarship. 2017. URL: https://escholarship.org/uc/item/8mw9t844 [accessed 2020-12-25]
35. Bishay LC, Nelson E, Goldmann D, Weitzman E, Sawicki GS. Pediatric Pulmonology. Pediatr Pulmonol 2017 Sep 19;52(S47):S1-S82. [doi: 10.1002/ppul.23837]

XSL•FO
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36. Harris PA, Taylor R, Thielke R, Payne J, Gonzalez N, Conde JG. Research electronic data capture (REDCap)--a metadata-driven methodology and workflow process for providing translational research informatics support. J Biomed Inform 2009 Apr;42(2):377-381 [FREE Full text] [doi: 10.1016/j.jbi.2008.08.010] [Medline: 18929686]
37. Harris PA, Taylor R, Minor BL, Elliott V, Fernandez M, O'Neal L, REDCap Consortium. The REDCap consortium: Building an international community of software platform partners. J Biomed Inform 2019;95:103208 [FREE Full text] [doi: 10.1016/j.jbi.2019.103208] [Medline: 31078660]
38. Flewelling KD, Sellers DE, Sawicki GS, Robinson WM, Dill EJ. Social support is associated with fewer reported symptoms and decreased treatment burden in adults with cystic fibrosis. J Cyst Fibros 2019 Jul;18(4):572-576 [FREE Full text] [doi: 10.1016/j.jcf.2019.01.013] [Medline: 30772245]
39. Flewelling KD, Sellers DE, Sawicki GS, Robinson WM, Dill EJ. Male gender and unemployment are associated with lower levels of perceived social support in adults with cystic fibrosis. J Psychosom Res 2019 Dec;127:109858. [doi: 10.1016/j.jpsychores.2019.109858] [Medline: 31706070]
40. Hausmann JS, Touloumtzis C, White MT, Colbert JA, Gooding HC. Adolescent and Young Adult Use of Social Media for Health and Its Implications. J Adolesc Health 2017 Jun;60(6):714-719. [doi: 10.1016/j.jadohealth.2016.12.025] [Medline: 28259620]
41. Serlachius A, Badawy SM, Thabrew H. Psychosocial Challenges and Opportunities for Youth With Chronic Health Conditions During the COVID-19 Pandemic. JMIR Pediatr Parent 2020 Oct 12;3(2):e23057 [FREE Full text] [doi: 10.2196/23057] [Medline: 33001834]
42. Badawy SM, Radovic A. Digital Approaches to Remote Pediatric Health Care Delivery During the COVID-19 Pandemic: Existing Evidence and a Call for Further Research. JMIR Pediatr Parent 2020 Jun 25;3(1):e20049 [FREE Full text] [doi: 10.2196/20049] [Medline: 32540841]

Abbreviations

AYA: adolescents and young adults
CF: cystic fibrosis
mHealth: mobile health
ppFEV1: percent predicted forced expiratory volume in 1 second

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Viewpoint

Internet Searches for Terms Related to Child Maltreatment During COVID-19: Infodemiology Approach

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Abstract

We examined internet searches indicative of abusive parental behaviors before and after the World Health Organization's declaration of COVID-19 as a pandemic (March 11, 2020) and subsequent lockdown measures in many countries worldwide. Using Google Trends, we inferred search trends between December 28, 2018, and December 27, 2020, for queries consisting of "mother," "father," or "parents" combined with each of the 11 maltreatment-related verbs used in the Conflict Tactics Scales, Parent-Child version. Raw search counts from the Google Trends data were estimated using Comscore. Of all 33 search terms, 28 terms showed increases in counts after the lockdowns began. These findings indicate a strong increase in internet searches relating to occurrence, causes, or consequences of emotional and physical maltreatment since the lockdowns began and call for the use of maltreatment-related queries to direct parents or children to online information and support.

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KEYWORDS

child; maltreatment; COVID-19; pandemic; internet searches; information-seeking; internet; abuse; trend; Google trends; infodemiology

Background

With social distancing measures, school closures, and mounting unemployment rates, the COVID-19 pandemic drastically impacted the lives of families across the globe. An accumulating number of studies show that pandemic-related stressors induce mental health problems, which in turn may impede parenting abilities. For example, job loss, financial insecurities, parental anxiety, and depressive symptoms during the pandemic have been associated with increased (risk for) emotional and physical abuse [1,2]. Although these findings give rise to widespread concerns about potential increases in child maltreatment [3], data on the scope of this problem are still scarce.

A previous study on the impact of the pandemic showed that mental health problems were reflected in internet searches in the early phase of the pandemic [4]. More specifically, internet searches indicative of acute anxiety spiked in March 2020. Another study also indicated a relationship between search traffic data from Google and cases and deaths in severely affected European countries [5]. These studies motivate surveillance of such internet searches in order to track outbreaks and monitor health threats during the pandemic; hence, infodemiology (ie, information epidemiology) metrics, in particular data from Google Trends, could be useful in tracking the virus and monitoring its impact. Here, we applied an infodemiology approach to examine whether increased risk for

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child maltreatment during the pandemic is reflected in internet searches. We examined internet searches indicative of abusive parental behaviors (emotional and physical abuse) before and after the World Health Organization's declaration of COVID-19 as a pandemic (March 11, 2020) and subsequent lockdown measures in many countries worldwide.

Increases in Child Maltreatment–Related Search Terms

We used Google Trends to infer search trends between December 28, 2018, and December 27, 2020, for queries consisting of "mother," "father," or "parents" combined with each of the 11 maltreatment-related verbs used in the Conflict Tactics Scales, Parent-Child version [6]: yelled, screamed, shouted, cursed, swore, threatened, pinched, hit, slapped, beat, and shook. We inferred weekly raw search counts from the Google Trends data using the R package *gtrendR* [7]. This package first obtains global relative search trends for a particular search term (using quotes) from the Google Trends database. It then uses Comscore estimates to retrieve the total number of weekly Google desktop searches within a particular time period. For our analysis, this number was 3044 million searches per week for desktop searches and 6088 million searches in total, assuming that desktop searches amount to 50% of total Google searches. We fitted a seasonal autoregressive integrated moving average model using the R package *gtrendsr* [8] to estimate the excess search count after the start of the pandemic compared to the years before, while taking into account seasonal patterns recurring throughout the year. The increase in search activity was estimated by taking the ratio of the average weekly number of searches before and after the interruption point (March 11, 2020, when the World Health Organization declared COVID-19 a pandemic). All analyses were conducted using R statistical software (version 3.5.0; R Foundation for Statistical Computing) and the script can be found on the Open Science Framework [9].

Figure 1 visualizes search trends before and after lockdowns began for the terms showing a pronounced increase: "mother slapped," "mother cursed," "father beat," "father shook," "parents cursed," and "parents beat." Several other search terms showed similar increases (see [9]). Of all 33 search terms, 28 showed increases in counts (range: 1.8%-196.1%; Table 1).

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Figure 1. Google search trends for abuse-related verbs. The blue solid lines indicate the raw estimated weekly search counts and the gold shaded lines indicate the fitted LOESS regression curve including a 95% CI. The red lines represent the average estimated search counts before and after the World Health Organization's declaration of COVID-19 as a pandemic (black dotted line). Differences in raw search counts should only be interpreted within (and not between) search terms. For more details, see the R script available on the Open Science Framework site [9].

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Table 1. Percentages indicating increases in search terms since the COVID-19 lockdown started.

Terms	Father, %	Mother, %	Parents, %
Psychological abuse			
Yelled	48.6	13.2	48.6
Screamed	51.2	57.4	—[a]
Shouted	87.0	54.6	23.6
Cursed	42.2	112.4	196.1
Swore	15.0	32.7	—
Threatened	29.0	28	82.3
Average change	45.5	49.7	87.7
Physical abuse			
Pinched	—	81.8	—
Hit	22.0	1.8	7.2
Slapped	17.9	101.6	4.6
Beat	—	7.7	26.5
Shook	82.2	51.9	45.5
Average change	40.7	49.0	21.0

[a]Not available; search terms without percentages did not yield sufficient data.

Conclusions

These findings indicate a strong increase in internet searches relating to occurrence, causes, or consequences of harsh parenting since the lockdowns began. We cannot determine who is searching (eg, parents, family members, children, others) and whether searches are directly linked to specific experiences, witnessing maltreatment, or acts of emotional and physical maltreatment. However, our findings suggest an increased risk for child maltreatment due to the COVID-19 pandemic and may motivate the development of novel data-driven family support strategies. Moreover, our findings extend previous studies showing that an infodemiology approach could be an integral part of the surveillance of the pandemic and its impact [4,5]. This topic warrants further research to examine how increases in maltreatment-related search queries are related to actual regional increases in prevalence of maltreatment during the pandemic.

During the pandemic, child maltreatment has progressed from unnoticed to invisible due to social distancing [10]. Observing maltreatment-related search activity may be a way to monitor increases in child maltreatment and inform policy makers to stimulate (preventive) intervention. Importantly, internet searches can also be used to target support to families at risk during pandemics. With the “OneBox” approach, Google refers searchers using suicide-related queries to links to local helplines that are highlighted at the top of search results. Maltreatment-related Google queries, however, do not result in any helpline referrals. As already implemented for search terms like suicide, it is time for maltreatment-related queries to direct parents or children to online information and support.

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Authors' Contributions

All authors contributed to the design of the study. PL analyzed the data, MMER and PL wrote the manuscript, and all authors reviewed and commented on the manuscript.

Conflicts of Interest

None declared.

References

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1. Brown SM, Doom JR, Lechuga-Peña S, Watamura SE, Koppels T. Stress and parenting during the global COVID-19 pandemic. Child Abuse Negl 2020 Dec;110(Pt 2):104699 [FREE Full text] [doi: 10.1016/j.chiabu.2020.104699] [Medline: 32859394]
2. Lawson M, Piel MH, Simon M. Child Maltreatment during the COVID-19 Pandemic: Consequences of Parental Job Loss on Psychological and Physical Abuse Towards Children. Child Abuse Negl 2020 Dec;110(Pt 2):104709 [FREE Full text] [doi: 10.1016/j.chiabu.2020.104709] [Medline: 32893003]
3. Addressing violence against children, women and older people during the COVID-19 Pandemic: Key actions World Health Organization. World Health Organization. URL: https://www.who.int/publications/i/item/WHO-2019-nCoV-Violence_actions-2020.1 [accessed 2020-10-30]
4. Ayers JW, Leas EC, Johnson DC, Poliak A, Althouse BM, Dredze M, et al. Internet Searches for Acute Anxiety During the Early Stages of the COVID-19 Pandemic. JAMA Intern Med 2020 Dec 01;180(12):1706-1707. [doi: 10.1001/jamainternmed.2020.3305] [Medline: 32832984]
5. Mavragani A. Tracking COVID-19 in Europe: Infodemiology Approach. JMIR Public Health Surveill 2020 Apr 20;6(2):e18941 [FREE Full text] [doi: 10.2196/18941] [Medline: 32250957]
6. Straus MA, Hamby SL, Finkelhor D, Moore DW, Runyan D. Identification of Child Maltreatment With the Parent-Child Conflict Tactics Scales: Development and Psychometric Data for a National Sample of American Parents. Child Abuse & Neglect 1998 Apr;22(4):249-270. [doi: 10.1016/s0145-2134(97)00174-9]
7. Caputi TL. gtrendspy and gtrendR: Packages for analyzing Google Trends Data. 2020. URL: https://gtrendr.readthedocs.io/en/latest/ [accessed 2021-07-06]
8. Massicotte P, Eddelbuettel D, Massicotte M. R package gtrendsR, version 1.4.7. URL: https://github.com/PMassicotte/gtrendsR [accessed 2020-06-18]
9. Lodder P. OSF | COVID-19 abuse related Google search trends. URL: https:/osf.io/af6uc/ [accessed 2021-07-06]
10. Thomas EY, Anurudran A, Robb K, Burke TF. Spotlight on child abuse and neglect response in the time of COVID-19. Lancet Public Health 2020 Jul;5(7):e371 [FREE Full text] [doi: 10.1016/S2468-2667(20)30143-2] [Medline: 32619538]

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Original Paper

Pediatric Coping During Venipuncture With Virtual Reality: Pilot Randomized Controlled Trial

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Abstract

Background: Virtual reality (VR) has shown promise in reducing children's pain and anxiety during venipuncture, but studies on VR lack objective observations of pediatric coping. Notably, the process of capturing objective behavioral coping data can be labor- and personnel-intensive.

Objective: The primary aims of this pilot trial were to assess the feasibility of conducting a trial of VR in a pediatric emergency department and the feasibility of documenting observed coping behaviors during pediatric procedures. Secondarily, this study examined whether VR affects child and caregiver coping and distress during venipuncture in the pediatric emergency department.

Methods: This stratified, randomized, controlled pilot trial compared coping and distress between child life–supported VR engagement and child life specialist support without VR during painful procedures in children aged 7-22 years in the pediatric emergency department. An external control (reference group) received no standardized support. Primary feasibility outcomes included rates of recruitment, rates of withdrawal from VR, and rates of completed Child Adult Medical Procedure Interaction Scale-Short Form (CAMPIS-SF) observations. Secondary clinical outcomes were applied to venipuncture procedures and included CAMPIS-SF coping and distress (range 0-1.0), pain and anxiety on a visual analog scale (range 0-10), and cybersickness symptoms.

Results: Overall recruitment was 93% (66/71), VR withdrawal rate was 27% (4/15), and of the completed procedures, 100% (63/63) CAMPIS-SF observations were completed. A total of 55 patients undergoing venipuncture in the pediatric emergency department were included in the analyses of clinical outcomes: 15 patients (15 caregivers) randomized to VR, 20 patients (15 caregivers) randomized to child life specialist support, and 20 patients (17 caregivers) in the reference group. Patient coping differed across groups with higher coping in the VR group and child life specialist group than in the reference group (P=.046). There were no significant differences in the distress and pain ratings for patients and caregivers between the groups. Caregivers rated the lowest perceived anxiety in the child life specialist group (P=.03). There was no apparent change in cybersickness symptoms before and after VR use (P=.37).

Conclusions: Real-time documentation of observed behaviors in patients and caregivers was feasible during medical procedures in which VR was utilized, particularly with the availability of research staff. VR and child life specialists improved coping in children during venipuncture procedures. Given the high participation rate, future studies to evaluate the efficacy of VR are recommended to determine whether an off-the-shelf VR headset can be a low-cost and low-risk tool to improve children's coping during venipuncture or other related procedures.

Trial Registration: ClinicalTrials.gov NCT03686176; https://clinicaltrials.gov/ct2/show/NCT03686176

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(JMIR Pediatr Parent 2021;4(3):e26040) doi:10.2196/26040

KEYWORDS

pediatrics; psychological distress; virtual reality; procedural pain; anxiety; phlebotomy

Introduction

Venipuncture is a common pediatric emergency department procedure; yet, optimal psychological interventions to promote coping remain undetermined. Standard of care ranges from no intervention to certified child life specialist support with use of a variety of cognitive or behavioral strategies. Data on virtual reality (VR) have overall demonstrated improved pediatric pain and anxiety during venipuncture [1-4], although 1 study found no change in pain [5]. These studies evaluated VR games that cater to a medical procedure (eg, a field of view that minimizes head movement or interaction that does not require a hand controller) [1-3,6]. Customized VR games for medical procedures are either designed locally and not available for dissemination or require a costly subscription. No prior studies have evaluated an off-the-shelf commercially available VR headset during pediatric procedures. Furthermore, prior VR studies evaluated outcomes of self-reported pain and anxiety, which are subjective and less meaningful in younger children. To our knowledge, no study to date has reported objective observations of children and caregiver's coping behaviors. Observational measures of coping behaviors offer several advantages, including objectivity, inclusion of all age ranges, and inclusion of caregiver behaviors [7]. Since a caregiver's response influences children's coping [8], understanding caregiver behaviors may also elucidate the potential benefits of VR during pediatric procedures. While objective observations of child and caregiver behaviors are informative, the process of capturing these data can be labor- and personnel-intensive.

We conducted a pilot study to understand the feasibility of conducting a trial with a commercially available VR headset in a pediatric emergency department and the feasibility of documenting observed coping behaviors during pediatric procedures. Secondarily, this study examined whether VR affects child and caregiver coping and distress during venipuncture in the pediatric emergency department. The results of this study provide preliminary data for the planning of subsequent studies.

Methods

Study Design, Setting, and Sampling Technique

A convenience sample of patients aged 7-22 years who required a painful procedure (eg, venipuncture, laceration repair, burn debridement) in the pediatric emergency department were recruited. The study design was a stratified, randomized, controlled pilot trial that compared coping and distress between child life specialist–supported VR engagement and child life specialist support (clinicaltrials.gov NCT03686176). This study was conducted in an academic, urban, tertiary care pediatric emergency department. Randomization allocation was 1:1, performed in randomized blocks of 2, 4, 6, and 8 (R, Version 3.2.2, 2018), and stratified by the type of procedure. The block randomization allocation was imported into REDCap (version 10.0.28, 2019) [9,10] and performed by research assistants upon recruitment. Consistent with the recommendations by Kraemer et al [11], this pilot trial will be used to refine the research protocol, establish the infrastructure, address pragmatic issues, and gather pilot data to answer key questions about the use of VR during pediatric procedures. An external control (reference) group was enrolled when VR and child life specialists were unavailable.

Inclusion and Exclusion Criteria

Inclusion criteria were patients aged 7-22 years who were in the pediatric emergency department and were undergoing any of the following procedures: burn debridement or dressing change, laceration repair, venipuncture (intravenous line or blood draw), abscess incision and drainage, fracture reduction or cast placement, or implanted central venous port placement. Exclusion criteria included severe developmental delays, seizures, blindness, trauma/infection on the head/face, altered mental status, medical urgency, and non-English speakers. Caregivers provided verbal consent and patients provided verbal assent to participate in the study.

Study Protocol

Patient eligibility was screened by research assistants. Eligible patients and caregivers were introduced to the study by research assistants. Research assistants discussed the aims, risks, and benefits of the study, described the VR intervention, and invited patients and caregivers to participate. If consent was obtained, patients were block randomized as described above. Patients randomized to VR played a game using a commercially available VR headset with child life specialist support (*Oculus Go*, version 6.0. Facebook Technologies, 2018). Prior to the study start, child life specialists selected and downloaded VR games and apps, including a variety of passive VR experiences and active game play. Specific VR games/apps included *Netflix* version 1.1, *Bait!* version 1.11.61278 (Resolution Games; 2016), *Epic Roller Coasters* version 6.22.0 (Balneário Camboriú), BR: B4T Games; 2017, *Temple Run* version 1.0.4 (Imangi Studios, 2015), and *Disney Movies VR* version 1.6.472 (Walt Disney Studios, 2017). For all patients in the child life specialist support and VR groups, child life specialists performed a psychosocial assessment that considered child, family, and health care variables to determine how to support the patient during the procedure. For the VR group, child life specialists offered simple descriptions of select developmentally appropriate VR experiences, thereby allowing the child to then make a choice of VR games based upon their personal interests. Prior to VR use, the device was cleaned with hospital-grade disinfectant wipes. A disposable paper face shield and a disposable surgical cap were used for infection control. VR play was limited to 30 minutes to minimize development of cybersickness symptoms. The duration of VR play was monitored by child life specialists and the research assistant. The duration of the procedure served

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as a surrogate for VR duration, as the VR headset was applied immediately before the procedure and VR play completed after the procedure. Patients did not pretrain on VR games/application prior to their procedure start. Patients were empowered to discontinue VR at any point during the procedure upon verbal request or by removing the headset. Patients explored and experienced the VR game/application independently during their procedure and could request technical support from child life specialists when needed. Child life specialists provided technical support, including headset fit, menu navigation, or selection of an alternate game/application. Patients immersed in VR could opt for a tactile or verbal prompt just prior to the painful part of the procedure.

The active control group received child life specialist support and distraction of the child's choosing. The reference group received no standardized support. During the procedures, an independent evaluator (ie, research assistant) logged the frequency of patient coping/distress and caregiver coping-promoting/distress-promoting behaviors in 1-minute increments by using a validated scale (Child Adult Medical Procedure Interaction Scale-Short Form [CAMPIS-SF], Multimedia Appendix 1) [12]. The work describing adult and child coping and distress behaviors was originally described by Blount et al [13]. CAMPIS-SF was selected because it is an abbreviated version of CAMPIS and is a validated and objective measure of coping and distress that has been studied in school-aged and adolescent-aged children [12,14,15]. Each independent evaluator was trained by the lead investigator (TC) to code behavior observations using the CAMPIS-SF scale. During training, the evaluators reviewed and coded prerecorded videos of children undergoing intravenous placement. They continued coding until interrater reliability (κ) was ≥0.8. In real time, during procedures, independent evaluators recorded behavior events of CAMPIS-SF on a paper scoring sheet. No video recordings were taken during this study. They utilized a 1-minute timer that provided a visual and audio cue to each minute interval. They later transcribed the CAMPIS-SF scores and other demographic or clinical data into REDCap.

Feasibility Outcomes

The primary (feasibility) outcomes were the recruitment rate, defined as the number of patients who enrolled divided by those invited to participate; the withdrawal rate of VR, defined as the number of patients who stopped VR engagement divided by the number of patients who completed a procedure and were randomized to VR; and completion percentage of CAMPIS-SF observations for each patient/caregiver dyad of completed procedures. Feasibility benchmarks for each outcome were set at 80% or higher, and the target sample size for the feasibility outcomes of this pilot trial was at least 12 patients in each arm [16]. This pilot study will inform us of the feasibility to perform a full randomized controlled trial that is powered to detect changes in patients' coping and distress (CAMPIS-SF) scores.

Clinical Outcomes

The secondary (clinical) outcomes were CAMPIS-SF coping and distress scores. Coping scores were calculated by summing all coping events divided by the sum of all the coded behaviors exhibited during the procedure and reported as a proportion of the total behaviors (range 0-1.0). Distress scores were calculated similarly—the sum of all distress events divided by the sum of all behavior events. Secondary clinical outcomes also included change in pain and anxiety on a 10-point visual analog scale from baseline to peak levels during the procedure (range –10 to 10) and cybersickness symptoms [17]. Research assistants showed patients the visual analog scale and asked them to select a number (0-10) that characterized their pain and anxiety. Patients reported a pain or anxiety score before the procedure, and after the procedure, they were asked to report their peak pain or anxiety score experienced during the procedure. The change in pain or anxiety was calculated as postprocedure minus preprocedure. Owing to limited enrollment of other procedures, this study reported clinical outcomes on the subgroup of venipuncture (blood draw or intravenous line).

Statistical Analysis

Feasibility outcomes were summarized. Demographics and clinical characteristics and outcomes were compared across groups by using analysis of variance for continuous variables and Fisher exact tests for categorical variables. A P value less than .05 was considered statistically significant. Patients whose procedures were not performed (eg, cancelled) or proceeded without study personnel present owing to medical urgency were excluded from the analysis. Analysis was performed with the intention-to-treat model.

Ethical Considerations

This study was approved by the Johns Hopkins University School of Medicine institutional review board (ID IRB00161331).

Results

Patient Enrollment

The eligibility, enrollment, and randomization procedures are shown in Figure 1. Recruitment occurred from June 2019 to March 2020 and was terminated owing to safety precautions during the COVID-19 pandemic.

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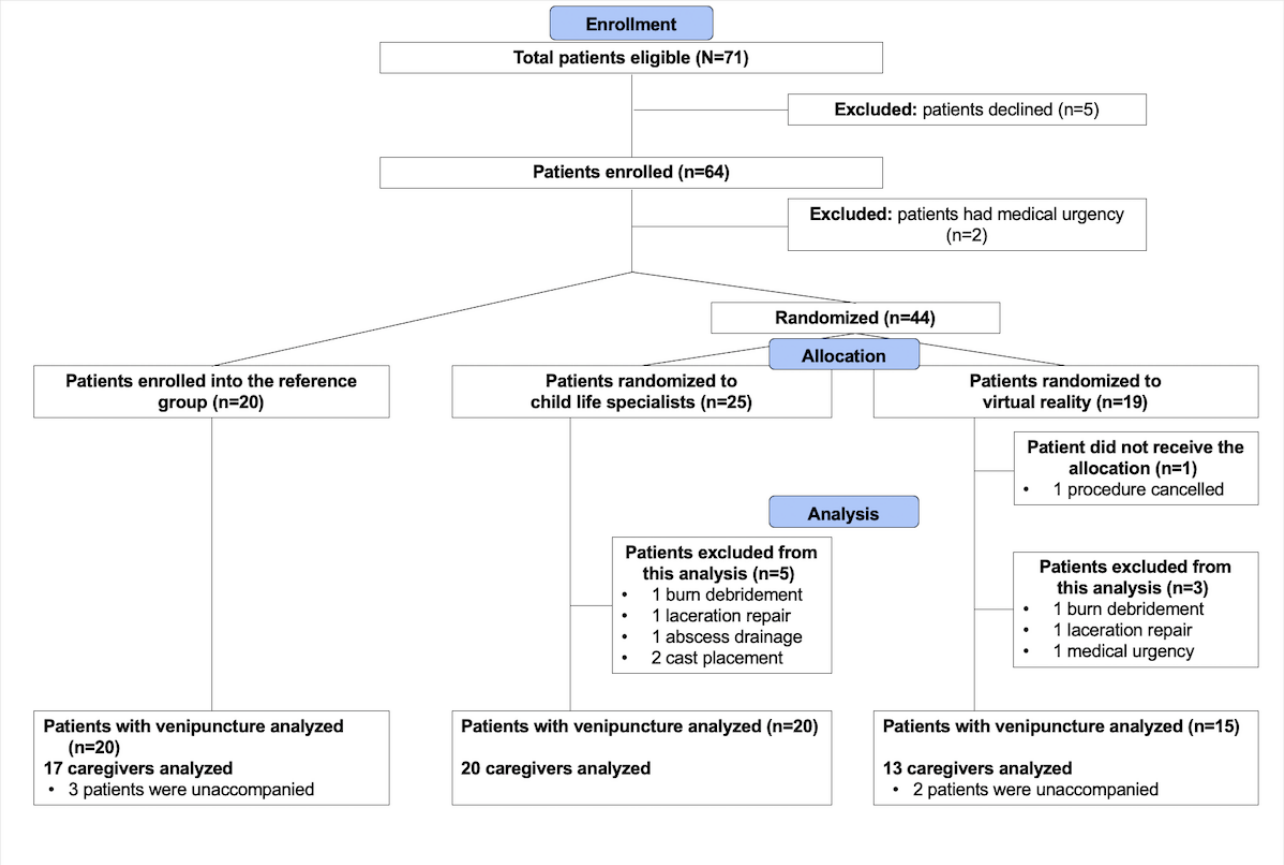
Figure 1. CONSORT patient flow diagram.



Feasibility

Out of the 71 patients invited to participate, 66 (93%) were recruited and 5 (7%) declined to enroll. Of the 15 patients in this study population who were randomized to VR, 4 patients (27%) withdrew from using VR. Patients who withdrew had issues related to the fit of the headset or distress with a preference to watch the procedure (Table 1). All completed procedures (63/63) had complete documentation of the parent and caregiver coping/distress behaviors.

Table 1. Characteristics of the patients who withdrew from the use of virtual reality during pediatric procedures.

Age (years)	Sex	Reason for virtual reality withdrawal	CAMPIS-SF[a] scores (range 0-1.00)		Visual analog scale (range 0-10)		Cybersickness symptoms
			Patient coping	Patient distress	Peak pain	Peak anxiety	
10	Female	Declined because of poor headset fit and sliding down too much, virtual reality withdrawn before the procedure	1.00	0.00	7	1	No
10	Female	Distressed during the procedure, removed virtual reality to watch the procedure, withdrew in the middle of the procedure	0.46	0.54	5	9	No
8	Female	Patient rolling and flailing, virtual reality removed for safety and because the child preferred to see the procedure, withdrew in the middle of the procedure	0.43	0.57	10	10	No
7	Female	The patient was moving, virtual reality headset was slipping, so virtual reality removed at child life specialists' and patient discretion, withdrawn near the end of the procedure.	0.89	0.11	6	3	No

[a]CAMPIS-SF: Child Adult Medical Procedure Interaction Scale-Short Form.

Clinical Outcomes

A total of 55 patients undergoing venipuncture comprised the analysis of the clinical outcomes: 15 patients (15 caregivers) were randomized to VR group, 20 patients (15 caregivers) were randomized to child life specialist group, and 20 patients (17 caregivers) were included in the reference group. The mean age of all the patients was 14.1 (SD 4.1) years. Patient demographics were well-balanced across groups (Table 2). The mean procedure duration in minutes was 5.1 (SD 5.0), 7.7 (SD 5.5),

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and 11.6 (SD 7.0) for the reference, child life specialist, and VR groups, respectively. Analysis of the venipuncture procedural resources (eg, needle success rate) are reported in a secondary analysis [18]. Topical treatments (either lidocaine cream or cold spray) were applied to 0, 2, and 2 patients in the VR, child life specialist, and reference groups, respectively. Opiates (fentanyl, oxycodone, or morphine) were administered to treat pain in 0, 2, and 1 patients in the VR, child life specialist, and reference groups, respectively.

Patient coping differed across groups with higher coping in VR group and child life specialist group than in the reference group (*P*=.05). There were no significant differences in distress and pain ratings for patients and caregivers between the groups. Caregivers' perception of their child's anxiety also differed with the lowest perceived anxiety in the child life specialist group (*P*=.03). There was no change in cybersickness symptoms before and after VR use (*P*=.37) (Table 3).

Table 2. Patient demographics for clinical outcomes.

Patient demographics	Reference group	Child life specialist group	Virtual reality group	Total patients	*P* value
Patients, n (%)	20 (36)	20 (36)	15 (27)	55 (100)	N/A[a]
Age (years), mean (SD)	14.5 (4.2)	15.2 (4.0)	12.1 (3.5)	14.1 (4.1)	.08
Age category (years), n (%)					.19
Child (age range 7-9 years)	4 (20)	1 (5)	3 (20)	9 (15)	
Early adolescent (age range 10-13 years)	3 (15)	7 (35)	6 (40)	19 (31)	
Middle adolescent (age range 14-17 years)	9 (45)	5 (25)	5 (33)	22 (35)	
Late adolescent/adult (age ≥18+ years)	4 (20)	7 (35)	1 (7)	12 (19)	
Sex, n (%)					.72
Female	12 (60)	12 (60)	11 (73)	35 (64)	
Male	8 (40)	8 (40)	4 (27)	20 (36)	
Race, n (%)					.20
Black or African American	11 (55)	11 (55)	4 (27)	26 (47)	
White	8 (40)	7 (35)	9 (60)	24 (44)	
Unknown or not reported	1 (5)	2 (10)	2 (13)	5 (9)	
Ethnicity, n (%)					.30
Hispanic or Latino	0 (0)	2 (10)	2 (13)	4 (7)	
Not Hispanic or Latino	20 (100)	18 (90)	13 (87)	51 (93)	

[a]N/A: not applicable.

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Table 3. Clinical outcomes of the patients and caregivers.[a]

Outcome	Reference group	Child life specialist group	Virtual reality group	*P* value
Patient outcomes				
Patients (N=55), n (%)	20 (36)	20 (36)	15 (27)	N/A[b]
Child Adult Medical Procedure Interaction Scale score, mean (SD)				
Patient coping score	0.70 (0.39)	0.90 (0.14)	0.88 (0.19)	.046[c]
Patient distress score	0.20 (0.31)	0.10 (0.14)	0.12 (0.19)	.36
Change in pain and anxiety scores, mean (SD)				
Pain	0.95 (2.35)	–1.20 (4.16)	–0.20 (4.31)	.19
Anxiety	1.45 (3.32)	–0.10 (1.74)	0.53 (2.77)	.20
Topical anesthetic used, n (%)	1 (5)	2 (10)	2 (13)	.21
Cybersickness symptoms in children, n (%)				.37
Before virtual reality use	N/A	N/A	3 (20)	
After virtual reality use	N/A	N/A	3 (20)	
Caregiver outcomes				
Caregivers, n (%)	17 (36)	15 (32)	15 (32)	N/A
Child Adult Medical Procedure Interaction Scale score, mean (SD)				
Caregiver coping promoting score	0.57 (0.43)	0.52 (0.48)	0.63 (0.38)	.76
Caregiver distress promoting score	0.20 (0.31)	0.08 (0.20)	0.24 (0.30)	.28
Change in pain and anxiety scores, mean (SD)				
Caregiver's perception of patient's pain	0.00 (2.52)	–2.47 (3.50)	–1.73 (3.49)	.09
Caregiver's perception of patient's anxiety	1.29 (2.47)	–1.60 (2.64)	–0.53 (3.85)	.03[c]
Caregiver's own anxiety	0.35 (1.32)	–0.73 (2.22)	–0.13 (3.09)	.41

[a]Change in pain and anxiety scores ranges from –10 to 10. A negative value signifies reduced pain or anxiety during the procedure.

[b]N/A: not applicable.

[c]Significant at $P<.05$.

Discussion

Principal Findings

This study found that real-time objective behavior observations of patient and caregiver coping were feasible to perform in a study of VR use in the pediatric emergency department. The addition of objective behavioral observations in this study is a novel addition within the VR literature and may provide a complementary endpoint for future VR studies. Observations of patients' behavior during medical procedures offer rich objective data that can support past studies on the effectiveness of VR on pain and anxiety [2,5,6,19]. Although real-time observations were feasible in this protocol, patient recruitment was slower than expected, in part due to patients who declined to enroll and research assistant availability. Future protocols may address this issue with augmented research staffing (eg, increased numbers of hours per day or days per week of active enrollment or increased numbers of research personnel) to maximize recruitment during the study period. The clinical outcomes offer preliminary data into the effect of VR on children's coping during venipuncture. Patients in the VR and child life specialist groups exhibited similar coping during venipuncture, and both had higher coping than the group with no standardized support (reference group). While past findings show conflicting data on the effect of VR on pain and anxiety during pediatric procedures [1,6,19,20], our study suggests that VR and child life specialists both improve children's coping during venipuncture. A study designed with coping as a primary endpoint is warranted to fully explore the link between coping and VR use.

Distraction is a psychological intervention that is effective at reducing pain in children undergoing needle-based interventions [21]. The immersive nature of VR makes it a deepened mode of distraction [22], and therefore, a possible modality to improve the experience of pediatric venipuncture. Given the recent physical distancing guidelines, alternate protocols can be explored whereby the staff can set up the VR headset remotely. A remote setup protocol for VR has considerable potential as a relatively hands-off and well-tolerated distraction tool. However, as caregivers perceived the lowest anxiety in the child life specialist group, the psychological benefit of a formal child life specialist support is evident.

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Our protocol offered VR to children as young as 7 years. Other study protocols that used standard-sized VR headsets included children as young as 7-10 years [1-3,5,19,23]. In addition, unique to our protocol was the study of an off-the-shelf headset with VR games that were not specifically designed for child use during medical procedures. Our study found fairly low recruitment of children younger than 13 years, in part owing to a lack of children undergoing eligible procedures during periods of enrollment. Of the 9 children younger than 13 years who were assigned to VR, 4 removed the mask due to distress or poor fit. One consideration for inclusion criteria with VR use in a young school-aged child is the measurement of the head circumference or interpupillary distance. Proper VR headset fit may be better predicted by a patient's head size than age alone. The efficacy of VR is influenced not only by its role as a distraction and immersion tool but also by the fit of the headset, maturity of the child, and their inherent ability to regulate their emotional state in an immersed environment. Effectiveness of a standard-sized VR headset and nonmedical games warrant deeper exploration to understand what key factors influence successful use of VR in children of different ages.

Another novel aspect of this protocol was the observation of caregiver behaviors during pediatric VR use. Caregivers' comments and actions (eg, reassuring comments, apology, or empathetic statements) are well described antecedents of children's distress [7,12,24]. VR as an immersive experience may reduce caregiver distressing behaviors, which may be, in part, due to the lack of need to overreassure a child who is distracted or due to the inability to observe facial expressions under a VR headset. Future studies may explore whether VR affects the child-caregiver dynamics, which may elucidate a new or evolving state of child-caregiver interactions during medical procedures.

Limitations

This pilot study has several limitations. First, as this was a pilot study, we were not powered to detect clinically meaningful differences in several patient outcomes. Furthermore, owing to the nature of the study interventions, blinding was not possible for patients or study personnel. Thus, the effect of the novelty of VR or biases through informed consent may have influenced the clinical outcomes (eg, objective coping/distress or subjective self-reported pain/anxiety). Nevertheless, we have demonstrated that the collection of observational measures during VR is feasible and results obtained from this study provide important preliminary data for the design of larger interventional investigations. Next, owing to pediatric emergency department procedures, child life specialists were present to support children during VR use. Therefore, it is not possible to separate the effect of child life specialists from VR, and this is of particular concern for the patients who discontinued VR. This can also limit generalizability to clinical sites that use VR without child life specialists present. Of note, the procedures were not video recorded for later reviews and accordingly, intraobserver reliability of the evaluators was not calculated after their training period. For a future large-scale study using CAMPIS-SF, recordings of the procedures could be included in the protocol to ensure evaluator consistency. Finally, we found that an individual who was not performing the venipuncture procedure was needed to support VR use. This may have broader implications for scalability of VR use as child life specialists assisted patients with the fitting of the VR headset, navigation of menus or games, helped remove the headset urgently when it was not tolerated, and observed for cybersickness symptoms.

Conclusion

The findings of this study demonstrate that real-time documentation of observed behaviors in pediatric patients and caregivers is feasible in a study protocol evaluating VR during medical procedures, particularly with sufficient research staff for recruitment. Better coping was observed in children receiving VR or child life specialist support during venipuncture procedures. Further studies including children/early adolescents is warranted to fully evaluate the benefits of VR on pediatric coping and on the child-caregiver dynamics.

Acknowledgments

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Authors' Contributions

The following authors have made substantial contributions to the content of this manuscript. TC, CP, AS, KK, and JM conceptualized this study. The methodology was performed by TC, CP, CS, AS, KK, KP, and JM. AB and KP performed formal analysis, while TC, CS, AB, and AS performed the investigations. Resources were provided by TC and AS, and data were curated by TC, CS, and AB. The original draft was written by TC, CS, and AB. TC, CP, CS, AB, AS, KK, KP, and JM wrote, reviewed, and edited the manuscript. Visualization was performed by TC, CS, and AB, while supervision was performed by CP and JM. The project was administered by CS and AB, and funds were acquired by TC, CP, AS, KK, and JM.

Conflicts of Interest

TC is the CEO and Founder of CurieDx, a medical software company. This company's work is unrelated to the content described in the present manuscript. The other authors have no conflicts of interest to declare.

Multimedia Appendix 1

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Child adult medical procedure interaction scale-short form codes.
[PNG File , 1926 KB - pediatrics_v4i3e26040_app1.png]

Multimedia Appendix 2
CONSORT-EHEALTH checklist (V. 1.6.1).
[PDF File (Adobe PDF File), 2346 KB - pediatrics_v4i3e26040_app2.pdf]

References

1. Gold J, Mahrer N. Is Virtual Reality Ready for Prime Time in the Medical Space? A Randomized Control Trial of Pediatric Virtual Reality for Acute Procedural Pain Management. J Pediatr Psychol 2018 Apr 01;43(3):266-275. [doi: 10.1093/jpepsy/jsx129] [Medline: 29053848]
2. Dumoulin S, Bouchard S, Ellis J, Lavoie KL, Vézina MP, Charbonneau P, et al. A Randomized Controlled Trial on the Use of Virtual Reality for Needle-Related Procedures in Children and Adolescents in the Emergency Department. Games Health J 2019 Aug;8(4):285-293. [doi: 10.1089/g4h.2018.0111] [Medline: 31135178]
3. Piskorz J, Czub M. Effectiveness of a virtual reality intervention to minimize pediatric stress and pain intensity during venipuncture. J Spec Pediatr Nurs 2017 Nov 20;23(1):e12201. [doi: 10.1111/jspn.12201]
4. Eijlers R, Utens EMWJ, Staals LM, de Nijs PFA, Berghmans JM, Wijnen RMH, et al. Systematic Review and Meta-analysis of Virtual Reality in Pediatrics. Anesthesia & Analgesia 2019;129(5):1344-1353. [doi: 10.1213/ane.0000000000004165]
5. Walther-Larsen S, Petersen T, Friis SM, Aagaard G, Drivenes B, Opstrup P. Immersive Virtual Reality for Pediatric Procedural Pain: A Randomized Clinical Trial. Hosp Pediatr 2019 Jul;9(7):501-507. [doi: 10.1542/hpeds.2018-0249] [Medline: 31160472]
6. Chan E, Hovenden M, Ramage E, Ling N, Pham JH, Rahim A, et al. Virtual Reality for Pediatric Needle Procedural Pain: Two Randomized Clinical Trials. J Pediatr 2019 Jun;209:160-167.e4. [doi: 10.1016/j.jpeds.2019.02.034] [Medline: 31047650]
7. Blount RL, Piira T, Cohen LL, Cheng PS. Pediatric procedural pain. Behav Modif 2006 Jan;30(1):24-49. [doi: 10.1177/0145445505282438] [Medline: 16330518]
8. Blount RL, Landolf-Fritsche B, Powers SW, Sturges JW. Differences between high and low coping children and between parent and staff behaviors during painful medical procedures. J Pediatr Psychol 1991 Dec;16(6):795-809. [doi: 10.1093/jpepsy/16.6.795] [Medline: 1798015]
9. Harris PA, Taylor R, Minor BL, Elliott V, Fernandez M, O'Neal L, REDCap Consortium. The REDCap consortium: Building an international community of software platform partners. J Biomed Inform 2019 Jul;95:103208 [FREE Full text] [doi: 10.1016/j.jbi.2019.103208] [Medline: 31078660]
10. Harris PA, Taylor R, Thielke R, Payne J, Gonzalez N, Conde JG. Research electronic data capture (REDCap)--a metadata-driven methodology and workflow process for providing translational research informatics support. J Biomed Inform 2009 Apr;42(2):377-381 [FREE Full text] [doi: 10.1016/j.jbi.2008.08.010] [Medline: 18929686]
11. Kraemer HC, Mintz J, Noda A, Tinklenberg J, Yesavage JA. Caution regarding the use of pilot studies to guide power calculations for study proposals. Arch Gen Psychiatry 2006 May;63(5):484-489. [doi: 10.1001/archpsyc.63.5.484] [Medline: 16651505]
12. Blount RL, Bunke V, Cohen LL, Forbes CJ. The Child–Adult Medical Procedure Interaction Scale-Short Form (CAMPIS-SF). Journal of Pain and Symptom Management 2001 Jul;22(1):591-599. [doi: 10.1016/s0885-3924(01)00303-7]
13. Blount RL, Corbin SM, Sturges JW, Wolfe VV, Prater JM, James LD. The relationship between adults' behavior and child coping and distress during BMA/LP procedures: A sequential analysis. Behavior Therapy 1989;20(4):585-601. [doi: 10.1016/S0005-7894(89)80136-4]
14. Sadhasivam S, Cohen LL, Hosu L, Gorman KL, Wang Y, Nick TG, et al. Real-Time Assessment of Perioperative Behaviors in Children and Parents. Anesthesia & Analgesia 2010;110(4):1109-1115. [doi: 10.1213/ane.0b013e3181d2a509]
15. Spagrud LJ, von Baeyer CL, Ali K, Mpofu C, Fennell LP, Friesen K, et al. Pain, distress, and adult-child interaction during venipuncture in pediatric oncology: an examination of three types of venous access. J Pain Symptom Manage 2008 Aug;36(2):173-184 [FREE Full text] [doi: 10.1016/j.jpainsymman.2007.10.009] [Medline: 18400458]
16. Julious SA. Sample size of 12 per group rule of thumb for a pilot study. Pharmaceut. Statist 2005 Oct;4(4):287-291. [doi: 10.1002/pst.185]
17. Kennedy RS, Lane NE, Berbaum KS, Lilienthal MG. Simulator Sickness Questionnaire: An Enhanced Method for Quantifying Simulator Sickness. The International Journal of Aviation Psychology 1993 Jul;3(3):203-220. [doi: 10.1207/s15327108ijap0303_3]
18. Canares TL, Parrish C, Santos C, Badawi A, Stewart A, Kleinman K, et al. Resource Use During Pediatric Venipuncture With Virtual Reality: Secondary Analysis of a Randomized Controlled Pilot Trial. Hosp Pediatr 2021 Jun 14:epub. [doi: 10.1542/hpeds.2020-003822] [Medline: 34127486]
19. Aydın A, Özyazıcıoğlu N. Using a Virtual Reality Headset to Decrease Pain Felt During a Venipuncture Procedure in Children. J Perianesth Nurs 2019 Dec;34(6):1215-1221. [doi: 10.1016/j.jopan.2019.05.134] [Medline: 31495558]

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20. Atzori B, Hoffman HG, Vagnoli L, Patterson DR, Alhalabi W, Messeri A, et al. Virtual Reality Analgesia During Venipuncture in Pediatric Patients With Onco-Hematological Diseases. Front Psychol 2018;9:2508 [FREE Full text] [doi: 10.3389/fpsyg.2018.02508] [Medline: 30618938]
21. Uman LS, Chambers CT, McGrath PJ, Kisely S. A systematic review of randomized controlled trials examining psychological interventions for needle-related procedural pain and distress in children and adolescents: an abbreviated cochrane review. J Pediatr Psychol 2008 Sep;33(8):842-854 [FREE Full text] [doi: 10.1093/jpepsy/jsn031] [Medline: 18387963]
22. Li A, Montaño Z, Chen VJ, Gold JI. Virtual reality and pain management: current trends and future directions. Pain Manag 2011 Mar;1(2):147-157 [FREE Full text] [doi: 10.2217/pmt.10.15] [Medline: 21779307]
23. Birnie KA, Kulandaivelu Y, Jibb L, Hroch P, Positano K, Robertson S, et al. Usability Testing of an Interactive Virtual Reality Distraction Intervention to Reduce Procedural Pain in Children and Adolescents With Cancer [Formula: see text]. J Pediatr Oncol Nurs 2018;35(6):406-416. [doi: 10.1177/1043454218782138] [Medline: 29950139]
24. Blount RL, Cohen LL, Frank NC, Bachanas PJ, Smith AJ, Manimala MR, et al. The Child-Adult Medical Procedure Interaction Scale-Revised: an assessment of validity. J Pediatr Psychol 1997 Feb;22(1):73-88. [doi: 10.1093/jpepsy/22.1.73] [Medline: 9019049]

Abbreviations

CAMPIS-SF: Child Adult Medical Procedure Interaction Scale-Short Form
VR: virtual reality

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Review

Digital Technologies for Monitoring and Improving Treatment Adherence in Children and Adolescents With Asthma: Scoping Review of Randomized Controlled Trials

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Abstract

Background: Inadequate pediatric asthma care has resulted in potentially avoidable unplanned hospital admissions and morbidity. A wide variety of digital technologies have been developed to monitor and support treatment adherence in children and adolescents with asthma. However, existing reviews need to be updated and expanded to provide an overview of the current state of research on these technologies and how they are being integrated into existing health care services and care pathways.

Objective: This study aims to provide an overview of the current research landscape and knowledge gaps regarding the use of digital technologies to support the care of children and adolescents with asthma.

Methods: This study was structured according to the PRISMA-ScR (Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for Scoping Reviews) and Population, Intervention, Comparator, Outcome, and Study frameworks. Five databases (PubMed, the Cochrane Central Register of Controlled Trials, Web of Science, Embase, and PsycINFO) were systematically searched for studies published in English from 2014 onward. Two reviewers independently screened the references and selected studies for inclusion based on the eligibility criteria. Data were systematically extracted per research question, which were synthesized in a descriptive analysis.

Results: A wide variety of study characteristics, including the number and age of participants, study duration, and type of digital intervention, were identified. There was mixed evidence for the effectiveness of the interventions. Of the 10 studies that evaluated treatment adherence, 7 (70%) found improvements, but the evidence was inconsistent for asthma control (6/9, 67% of studies reported improvement or maintenance, but only 1 was significantly different between groups) and health outcome variables (5/9, 56% of studies found no evidence of effectiveness). The 6 studies that examined patient perceptions and assessments of acceptability and usability generally had positive findings.

Conclusions: A wide range of digital interventions are being developed and evaluated to support the monitoring and treatment adherence of children and adolescents with asthma. Meta-analyses are inhibited by the use of samples with a variety of overlapping age ranges; a theoretical framework for evaluating specific age groups would aid comparison between studies. Most studies found significant evidence for improved adherence to treatment or medications, but there was mixed evidence of the impact of the digital interventions on asthma control and other health outcomes. There are gaps in the literature relating to cost-effectiveness and integration with existing clinical care pathways. This study will be necessary to determine which digital interventions for children and young people with asthma are worth supporting and adopting in the clinical care pathways.

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KEYWORDS

asthma; disease management; child; adolescent; telemedicine

Introduction

Background

Globally, asthma is the most common chronic illness affecting children [1-3] and can have serious health consequences. It is one of the key causes of urgent hospital admissions and morbidity in children [3,4]. This is a particularly urgent problem in the United Kingdom. Out of all the Organization for Economic Co-operation and Development countries, the United Kingdom has the third highest risk of death because of pediatric asthma [3,4]. Although specific data are not available for many countries, asthma has high costs worldwide [5]. The variation in mortality across countries suggests that many of the negative outcomes of childhood asthma, for patients and health care systems, are potentially avoidable [6]. Effective management programs are likely to be a cost-effective means of improving asthma control and reducing the economic burden across countries by enabling early and preventive measures to be taken [5].

A growing number of digital technologies are being developed to help the self-management of people with asthma [7-9]. Broadly, digital technologies are electronic systems that can collect, analyze, and share data, and common examples include mobile or web apps, smart devices, and other phone or internet-based interventions. Some evidence suggests that digital interventions can help support asthma health management, particularly by improving medication adherence [10,11]. However, other results, particularly in terms of effectiveness (depending on the outcome examined) [9] and app quality [8], are mixed. Research has also identified limitations in the studies examining these interventions, including inadequate descriptions of digital interventions, a lack of economic analyses, and small sample sizes [10,12].

For digital interventions to be effective, people need to be willing to use them. Although digital interventions have been shown to be generally acceptable to a wider population [11], special consideration is needed when evaluating digital interventions for children and young people. Adolescents are a particularly challenging group to treat, and poor health literacy and self-management skills can affect their treatment adherence and health outcomes [2]. Attitudes toward electronic monitoring devices were found to be mixed in adolescents, depending on how they perceived the intervention [13]. Among those who viewed asthma as a serious threat, the monitoring device was viewed as reassuring. However, many adolescents were suspicious of the device, reporting concerns that it would get them in trouble if they did not adhere properly to their medication and beliefs that their health care providers did not trust them to take the medication [13]. This demonstrates the need to examine digital interventions tailored specifically for children and young people, as their needs and responses to the interventions may not be the same as the general population.

Rationale

Although several systematic reviews have examined various topics related to digital interventions for asthma management, there is a need for a comprehensive overview of the evidence being gathered to assess the effectiveness of various types of digital interventions for children and young people with asthma. No previous reviews have been identified that are specific to this population but are broad in terms of the digital interventions examined.

Of the systematic reviews that have focused specifically on children and young people, the scope was limited with respect to either outcome (eg, a focus on treatment adherence [14]) or type of digital technology (eg, only mobile apps [10] or smart devices [15]). One review provided a comprehensive assessment of other systematic reviews [12]. However, this review was published in 2014; given the rapid evolution of digital technology [16], the state of the field has changed since the review was conducted. For instance, electronic inhaler monitoring is a relatively new development [17,18], with smart inhalers only recently becoming commercially available [19]. Another review analyzed studies of children with a wide range of outcomes—adherence, health outcomes, and user perceptions—but only searched PubMed and Embase databases for the study, which raises the concern that some relevant studies might have been missed [9]. To determine if any relevant reviews were in progress, PROSPERO was searched using several combinations of keywords (*asthma* AND *child* OR *paediatric* OR *pediatric* AND *digital* OR *technology* OR *mHealth* OR *eHealth*). These searches identified one relevant registration: a review that was planned, but not executed, by academics associated with the current research team [20].

No reviews were found that examined how the technologies are integrated into current clinical care pathways for children and adolescents with asthma. This is an important area to examine because digital technologies can provide health care professionals with a large body of information that enables them to personalize asthma care plans and focus on preventive measures [21]. A small study by American physicians identified a mix of perceived benefits, barriers, and concerns about integrating digital technologies in asthma care for adolescents [22]. Further research is needed into how digital interventions are currently integrated with health care services [21], to inform the development of integrated clinical care pathways. An overview of the different types of digital technologies and the different ways they are being integrated with health care systems will help inform the development of effective, technologically enhanced care pathways for children with asthma.

Objectives and Research Questions

The primary objectives of the scoping review are to assess and summarize the current state of the literature on digitally enhanced asthma care for young people and identify any gaps [23]. Three research questions were developed to focus on the review:

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1. How are randomized controlled trials (RCTs) of technologically supported asthma pathways being conducted?
2. What is known about the effectiveness of digital technologies in supporting treatment adherence and remote symptom monitoring in children and adolescents?
3. How are studies examining the integration of digital technology into clinical care pathways for pediatric asthma?

Methods

Overview

The review was structured following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses Extension for Scoping Reviews (PRISMA-ScR; Multimedia Appendix 1) [24], and the search strategy was developed using the Population, Intervention, Comparator, Outcome, and Study framework (Textbox 1). No protocol was registered or published for this review. A preliminary review of the literature was conducted to extract Medical Subject Headings (MeSH) terms and keywords for the search. The search was performed in five databases (PubMed, the Cochrane Central Register of Controlled Trials [CENTRAL], Web of Science, Embase, and PsycINFO) using the University of Plymouth's search tool Primo, with slightly adjusted search terms to fit the specific structure of each database. The search terms were grouped into four themes joined in this structure: asthma (MeSH OR Keywords) AND asthma management (MeSH OR Keywords) AND children (MeSH OR Keywords) AND digital technology (MeSH OR Keywords). Multimedia Appendix 2 lists a complete record of the specific search terms and strings used for each database and the number of references returned. The database searches were completed on December 30, 2020, except for the CENTRAL database, which was searched on December 31, 2020.

The inclusion and exclusion criteria are shown in Textbox 2.

Textbox 1. The Population, Intervention, Comparator, Outcome, and Study framework.

Population

- Children and young people under 18 years of age with asthma.

Intervention

- Any digital health technology aiming to support monitoring or treatment adherence of children and adolescents with asthma.

Comparator

- No comparator is required.

Outcome

- The primary outcome was the evidence for the digital interventions at improving monitoring or treatment adherence. Secondary outcomes included how the research was conducted, evidence for improved health outcomes, cost-effectiveness, and integration of the technology with health care systems.

Study types

- Randomized controlled trials that evaluate at least one digital technology to support the care of children with asthma.

Textbox 2. Inclusion and exclusion criteria of the study.

Inclusion criteria

- The review included studies evaluating digital technologies that aim to support the monitoring or treatment adherence of children and adolescents aged below 18 years with asthma.
- Digital technologies included, but were not limited to, mobile or web apps, smart devices, and other phone or internet-based interventions.
- Initially, randomized controlled trials, quantitative, qualitative, cohort, and case study types were eligible for inclusion.
- Given the number of studies identified, only randomized controlled trials were included in the review.
- As the scope of the review was focused on assessing evidence of the effectiveness of digital technologies for asthma monitoring and treatment adherence, it was appropriate to limit the included studies to randomized controlled trials that can evaluate effectiveness.

Exclusion criteria

- Studies with adult participants were excluded during screening, and studies that only included adults were excluded during the full-text review.
- Studies published before 2014 were excluded to limit the review to the current technologies.
- Studies that merely described an intervention without evaluation were excluded.
- Studies published in languages other than English were also excluded, as the review team did not have the necessary resources to assess them.

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Screening and Article Selection

References were exported to the citation management software EndNote X9 (Clarivate Analytics) for storage and duplicate removal. Owing to the returning of the large number of references, an initial screening was conducted by inputting keywords relating to the inclusion and exclusion criteria into the EndNote X9 search function. This was done in several stages, with each subsequent screening being conducted on the subset of studies retrieved in the previous stage. For example, keywords relating to digital technologies were searched for in any field, and studies that did not contain at least one of those keywords were excluded. Subsequent searches used keywords to exclude studies that used terms unrelated to the topic (eg, cancer, diabetes, and enzyme). Multimedia Appendix 3 contains a full description of the searches conducted. Searches of keywords to exclude were based on common features of irrelevant studies that were identified in a manual search. The remaining titles and abstracts were screened by 2 reviewers (MMI and CL) independently (with articles excluded with reasons), and the final eligibility was determined by full-text reviews of the remaining references. Any discrepancies between the reviewers were discussed until a consensus was reached.

Data Extraction

Outcomes were extracted by a reviewer (MMI) into a table structured according to the 3 research questions (Multimedia Appendix 4) and verified by a second reviewer (CL). Key outcomes were predetermined based on a preliminary review of the literature; however, because of the expected variety of reported outcomes, relevant outcomes that were not prespecified in the Population, Intervention, Comparator, Outcome, and Study framework or data extraction tables were also considered for inclusion in the final review (Textbox 3).

Textbox 3. Article information and data extraction.

General study information

- Year of publication
- Sample size
- Age of participants

Digital technology

- Type of digital technology
- Health care setting used in

Evaluation

- Effect of technology on behavioral outcomes (eg, medication adherence and symptom monitoring and reporting)
- Effect of technology on health outcomes
- Cost-effectiveness of the intervention
- Integration of the technology with a health system or care pathway
- Participant perceptions
- Acceptability
- Usability
- Other key performance indicators reported

Data Analysis and Synthesis

The data extracted from the studies about the key outcomes listed in Textbox 3 were assessed using descriptive analysis and summarized to provide an overview of the state of the literature. For outcomes related to effectiveness, the number of studies that found strong evidence of effectiveness was compared with the number of studies that assessed the outcome to provide a synthesis of the state of the evidence for that outcome. Implications of the findings were examined in the discussion.

Results

Included Studies

A total of 6314 articles were retrieved from the search of the 5 databases (Multimedia Appendix 2). A total of 1029 duplicates were removed by the EndNote X9 software, and a further 5193 were screened using keyword searches in EndNote (Multimedia Appendix 3). The titles and abstracts of 92 studies were screened and articles were excluded with reasons. Of these articles, 25 were selected for full-text review, and 20 were selected for inclusion in the review. Of the total references, 6 referred to one study and were either conference abstracts or did not include the final results of the RCT. The paper with published results of the RCT of that study was identified and included [25]. Three references that only provided abstracts subsequently had full texts identified; these full texts were cited and used for data extraction and analysis. The reasons for exclusion in the full-text review stage are shown in Figure 1.

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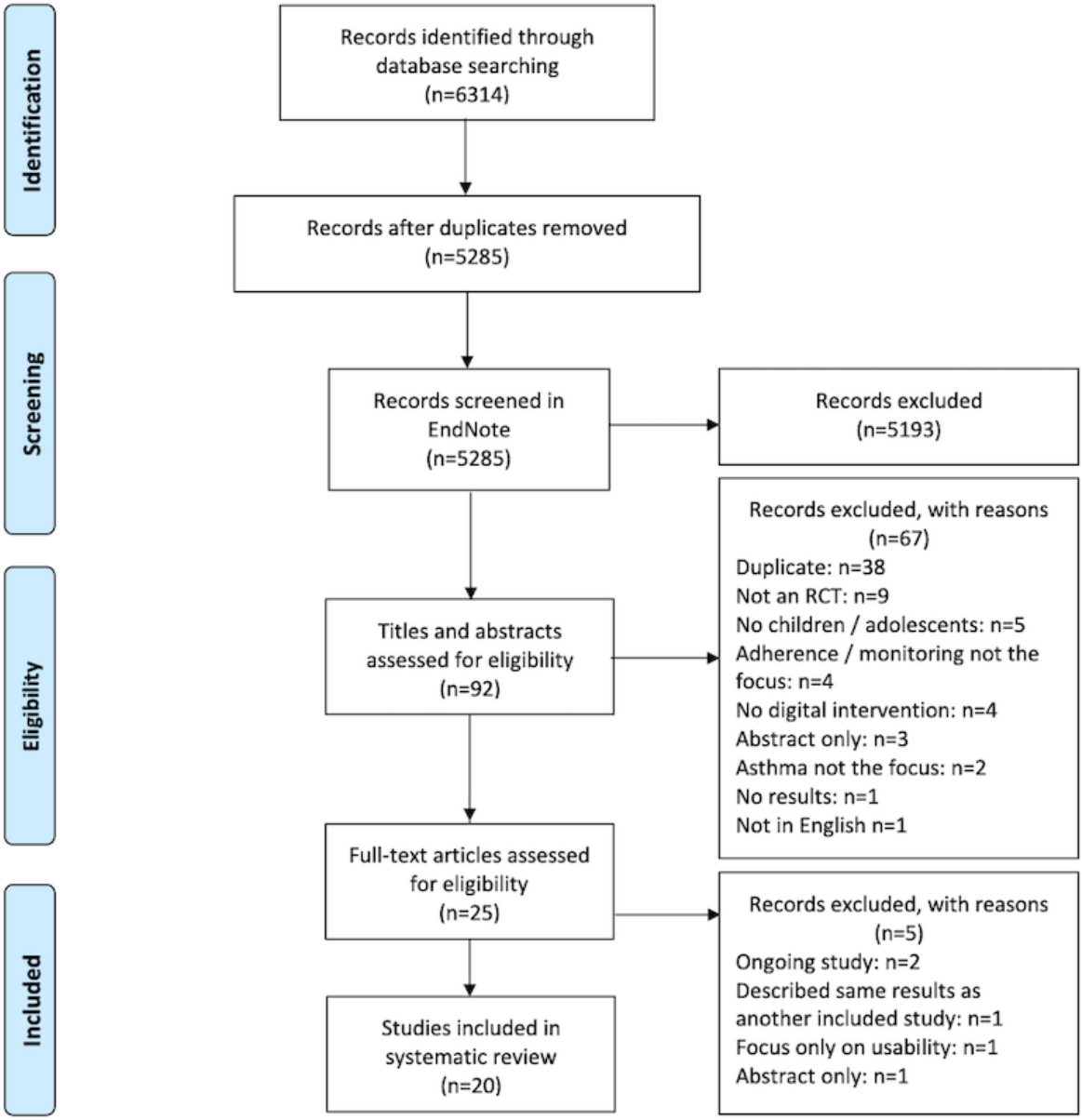
Figure 1. PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flow diagram. RCT: randomized controlled trial.



Study Characteristics

All the studies included in the review were RCTs and limited to those that included monitoring or adherence functions and aims. Despite these restrictions to the scope of the review, the included studies had a wide variety of study durations, sample sizes, age ranges, and types of digital intervention (Table 1).

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Table 1. Summary of characteristics of 20 identified studies (N=20).

Study	Year	Study duration	Number of participants	Age of participants (years)	Type of digital intervention
Beerthuizen et al [26]	2016	12 months	272 (280 enrolled)	4-18	Web-based monitoring
Bender et al [27]	2015	24 months	1187	3-12	Speech recognition automated telephone program
Britto et al [28]	2017	6 months	64	12-22	Text message reminders
Chan et al [29,30]	2015, 2017	6 months	220	6-15	Inhaler EMD[a] with audiovisual reminders
Goossens et al [31]	2014	12 months	209	4-11	EMD with text messages
Johnson et al [32]	2016	3 weeks	98	12-17	Website and text-based reminder system (MyMediHealth)
Kosse et al [25,33,34]	2019	6 months	234 (66 pharmacies)	12-18	App (ADAPT[b])
Merchant et al [35]	2014	100 days	368 (490 enrolled)	5-80	EMD with feedback and educational content
Morton et al [36]	2017	12 months	77 (90 enrolled)	6-16	EMD with alarms and feedback
Perry et al [37]	2018	3 months	393	7-14	School-based educational telemedicine intervention
Real et al [38]	2019	4 months	40	4-11	Gamified app (CHANGE Asthma)
Reece et al [39]	2017	4 months	48	13-60	App (AsthmaWin)
Shields et al [40]	2017	12 weeks	22	2-16	MDOT[c]
Simoneau et al [41]	2019	6 months	43	8-17	EMD with reminders
van den Wijngaart et al [42]	2017	16 months	210	6-16	Web-based monitoring (Virtual Asthma Clinic)
Vasbinder et al [43]	2016	12 months	209	4-11	EMD with text messages
Voorend-van Bergen et al [44]	2015	12 months	268 (280 enrolled)	4-18	Web-based monitoring

[a]EMD: electronic monitoring device.

[b]ADAPT: ADolescent Adherence Patient Tool.

[c]MDOT: mobile directly observed therapy.

Over a third of the references identified as eligible during title and abstract screening only had abstracts available (7/20, 35%) [31,35,36,39-42]. They were included in the analysis where relevant data were available; one of the abstracts only presented interim results [35]. Full texts were found for 3 of these 7 references [36,40,42], and data from those papers were used. A total of 4 studies were analyzed by 9 separate articles and abstracts: the ADolescent Adherence Patient Tool (ADAPT) study [25,33,34], a study comparing web-based Asthma Control Test and fractional exhaled nitric oxide monitoring with standard care [26,44], a study of inhaler electronic monitoring devices (EMDs) with audiovisual reminders [29,30], and a study of a real-time medication monitoring device with SMS text messaging reminders [31,43].

There was a wide range in study durations, from 3 weeks [32] to 24 months [27], with the most common length of follow-up being 6 or 12 months (n=4 [25,28-30,33,34,41] and n=3 studies [26,31,36,43,44] for each). There was also a wide range of numbers of participants included in the 15 studies, ranging from 22 [40] to almost 1200 [27], with an average of approximately 230 participants and a median of 209 [31,43].

There were no distinctive age categories that emerged from these studies. Of the 15 distinct studies, only 2 pairs used the same age range (4-11 years [31,38,43] and 6-16 years [36,42]). A total of 3 studies included adult participants, as well as child or adolescent participants [28,35,39]. The youngest participants included in the study were aged 3 years [27]. Of the studies that focused on participants under 18 years, the age range eligible for inclusion in each study ranged from 6 years (age 12-17 years [32]) to 15 years (age 4-18 years [26,44]).

A total of 4 studies took place across multiple centers [26,31,35,42-44], and most of the rest were associated with large medical centers [27,28,32,38] or clinics [36,41]. The remaining 5 studies were recruited from or associated with a hospital emergency department [29,30], community pharmacies [25,33,34], outpatient appointments in a hospital or Asthma Clinic [40], Howard University [39], and impoverished, rural school districts [37].

Types of Digital Interventions

Various types of digital interventions for monitoring or improving medication adherence examined in the studies were included in this review (Table 1). The most common type of

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intervention, evaluated by a third of the studies (5/15, 33%), was EMDs. However, these EMDs varied in their features, which included audiovisual reminders [29,30], text messages [31,43], alarms [36], and app or web-based sources that could be synced to provide personal feedback [35,36], educational content [35], reminders [41], and capture adherence data [41].

Apps were another common intervention evaluated; 3 studies specifically evaluated three different app-based interventions. These included the ADAPT app that connects adolescents to their community pharmacist through a desktop application and enables them to monitor symptoms and adherence, chat with peers and their pharmacist, watch short educational movies, and set medication alarms [25,33,34]. Another app, CHANGE Asthma, was developed for children by 5 pediatricians and modified based on feedback from a pilot of 24 caregivers. It used short videos and games and an asthma action plan to improve asthma knowledge and control [38]. The third app evaluated (AsthmaWin) also included an asthma action plan but focused more on monitoring symptoms and medication adherence [39].

Other types of interventions evaluated included web-based monitoring programs [26,44] (one of which was a component of a Virtual Asthma Clinic [42]), a speech recognition automated telephone program to improve medication adherence [27], text message medication reminders [28], a website and text-based reminder system (MyMediHealth) [32], a remote directly observed therapy tool to improve inhaler use and adherence [40], and a school-based educational telemedicine intervention that provided interactive video sessions for children, caregivers, and school nurses [37].

Evidence of Effectiveness

Overview

Several different outcome measures were used in the studies to evaluate the interventions, but the results regarding effectiveness were inconsistent. The outcome with the highest proportion of studies finding a significant, positive effect was for improving medication adherence. The reported effectiveness of interventions and improvement in asthma control and health outcomes were mixed. Patient feedback regarding acceptability and usability was generally high.

Treatment or Medication Adherence

A total of 10 studies evaluated the effectiveness of their interventions in improving treatment or medication adherence. Over two-thirds (7/10, 70%) reported significantly higher adherence in the intervention group compared with the control group [25,27,29,31,32,36,41,43]. Of the remaining 3 studies, one reported higher adherence in the intervention group compared with the control group, but no analysis of significance was provided [37], and one reported a trend toward improvement over time [40]. The final study, which evaluated an SMS text messaging reminder system, found a decline in adherence over the intervention and control periods in both groups [28].

Of the 7 studies that found a significant difference in adherence between groups, 4 were evaluating EMDs [29,31,41,43]. The others evaluated the speech recognition automated telephone program [27], the website and text-based reminder system (MyMediHealth) [32], and the ADAPT app [25].

Only one study each evaluated the effectiveness of improving inhaler use and symptom monitoring, both of which found improvements. Shields et al [40] found that remote directly observed therapy improved the inhaler technique equally in the immediate and delayed intervention groups. Perry et al [37] found significantly higher self-reports of peak flow meter use in the intervention group compared with the control group.

Asthma Control and Health Care Visits

There were very mixed results in 9 studies that evaluated asthma control as an outcome. Of the 9 studies, 4 found either no effect of the intervention on asthma control [25,35,43] or no significant difference between groups [38]. However, Real et al [38] found a significant positive association between the degree of app use and asthma control.

Another 4 studies reported improved asthma control in the intervention group compared with the control group [36,39,40,42], although only one of these studies demonstrated statistical significance [42]. Another one of these studies analyzed the 2 groups together and reported a significant improvement in asthma control over time [40]. The final study found that asthma control could be maintained after a clinically relevant reduction in inhaled corticosteroids in the web-based monitoring condition [26,44].

Only 2 studies evaluated the effect of the intervention on health care visits, but neither found any differences [27,42].

Health and Quality of Life Outcomes

The overall effect of the digital interventions on health outcomes remains unclear. Of the 9 studies that evaluated health outcomes (including quality of life or symptom-free days), 5 found no significant improvement [25,26,31,36,37,43,44]. However, 3 studies reported significant improvements in self-reported quality of life [32], asthma morbidity scores [29], and number of symptom-free days [42]. One study reported a significant improvement in parents' self-reported quality of life over time and a nonsignificant trend toward improvement of the children's quality of life [40].

Patient Perceptions, Acceptability, and Usability

A total of 6 studies examined outcomes related to patient perceptions, acceptability, or usability. These studies reported generally high satisfaction and acceptability [30,32-34], a desire to continue using the intervention [39,41], or positive feedback [40].

Cost-effectiveness

Only 1 study (2 articles) explicitly assessed cost-effectiveness [31,43]. The authors found that costs were higher in the intervention group, and although this difference was not statistically significant [43], the technology was deemed not cost-effective because it was not associated with significant improvements in health outcomes [31]. Upon closer inspection of reported mean adjusted costs per patient, although the hospital costs in the intervention arm are lower, the medication cost and parental production loss because of absence from paid work to

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care for children have been calculated to be higher by 16% and 141.8%, respectively [31]. Another study discussed the potential cost savings but did not analyze them as part of the study [36].

Integration With Clinical Care Pathways

Half of the studies included in the review (8/15, 53% of studies or 10/20, 50% of articles) did not explicitly discuss how the digital intervention they were evaluating was integrated with clinical care pathways [28-32,38-41,43]. A few studies described sending data from the interventions back to physicians to update the patients' health records or inform care, although this potential would likely be feasible for many of them. For the few that did, integration of the intervention with the health care system was generally reported positively.

Even among those that described a specific link between the intervention and the health care system, the specific details about integration were not the primary focus of the paper. For instance, one study noted that the intervention was built into routine clinical care in the study and described how data could be uploaded to a website for patients, parents or caregivers, and clinicians to review adherence data together at appointments [36]. Some of the studies that monitored symptoms or adherence produced treatment advice based on data analysis from the system algorithms [26,44] or sent physicians warnings if a patient was out of a certain threshold [35]. The Virtual Asthma Clinic, which also sent feedback to physicians if a patient's asthma control scores were low, was found to be successful in increasing asthma control and symptom-free days and was proposed by the authors as a partial replacement for outpatient visits [42]. Details of how these systems were integrated with the health care system have not been described.

One study whose intervention was significantly integrated with the health care system was the ADAPT app study [25,33,34]. One of the aims of the intervention was to increase collaboration and communication between adolescents and pharmacists because of the increasing role of pharmacists as health care providers in the Netherlands [25]. Pharmacists involved in the intervention reported valuing the improved contact with patients and found the intervention satisfactory, useful in fulfilling their role, and not time-consuming [34]. This contrasted with the perceptions of pharmacists who did not participate in the intervention, who identified time constraints as a barrier to the use of mobile health [34]. However, a barrier was identified because the *stand-alone* desktop interface of the ADAPT app for pharmacists was not integrated with the pharmacy's general information system [34]. This study highlights the potential value of deliberate and considers efforts to integrate new digital health technologies for asthma management with existing health systems.

Speech recognition telemedicine intervention was another study that demonstrated integration with the health care system, which was integrated with the hospital's electronic health record (EpicCare) to provide personalized calls to patients and is compatible with all standard electronic health record systems [27].

The attempt of one study [37] to involve primary care providers in the intervention was not successful. Treatment prompts with medication recommendations based on caregiver reports and guidelines were provided to the participants' primary care providers. These were found to be ineffective; of the 141 prompts sent out for individual participants, the request for feedback received a response from only 1 primary care provider [37].

Discussion

Principal Findings

Different varieties of studies were examined in this review; the study duration ranged from 3 weeks to 2 years, the number of participants ranged from 22 to 1187, and although the review was focused on children and adolescents, the range of ages studied was wide, with no distinct age groups emerging from the studies. There were also several different types of digital interventions analyzed in the RCTs, with EMDs and mobile apps being the most common. Moreover, the integration of these technologies with existing clinical care pathways and health systems has not been extensively discussed in most studies.

The review found inconsistent evidence for the effectiveness of digital technologies in achieving their various aims. Most support was found for the effectiveness of the interventions in improving treatment or medication adherence (7/10, 70% of studies found significant evidence of effectiveness). The results of studies assessing the impact of the intervention on asthma control and health outcomes were mixed, with some studies reporting positive effects and others showing no significant effect. Across the studies, evaluations of patient perceptions, acceptability, and usability were generally positive. Only one study evaluated the cost-effectiveness of these solutions, but because of insignificant improvement in health outcomes, the intervention was not found to be cost-effective [31].

Limitations

One limitation of this review is that a risk of bias assessment was not performed on the studies. Although this is not a standard requirement for scoping reviews, it is a limitation of the study, as it would have contributed to the assessment of the first research question by providing an analysis of the quality of the research being conducted on technologically supported asthma pathways.

Another limitation is that the research questions and aims were adjusted after the search was performed. They were changed before any screening or selection took place but may have resulted in relevant articles being missed because the search terms were established for a slightly different scope. Owing to time limitations, no manual searches of the references of reviews retrieved in the initial search were performed, which could have resulted in eligible articles being overlooked.

Meaning and Future Research

The large number of studies identified in the initial search and the variety of technological interventions to support pediatric asthma care demonstrate the broad scope of this research area. This review identified a few strong trends regarding how technologically supported asthma pathways for children and young people are being researched. The studies used a large

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range of sample sizes and participants of varying ages, which makes it difficult to make valid comparisons or conduct meta-analyses across different studies. A theoretical framework for determining what ages to study or how to stratify children and young people into age groups would be useful for the future. Currently, there is no consensus in the literature on how to group children of various ages for research, which is a significant limitation in the field.

This review found that there is a wide variety of digital interventions being explored. Although many of the studies examined reported positive results, strong evidence of their effectiveness in achieving various aims is still lacking. The strongest evidence was for improving treatment and medication adherence. However, the mixed evidence of asthma control, health, and quality of life outcomes suggests that there might be a disconnect between behavioral change and health outcomes. As asthma is a long-term condition, the study duration of included studies (from 3 weeks to 24 months) may not be long enough to observe significant health impacts, or there may be other factors influencing the relationship between treatment adherence and health outcomes (eg, technique). Understanding why this discrepancy was observed could help inform the design of more effective digital interventions and better study designs.

Another notable area that was missing from many of the studies was an assessment of the cost-effectiveness of the intervention. Considering that a key aim of many digital health technologies is to reduce the burden on health care systems by improving patient self-management, the benefit and cost of the intervention compared with the current standard of care is essential in the decision to integrate digital interventions into clinical care pathways. This will be a key area to consider for future evaluations of these technologies so that limited health care resources can be deployed to create the greatest value [45].

The overall findings are generally consistent with the previous reviews described in the *Introduction* section. Collectively, they identified at least some evidence of the benefits (depending on outcomes) of various digital health technologies on asthma-related outcomes [9-12,14]. One review also noted a lack of data regarding the cost-effectiveness of the digital asthma self-management interventions and patient perspectives [12]. This is also consistent with this review; patient perspectives were generally high when reported but were only examined in about a quarter (6/20, 30%) of the included studies.

Another key area for future research will be around the integration of these digital solutions into clinical pathways. As with cost-effectiveness, this review found that most studies did not explicitly consider or evaluate how the technology they were examining would interact with existing health systems. The potential benefit of integrating patient-reported data with patients' health records to inform care plans and pathways is likely feasible for many of the technologies assessed but was not examined as a key outcome of the technology. Similarly, acceptability and usability data focused primarily on patient users. Understanding how these technologies can best support and interact with existing clinical pathways could help inform their design, improvement, and sustainable adoption.

Conclusions

The purpose of this scoping review was to summarize the literature on technologically enhanced asthma care pathways for children and young people. A large body of research is ongoing in this area and spans a wide range of technologies and ages. Although there was some evidence for the effectiveness of the digital interventions examined, particularly for improving treatment and medication adherence, further research is needed to establish the effectiveness of the interventions in improving asthma control and other health outcomes. This apparent discrepancy between significant evidence for behavior change and a lack of significant evidence for subsequent health impacts should be further examined, as it could indicate factors other than treatment adherence that affect health outcomes and could also be targeted for intervention. A couple of gaps in the literature were identified in terms of cost-effectiveness and integration with existing care pathways. Both of these aspects are essential for the successful adoption, scale-up, and sustained use of digital health interventions and are key areas for future research.

Authors' Contributions

IW conceived the key research questions. JG and KH developed and submitted the previous PROSPERO registration, which was used as the basis for the protocol. The scoping review was executed by MMI and CL and drafted by MMI with revisions from CL and EM.

Conflicts of Interest

None declared.

Multimedia Appendix 1
PRISMA-ScR (Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for scoping reviews) checklist.
[PDF File (Adobe PDF File), 86 KB - pediatrics_v4i3e27999_app1.pdf]

Multimedia Appendix 2
Search record.
[DOCX File , 16 KB - pediatrics_v4i3e27999_app2.docx]

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Multimedia Appendix 3
EndNote search criteria.
[DOCX File , 15 KB - pediatrics_v4i3e27999_app3.docx]

Multimedia Appendix 4
Data extraction table.
[DOCX File , 25 KB - pediatrics_v4i3e27999_app4.docx]

References

1. Mallol J, Crane J, von Mutius E, Odhiambo J, Keil U, Stewart A, ISAAC Phase Three Study Group. The International Study of Asthma and Allergies in Childhood (ISAAC) Phase Three: a global synthesis. Allergol Immunopathol (Madr) 2013;41(2):73-85. [doi: 10.1016/j.aller.2012.03.001] [Medline: 22771150]
2. Lenney W, Bush A, Fitzgerald DA, Fletcher M, Ostrem A, Pedersen S, et al. Improving the global diagnosis and management of asthma in children. Thorax 2018 May 14;73(7):662-669. [doi: 10.1136/thoraxjnl-2018-211626]
3. Childhood asthma. NHS England. URL: https://www.england.nhs.uk/childhood-asthma/ [accessed 2020-12-29]
4. Why asthma still kills National Review of Asthma Deaths. National Review of Asthma Deaths - Royal College of Physicians. 2015 Aug. URL: https://www.rcplondon.ac.uk/projects/outputs/why-asthma-still-kills [accessed 2020-12-29]
5. Nunes C, Pereira AM, Morais-Almeida M. Asthma costs and social impact. Asthma Res Pract 2017;3:1 [FREE Full text] [doi: 10.1186/s40733-016-0029-3] [Medline: 28078100]
6. Wolfe I, Thompson M, Gill P, Tamburlini G, Blair M, van den Bruel A, et al. Health services for children in western Europe. Lancet 2013 Apr 06;381(9873):1224-1234. [doi: 10.1016/S0140-6736(12)62085-6] [Medline: 23541056]
7. Lycett HJ, Raebel EM, Wildman EK, Guitart J, Kenny T, Sherlock J, et al. Theory-based digital interventions to improve asthma self-management outcomes: systematic review. J Med Internet Res 2018 Dec 12;20(12):e293 [FREE Full text] [doi: 10.2196/jmir.9666] [Medline: 30541741]
8. Tinschert P, Jakob R, Barata F, Kramer J, Kowatsch T. The potential of mobile apps for improving asthma self-management: a review of publicly available and well-adopted asthma apps. JMIR Mhealth Uhealth 2017 Aug 02;5(8):e113 [FREE Full text] [doi: 10.2196/mhealth.7177] [Medline: 28768606]
9. Unni E, Gabriel S, Ariely R. A review of the use and effectiveness of digital health technologies in patients with asthma. Ann Allergy Asthma Immunol 2018 Dec;121(6):680-691. [doi: 10.1016/j.anai.2018.10.016] [Medline: 30352288]
10. Alquran A, Lambert KA, Farouque A, Holland A, Davies J, Lampugnani ER, et al. Smartphone applications for encouraging asthma self-management in adolescents: a systematic review. Int J Environ Res Public Health 2018 Dec 29;15(11):2403 [FREE Full text] [doi: 10.3390/ijerph15112403] [Medline: 30380692]
11. Jeminiwa R, Hohmann L, Qian J, Garza K, Hansen R, Fox BI. Impact of eHealth on medication adherence among patients with asthma: A systematic review and meta-analysis. Respir Med 2019 Mar;149:59-68. [doi: 10.1016/j.rmed.2019.02.011] [Medline: 30803887]
12. Morrison D, Wyke S, Agur K, Cameron EJ, Docking RI, Mackenzie AM, et al. Digital asthma self-management interventions: a systematic review. J Med Internet Res 2014;16(2):e51 [FREE Full text] [doi: 10.2196/jmir.2814] [Medline: 24550161]
13. Stewart AC, Gannon KN, Beresford F, Fleming L. Adolescent and caregivers' experiences of electronic adherence assessment in paediatric problematic severe asthma. J Child Health Care 2018 Jun;22(2):238-250. [doi: 10.1177/1367493517753082] [Medline: 29325420]
14. Ramsey RR, Plevinsky JM, Kollin SR, Gibler RC, Guilbert TW, Hommel KA. Systematic review of digital interventions for pediatric asthma management. J Allergy Clin Immunol Pract 2020 Apr;8(4):1284-1293. [doi: 10.1016/j.jaip.2019.12.013] [Medline: 31870809]
15. Betz CL, Lewinter K, Kysh L, Hudson S, Espinoza J. Smart devices for the management of pediatric asthma: a scoping review protocol. JBI Database System Rev Implement Rep 2019 Nov;17(11):2308-2316. [doi: 10.11124/JBISRIR-D-19-00083] [Medline: 31592979]
16. Steinhubl SR, Muse ED, Topol EJ. The emerging field of mobile health. Sci Transl Med 2015 Apr 15;7(283):283rv3. [doi: 10.1126/scitranslmed.aaa3487] [Medline: 25877894]
17. Gaga M, Samitas K, Zervas E. Inhaler adherence in severe asthma: is there an electronic solution? Eur Respir J 2018 Jan;51(1):1702219 [FREE Full text] [doi: 10.1183/13993003.02219-2017] [Medline: 29301915]
18. Attaway AH, Alshabani K, Bender B, Hatipoğlu US. The utility of electronic inhaler monitoring in COPD management: promises and challenges. Chest 2020 Jun;157(6):1466-1477. [doi: 10.1016/j.chest.2019.12.034] [Medline: 31981565]
19. Learn about the launch of the innovative smart inhaler device in Europe. Health Europa. 2019. URL: https://www.healtheuropa.eu/smart-inhaler-device/91492/ [accessed 2021-01-07]
20. Harris K, Grigg J. The use of electronic monitoring devices in adherence with asthma medications, and their impact on patient outcomes: a systematic review PROSPERO registration. PROSPERO. 2019. URL: https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=127361 [accessed 2020-12-29]

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21. Blakey JD, Bender BG, Dima AL, Weinman J, Safioti G, Costello RW. Digital technologies and adherence in respiratory diseases: the road ahead. Eur Respir J 2018 Nov;52(5):1801147 [FREE Full text] [doi: 10.1183/13993003.01147-2018] [Medline: 30409819]
22. Schneider T, Panzera AD, Martinasek M, McDermott R, Couluris M, Lindenberger J, et al. Physicians' perceptions of mobile technology for enhancing asthma care for youth. J Child Health Care 2016 Jun;20(2):153-163. [doi: 10.1177/1367493514556555] [Medline: 25427556]
23. Munn Z, Peters MD, Stern C, Tufanaru C, McArthur A, Aromataris E. Systematic review or scoping review? Guidance for authors when choosing between a systematic or scoping review approach. BMC Med Res Methodol 2018 Nov 19;18(1):143 [FREE Full text] [doi: 10.1186/s12874-018-0611-x] [Medline: 30453902]
24. Tricco AC, Lillie E, Zarin W, O'Brien KK, Colquhoun H, Levac D, et al. PRISMA extension for Scoping Reviews (PRISMA-ScR): checklist and explanation. Ann Intern Med 2018 Oct 02;169(7):467-473. [doi: 10.7326/M18-0850] [Medline: 30178033]
25. Kosse RC, Bouvy ML, de Vries TW, Koster ES. Effect of a mHealth intervention on adherence in adolescents with asthma: A randomized controlled trial. Respir Med 2019 Mar;149:45-51. [doi: 10.1016/j.rmed.2019.02.009] [Medline: 30803885]
26. Beerthuizen T, Voorend-van Bergen S, van den Hout WB, Vaessen-Verberne AA, Brackel HJ, Landstra AM, et al. Cost-effectiveness of FENO-based and web-based monitoring in paediatric asthma management: a randomised controlled trial. Thorax 2016 Jul;71(7):607-613. [doi: 10.1136/thoraxjnl-2015-207593] [Medline: 27048197]
27. Bender BG, Cvietusa PJ, Goodrich GK, Lowe R, Nuanes HA, Rand C, et al. Pragmatic trial of health care technologies to improve adherence to pediatric asthma treatment: a randomized clinical trial. JAMA Pediatr 2015 Apr;169(4):317-323. [doi: 10.1001/jamapediatrics.2014.3280] [Medline: 25664620]
28. Britto MT, Rohan JM, Dodds CM, Byczkowski TL. A randomized trial of user-controlled text messaging to improve asthma outcomes: a pilot study. Clin Pediatr (Phila) 2017 Dec;56(14):1336-1344. [doi: 10.1177/0009922816684857] [Medline: 28056541]
29. Chan AH, Stewart AW, Harrison J, Camargo CA, Black PN, Mitchell EA. The effect of an electronic monitoring device with audiovisual reminder function on adherence to inhaled corticosteroids and school attendance in children with asthma: a randomised controlled trial. Lancet Respir Med 2015 Mar;3(3):210-219. [doi: 10.1016/S2213-2600(15)00008-9] [Medline: 25617215]
30. Chan AH, Stewart AW, Harrison J, Black PN, Mitchell EA, Foster JM. Electronic adherence monitoring device performance and patient acceptability: a randomized control trial. Expert Rev Med Devices 2017 May;14(5):401-411. [doi: 10.1080/17434440.2017.1322505] [Medline: 28434269]
31. Goossens LM, Vasbinder EC, Van den Bemt PM, Rutten-van Mölken MP. Cost-effectiveness of real-time medication monitoring in children with asthma. Value Health 2014 Nov;17(7):A329 [FREE Full text] [doi: 10.1016/j.jval.2014.08.605] [Medline: 27200563]
32. Johnson KB, Patterson BL, Ho Y, Chen Q, Nian H, Davison CL, et al. The feasibility of text reminders to improve medication adherence in adolescents with asthma. J Am Med Inform Assoc 2016 May;23(3):449-455. [doi: 10.1093/jamia/ocv158] [Medline: 26661717]
33. Kosse RC, Bouvy ML, Belitser SV, de Vries TW, van der Wal PS, Koster ES. Effective engagement of adolescent asthma patients with mobile health-supporting medication adherence. JMIR Mhealth Uhealth 2019 Mar 27;7(3):e12411 [FREE Full text] [doi: 10.2196/12411] [Medline: 30916664]
34. Kosse RC, Bouvy ML, de Vries TW, Koster ES. Evaluation of a mobile health intervention to support asthma self-management and adherence in the pharmacy. Int J Clin Pharm 2019 Apr;41(2):452-459 [FREE Full text] [doi: 10.1007/s11096-019-00798-3] [Medline: 31028598]
35. Merchant R, Inamdar R, Quade R, Van Sickle D, Maenner M, Patmas M. Interim results from a randomized, controlled trial of remote monitoring of inhaled bronchodilator use on asthma control and management. Chest 2013 Oct;144(4):71A. [doi: 10.1378/chest.1703281]
36. Morton RW, Elphick HE, Rigby AS, Daw WJ, King DA, Smith LJ, et al. STAAR: a randomised controlled trial of electronic adherence monitoring with reminder alarms and feedback to improve clinical outcomes for children with asthma. Thorax 2017 Apr;72(4):347-354. [doi: 10.1136/thoraxjnl-2015-208171] [Medline: 27815524]
37. Perry TT, Halterman JS, Brown RH, Luo C, Randle SM, Hunter CR, et al. Results of an asthma education program delivered via telemedicine in rural schools. Ann Allergy Asthma Immunol 2018 Dec;120(4):401-408 [FREE Full text] [doi: 10.1016/j.anai.2018.02.013] [Medline: 29471032]
38. Real FJ, Beck AF, DeBlasio D, Zackoff M, Henize A, Xu Y, et al. Dose Matters: A smartphone application to improve asthma control among patients at an urban pediatric primary care clinic. Games Health J 2019 Oct;8(5):357-365. [doi: 10.1089/g4h.2019.0011] [Medline: 31157983]
39. Reece ER, Burnette AF, Lewis-Land CJ. Pilot study of Asthmawin mobile iphone app in the management of asthma. J Allerg Clin Immunol 2017 Feb;139(2):AB382. [doi: 10.1016/j.jaci.2016.12.918]
40. Shields MD, ALQahtani F, Rivey MP, McElnay JC. Mobile direct observation of therapy (MDOT) - A rapid systematic review and pilot study in children with asthma. PLoS One 2018;13(2):e0190031 [FREE Full text] [doi: 10.1371/journal.pone.0190031] [Medline: 29401500]

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41. Simoneau T, Sun Y, Gherlone N, Almeida S, Manice M, Hollenbach J. A prospective, randomized, controlled study of inhaler electronic monitoring devices to improve adherence in children with asthma. In: Proceedings of the American Thoracic Society International Conference.: American Journal of Respiratory and Critical Care Medicine; 2019 Presented at: American Thoracic Society International Conference; May 17-22, 2019; Dallas, Texas URL: https://www.atsjournals.org/doi/abs/10.1164/ajrccm-conference.2019.199.1_MeetingAbstracts.A7177 [doi: 10.1164/ajrccm-conference.2019.199.1_meetingabstracts.a7177]
42. van den Wijngaart LS, Roukema J, Boehmer AL, Brouwer ML, Hugen CA, Niers LE, et al. A virtual asthma clinic for children: fewer routine outpatient visits, same asthma control. Eur Respir J 2017 Oct;50(4):1700471 [FREE Full text] [doi: 10.1183/13993003.00471-2017] [Medline: 28982775]
43. Vasbinder EC, Goossens LM, Rutten-van Mölken MP, de Winter BC, van Dijk L, Vulto AG, et al. e-Monitoring of Asthma Therapy to Improve Compliance in children (e-MATIC): a randomised controlled trial. Eur Respir J 2016 Sep;48(3):758-767 [FREE Full text] [doi: 10.1183/13993003.01698-2015] [Medline: 27230437]
44. Voorend-van Bergen S, Vaessen-Verberne AA, Brackel HJ, Landstra AM, van den Berg NJ, Hop WC, et al. Monitoring strategies in children with asthma: a randomised controlled trial. Thorax 2015 Jun;70(6):543-550. [doi: 10.1136/thoraxjnl-2014-206161] [Medline: 25825006]
45. Jamison D, Breman J, Measham A, Alleyne G, Claeson M, Evans D, et al, editors. Cost-effectiveness analysis. In: Priorities in Health. Washington, DC: The International Bank for Reconstruction and Development / The World Bank; 2006:39-58.

Abbreviations

ADAPT: ADolescent Adherence Patient Tool
CENTRAL: Cochrane Central Register of Controlled Trials
EMD: electronic monitoring device
MeSH: Medical Subject Headings
PRISMA-ScR: Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for scoping reviews
RCT: randomized controlled trial

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Original Paper

Changes in Anxiety and Stress Among Pregnant Women During the COVID-19 Pandemic: Content Analysis of a Japanese Social Question-and-Answer Website

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Abstract

Background: The changing pattern of anxiety and stress experienced by pregnant women during the COVID-19 pandemic is unknown.

Objective: We aimed to examine the sources of anxiety and stress in pregnant women in Japan during the COVID-19 pandemic.

Methods: We performed content analysis of 1000 questions posted on the largest social website in Japan (Yahoo! Chiebukuro) from January 1 to May 25, 2020 (end date of the national state of emergency). The Gwet AC1 coefficient was used to verify interrater reliability.

Results: A total 12 categories were identified. Throughout the study period, anxiety related to going outdoors appeared most frequent, followed by anxiety regarding employment and infection among family and friends. Following the declaration of the state of national emergency at the peak of the infection, infection-related anxiety decreased, whereas anxiety about social support and mood disorders increased. Stress regarding relationships appeared frequent throughout the pandemic.

Conclusions: The sources of anxiety and stress in pregnant women in Japan changed during the pandemic. Our results suggest the need for rapid communications in the early phase of a pandemic as well as long-term psychosocial support to provide optimal support to pregnant women in Japan. Health care professionals should understand the changing pattern of requirements among pregnant women.

(JMIR Pediatr Parent 2021;4(3):e27733) doi:10.2196/27733

KEYWORDS

anxiety; content analysis; COVID-19; health communication; health information; mental health; pregnancy; social question-and-answer website; social support; stress

Introduction

During a pandemic, public health professionals have to communicate health information to vulnerable people [1]. Pregnant women are considered vulnerable because of the unknown risks to their health and to the fetus, pregnancy-related treatment restrictions, and restrictions on the number of prenatal hospital visits. In addition to specific infection prevention measures, pregnant women also require psychological care during a pandemic as excessive anxiety and stress adversely affect both maternal and infant health [2]. Many previous studies have reported a higher incidence of anxiety among pregnant

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women during the ongoing COVID-19 pandemic, with a prevalence rate of 30% for severe depressive and anxiety symptoms [3,4].

Effective risk communication campaigns require a "social constructionist approach," which sees risk as being constructed through social and cultural processes [5-7]. In this concept, perceived risk may fluctuate through social processes and professionals have to understand what the stakeholders may ask and expect at each stage of a pandemic. Previous studies have reported that increased distress among pregnant women has been attributed to various factors, such as the risk of perinatal infection, unpreparedness for delivery, and altered support relationships [8,9]. However, little is known about changes in the sources of anxiety and stress over time.

To cope with isolation associated with enforced lockdown measures during the COVID-19 pandemic, many individuals began to use the internet to search for information and connect with others [10]. There are many social question-and-answer (Q&A) websites, where individuals may freely post questions to be answered by other community members. In the past, content analysis of questions posted to Q&A websites provided valuable insights into negative feelings and anxiety [11]. Social Q&A sites can be similarly useful for rapid investigation of anxiety and stress in dynamic situations such as the COVID-19 pandemic.

This study aimed to identify whether and how the sources of anxiety and stress in pregnant women in Japan changed during the COVID-19 pandemic, by carrying out content analysis of a social Q&A website, to inform preparations for timely essential support for this population in the ongoing and future pandemics.

Methods

Material Collection

The data were extracted from all questions posted in Japanese on Yahoo! Chiebukuro [12], the largest social Q&A website in Japan, from January 1 to May 25, 2020 (the end date of the national state of emergency). The questions were identified through a web-based search of the Japanese terms "corona AND ninnpu (pregnant woman)," "corona AND ninnshin (pregnancy)," "corona AND syussann (birth)," "corona AND bunnben (delivery)," and "corona AND osann (delivery)." We included only questions posted by pregnant women and excluded those posted by women with intrauterine fetal death, those unrelated to anxiety or stress about COVID-19, and duplicate questions. Because the data were publicly available, the requirement for informed consent was waived.

Coding Procedure

The unit of coding was each question. Author RS read the text of all questions and then inductively assigned codes and categories to the extracted descriptions of anxiety and stress. When a question expressed multiple sources of distress, we coded it into all applying categories or codes. We enumerated the questions in each category or code. RS also tabulated the posting dates to correlate changes with weekly events in Japan.

Two independent coders (RS an RY) coded 20% of the questions, and interrater reliability was measured using the Gwet AC1 coefficient [13]. We used the coding carried out by RS for the analysis.

Coding procedures were conducted using Excel (version 2011, Microsoft Inc). Statistical analyses were performed using R for Windows (version 4.0.2, R Foundation for Statistical Computing).

Results

Material and Coding

A total of 4200 "hits" were obtained from the search terms, of which 2040 were questions posted by pregnant women. After excluding questions by women with intrauterine fetal death (n=5), unrelated questions (n=376), and duplicate questions (n=659), we retained 1000 questions for analysis (Figure 1). The questions had a median of 360 (IQR 228-546) Japanese characters and yielded 12 categories and 20 constituent codes (Table 1), which demonstrated strong interrater reliability (Gwet AC1=0.93, 95% CI 0.92-0.94). The total number of the codes was 1677 (median 1, IQR 1-2).

Figure 1. Flow chart for the selection of questions for analysis on Yahoo! Chiebukuro (the Japanese version of Yahoo! Answers), the largest social question-and-answer website in Japan.

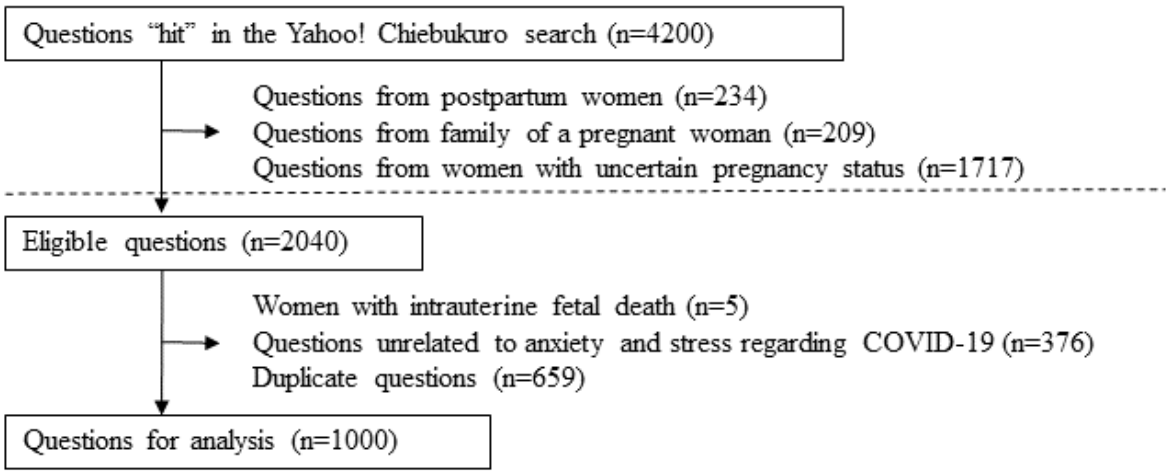

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Table 1. Definitions of the assigned anxiety and stress categories and constituent codes.

Codes	Content	Example terms[a] extracted from the questions
Personal infection		
Risk of maternal infection	Anxiety about personal infection as a pregnant woman	Infection, coronavirus, maternal death, become severe, risk to pregnant women, immune weakness, and contraindications
Pre-existing conditions or contacts with infected persons	Among nonpregnant women; anxiety about personal infection related to current symptoms, chronic conditions, or contact with a suspected infectious person	Fever, sore throat, asthma, and diabetes mellitus
Infecting others	Anxiety about infecting others if asymptomatically infected	Family, other pregnant women, and canceled consultation
Fetal well-being		
Adverse effects of infection	Anxiety about transmitting SARS-CoV-2 to the fetus or neonate	Baby, fetus, newborn, protect, mother-to-child transmission, abortion, stillbirth, disability, and sequelae
Adverse effects of stress	Anxiety about adverse effects of maternal stress	Baby, fetus, newborn, and stress
Going outdoors		
Daily life (eg, going to work)	Anxiety or stress about going outdoors for each purpose	Shopping, supermarket, workplace, and government offices
Hospital visits (including prenatal check-ups)	Anxiety or stress about going outdoors for each purpose	Hospital, prenatal check-up, parents' or mother's class, and dentist
Family events	Anxiety or stress about going outdoors for each purpose	Wedding, graduation ceremony, and funeral
Social or leisure activities	Anxiety or stress about going outdoors for each purpose	Party, travel, beauty salon, and zoo
Infection among family and friends		
Daily life (eg, going to work or school)	Anxiety or stress about contacts, including the behavior of those close to the person, such as partner, family members, and friends and workplace colleagues	Workplace, business trip, school, kindergarten, hospital, and daily shopping
Family events	Anxiety or stress about contacts, including the behavior of those close to the person, such as partner, family members, and friends and workplace colleagues	Wedding, graduation ceremony, and funeral
Undesirable outings or behaviors	Anxiety or stress about contacts, including the behavior of those close to the person, such as partner, family members, and friends and workplace colleagues	Dinner party, drinking party, travel, gambling, unwelcome visit, visit after delivery, not washing hands, and not wearing a mask
Relationships		
Discord from risk behavior	Anxiety or stress and deteriorating relationships over others' risk behaviors	Quarrel, get angry or annoyed, dislike, untrustworthy, divorce, and yelling at child
Discord from each other factors (eg, spending more time together, estrangement, and stress)	Distress about relationships with partners, children, family, or friends, apart from risk behavior	Quarrel, get angry or annoyed, dislike, untrustworthy, divorce, and yelling at child
Mood disorders	Extreme feelings of sadness or depression	Cry, depressed, anxiety, finding it hard to live, wanting to die, abuse, symptoms of stress (eg, arrhythmia, stomach ache), and postponed or canceled ceremony
Employment		
Financial insecurity	Anxiety about money	Salary, unpaid, allowance, retirement, unemployment,
Treatment as pregnant woman at the workplace	Anxiety or stress about workplace practices or system, such as being forced to work or forced retirement	Forced to work, reduced working hours, maternity or child care leave, intends to leave, harassment, and revealing the pregnancy

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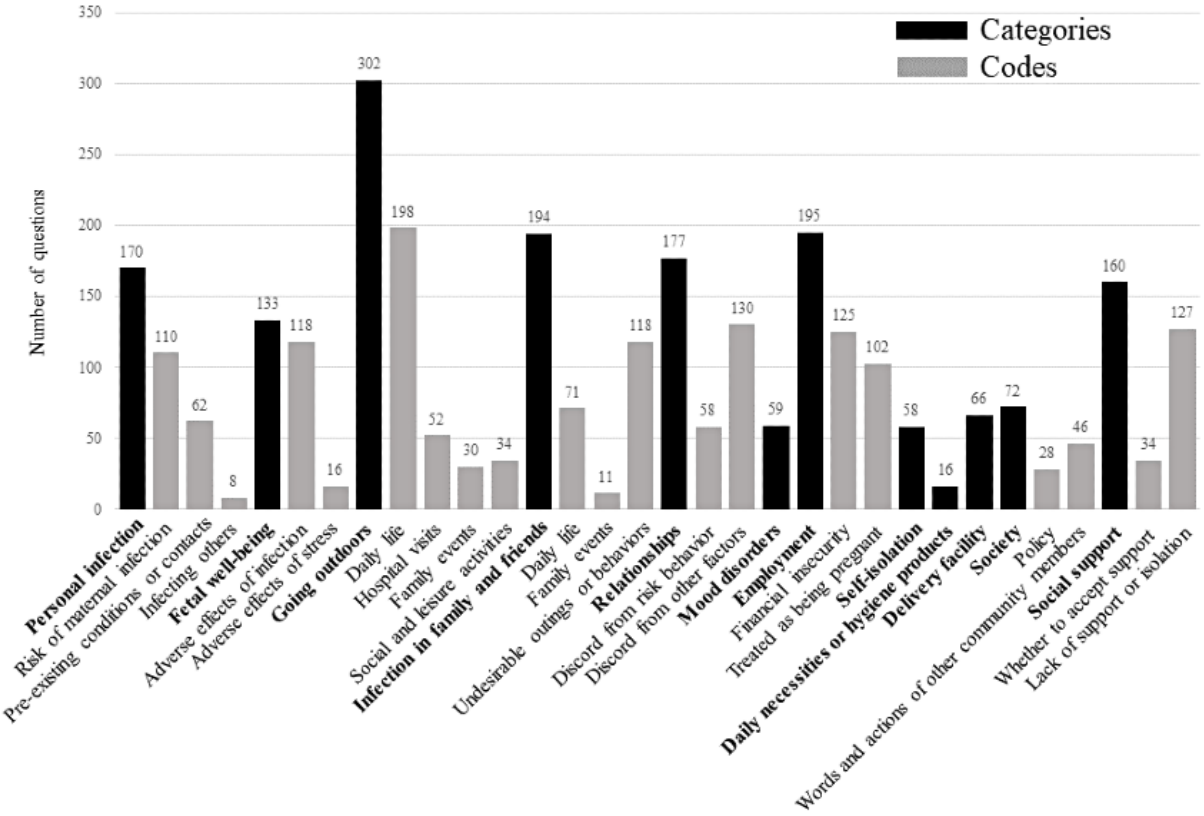
Codes	Content	Example terms[a] extracted from the questions
Self-isolation	Anxiety or stress about self-isolation or staying at home	Stay at home, all day long, meal, exercise, and neighborhood noise
Daily necessities or hygiene products	Anxiety about daily necessities or hygiene	Lifeline, mask, thermometer, baby supplies, hoarding, and lack
Delivery facility	Anxiety or stress about the delivery facility (excluding prenatal visits and delivering alone)	Delivery facility, large hospital, returning to parental home around delivery (*Satogaeri*), transfer, waiting time, postponed check-ups, and cesarean section
Society		
Policy	Anxiety or stress about social policies or systems	Government, municipality, system, emergency economic measures, and cash payment
Actions or words of community members	Anxiety or stress about talk or actions of people in the community	Hoarding, blame, and public
Social support		
Whether to accept support	Stress about accepting support	Self-restraint, parents, friends, homecoming, school, and kindergarten
Lack of support or isolation	Anxiety about lack of support or isolation	Alone, no one to talk to, lonely, delivering alone, visit restrictions, father, refused consultation, and caring for a child alone

[a]Terms have been translated from Japanese into English in this publication.

Distribution of Categories and Codes

Figure 2 shows the frequency distribution of the assigned categories and codes throughout the study period. The number for each category refers to the total number of applied questions and not the sum of the codes. Anxiety about going outdoors was most frequent, followed by anxiety related to work and anxiety related to infection among family and friends. Within the category "going outdoors," anxiety or stress about infection in daily life (including going to work) was predominant, while in the category "infection in family and friends," anxiety or stress related to undesirable activity was most frequent.

Figure 2. Frequency distribution of the anxiety and stress categories and codes described in the search questions. Some questions included multiple categories and codes.



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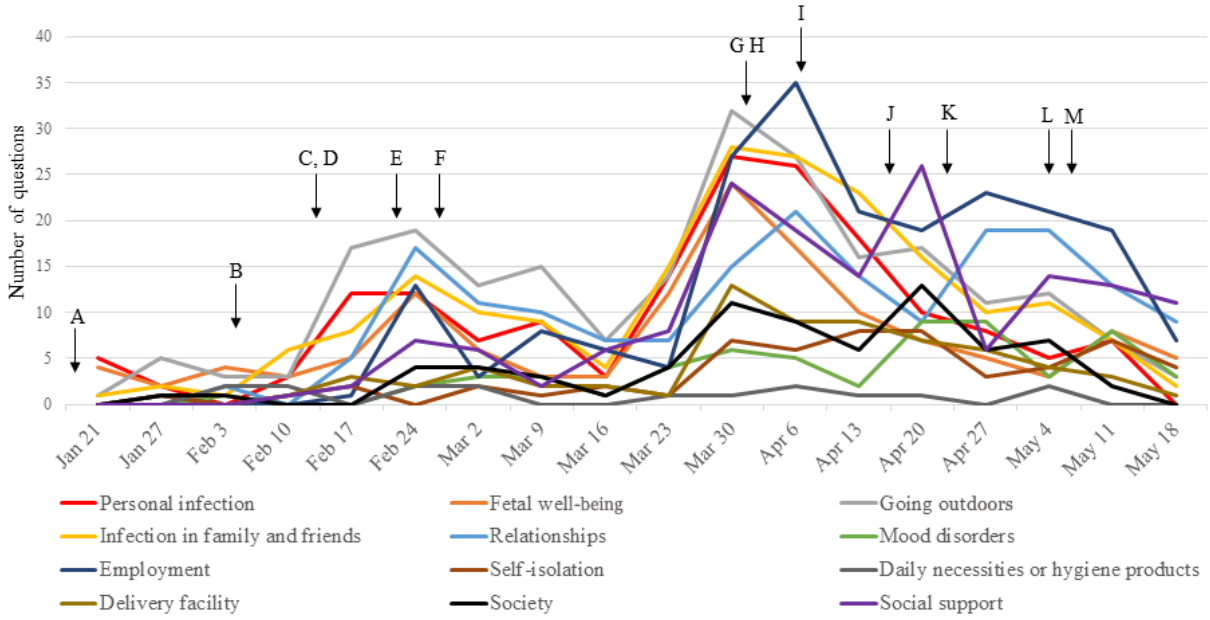
Changes in the Frequency Distribution of Categories

Figure 3 shows the weekly changes in the frequency distribution of the anxiety categories and the chronology of key events among pregnant women in Japan. There were 3 peaks in the number of questions: the first peak occurred in the week of February 24, 2020, which coincided with the first wave of infection spreading from China; the second peak occurred in the week of April 6, 2020, which coincided with the sharp increase in infection with the second wave spreading from Europe and the United States; and the third peak occurred in the week of May 4, which coincided with the extended state of emergency. Infection-related anxiety (maternal infection, going outdoors, and infection among family and friends), and contributed to the first and the second peaks but declined during the third peak, during which anxiety about social support and mood disorders increased. Questions related to relationship stress were frequent throughout the study period.

Figure 3. Weekly changes in the distribution of anxiety and stress categories and the chronology of key events among pregnant women in Japan. A: first COVID-19 case in Japan (January 16, 2020), B: report of a neonate infected with COVID-19 in China (February 5, 2020), C: first COVID-19 death in Japan (February 13, 2020), D: infection in a male physician; community infection suspected (February 13, 2020), E: official announcement of candidate drugs, including one contraindicated among pregnant women (February 22, 2020), F: nationwide school closures in Japan (February 27, 2020), G: report of a severe infection in a neonate in Japan (April 1, 2020), H: official request to economic and labor organizations for special considerations for pregnant workers (April 1, 2020), I: declaration of a state of emergency in 7 prefectures (April 7, 2020), J: declaration of a national state of emergency (April 16, 2020), K: reports of refusal among rural hospitals to accept transfers of pregnant women (April 23, 2020), L: extension of the national state of emergency (May 4, 2020), and M: amendment of labor guidelines that enable leave of absence from work among pregnant workers during the COVID-19 pandemic (May 7, 2020).



Discussion

Principal Findings

During the COVID-19 pandemic, pregnant women in Japan expressed anxiety about infection and work as well as stress regarding relationships and social support. The sources of anxiety and stress changed over time; infection-related anxiety increased during the early phase of rapid pandemic growth, while anxiety about social support and mood disorders increased with the extending period of self-isolation.

Anxiety and Stress in Pregnant Women

According to previous studies, many pregnant women were anxious about using public transportation and about infection among family members during the COVID-19 pandemic [14,15]. Pregnant women and new mothers were also uniquely impacted by stress related to reduced social and emotional support or family conflicts during the pandemic [9]. The anxiety and stress identified in this study is consistent with those reported previously. During the outbreak, the prevalence of severe depressive or anxiety symptoms among pregnant women increased with an increase in the number of cases and deaths [4]. Further, up to 30% of pregnant women reported experiencing anxiety symptoms even during the remission phase [16]. Given these findings, our study suggests the need for different types of care for pregnant women during a pandemic; rapid intervention is warranted during early stages of infection spread, whereas long-term support is needed even as the infection begins to wane.

At the beginning of the COVID-19 pandemic, risk communication campaigns seemed to fail owing to their reliance on a realist approach [6,7]. In this study, pregnant women frequently posted questions regarding their anxiety and stress related to maternal infection during the early spread of the pandemic. It has been reported that among expectant mothers, anxiety was heightened upon learning about infected new mothers and the lack of information from their physicians [17]. Pregnant women may perceive risk uniquely because the risk of affecting offspring is likely to be deemed greater [18]. Health care providers should use a variety of communication channels and added information resources to distribute audience-specific health messages [1]. Our results further suggest the need for

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specific messages that target family and friends—undesirable behaviors in the people surrounding the pregnant woman account for most of the anxiety about infection in family and friends as well as for relationship stress. Through a constructionist approach, there may be a need for different types of messages for partners and family members.

As the period of self-isolation became prolonged, pregnant women frequently posted about mood disorders and the lack of social support. A previous study reported that minimal contact with health care providers and the lack of routine nursing care during the outbreak contributes to social isolation [17]. Provision of perinatal care as part of infection prevention protocols requires ingenuity. Our results also suggest that health care providers should continue to evaluate psychological distress in pregnant women during this extended period.

Implication in Practice and Future Studies

During a crisis, professionals must attempt to identify the needs of people in a timely manner because their needs may change with time or through social processes. In the expanding phase of a pandemic, health care providers should provide information regarding infection to pregnant women and their support network. With prolonging self-isolation, pregnant women also require psychosocial support. Evaluation of interventions including virtual perinatal care [19] will be needed in future.

Limitations

This study has some limitations. Although our results do not differ from those of previous studies, the descriptions of anxiety and stress posted on the social Q&A site may have been biased. Although Yahoo! Chiebukuro is widely used (resolving approximately 75,000 questions per month), its use as the data source may have introduced a selection bias, given that the characteristics of the user population were unknown. The assigned codes may also have reflected an author bias. Despite these limitations, to our knowledge, this is the first study to show the changing pattern of anxiety and stress in pregnant women during the COVID-19 pandemic.

Conclusions

Our findings show that pregnant women in Japan experienced anxiety and stress about infection in the early stages of the COVID-19 pandemic; however, over time, they increasingly experienced mood disorders and distress about the lack of social support. Professionals must understand these changing needs of vulnerable populations for effective communication during a crisis.

Acknowledgments

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Authors' Contributions

RS, TO, and RY designed the study. RS collected the data. RS and RY analyzed the data. RS drafted the manuscript. TO critically revised the manuscript. HO, EG, and TK made substantial contributions to strengthening the interpretation. RS was responsible for the final content. All authors read and approved the final manuscript.

Conflicts of Interest

None declared.

References

1. Interim Guidance: Public Health Communicators Get Your Community Ready for Coronavirus Disease 2019 (COVID-19). Centers for Disease Control and Prevention. URL: https://www.cdc.gov/healthcommunication/phcomm-get-your-community-ready.html [accessed 2021-05-13]
2. Dunkel Schetter C, Tanner L. Anxiety, depression and stress in pregnancy: implications for mothers, children, research, and practice. Curr Opin Psychiatry 2012 Mar;25(2):141-148 [FREE Full text] [doi: 10.1097/YCO.0b013e3283503680] [Medline: 22262028]
3. Berthelot N, Lemieux R, Garon-Bissonnette J, Drouin-Maziade C, Martel É, Maziade M. Uptrend in distress and psychiatric symptomatology in pregnant women during the coronavirus disease 2019 pandemic. Acta Obstet Gynecol Scand 2020 Jul;99(7):848-855. [doi: 10.1111/aogs.13925] [Medline: 32449178]
4. Wu Y, Zhang C, Liu H, Duan C, Li C, Fan J, et al. Perinatal depressive and anxiety symptoms of pregnant women during the coronavirus disease 2019 outbreak in China. Am J Obstet Gynecol 2020 Aug;223(2):240.e1-240.e9 [FREE Full text] [doi: 10.1016/j.ajog.2020.05.009] [Medline: 32437665]
5. Joffe H. Risk: from perception to social representation. Br J Soc Psychol 2003 Mar;42(Pt 1):55-73. [doi: 10.1348/014466603763276126] [Medline: 12713756]
6. Smith RD. Responding to global infectious disease outbreaks: lessons from SARS on the role of risk perception, communication and management. Soc Sci Med 2006 Dec;63(12):3113-3123 [FREE Full text] [doi: 10.1016/j.socscimed.2006.08.004] [Medline: 16978751]

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7. White SJ, Barello S, Cao di San Marco E, Colombo C, Eeckman E, Gilligan C, et al. Critical observations on and suggested ways forward for healthcare communication during COVID-19: pEACH position paper. Patient Educ Couns 2021 Feb;104(2):217-222 [FREE Full text] [doi: 10.1016/j.pec.2020.12.025] [Medline: 33419600]
8. Preis H, Mahaffey B, Lobel M. Psychometric properties of the Pandemic-Related Pregnancy Stress Scale (PREPS). J Psychosom Obstet Gynaecol 2020 Sep;41(3):191-197. [doi: 10.1080/0167482X.2020.1801625] [Medline: 32838629]
9. Chivers BR, Garad RM, Boyle JA, Skouteris H, Teede HJ, Harrison CL. Perinatal Distress During COVID-19: Thematic Analysis of an Online Parenting Forum. J Med Internet Res 2020 Sep 07;22(9):e22002 [FREE Full text] [doi: 10.2196/22002] [Medline: 32857707]
10. Taylor S, Landry CA, Paluszek MM, Fergus TA, McKay D, Asmundson GJG. COVID stress syndrome: Concept, structure, and correlates. Depress Anxiety 2020 Aug;37(8):706-714 [FREE Full text] [doi: 10.1002/da.23071] [Medline: 32627255]
11. Hodgson NS, Yom-Tov E, Strong WF, Flores PL, Ricoy GN. Concerns of Female Adolescents About Menarche and First Sexual Intercourse: Mixed Methods Analysis of Social Media Questions. JMIR Pediatr Parent 2019 Jun 04;2(1):e13158 [FREE Full text] [doi: 10.2196/13158] [Medline: 31518326]
12. Yahoo Japan. URL: https://chiebukuro.yahoo.co.jp/ [accessed 2021-06-30]
13. Gwet KL. Computing inter-rater reliability and its variance in the presence of high agreement. Br J Math Stat Psychol 2008 May;61(Pt 1):29-48. [doi: 10.1348/000711006X126600] [Medline: 18482474]
14. Corbett GA, Milne SJ, Hehir MP, Lindow SW, O'connell MP. Health anxiety and behavioural changes of pregnant women during the COVID-19 pandemic. Eur J Obstet Gynecol Reprod Biol 2020 Jun;249:96-97 [FREE Full text] [doi: 10.1016/j.ejogrb.2020.04.022] [Medline: 32317197]
15. Taubman-Ben-Ari O, Chasson M, Abu Sharkia S, Weiss E. Distress and anxiety associated with COVID-19 among Jewish and Arab pregnant women in Israel. J Reprod Infant Psychol 2020 Jul;38(3):340-348. [doi: 10.1080/02646838.2020.1786037] [Medline: 32573258]
16. Zeng X, Li W, Sun H, Luo X, Garg S, Liu T, et al. Mental Health Outcomes in Perinatal Women During the Remission Phase of COVID-19 in China. Front Psychiatry 2020;11:571876 [FREE Full text] [doi: 10.3389/fpsyt.2020.571876] [Medline: 33132935]
17. Dodgson JE, Tarrant M, Chee Y, Watkins A. New mothers' experiences of social disruption and isolation during the severe acute respiratory syndrome outbreak in Hong Kong. Nurs Health Sci 2010 Jun;12(2):198-204. [doi: 10.1111/j.1442-2018.2010.00520.x] [Medline: 20602692]
18. Slovic P. Perception of risk. Science 1987 Apr 17;236(4799):280-285. [doi: 10.1126/science.3563507] [Medline: 3563507]
19. Marshall J, Kihlström L, Buro A, Chandran V, Prieto C, Stein-Elger R, et al. Statewide Implementation of Virtual Perinatal Home Visiting During COVID-19. Matern Child Health J 2020 Oct;24(10):1224-1230 [FREE Full text] [doi: 10.1007/s10995-020-02982-8] [Medline: 32691359]

Abbreviations

Q&A: question-and-answer

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Review

Telemedical Approaches to Managing Gestational Diabetes Mellitus During COVID-19: Systematic Review

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Abstract

Background: In 2019, a new coronavirus emerged in China, and the disease caused by the virus (COVID-19) was rapidly classified as a pandemic. Pregnant women with gestational diabetes mellitus (GDM) are considered to be at risk for severe COVID-19. In the context of the pandemic, there are serious concerns regarding adverse effects on maternal and neonatal outcomes for women with GDM. Effective treatments for patients with GDM are therefore particularly important. Due to contact restrictions and infection risks, digital approaches such as telemedicine are suitable alternatives.

Objective: This systematic review aims to summarize currently available evidence on maternal and offspring outcomes of pregnant women with GDM and COVID-19 and to examine telemedical interventions to improve maternal glycemic control during the COVID-19 pandemic.

Methods: Publications were systematically identified by searching the Cochrane Library, MEDLINE via PubMed, Web of Science Core Collection, Embase, and CINAHL databases for studies published up to March 2021. We sorted the COVID-19 studies by outcome and divided the telemedical intervention studies into web-based and app-based groups. We analyzed case reports (COVID-19) and both randomized and nonrandomized controlled clinical trials (telemedicine). To determine the change in glycated hemoglobin A_{1c} (HbA_{1c}), we pooled appropriate studies and calculated the differences in means, with 95% CIs, for the intervention and control groups at the end of the interventions.

Results: Regarding COVID-19 studies, we identified 11 case reports, 3 letters, 1 case series, and 1 retrospective single-center study. In total, 41 patients with GDM and COVID-19 were analyzed. The maternal and neonatal outcomes were extremely heterogeneous. We identified adverse outcomes for mother and child through the interaction of GDM and COVID-19, such as cesarean deliveries and low Apgar scores. Furthermore, we selected 9 telemedicine-related articles: 6 were randomized controlled trials, 2 were clinical controlled trials, and 1 was a quasi-experimental design. In total, we analyzed 480 patients with GDM in the intervention groups and 494 in the control groups. Regarding the quality of the 9 telemedical studies, 4 were rated as strong, 4 as moderate, and 1 as weak. Telemedical interventions can contribute to favorable impacts on HbA_{1c} and fasting blood glucose values in the context of the COVID-19 pandemic. Meta-analysis revealed a mean difference in HbA_{1c} of –0.19% (95% CI 0.34% to 0.03%) for all telemedical interventions, –0.138% (95% CI –0.24% to –0.04%) for the web-based interventions, and –0.305% (96% CI –0.88% to 0.27%) for the app-based interventions.

Conclusions: Telemedicine is an effective approach in the context of COVID-19 and GDM because it enables social distancing and represents optimal care of patients with GDM, especially with regard to glycemic control, which is very important in view of the identified adverse maternal and neonatal outcomes. Further research is needed.

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KEYWORDS

gestational diabetes; telemedicine; mobile applications; COVID-19; systematic review; digital health; diabetes

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Introduction

In 2019, a new coronavirus known as SARS-CoV-2 emerged in Wuhan, China; the disease caused by this virus, COVID-19, was classified as a pandemic in a short period of time [1]. Since then, 115,289,961 million people have been diagnosed with COVID-19 worldwide, and 2.564.560 million people had died as of March 5, 2021 [1]. The most vulnerable populations to this virus are those with chronic diseases such as diabetes mellitus (DM) [2-4]. Pregnant women are also considered risk patients [5]. In addition, pregnancy complications such as DM and hypertension have known as a double risk factor of COVID-19 for pregnant women [6]. Pregnant women become particularly vulnerable if they are diagnosed with gestational diabetes mellitus (GDM) and also infected with SARS-CoV-2.

In 2019, in approximately 16% of live births, the mother suffered from hyperglycemia during pregnancy [7]. In total, 84% of those births were associated with GDM [7]. The global prevalence of GDM was between 2.1% and 37.5% as of 2019, depending on screening methods and diagnostic criteria [7]. GDM is diagnosed in the second or third trimester and is defined as not overt diabetes prior to gestation [8]. GDM-complicated pregnancies are associated with adverse maternal and offspring outcomes such as preeclampsia, hypoxia, pregnancy-induced hypertension, type 2 diabetes, obesity and macrosomia, neonatal hypoglycemia, large for gestational age, and adult type 2 diabetes, and cardio-metabolic diseases [9-11]. Intrauterine exposure to increased levels of hyperglycemia "program" the offspring for these lifelong consequences [12-16]. This concept of transgenerational programming (also known as "fetal programming" or "perinatal programming") leads to perturbation during the development phase by molecular mechanisms, which may lead to dysfunctions in organs and metabolism [12-16].

In the context of the COVID-19 pandemic and with a focus on women with GDM, there are serious concerns regarding adverse effects on maternal, fetal, and neonatal outcomes [17]. Patients with GDM need special care; however, clinical implications of COVID-19 are unexplored [5]. Social distancing and quarantine have ensured that clinic visits are reduced owing to fear of infection [18]. Furthermore, the measures ensure that physical activity is reduced and eating habits change negatively; moreover, there are overall effects on health care and access to medication [18].

Owing to the contact restrictions and restrictions in the health care area, digital solutions are available to ensure close treatment of patients with GDM. National guidelines explicitly recommend telemedicine for the management of GDM during the COVID-19 pandemic in countries such as Canada, Australia, New Zealand, United Kingdom, Germany, Italy, and India [19-22]. The national guidelines advise alternative opportunistic screening strategies with a focus on glycated hemoglobin A_{1c} (HbA_{1c}), random plasma glucose (RPG), and fasting blood glucose (FBG).

Diabetes technology includes hardware, software, and technical devices that help to control the disease [23]. Telemedical treatments show great potential in clinical diabetes management [24-26]. Telemedicine characterizes the use of communication technologies to improve patient outcomes by increasing access to care and medical information [23]. Telemedicine overcomes geographical and physical limits, improves health care access, and enhances health-related outcomes [23].

More clinical and scientific analyses are urgently needed in the context of COVID-19, GDM, and digital therapy options. It is generally advisable to draw up a general pandemic guideline with corresponding recommendations for action, which can also be useful in case of other pandemics.

Against this background, we examine pregnant women with GDM in more detail in the context of the COVID-19 pandemic and with a view to telemedical treatment options. In this new research area, it is important to analyze previous literature and provide suggestions for further research. Due to the limited evidence in this field, this systematic review aimed to summarize currently available evidence on maternal and offspring outcomes of pregnant women with GDM and COVID-19 and to examine telemedical treatment methods to improve maternal glycemic control during the COVID-19 pandemic. With this analysis, we would like to set a first milestone and a starting point for further research in the areas of gestational diabetes, COVID-19, and telemedicine.

Methods

Search Strategy

In general, when creating the systematic review and analyzing the studies, we followed the *Cochrane Handbook for Systematic Reviews of Interventions* [27].

Publications were systematically identified by searching the Cochrane Library, MEDLINE via PubMed, Web of Science Core Collection, Embase, and CINAHL databases for studies published up to March 2021.

We conducted two systematic searches to address our respective research questions. One focused on the topic of GDM and COVID-19, and the other focused on the topic of GDM and telemedicine. The search strategies are shown in Multimedia Appendix 1. After searching the databases, we removed duplicate entries, screened titles and abstracts for suitability, and then read the full texts. The studies were selected by two independent reviewers.

COVID-19

This search was conducted using the following keywords: ("gestational diabetes mellitus") AND ("COVID-19" OR "coronavirus" OR "SARS-CoV-2"). Medical Subject Headings (MeSH) and Embase Subject Headings terms as well as title/abstract terms were searched. In addition, we manually searched reference lists.

Telemedicine

This search was conducted using the following keywords: ("gestational diabetes mellitus") AND ("telemedicine" OR "telemonitoring" OR "telemetry" OR "mHealth" OR "mobile applications" OR "smartphone"). MeSH and Embase Subject Headings terms as well as title/abstract terms were searched. In addition, we manually searched reference lists.

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Inclusion Criteria

COVID-19

Studies that met the following inclusion criteria were selected: peer-reviewed; published in English or German; observational, cohort, and clinical studies, case reports/series, letters, and comments reporting maternal, fetal, and neonatal outcomes in pregnant women with GDM and COVID-19.

Because COVID-19 emerged at the end of 2019 and is still quite unexplored, especially for special target groups such as patients with GDM, few studies are available to date. We therefore considered various study designs (including case reports and letters) and included all papers that reported on patients with COVID-19 and GDM, even if GDM was not explicitly the focus of the respective paper. Furthermore, we did not impose any restrictions on maternal, fetal, or neonatal outcomes.

Telemedicine

Articles that met the following inclusion criteria were selected: peer-reviewed; published in English or German; clinical controlled trials (CCTs) and randomized controlled trials (RCTs) examining telemedical treatment for pregnant women with GDM and reporting on the maternal outcomes of HbA_{1c}, FBG, or RBG.

Telemetry was defined as remote recording and transmission of patient data via a telecommunications system to a health care provider to provide clinical support and improve health outcomes [23]. We included video calls, telephone calls, internet/web-based platforms, and smartphone/mobile app–based interventions.

Exclusion Criteria

COVID-19

The following articles were excluded: papers that did not specify the type of diabetes, and articles that did not report maternal or offspring outcomes and focused instead on GDM screening/management/diagnosis, anxiety and stress during lockdown, or prevention of GDM.

Telemedicine

The following articles were excluded: posters, comments, letters, study protocols, and proceedings; studies that did not specify the type of diabetes; studies that described technologies only; studies that pooled data with other technologies and other diseases; and papers focusing on GDM prevention or GDM diagnosis.

Data Extraction

We extracted the year of publication, study region, study design, patient characteristics, intervention details for the telemedical studies (type of technology, intervention and control group, sample size), outcomes, and main results.

Data Synthesis and Analysis

COVID-19

We sorted the COVID-19 studies by outcome. We divided the outcomes into maternal and offspring outcomes. Where possible, we calculated the means (Apgar score and birth weight).

Telemedicine

We divided the telemedical studies into two groups: web-based and app-based. Web-based interventions are based on websites and the internet, with asynchronous communication between patients and health care providers. App-based interventions are based on smartphone and mobile phone apps.

We conducted a meta-analysis to determine the impact of the interventions on the HbA_{1c} concentration because this was the most frequently studied outcome. We used Excel (Microsoft Corporation) for pooling the data and for calculations, and we took into account all studies that provided complete information. In the calculations, we considered web-based and app-based studies. For determining the change in HbA_{1c} (%), we pooled appropriate studies and calculated the differences in the means, with 95% confidence intervals, for the intervention and control groups at the end of the interventions. Based on the HbA_{1c} mean differences and confidence intervals, we created a forest plot.

Assessment of Risk of Bias

A quality appraisal of the studies was performed to determine the risk of bias. We used the valid and reliable Effective Public Health Practice Project (EPHPP) tool [28] for the appraisal of randomized and nonrandomized clinical trials on health-related topics. EPHPP consists of the following components: selection bias, study design, confounders, blinding, data collection methods, and withdrawals and dropouts. The tool classifies the study quality ranging between strong, moderate, and weak. The assessment was performed by one reviewer.

Results

COVID-19

Search Results and Trial Flow

The literature search yielded 228 citations, of which 136 unique citations were screened based on title and abstract (Multimedia Appendix 2). Then, we screened 19 articles based on the full texts. We added 1 study (Blauvelt et al [29]) through manual research. Finally, we selected 16 appropriate COVID-19–related articles in this systematic review [3,5,29-42]. Of these, 11 were case reports, 3 were letters, 1 was a case series, and 1 was a retrospective single-center study. In total, 41 patients with GDM were analyzed. An overview of all included studies is provided in Multimedia Appendix 3.

Overall, the reported outcomes are very heterogeneous, and there are only a few overlaps of the same outcomes in the studies. It should also be noted that the papers are not very detailed and generally contain little information, which is related to the fact that at the beginning of the COVID-19 pandemic, papers can be published more quickly and in a simplified form.

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Maternal Outcomes

The most common COVID-19 symptoms were fever (n=12), cough (n=13), dyspnea (n=4), shortness of breath (n=3), diarrhea (n=2), and vomiting (n=2).

In total, 22 women required cesarean deliveries, including 3 emergency cesarean deliveries. Furthermore, 5 women were admitted to the intensive care unit (ICU). Blauvelt et al [29] reported lung overdistension, hyperglycemia after antenatal corticosteroid administration, and mechanical ventilation (n=1 woman). Cooke et al [30] noticed that 1 patient experienced psychiatric sequelae postoperatively. In addition, Vlachodimitropoul et al [31] outlined a postpartum hemorrhage of 1.5 L, controlled with uterine artery ligation and B-Lynch compression alongside uterotonics and blood products (n=1 patient). Govind et al [32] reported that, following delivery, a patient continued to desaturate (80%-85%) on 100% oxygen.

Kleinwechter and Laubner [3] noted vaginal bleeding and that the patient was fever-free at day 6, with oxygen requirement declining (n=1 woman).

Moreover, Oliva et al [33] outlined that the status of the patient (fever, dyspnea, respiratory status worsening from oxygen saturation as measured by pulse oximetry [SpO_2] 95%-92%) improved rapidly post–cesarean delivery (2 hours after delivery SpO_2 to the low 90th percentile on room air, which improved to 100% on 15 L/min of oxygen) and that the cesarean delivery was uncomplicated. Uzel and Lakhno [35] found leukocytosis (19 × 103/µL) and mild anemia (hemoglobin 10.1 g/dL) in the clinical blood test (n=1 woman). Activated partial thromboplastin time, prothrombin time, and international normalized ratio were prolonged, levels of C-reactive protein (179.7 mg/L) and procalcitonin (0.15 ng/ml) were raised, and low oxygen saturation (75%) was detected. The authors reported that the patient died (35 year-old woman with obesity, lymphopenia, hyperglycemia, and trace proteinuria). D'Ambrosi et al [37] outlined that 6 women (in a retrospective single-center study) did not require ICU admission or mechanical ventilation, but 4 women underwent elective cesarean deliveries.

In addition, Fontanella et al [38] reported that there were no adverse maternal outcomes and the women left the hospital after 5 days because of rapid improvement. Kleinwechter et al [5] (n=21 women) showed 2 admissions to the ICU, and in 4 cases, the women received symptomatic COVID-19–associated therapy. No invasive ventilation was required, and 56% of the cases were cesarean deliveries (9/16).

Offspring Outcomes

The mean Apgar score (1 minute) was 5.9 in 8 newborns [29-33,35,36,39]. In addition, the mean Apgar score (5 minutes) was 7.5 in 17 newborns (including 1 set of triplets) [29-33,35-37,39]. The birth weight [30,32-37,40,41] was between 1250 g [40] (a triplet) and 4165 g [32], whereas the mean birth weight was approximately 2621.29 g (n=17 newborns).

Blauvelt et al [29] reported that the newborn evolved respiratory distress syndrome and laboratory test results showed leukopenia, neutropenia, lymphopenia, mild acidosis, and normal lactate. A SARS-CoV-2 test was negative, and at 16 days of life, the neonate was clinically stable on high-flow nasal cannula at 3 L/min and 21% fraction of inspired oxygen (FiO_2). Govind et al [32] observed viral pneumonia on day 6, but the newborn recovered well and was ventilated for 10 days.

Moreover, Oliva et al [33], Uzel and Lakhno [35], Majachani et al [39] and Rabiei et al [40] noted admissions to the neonatal intensive care unit (NICU) (n=6 newborns). In total, 2 newborns needed supplement oxygen [32,33]. Additionally, 7 children tested negative for COVID-19 after their births [29,35,36,40,41], and 1 had a positive nasopharyngeal swab after 24 hours. Pulinx et al [42] reported the death of 2 fetuses (twins). Cooke et al [30] and Fontanella et al [38] observed no adverse neonatal outcomes.

Telemedicine

Search Results and Trial Flow

The literature search yielded 408 citations, of which 348 unique citations were screened based on title and abstract (Multimedia Appendix 3). Then, we screened 14 articles based on the full texts. Finally, we selected 9 appropriate telemedicine-related articles [43-51] in this systematic review. Of these, 6 were RCTs, 2 were CCTs, and 1 was a quasi-experimental design.

In total, we analyzed 480 patients in the intervention groups and 494 in the control groups. The interventions were grouped into web-based and app-based interventions. We did not find any video consultations, and we did not find studies that reported on the outcome of RPG. In total, we analyzed 6 web-based and 3 app-based telemedicine interventions.

Quality Assessment

Furthermore, 4 studies were rated as strong, 4 were rated as moderate and 1 as weak quality studies. The quality appraisals are presented in Multimedia Appendix 4.

Table 1 provides an overview of the characteristics and outcomes of the telemedicine studies. Multimedia Appendix 5 contains a PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) checklist for the systematic review.

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Table 1. Characteristics and outcomes of telemedicine studies.

Author (year), location	Design	Patients, n	Type of technology	Intervention details	Outcomes	*P* value
Homko et al (2007), United States [44]	RCT[a]	• IG[b]: 34 • CG[c]: 29	Web-/internet-based	• Web-based management interactive system: data transmission at least 3 times/week, messaging between health care professionals and patients • Control: usual care + paper logbooks	HbA_{1c}[d] (change) (%) • IG: 6.1 (0.8) • CG: 6.2 (2.2) FBG[e] (change) (mg/dL) • IG: 90.8 (11.8) • CG: 88.6 (9.5)	Not reported
Homko et al (2012), United States [43]	RCT	• IG: 40 • CG: 40	Web-/internet-based	• Web-based management interactive system: interactive voice response telephone communication, auto-reminders • Control: logbook reviewed at clinic visits • Both: clinic visits every 2 weeks and weekly from 36 weeks' gestation	FBG (change) (mg/dL) • IG: 91.5 (10.5) • CG: 94.3 (0.26)	.26
Kim et al (2019), Korea [46]	Quasi-experimental study	• IG: 22 • CG: 22	Web-/internet-based	• Web-based system: weekly web-based health diary, transmitting daily FBG, individual education • Control: received nutrition education	HbA_{1c} (change) (%) • IG: 5.0 (0.2) • CG: 5.3 (0.2) FBG (change) (mg/dL) • IG: 78.8 (8.4) • CG: 80.9 (8.4)	Both not significant (not reported)
Given et al (2015), United Kingdom [45]	RCT	• IG: 24 • CG: 26	Web-/internet-based	• Usual care and telemedicine hub with website, mobile phone and landline, weekly feedback • Control: face-to-face clinic visits every 2 weeks, self-monitoring of blood glucose	HbA_{1c} • IG: 34.04 (3.23) mmol/mol (5.26% increase) • CG: 33.84 (2.88) mmol/mol (5.25% increase)	Not reported
Dalfra et al (2009), Italy [47]	CCT[f]	• IG: 88 • CG: 115	Web-/internet-based	• Transmission of glycemic data through telephone receiver and feedback, monthly clinic visits • Control: not adequately described	HbA_{1c} (change) (%) • IG: 5.1 (0.6) • CG: 5.3 (0.5)	.008
Perez-Ferre et al (2010), Spain [48]	RCT	• IG: 50 • CG: 50	Web-/internet-based	• Internet and app–based via SMS text messaging: transmission of self-monitoring data, SMS text message communication between patients and health care professionals • Control: face-to-face outpatient clinic visits	HbA_{1c} (change) (%) • IG: 5.3 (0.4) • CG: 5.4 (0.4)	Not reported

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Author (year), location	Design	Patients, n	Type of technology	Intervention details	Outcomes	*P* value
Mackillop et al (2014), United Kingdom [50]	RCT	• IG: 101 • CG: 102	App-based	• Mobile phone-based blood glucose management solution: health app (data transmission and feedback at least 3 times per week), clinic visits every 4 to 8 weeks	HbA_{1c} • 0.02% rise per 28 days in the intervention group and 0.03% rise per 28 days in the control group • No statistically significant difference (intervention vs control: –0.01%, 95% CI –0.05 to 0.03)	Not reported
Guo et al (2019), China [49]	RCT	• IG: 64 • CG: 60	App-based	• Usual care + app "Dnurse" for data transmission and feedback, and educational information about gestational diabetes mellitus • Control: standard outpatient treatment	HbA_{1c} (change) (%) • IG: 4.7 (0.2) • CG: 5.3 (0.3) FBG (change) (mg/dL) • IG: 4.9 (0.9) • CG: 5.0 (0.8)	HbA_{1c}: .001 FBG: .74
Yang et al (2018), China [51]	CCT	• IG: 57 • CG: 50	Smartphone/WeChat	• Smartphone-based telemedicine system (WeChat app) and articles providing continuous health education, cloud database • Control: Usual care and health education	FBG (change) (mg/dL) • IG: 4.31 (0.75) • CG: 5.31 (1.29)	<.001

[a]RCT: randomized controlled trial.

[b]IG: intervention group.

[c]CG: control group.

[d]HbA_{1c}: glycated hemoglobin A_{1c}.

[e]FBG: fasting blood glucose.

[f]CCT: controlled clinical trial.

Effects on HbA_{1c} Values

The effects of the interventions based on the comparison of HbA_{1c} levels between the intervention and control groups at the end of the study were analyzed. Dalfra et al [47] (strong quality) and Guo et al [49] (strong quality) indicated clear and significant improvements of HbA_{1c} values (P<.001 [49] and P=.008 [47]). Our findings are based on 5 web-based [44-48] and 2 app-based [49,50] studies. The meta-analysis revealed a mean effect size of –0.138% (95% CI –0.24 to –0.04) for web-based interventions and –0.305% (95% CI –0.88 to 0.27) for app-based interventions.

In general, the meta-analysis demonstrated a mean difference in HbA_{1c} of –0.19% (95% CI 0.34 to 0.03) for all telemedical interventions (n=7). Multimedia Appendix 6 provides the forest plot and the calculations and data for the meta-analysis.

Effects on FBG Values

In total, 2 of 3 studies reported improvements in FBG levels by web-based interventions. Homko et al [43] (moderate quality) outlined enhancements in their intervention group compared to the control group (intervention, 91.5 [10.5] mg/dL, vs control, 94.3 [0.26] mg/dL, P=.26). In addition, Kim et al [46] (weak quality) showed lower FBGs in the intervention group (78.8 [8.4] mg/dL) compared to the control group (80.9 [8.4] mg/dL), but the difference was not significant between groups (P value not reported).

Furthermore, all app-based studies (n=2) reported clear decreases in FBG values by implementing app-based telemedicine interventions. Yang et al [51] (moderate quality) indicated that a smartphone-based telemedicine system (WeChat app) was significantly associated with an obvious improvement in FBG (intervention 4.31% (0.75) versus control 5.31% (1.29), P<.001). Moreover, Guo et al [49] (strong quality) showed improvements compared to the controls (intervention 4.9% (0.9) vs control 5.0% (0.8), P=.74).

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Discussion

Principal Results

This systematic review indicated that telemedicine interventions, especially web-based and app-based treatments, can contribute to favorable impacts on HbA_{1c} and FBG values in the context of the COVID-19 pandemic. Meta-analysis revealed a mean difference in HbA_{1c} of –0.19% (95% CI 0.34 to 0.03) for all telemedical interventions (n=7), –0.138% (95% CI –0.24 to –0.04) for web-based interventions (n=5), and –0.305% (96% CI –0.88 to 0.27) for app-based interventions (n=2).

Digital treatment approaches are an innovative alternative to conventional GDM therapies, particularly due to social distancing, quarantine, and the risk of infection for pregnant women. Interest in telemedicine is increasing because of benefits such as the lack of need for physical presence as well as cost and time benefits [52,53].

Optimal glycemic control through telemedicine can mitigate negative consequences for mother and child. Patients with GDM and COVID-19 need special care. Using the current evidence, we were able to identify and analyze adverse outcomes for mother and child through the interaction of GDM and COVID-19, such as cesarean deliveries, admission to the (neonatal) ICU, ventilation, and low Apgar scores. The long-term consequences for mother and child are currently unknown. Unfortunately, the papers available are often short, are not detailed, and only consist of case reports and letters. Clinical studies with strong methodologies on various outcomes in the context of COVID-19 and GDM are urgently needed to determine the clinical implications. Furthermore, the outcomes were extremely heterogeneous, and quantitative analysis and synthesis were therefore difficult.

As mentioned, early therapeutic strategies are required to improve the management of GDM effectively, because GDM can contribute to long-term consequences for the child by transgenerational programming. Transgenerational programming is a perturbation during development phases that can lead to “programming errors” in organ functions and metabolic regulation. This leads to diseases in later life, such as non–insulin-dependent diabetes, obesity, hypertension, and cardiovascular disease [54]. GDM can contribute to these programming errors and to long-term consequences for the child [54]. GDM occurs over a limited period of time; therefore, little time is available to understand and treat the condition, and effective therapies targeting glycemic and metabolic control are thus necessary [55].

Overall, telemedical approaches clearly improve the glycemic control of women with GDM and thus lead to a positive effect regarding transgenerational programming. Telemedicine effectively enhances the management of GDM in the context of the COVID-19 pandemic. Other reviews by us [24-26] showed that telemedical therapy can improve glycemic control, decrease the number of scheduled and unscheduled hospital visits, and improve several fetal and neonatal short-term outcomes.

With regard to different types of telemedical interventions, we were able to identify web-based and app-based interventions; however, fewer interventions were app-based despite the good operability of smartphone apps. We could not find other technologies, such as videoconferences, in the GDM context, but these technologies show great potential for other types of diabetes.

Limitations

The national guidelines as well as different threshold values for the diagnosis of GDM affect the findings and must be considered. With different definitions of GDM, participants may not be precisely comparable. A uniform global definition of GDM would be necessary to address this issue.

Moreover, because COVID-19 is a rapidly developing topic, the COVID-19 studies are methodologically weak, are not very detailed, and were published quickly to rapidly publish important findings and generate evidence in the context of the new COVID-19 pandemic. This limitation must be taken into account regarding this analysis.

In addition, the maternal and offspring outcomes were extremely heterogeneous. More studies are needed that bring patients with GDM into focus. Larger sample sizes are necessary. In further studies, it is advisable to focus on maternal, fetal, and neonatal outcomes, which are also considered in other GDM studies (when analyzing therapy options for patients with GDM) and are particularly relevant to health, such as cesarean deliveries, admission to the ICU, glycemic control (hyperglycemia, fasting blood glucose, HbA_{1c}, etc), fetal distress, Apgar score, birth weight, and respiratory distress syndrome. With regard to COVID-19–relevant outcomes, our analysis has shown that in particular, COVID-19 symptoms, ventilation, COVID-19 therapy, SARS-CoV-2 testing of newborns, nasopharyngeal swabs, and supplemental oxygen are relevant and need further investigation.

Comparison With Prior Work

To our knowledge, we are the first group to perform a systematic review on GDM, COVID-19, and telemedicine. Other reviews and meta-analyses on GDM and telemedicine reported similar positive results regarding telemedicine care in gestational diabetes management [56,57]. Xie et al [58] showed that, compared to usual care, telemedicine interventions can decrease the glycemic levels of patients with GDM effectively and reduce the risk complications (HbA_{1c}, mean difference −0.70, $P<.01$, and FBG, mean difference –0.52, $P<.01$). However, evidence of clinical effectiveness of telemetric interventions on GDM management is still lacking, particularly regarding app-based and video-based interventions.

Conclusions

This review sets a first milestone and a starting point for further research in managing patients with GDM during the COVID-19 pandemic.

Pregnant women with GDM are at increased risk of a severe course of COVID-19. In the context of the COVID-19 pandemic, a shift to digital therapy approaches, such as telemedicine, is taking place in medical practices. At this point in time, analyses

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and conclusions on adverse outcomes in women with COVID-19 and GDM are scarce; however, we have summarized the first evidence available. Further research is needed to understand the epidemiology and health care interventions needed to most effectively treat women with COVID-19 and GDM. In addition, the transgenerational interplay between COVID-19 and GDM needs further investigation, and evidence-based recommendations are needed.

Telemedicine is a modern and effective approach in the context of COVID-19 and GDM, as it reduces contact and enables patients with GDM to still be optimally treated with regard to adverse pregnancy outcomes. Other telemetric approaches, such as video consultations, need to be investigated more closely, and intervention effects in relation to RPG should also be examined.

Conflicts of Interest

None declared.

Multimedia Appendix 1
Search strategies.
[DOCX File , 24 KB - pediatrics_v4i3e28630_app1.docx]

Multimedia Appendix 2
PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flow charts.
[DOCX File , 69 KB - pediatrics_v4i3e28630_app2.docx]

Multimedia Appendix 3
Overview of COVID-19 studies.
[PDF File (Adobe PDF File), 470 KB - pediatrics_v4i3e28630_app3.pdf]

Multimedia Appendix 4
Quality assessments.
[PDF File (Adobe PDF File), 431 KB - pediatrics_v4i3e28630_app4.pdf]

Multimedia Appendix 5
PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) checklist.
[PDF File (Adobe PDF File), 401 KB - pediatrics_v4i3e28630_app5.pdf]

Multimedia Appendix 6
Forest plot.
[PDF File (Adobe PDF File), 498 KB - pediatrics_v4i3e28630_app6.pdf]

References

1. WHO coronavirus (COVID-19) dashboard. World Health Organization. URL: https://covid19.who.int/ [accessed 2021-07-21]
2. Verity R, Okell LC, Dorigatti I, Winskill P, Whittaker C, Imai N, et al. Estimates of the severity of coronavirus disease 2019: a model-based analysis. Lancet Infect Dis 2020 Jun;20(6):669-677. [doi: 10.1016/S1473-3099(20)30243-7]
3. Kleinwechter H, Laubner K. Coronavirus disease 2019 (COVID-19) and pregnancy: overview and report of the first German case with COVID-19 and gestational diabetes. Diabetologe 2020 Apr 06;16(3):242-246. [doi: 10.1007/s11428-020-00611-0]
4. Bajgain KT, Badal S, Bajgain BB, Santana MJ. Prevalence of comorbidities among individuals with COVID-19: a rapid review of current literature. Am J Infect Control 2021 Feb;49(2):238-246 [FREE Full text] [doi: 10.1016/j.ajic.2020.06.213] [Medline: 32659414]
5. Kleinwechter H. Diabetes and pregnancy—update 2020. Diabetologe 2020 May 22;16(5):470-477. [doi: 10.1007/s11428-020-00629-4]
6. Mimouni F, Lakshminrusimha S, Pearlman SA, Raju T, Gallagher PG, Mendlovic J. Perinatal aspects on the covid-19 pandemic: a practical resource for perinatal-neonatal specialists. J Perinatol 2020 May;40(5):820-826 [FREE Full text] [doi: 10.1038/s41372-020-0665-6] [Medline: 32277162]
7. IDF Diabetes Atlas, 9th edition. International Diabetes Federation. 2019. URL: https://www.diabetesatlas.org/en/resources/ [accessed 2021-07-21]
8. American Diabetes Association. 2. Classification and diagnosis of diabetes: standards of medical care in diabetes–2020. Diabetes Care 2020 Jan;43(Suppl 1):S14-S31. [doi: 10.2337/dc20-S002] [Medline: 31862745]
9. Chiefari E, Arcidiacono B, Foti D, Brunetti A. Gestational diabetes mellitus: an updated overview. J Endocrinol Invest 2017 Sep;40(9):899-909. [doi: 10.1007/s40618-016-0607-5] [Medline: 28283913]

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10. Bellamy L, Casas J, Hingorani AD, Williams D. Type 2 diabetes mellitus after gestational diabetes: a systematic review and meta-analysis. Lancet 2009 May 23;373(9677):1773-1779. [doi: 10.1016/S0140-6736(09)60731-5] [Medline: 19465232]
11. Kozhimannil KB, Pereira MA, Harlow BL. Association between diabetes and perinatal depression among low-income mothers. JAMA 2009 Feb 25;301(8):842-847. [doi: 10.1001/jama.2009.201] [Medline: 19244191]
12. Eberle C, Ament C. Diabetic and metabolic programming: mechanisms altering the intrauterine milieu. ISRN Pediatr 2012;2012:975685 [FREE Full text] [doi: 10.5402/2012/975685] [Medline: 23213562]
13. Eberle C. Fetale Programmierung des Diabetes mellitus Typ 2 : Intrauterine Wachstumsretardierung als Risikofaktor? MMW Fortschr Med 2010 Jan;152(Suppl 3):76-82. [doi: 10.1007/BF03373173] [Medline: 27659957]
14. Eberle C, Merki E, Yamashita T, Johnson S, Armando AM, Quehenberger O, et al. Maternal immunization affects in utero programming of insulin resistance and type 2 diabetes. PLoS One 2012;7(9):e45361 [FREE Full text] [doi: 10.1371/journal.pone.0045361] [Medline: 23028961]
15. Yamashita T, Freigang S, Eberle C, Pattison J, Gupta S, Napoli C, et al. Maternal immunization programs postnatal immune responses and reduces atherosclerosis in offspring. Circ Res 2006 Sep 29;99(7):e51-e64. [doi: 10.1161/01.RES.0000244003.08127.cc] [Medline: 16946133]
16. Eberle C, Stupin J. 5 Perinatale Programmierung. In: Stupin J, Schäfer-Graf U, Hummel M, editors. Praxisorientiertes Wissen zu Gestationsdiabetes, Diabetes mellitus Typ 1 und 2, MODY. Berlin, Germany: De Gruyter; Feb 24, 2020:242.
17. Yang H, Wang C, Poon LC. Novel coronavirus infection and pregnancy. Ultrasound Obstet Gynecol 2020 Apr;55(4):435-437 [FREE Full text] [doi: 10.1002/uog.22006] [Medline: 32134165]
18. Scott ES, Jenkins AJ, Fulcher GR. Challenges of diabetes management during the COVID-19 pandemic. Med J Aust 2020 Jul;213(2):56-57.e1. [doi: 10.5694/mja2.50665] [Medline: 32583422]
19. Ghosh A, Gupta R, Misra A. Telemedicine for diabetes care in India during COVID19 pandemic and national lockdown period: Guidelines for physicians. Diabetes Metab Syndr 2020;14(4):273-276 [FREE Full text] [doi: 10.1016/j.dsx.2020.04.001] [Medline: 32283497]
20. Rocha ADS, Bernardi JR, de Matos S, Kretzer DC, Schöffel AC, Moretti F, et al. Gestational diabetes mellitus screening in pandemic times: Are there viable alternatives? Aust N Z J Obstet Gynaecol 2020 Oct 11;60(5):E14-E15 [FREE Full text] [doi: 10.1111/ajo.13235] [Medline: 33043438]
21. Dodesini AR, Galliani S, Ciriello E, Bellante R, Trevisan R. Pre-gestational diabetes during the COVID-19 pandemic in Bergamo, Italy. Int J Gynaecol Obstet 2020 Nov 08;151(2):295-296. [doi: 10.1002/ijgo.13306] [Medline: 32657430]
22. Nouhjah S, Jahanfar S, Shahbazian H. Temporary changes in clinical guidelines of gestational diabetes screening and management during COVID-19 outbreak: a narrative review. Diabetes Metab Syndr 2020;14(5):939-942 [FREE Full text] [doi: 10.1016/j.dsx.2020.06.030] [Medline: 32593935]
23. Telemedicine: Opportunities and Developments in Member States. Report on the Second Global Survey on eHealth. World Health Organization. 2009. URL: https://apps.who.int/iris/bitstream/handle/10665/44497/9789241564144_eng.pdf?sequence=1&isAllowed=y [accessed 2021-07-21]
24. Eberle C, Löhnert M, Stichling S. Effectiveness of disease-specific mHealth apps in patients with diabetes mellitus: scoping review. JMIR mHealth uHealth 2021 Feb 15;9(2):e23477 [FREE Full text] [doi: 10.2196/23477] [Medline: 33587045]
25. Eberle C, Stichling S. Clinical improvements by telemedicine interventions managing type 1 and type 2 diabetes: systematic meta-review. J Med Internet Res 2021 Feb 19;23(2):e23244 [FREE Full text] [doi: 10.2196/23244] [Medline: 33605889]
26. Eberle C, Stichling S. Effect of telemetric interventions on glycated hemoglobin A1c and management of type 2 diabetes mellitus: systematic meta-review. J Med Internet Res 2021 Feb 17;23(2):e23252 [FREE Full text] [doi: 10.2196/23252] [Medline: 33595447]
27. Cochrane Handbook for Systematic Reviews of Interventions. Cochrane Training. URL: https://training.cochrane.org/handbook [accessed 2021-07-22]
28. Thomas BH, Ciliska D, Dobbins M, Micucci S. A process for systematically reviewing the literature: providing the research evidence for public health nursing interventions. Worldviews Evid Based Nurs 2004;1(3):176-184. [doi: 10.1111/j.1524-475X.2004.04006.x] [Medline: 17163895]
29. Blauvelt C, Chiu C, Donovan A, Prahl M, Shimotake T, George R, et al. Acute respiratory distress syndrome in a preterm pregnant patient with coronavirus disease 2019 (COVID-19). Obstet Gynecol 2020;136(1):46-51. [doi: 10.1097/aog.0000000000003949]
30. Cooke WR, Billett A, Gleeson S, Jacques A, Place K, Siddall J, et al. SARS-CoV-2 infection in very preterm pregnancy: experiences from two cases. Eur J Obstet Gynecol Reprod Biol 2020 Jul;250:259-260 [FREE Full text] [doi: 10.1016/j.ejogrb.2020.05.025] [Medline: 32425297]
31. Vlachodimitropoulou Koumoutsea E, Vivanti AJ, Shehata N, Benachi A, Le Gouez A, Desconclois C, et al. COVID-19 and acute coagulopathy in pregnancy. J Thromb Haemost 2020 Jul;18(7):1648-1652. [doi: 10.1111/jth.14856] [Medline: 32302459]
32. Govind A, Essien S, Karthikeyan A, Fakokunde A, Janga D, Yoong W, et al. Re: Novel Coronavirus COVID-19 in late pregnancy: outcomes of first nine cases in an inner city London hospital. Eur J Obstet Gynecol Reprod Biol 2020 Aug;251:272-274 [FREE Full text] [doi: 10.1016/j.ejogrb.2020.05.004] [Medline: 32402627]

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33. Oliva M, Hsu K, Alsamarai S, Chavez VD, Ferrara L. Clinical improvement of severe COVID-19 pneumonia in a pregnant patient after caesarean delivery. BMJ Case Rep 2020 Jul 16;13(7):e236290 [FREE Full text] [doi: 10.1136/bcr-2020-236290] [Medline: 32675129]
34. Smati S, Mahot P, Bourdiol A, Ploteau S, Hadjadj S, Cariou B. Euglycaemic ketoacidosis during gestational diabetes with concomitant COVID-19 infection. Diabetes Metab 2021 Mar;47(2):101181 [FREE Full text] [doi: 10.1016/j.diabet.2020.07.008] [Medline: 32738403]
35. Uzel K, Lakhno IV. A mortal case of coronavirus disease in a pregnant diabetic woman. Clinical case. Reprod Endocrinology 2020 May 13(52):33-36. [doi: 10.18370/2309-4117.2020.52.33-36]
36. Tang J, Song W, Xu H, Wang N. No evidence for vertical transmission of SARS-CoV-2 in two neonates with mothers infected in the second trimester. Infect Dis (Lond) 2020 Jul 28;52(12):913-916. [doi: 10.1080/23744235.2020.1798499] [Medline: 32721199]
37. D'Ambrosi F, Rossi G, Soldavini CM, Maggi V, Cetera GE, Carbone IF, et al. Management of gestational diabetes in women with a concurrent severe acute respiratory syndrome coronavirus 2 infection, experience of a single center in Northern Italy. Int J Gynaecol Obstet 2021 Mar;152(3):335-338. [doi: 10.1002/ijgo.13434] [Medline: 33099770]
38. Fontanella F, Hannes S, Keating N, Martyn F, Browne I, Briet J, et al. COVID-19 infection during the third trimester of pregnancy: current clinical dilemmas. Eur J Obstet Gynecol Reprod Biol 2020 Aug;251:268-271 [FREE Full text] [doi: 10.1016/j.ejogrb.2020.05.053] [Medline: 32505514]
39. Majachani N, Francois JLM, Fernando AK, Zuberi J. A case of a newborn baby girl infected with SARS-CoV-2 due to transplacental viral transmission. Am J Case Rep 2020 Oct 25;21:e925766 [FREE Full text] [doi: 10.12659/AJCR.925766] [Medline: 33099570]
40. Rabiei M, Soori T, Abiri A, Farsi Z, Shizarpour A, Pirjani R. Maternal and fetal effects of COVID-19 virus on a complicated triplet pregnancy: a case report. J Med Case Rep 2021 Feb 18;15(1):87 [FREE Full text] [doi: 10.1186/s13256-020-02643-y] [Medline: 33602315]
41. Gidlöf S, Savchenko J, Brune T, Josefsson H. COVID-19 in pregnancy with comorbidities: more liberal testing strategy is needed. Acta Obstet Gynecol Scand 2020 Jul;99(7):948-949. [doi: 10.1111/aogs.13862] [Medline: 32249924]
42. Pulinx B, Kieffer D, Michiels I, Petermans S, Strybol D, Delvaux S, et al. Vertical transmission of SARS-CoV-2 infection and preterm birth. Eur J Clin Microbiol Infect Dis 2020 Dec;39(12):2441-2445 [FREE Full text] [doi: 10.1007/s10096-020-03964-y] [Medline: 32661809]
43. Homko CJ, Deeb LC, Rohrbacher K, Mulla W, Mastrogiannis D, Gaughan J, et al. Impact of a telemedicine system with automated reminders on outcomes in women with gestational diabetes mellitus. Diabetes Technol Ther 2012 Jul;14(7):624-629 [FREE Full text] [doi: 10.1089/dia.2012.0010] [Medline: 22512287]
44. Homko CJ, Santamore WP, Whiteman V, Bower M, Berger P, Geifman-Holtzman O, et al. Use of an internet-based telemedicine system to manage underserved women with gestational diabetes mellitus. Diabetes Technol Ther 2007 Jun;9(3):297-306. [doi: 10.1089/dia.2006.0034] [Medline: 17561800]
45. Given JE, Bunting BP, O'Kane MJ, Dunne F, Coates VE. Tele-Mum: a feasibility study for a randomized controlled trial exploring the potential for telemedicine in the diabetes care of those with gestational diabetes. Diabetes Technol Ther 2015 Dec;17(12):880-888. [doi: 10.1089/dia.2015.0147] [Medline: 26394017]
46. Kim Y, Kim H, Kim Y. Effects of a web-based self-management program on the behavior and blood glucose levels of women with gestational diabetes mellitus. Telemed J E Health 2019 May;25(5):407-414. [doi: 10.1089/tmj.2017.0332] [Medline: 30036165]
47. Dalfrà MG, Nicolucci A, Lapolla A, TISG. The effect of telemedicine on outcome and quality of life in pregnant women with diabetes. J Telemed Telecare 2009;15(5):238-242. [doi: 10.1258/jtt.2009.081213] [Medline: 19590029]
48. Pérez-Ferre N, Galindo M, Fernández MD, Velasco V, de la Cruz MJ, Martín P, et al. A Telemedicine system based on Internet and short message service as a new approach in the follow-up of patients with gestational diabetes. Diabetes Res Clin Pract 2010 Feb;87(2):e15-e17. [doi: 10.1016/j.diabres.2009.12.002] [Medline: 20044162]
49. Guo H, Zhang Y, Li P, Zhou P, Chen L, Li S. Evaluating the effects of mobile health intervention on weight management, glycemic control and pregnancy outcomes in patients with gestational diabetes mellitus. J Endocrinol Invest 2019 Jun;42(6):709-714. [doi: 10.1007/s40618-018-0975-0] [Medline: 30406378]
50. Mackillop L, Loerup L, Bartlett K, Farmer A, Gibson OJ, Hirst JE, et al. Development of a real-time smartphone solution for the management of women with or at high risk of gestational diabetes. J Diabetes Sci Technol 2014 Nov;8(6):1105-1114 [FREE Full text] [doi: 10.1177/1932296814542271] [Medline: 25004915]
51. Yang P, Lo W, He Z, Xiao X. Medical nutrition treatment of women with gestational diabetes mellitus by a telemedicine system based on smartphones. J Obstet Gynaecol Res 2018 Jul;44(7):1228-1234. [doi: 10.1111/jog.13669] [Medline: 29797375]
52. Caballero-Ruiz E, García-Sáez G, Rigla M, Villaplana M, Pons B, Hernando ME. A web-based clinical decision support system for gestational diabetes: automatic diet prescription and detection of insulin needs. Int J Med Inform 2017 Jun;102:35-49. [doi: 10.1016/j.ijmedinf.2017.02.014] [Medline: 28495347]

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53. Lemelin A, Paré G, Bernard S, Godbout A. Demonstrated cost-effectiveness of a telehomecare program for gestational diabetes mellitus management. Diabetes Technol Ther 2020 Mar;22(3):195-202. [doi: 10.1089/dia.2019.0259] [Medline: 31603351]
54. Eberle C, Ament C. Diabetic and metabolic programming: mechanisms altering the intrauterine milieu. ISRN Pediatr 2012;2012:975685 [FREE Full text] [doi: 10.5402/2012/975685] [Medline: 23213562]
55. American Diabetes Association. 14. Management of Diabetes in Pregnancy: Standards of Medical Care in Diabetes–2020. Diabetes Care 2020 Jan;43(Suppl 1):S183-S192. [doi: 10.2337/dc20-S014] [Medline: 31862757]
56. Rasekaba TM, Furler J, Blackberry I, Tacey M, Gray K, Lim K. Telemedicine interventions for gestational diabetes mellitus: a systematic review and meta-analysis. Diabetes Res Clin Pract 2015 Oct;110(1):1-9. [doi: 10.1016/j.diabres.2015.07.007] [Medline: 26264410]
57. Ming W, Mackillop LH, Farmer AJ, Loerup L, Bartlett K, Levy JC, et al. Telemedicine technologies for diabetes in pregnancy: a systematic review and meta-analysis. J Med Internet Res 2016 Nov 09;18(11):e290 [FREE Full text] [doi: 10.2196/jmir.6556] [Medline: 27829574]
58. Xie W, Dai P, Qin Y, Wu M, Yang B, Yu X. Effectiveness of telemedicine for pregnant women with gestational diabetes mellitus: an updated meta-analysis of 32 randomized controlled trials with trial sequential analysis. BMC Pregnancy Childbirth 2020 Apr 06;20(1):198 [FREE Full text] [doi: 10.1186/s12884-020-02892-1] [Medline: 32252676]

Abbreviations

CCT: clinical controlled trial
DM: diabetes mellitus
EPHPP: Effective Public Health Practice Project
FBG: fasting blood glucose
FiO2: fraction of inspired oxygen
GDM: gestational diabetes mellitus
HbA_{1c}: glycated hemoglobin A_{1c}
ICU: intensive care unit
MeSH: Medical Subject Headings
NICU: neonatal intensive care unit
PRISMA: Preferred Reporting Items for Systematic Reviews and Meta-Analyses
RCT: randomized controlled trial
RPG: random plasma glucose
SpO_2: oxygen saturation as measured by pulse oximetry

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Original Paper

Analysis of Social Determinants and the Utilization of Pediatric Tele–Urgent Care During the COVID-19 Pandemic: Cross-sectional Study

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Abstract

Background: Telehealth is increasingly used to provide specialty consultations to infants and children receiving care. However, there is uncertainty if the COVID-19 pandemic has influenced the use of telehealth among vulnerable populations.

Objective: This research aims to compare the overall use of tele–urgent care visits for pediatric patients before and after the pandemic, especially among vulnerable populations.

Methods: We conducted a cross-sectional analysis of pediatric tele–urgent care visits at a virtual care center at a southeastern health care center. The main outcome of this study was the use of pediatrics tele–urgent visits across geographical regions with different levels of social disparities and between 2019 and 2020.

Results: Of 584 tele–urgent care visits, 388 (66.4%) visits occurred in 2020 during the pandemic compared to 196 (33.6%) visits in 2019. Among 808 North Carolina zip codes, 181 (22%) consisted of a high concentration of vulnerable populations, where 17.7% (56/317) of the tele–urgent care visits originated from. The majority (215/317, 67.8%) of tele–urgent care visits originated from zip codes with a low concentration of vulnerable populations. There was a significant association between the rate of COVID-19 cases and the concentration level of social factors in a given Zip Code Tabulation Area.

Conclusions: The use of tele–urgent care visits for pediatric care doubled during the COVID-19 pandemic. The majority of the tele–urgent care visits after COVID-19 originated from regions where there is a low presence of vulnerable populations. In addition, our geospatial analysis found that geographic regions with a high concentration of vulnerable populations had a significantly higher rate of COVID-19–confirmed cases and deaths compared to regions with a low concentration of vulnerable populations.

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KEYWORDS

telehealth; pediatrics; social; determinants; COVID-19; use; children; infant; consultation; telemedicine; urgent care; vulnerable population; cross-sectional; minority

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Introduction

Pediatricians have used telehealth to provide a broad range of health care services among primary and specialty care [1-3]. Telehealth is increasingly used to provide specialty consultations to infants and children receiving care [4]. However, since the pandemic, the use of tele–urgent care for pediatrics has grown to include telephone consultations and remote surveillance or to replace in-person primary care visits without sufficient evaluation of these interventions [5-7]. There is limited knowledge on the use of tele–urgent care to respond to urgent care needs in pediatrics during the COVID-19 pandemic.

Social determinants of health are paramount to the use of tele–urgent care because of the widening digital divide that occurs as a result of differences in individual socioeconomic characteristics [8]. Social determinants of health can be defined as the circumstances that impact the health of individuals from birth to death including socioeconomic, educational, and access to health care [9]. Prior to the COVID-19 pandemic, health disparities and inequity demonstrated differences in adoption of tele–urgent care among vulnerable populations [10,11]. Vulnerable populations are defined as populations that are at risk for health access because of economic, ethnic, or health characteristics [12]. In this study, we refer to vulnerable populations based on race, socioeconomic status, and health insurance status. There is uncertainty if the COVID-19 pandemic has influenced the use of tele–urgent care among vulnerable populations. Therefore, the objective of this study was to compare the overall use of tele–urgent care services for pediatric patients before and after the COVID-19 pandemic especially among vulnerable populations.

Methods

Overview

We conducted a cross-sectional analysis of pediatric tele–urgent care visits at a virtual care center at a southeastern health care center. The virtual care center offers on-demand services to all patients older than 2 years regardless of their geographic location or medical affiliation to a health care system. Patients are required to create a profile through the virtual care center web portal and provide personal information such as age, gender, residential address, and insurance coverage. The web portal allows patients to choose from a list of providers based on the patient's chief complaint. In addition, patients can choose between having an on-demand visit or scheduling a visit based on their preference and the availability of the provider. Board-certified physicians are available for on-demand televisits with the ability to prescribe and send medications to the patient's choice of pharmacy.

Data and Materials

The tele–urgent care visit data were received and preprocessed in Excel (Microsoft Corporation). All patients between the ages of 2 and 18 years were included in the analysis. Gender had three categories that were male, female, and nonbinary. Insurance type included the member ID and group ID if the patient provided insurance coverage information. Otherwise, the insurance field was empty, indicating the patient reported no insurance coverage.

Outcomes

The main outcome of this study was the use of pediatrics tele–urgent care visits across geographical regions with different levels of social disparities and between 2019 and 2020.

Data Analysis

Since tele–urgent care participant data were collected and available at the zip level, Zip Code Tabulation Areas (ZCTAs; generalized areal representations of United States Postal Service zip code service areas) were used as the unit of analysis [13]. A variety of detailed demographic data are available at the ZCTA level from the American Community Survey (ACS) [14]. We developed social factors based on previous social determinants of health models [15], including our own model to assess health disparities in the use of tele–urgent care [10]. We collected daily COVID-19 case counts in North Carolina zip codes during the study period to assess if there was a relationship between the prevalence of COVID-19 and the use of tele–urgent care for pediatrics within North Carolina zip codes.

For this research, ZCTA-level social factors data was obtained from the ACS 2014-2018 5-year estimates, the most current 5-year data available from the ACS. We used percentages to account for population density for each social factor. Social factors used in the analyses were percent American Indian or Alaska Native people, percent of Black or African American people, percent in poverty, percent of single female headed households receiving Supplemental Nutrition Assistance Program (SNAP; formerly known as food stamps) with children younger than 18 years, percent of households receiving food stamps or SNAP with a person older than 60 years, total population receiving Medicare, and total population receiving Medicaid. For each variable, a threshold was set to determine if a ZCTA was *at risk* (ZCTAs with a value over the threshold were coded with a 1, while those below were coded with a 0). Scores across all factors were tabulated for all ZCTAs to create an aggregate risk and deprivation score (higher aggregate scores indicate greater risk and deprivation). The coding of ZCTA counts were not mutually exclusive among social factors.

The maps were created using the computed social score for each ZCTA, as well as data on 2019 and 2020 telemedicine visits, and COVID-19 cases, aggregated to ZCTAs. For reference, urban centers throughout the state are labeled on the 2019 visits map. We used dot plot graphs to represent the number of visits coming from zip codes tagged with each of the listed social factors, tagged with no social factors, and the overall visit counts for each year for reference. Factors were sorted by the overall frequency of visits across both years.

Descriptive statistics, visualizations, and statistical tests were all performed in R (R Foundation for Statistical Computing) using ggplot2, version 4.0.2., and the maps were created using ArcGIS Pro 2.6.0 (Esri; July 28, 2020) and Illustrator 2020 24.3.0 (Adobe Inc; August 1, 2020). We primarily analyzed the data through descriptive tables and visualizations.

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Results

Of 584 tele–urgent care visits, 388 (66.4%) visits occurred in 2020 during the pandemic compared to 196 (33.6%) visits in 2019. Over half of the patients were male (112/196, 57%) in 2019, versus in 2020 when over half of the patients were female (202/388, 52%). For both years, the majority of patients reported having health insurance coverage. There was a larger gap between insured and uninsured patients in 2020, such that there were 265 (68%) insured patients and 123 (32%) uninsured patients (Table 1).

Table 1. Patient characteristics of tele–urgent care visits between March 1 and September 30, 2019 and 2020.

Variables	2019 visits (n=196), n (%)	2020 visits (n=388), n (%)	Ratio of increase	Total (N=584), n (%)
Gender				
Male	112 (57.1)	183 (47.2)	1.63	295 (50.5)
Female	84 (42.9)	202 (52.1)	2.4	286 (49.0)
Nonbinary	0 (0.0)	3 (0.8)	3	3 (0.5)
Insurance coverage				
Insured	108 (55.1)	265 (68.3)	2.45	373 (63.9)
Uninsured	88 (44.9)	123 (31.7)	1.39	211 (36.1)

The line graph in Figure 1 shows the tele–urgent care use over time broken up by social flag categories. Most of the groups show a clear increase in visits in 2020 compared to 2019. The pattern of visits over time (a peak in March followed by variable but generally lower visit counts in April through August) were not remarkably different between the time periods.

This line graph shows the percentage of the total visits across the March to August time frame within the overall telemedicine population and broken down into the zip codes with and without social factors identified. The year 2019 shows more variation around the overall trend, reflecting a year not driven by COVID-19 and the smaller overall number of visits.

Visits from 2019 and 2020 were also compared geographically. Figure 2 compares the number of tele–urgent care visits in each year against the social deprivation score for each zip code. Overall, there was an increase in visits in 2020, with much of the increase occurring in the central part of the state (Raleigh–Durham–Cary), an area with high population density with large medical centers. The areas colored in light gray demonstrate the ZCTAs with low social factors, while the areas colored in dark green represent areas with high social factors.

The number of tele–urgent care visits doubled in 2020 (n=317) compared to 2019 (n=155). In addition, the distribution of the visits covered more geographic locations in North Carolina compared to the visits in 2019 that were primarily around the Durham-Raleigh area (Figure 3A and B). The northern and southern part of the North Carolina include zip codes with higher social flag scores, which indicates areas of higher social vulnerability (Figure 3C). The same regions with a high social flag score also experienced high rates of COVID-19–confirmed cases, which presents a challenge to an already vulnerable population (Figure 3D).

Figure 1. Line graph presenting pediatrics tele–urgent care visits falling under each social flag during 2019 and 2020.

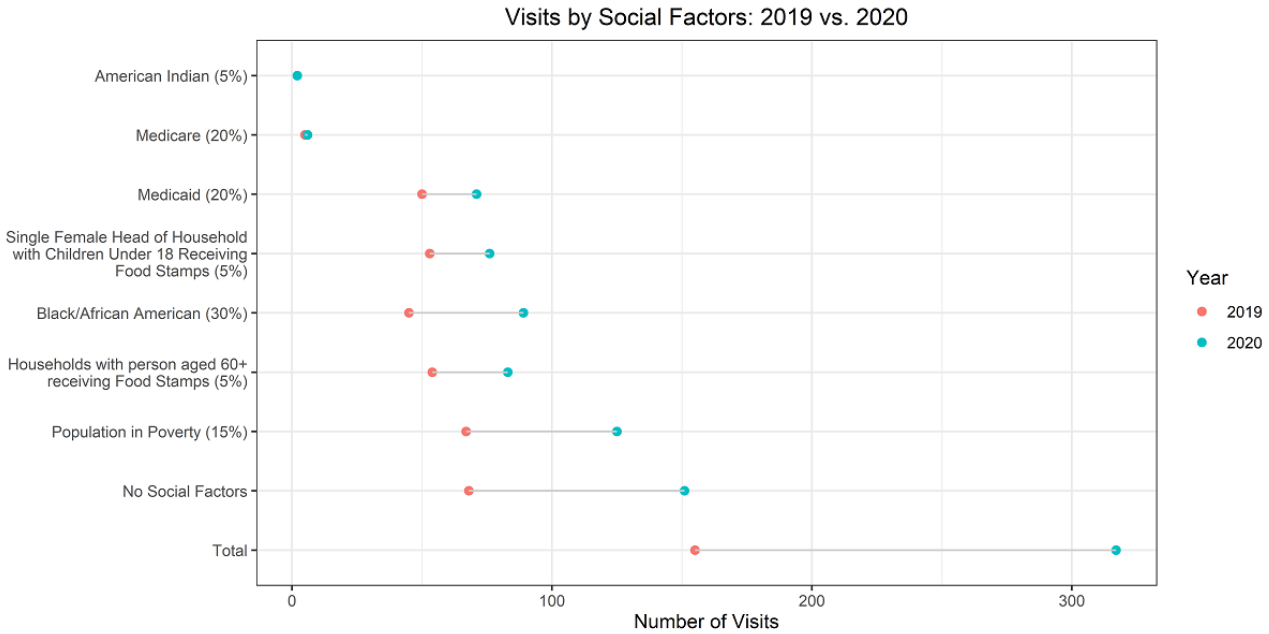

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Figure 2. Bivariate choropleth map comparing the number of visits per 10,000 residents younger than 17 years against social flag scores. Figure produced using ArcGIS Pro 2.6.0 (July 28, 2020) and Illustrator 2020 24.3.0 (August 1, 2020).

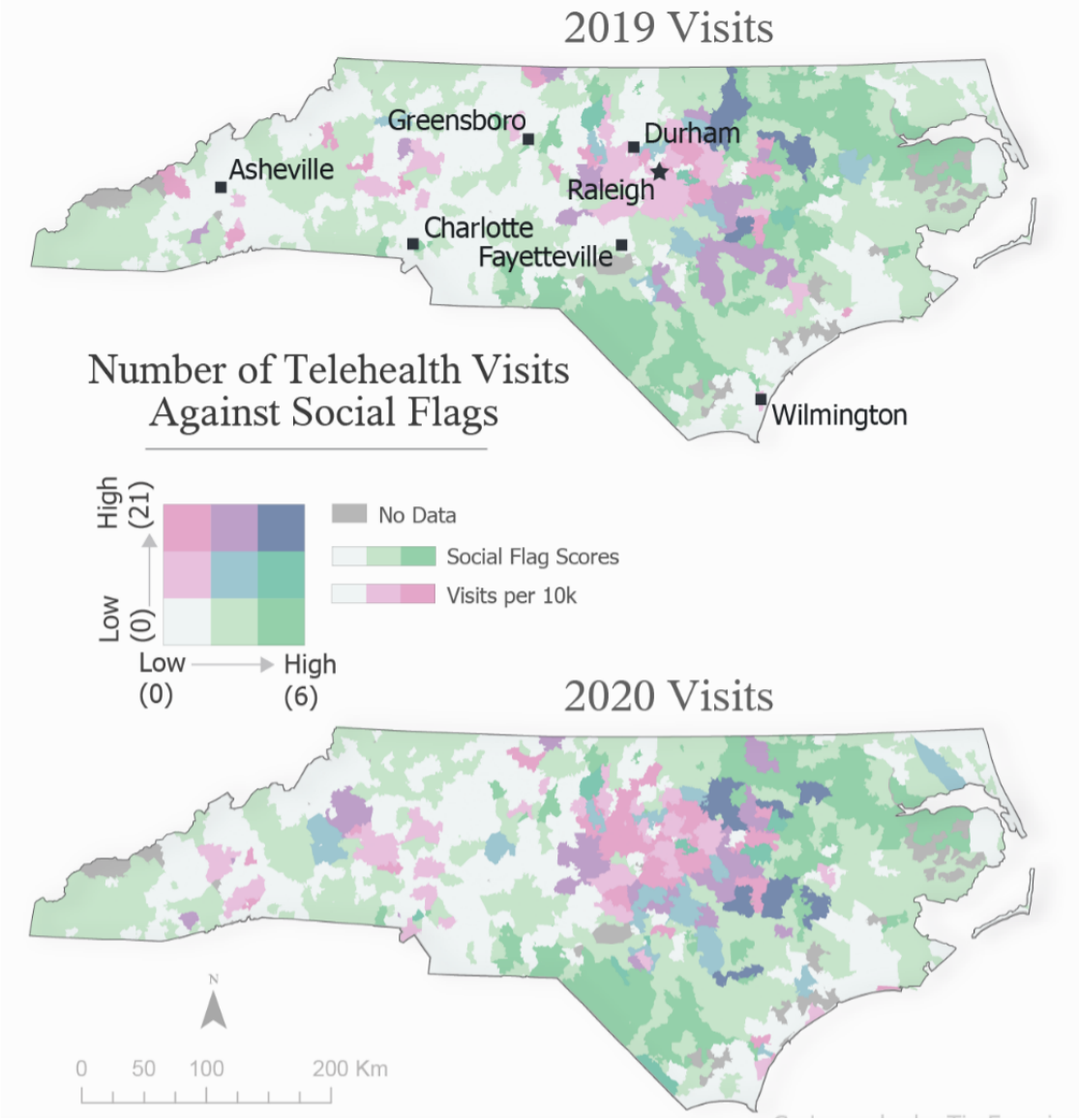

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Figure 3. (A) Map of the tele–urgent care pediatric visits in 2019, (B) the tele–urgent care pediatric visits in 2020, (C) the categorization of North Carolina zip codes based on social vulnerability factors, (D) map of North Carolina with COVID-19–confirmed cases in 2020 by zip code.

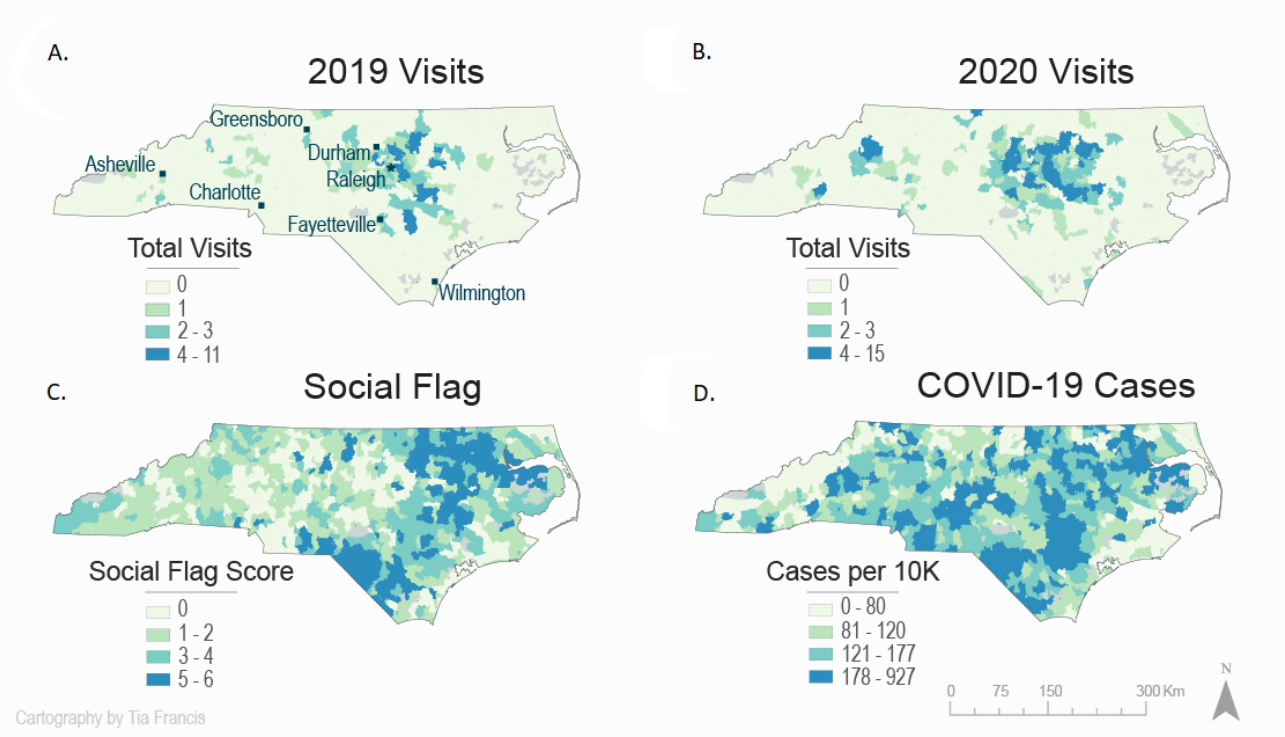

Table 2 shows that relatively more visits came from zip codes with lower social deprivation scores during the COVID-19 time period, even though the raw number of visits from *high* deprivation zip codes did increase. Using a Pearson chi-square test, we rejected the null hypothesis of independence between the COVID-19 time period and deprivation grouping at the 0.05 level with a *P* value of .04. Therefore, there was a significant association between the rate of COVID-19 cases and the concentration level of social factors in a given ZCTA.

Among 808 North Carolina zip codes, 181 (22%) consisted of a high concentration of vulnerable populations, where 17.7% (56/317) of the tele–urgent care visits originated from. The majority (215/317, 67.8%) of tele–urgent care visits originated from zip codes with a low concentration of vulnerable populations. Areas of high concentration of vulnerable populations experienced the highest rates of COVID-19 cases (214.8) and deaths (4.1) per 10,000.

Table 2. The use of tele–urgent care services among low, medium, and high regions of social vulnerability between March 1 and September 30, 2019 and 2020.

Social flag score	Zip codes, n	Telehealth visits Mar-Aug 2019, n (%)	Telehealth visits per 10,000 Mar-Aug 2019, n	Telehealth visits Mar-Aug 2020, n (%)	Telehealth visits per 10,000 Mar-Aug 2020, n	COVID-19–confirmed cases per 10,000, n	COVID-19 deaths per 10,000, n
Low (0-1)	417	93 (60)	0.71	215 (67.8)	1.64	138.2	2.0
Medium (2-3)	210	19 (12.3)	0.4	46 (14.5)	0.98	175.6	2.8
High (4-7)	181	43 (27.7)	0.9	56 (17.7)	1.18	214.8	4.1
Total	808	155	0.69	317	1.4	161.4	2.6

Discussion

Principal Findings

We conducted a cross-sectional study of pediatrics tele–urgent care visits before and after the COVID-19 pandemic among vulnerable populations. We found that the volume of tele–urgent care visits for pediatrics doubled after the pandemic when compared to the year before, which can be explained by the shutdown of health care systems during the initial phases of the COVID-19 pandemic. Post–COVID-19, there was a substantial shift in patient characteristics seeking tele–urgent care for pediatrics such that there were more women and patients with health insurance coverage compared to pre–COVID-19. Most of the postpandemic visits originated from the metropolitan Raleigh region. This could be explained by several reasons including the shutdown of in-person nonessential visits during the initial phases of the pandemic. In addition, there were strict COVID-19 social gathering restrictions, and many schools transitioned to virtual classrooms.

Post–COVID-19, we reported an overall substantial increase in pediatric tele–urgent care visits. Regions characterized with high poverty and high concentration of African American people encountered the highest increase in visits among all examined social factors. The high demand among these communities

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demonstrates high disparities for tele–urgent care use for pediatrics care during the pandemic. We recommend future exploration of current infrastructure and culture to adopting tele–urgent care such that patients living in areas of high poverty may not have access to tele–urgent care equipment or internet access, which may hinder the adoption of tele–urgent care among vulnerable populations. Therefore, offering video and telephone visit options to vulnerable populations may improve adoption and use levels of tele–urgent care.

Telehealth provides a convenient delivery method of health care for pediatric patients; however, the impact of tele–urgent care on patient outcomes remains unknown. We recommend future investigation of the effect of tele–urgent care on emergency department visits and urgent care clinic visits. Moreover, prescription rates were found to be different between video and telephone visits [16]. We recommend more investigations around medication prescription rates in tele–urgent care visits compared to in-person visits for pediatric care.

There appears to be a relationship between social flags and change in tele–urgent care visits between 2019 and 2020. Although a sizeable minority of tele–urgent care visits occurred in regions with high concentrations of vulnerable populations, the majority of tele–urgent care visits occurred in zip codes with low concentrations of vulnerable populations. Post–COVID-19, the volume of tele–urgent care visits in regions with a high concentration of vulnerable populations was less when compared to pre–COVID-19. Although the reason for such a drop in visits is unclear, it is possible that loss of employment and new local tele–urgent care clinics may have attributed to the decrease in visits. In addition, it is possible that during the pandemic newly established tele–urgent care services available through local clinics and primary care providers were preferred by patients.

Post–COVID-19, there were two spikes in the volume of tele–urgent care visits in March and July 2020 among most vulnerable population groups compared to two spikes in 2019 in March and April. The post–COVID-19 increase may be associated with the sudden shutdown of in-person appointments across the state in March 2020 in response to the World Health Organization announcing COVID-19 as a global pandemic [17]. Another explanation to the spikes in the month of March could be associated with the influenza and allergy seasons, which may explain the spike in both 2019 and 2020. In addition, the end of the school year coupled with the national holiday the Fourth of July may have attributed to the increase in visit volume during the month of July. Telehealth can be a suitable intervention to manage chronic care conditions within pediatric patients who may lack access during a pandemic due to the shutdown of schools and clinics [18].

Telehealth use in pediatric care has shown major increase in use during the pandemic. In the future, telehealth may be a suitable health care delivery modality that complements in-office pediatric visits for established patients. Although telehealth use has increased during the pandemic, there remains unanswered questions around the effectiveness of telehealth in pediatric care and the quality of care [19]. We recommend the integration of geospatial technologies to evaluate access factors such as broadband access, clinical effectiveness, medication prescription rates, and the acceptance of telehealth among pediatric patients and providers.

Limitations

This study has several limitations. Although the tele–urgent care data is statewide, it represents a single virtual care center. The patient demographic form did not include ethnic or racial information, which limited our ability to map patient-level ethnic data to zip code–level ethnic data. In the future, patient demographic forms will include ethnic and racial fields. The comparison of 2019 and 2020 data may include confounding factors including the effect of marketing campaigns and word of mouth in increasing the volume of tele–urgent care visits.

Conclusion

The use of tele–urgent care visits for pediatric care doubled during the COVID-19 pandemic. The majority of the tele–urgent care visits after COVID-19 originated from regions where there is a low presence of vulnerable populations. In addition, our geospatial analysis found that geographic regions with a high concentration of vulnerable populations had a significantly higher rate of COVID-19–confirmed cases and deaths compared to regions with a low concentration of vulnerable populations.

Conflicts of Interest

None declared.

References

1. Hakim A, Gaviria-Agudelo C, Edwards K, Olson D, PIDS Telehealth Working Group. Pre-coronavirus disease 2019 telehealth practices among pediatric infectious diseases specialists in the United States. J Pediatric Infect Dis Soc 2021 Apr 30;10(4):485-491 [FREE Full text] [doi: 10.1093/jpids/piaa146] [Medline: 33196843]
2. Gali K, Joshi S, Hueneke S, Katzenbach A, Radecki L, Calabrese T, et al. Barriers, access and management of paediatric epilepsy with telehealth. J Telemed Telecare 2020 Nov 12:1357633X20969531 [FREE Full text] [doi: 10.1177/1357633X20969531] [Medline: 33183129]
3. Allen HI, Gillespie P, Vazquez-Ortiz M, Murphy AW, Moylett EM. A cost-analysis of outpatient paediatric penicillin allergy de-labelling using telemedicine. Clin Exp Allergy 2021 Mar;51(3):495-498. [doi: 10.1111/cea.13782] [Medline: 33170984]
4. Burke BL, Hall RW, Section on Telehealth Care. Telemedicine: pediatric applications. Pediatrics 2015 Jul;136(1):e293-e308 [FREE Full text] [doi: 10.1542/peds.2015-1517] [Medline: 26122813]

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5. Sharawat IK, Panda PK. Caregiver satisfaction and effectiveness of teleconsultation in children and adolescents with migraine during the ongoing COVID-19 pandemic. J Child Neurol 2021 Mar;36(4):296-303. [doi: 10.1177/0883073820968653] [Medline: 33170754]
6. Ozsezen B, Emiralioglu N, Tural D, Sunman B, Buyuksahin H, Yalcin E, et al. Telephone surveillance during 2019 novel coronavirus disease: is it a helpful diagnostic tool for detecting acute pulmonary exacerbations in children with chronic lung disease? J Telemed Telecare 2020 Nov 12:1357633X20972008. [doi: 10.1177/1357633X20972008] [Medline: 33183128]
7. Macy ML, Huetteman P, Kan K. Changes in primary care visits in the 24 weeks after COVID-19 stay-at-home orders relative to the comparable time period in 2019 in metropolitan Chicago and northern Illinois. J Prim Care Community Health 2020;11:2150132720969557 [FREE Full text] [doi: 10.1177/2150132720969557] [Medline: 33174495]
8. Marcin JP, Shaikh U, Steinhorn RH. Addressing health disparities in rural communities using telehealth. Pediatr Res 2016 Jan;79(1-2):169-176. [doi: 10.1038/pr.2015.192] [Medline: 26466080]
9. Braveman P, Gottlieb L. The social determinants of health: it's time to consider the causes of the causes. Public Health Rep 2014;129 Suppl 2:19-31 [FREE Full text] [doi: 10.1177/00333549141291S206] [Medline: 24385661]
10. Khairat S, Haithcoat T, Liu S, Zaman T, Edson B, Gianforcaro R, et al. Advancing health equity and access using telemedicine: a geospatial assessment. J Am Med Inform Assoc 2019 Aug 01;26(8-9):796-805 [FREE Full text] [doi: 10.1093/jamia/ocz108] [Medline: 31340022]
11. Hamad J, Fox A, Kammire MS, Hollis AN, Khairat S. Evaluating the experiences of new and existing teledermatology patients during the COVID-19 pandemic: cross-sectional survey study. JMIR Dermatol 2021;4(1):e25999 [FREE Full text] [doi: 10.2196/25999] [Medline: 34028471]
12. Waisel D. Vulnerable populations in healthcare. Curr Opin Anaesthesiol 2013 Apr;26(2):186-192. [doi: 10.1097/ACO.0b013e32835e8c17] [Medline: 23385323]
13. ZIP Code Tabulation Areas (ZCTAs). United States Census Bureau. 2020. URL: https://www.census.gov/programs-surveys/geography/guidance/geo-areas/zctas.html [accessed 2020-11-28]
14. About the American Community Survey. United States Census Bureau. 2020. URL: https://www.census.gov/programs-surveys/acs/about.html [accessed 2020-08-18]
15. Kolak M, Bhatt J, Park YH, Padrón NA, Molefe A. Quantification of neighborhood-level social determinants of health in the continental United States. JAMA Netw Open 2020 Jan 03;3(1):e1919928 [FREE Full text] [doi: 10.1001/jamanetworkopen.2019.19928] [Medline: 31995211]
16. Khairat S, Liu S, Zaman T, Edson B, Gianforcaro R. Factors determining patients' choice between mobile health and telemedicine: predictive analytics assessment. JMIR Mhealth Uhealth 2019 Jun 08;7(6):e13772 [FREE Full text] [doi: 10.2196/13772] [Medline: 31199332]
17. Khairat S, Meng C, Xu Y, Edson B, Gianforcaro R. Interpreting COVID-19 and virtual care trends: cohort study. JMIR Public Health Surveill 2020 Apr 15;6(2):e18811 [FREE Full text] [doi: 10.2196/18811] [Medline: 32252023]
18. Serlachius A, Badawy SM, Thabrew H. Psychosocial challenges and opportunities for youth with chronic health conditions during the COVID-19 pandemic. JMIR Pediatr Parent 2020 Oct 12;3(2):e23057 [FREE Full text] [doi: 10.2196/23057] [Medline: 33001834]
19. Badawy SM, Radovic A. Digital approaches to remote pediatric health care delivery during the COVID-19 pandemic: existing evidence and a call for further research. JMIR Pediatr Parent 2020 Jun 25;3(1):e20049 [FREE Full text] [doi: 10.2196/20049] [Medline: 32540841]

Abbreviations

ACS: American Community Survey
SNAP: Supplemental Nutrition Assistance Program
ZCTA: Zip Code Tabulation Area

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Original Paper

Exploring Mothers' Experience of a Linguistic Feedback Technology for Children at Risk of Poor Language Development: Qualitative Pilot Study

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Abstract

Background: The early language environment is important for language development and a child's life-course trajectory. Risk factors associated with poor language development outcomes in children include maternal anxiety and depression, low educational attainment, substance misuse, and low socioeconomic status. Language Environment Analysis (LENA) is a wearable technology designed to promote caregivers' engagement in supporting their children's language development. LENA provides quantitative linguistic feedback, which has been shown to improve caregiver language output, thus enhancing a child's language environment. There is limited research on the uptake of this technology by families with developmentally at-risk children.

Objective: This qualitative study aims to explore the conditions under which mothers with children at risk of poor developmental outcomes are willing to adopt the use of LENA to monitor and improve caregiver language output.

Methods: Using a qualitative interpretive design, semistructured, in-depth interviews were conducted with 8 mothers. Participants were recruited purposively to select the maximal variation of socioeconomic and ethnodemographic backgrounds. The transcribed interview data were analyzed thematically and interpretatively. Themes were mapped abductively to an extended Unified Theory of Acceptance and Use of Technology, which included contextual factors for LENA acceptance.

Results: Factors that influenced the intention to use LENA included both technology-specific acceptance factors and contextual factors. Technology acceptance themes included reassurance, feeling overwhelmed, and trust. These themes were mapped to performance expectancy, effort expectancy, and social influence. Contextual themes included emergent success and the intrusion of past difficulties. These were mapped to parenting self-efficacy and perceived risk. The theme of building on success described behavioral intention. Mothers were more likely to adopt LENA when the technology was viewed as acceptable, and this was influenced by parenting self-efficacy and perceived risk.

Conclusions: LENA is a technology that is acceptable to mothers with children who are at risk of poor language development outcomes. Further studies are needed to establish LENA's effectiveness as an adjunct to strategies to enrich a child's early language environment.

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KEYWORDS

language development; technology; feedback; socioeconomic factors

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Introduction

Background

Given the importance of language skills at school entry, successful language development in early childhood is foundational. Parental engagement in language activities has been shown to improve language development and optimize school readiness [1]. The communication environment is reported to be a more dominant predictor of early language than social background [2]. Hoff [3] found that the effect of socioeconomic status (SES) on language development was fully mediated by maternal speech: "[The] critical role of early home language environment as a root cause of academic achievement disparity between children from low and high socioeconomic status homes is undeniable." A rich language environment is considered foundational to early language development and for future academic performance [2,4,5].

Local research has shown that 1 in 5 Australian children start school behind and are not ready to take advantage of learning opportunities at school [6]. There is a snowball effect, with children beginning school with delayed language being at greater risk of school failure [7]. Early language is an important precursor for development and is linked to literacy, cognitive and educational outcomes, and economic opportunities [7,8]. In particular, early interventions aimed at enriching home language environments and targeting early childhood development are argued to reduce social and health inequities across the life course [9,10] and ameliorate cumulative disadvantage [11].

Multiple factors have been implicated in language development. In particular, barriers to parent-child interaction negatively affect child language development [12]. These risk factors include social disadvantage, parental mental health, and maternal drug use [7,13-18].

The correlation between low SES (as defined by income level and level of education) and poor outcomes has been linked to the quantity and quality of language that a child is exposed to in their home environment [11,19,20]. SES is also a predictor of children's school readiness and future academic achievement [3,21,22]. Mothers with low SES backgrounds are more likely to use more directives, use fewer open-ended questions, speak in less complex sentences, and produce less speech and gesture. Children from low SES homes often start kindergarten with lower language and literacy skills than those from high SES homes [23,24]. This disparity has been shown to persist throughout school years and is predictive of lower high school graduation rates and economic opportunities [25].

Parental mental health affects the interaction between children and their caregivers. Maternal depression has a negative influence on children's language development [26,27]. A mother's engagement in their child's learning can be undermined by maternal depression, as mothers experiencing more stress and depression speak less to their children [13,28]. Depressed mothers are less sensitively attuned to their children and tend to be less emotionally responsive and less contingent [12,29]. Maternal depression and anxiety are independently associated with language delay [15]. Maternal psychological distress during the perinatal period is negatively associated with language outcomes [10]. Paternal depression has been linked to expressive vocabulary development through reduced parent-to-child reading [30].

Children who are exposed to continuing parental drug use are at risk of developmental delay [17]. Drug abuse is characterized as a chronic relapsing disease [31]. Maternal drug use affects mother-child behavior and the ability to provide a consistent and nurturing environment [32]. A review of studies has shown that prenatal exposure to cocaine is associated with language deficits [18].

A rich language environment, particularly through child-directed speech and adult-child conversations, is positively associated with language development. Infants with more child-directed speech show faster language processing speed and larger vocabularies [33,34]. Conversational turns between children and adults are essential for language development [35].

One approach to supporting early language is through a parent intervention targeted at enriching the home language environment by increasing child-directed speech and contingent responses [14]. Language interventions are often directed at parents as the primary contributors to their children's language environment. A meta-analysis of 18 studies showed that parent-implemented language interventions can improve language skills in children aged between 18 and 60 months [36]. The most common focus of these interventions was directed at "what the parents say and how much the parents say" [36]. However, adults are generally rarely conscious of the quantity of talk provided to children and frequently overestimate the amount they talk with their children [37]. Studies have demonstrated that caregiver language output can be increased by providing feedback on the linguistic environment, thus enhancing the child's language environment [37,38]. Therefore, having a tool to measure the linguistic environment and providing feedback over time may complement and enhance parent-implemented language interventions and support caregivers in improving the home language environment.

Language Environment Analysis

Language Environment Analysis (LENA) provides quantitative feedback on the home language environment and has been likened to a "linguistic pedometer" [37]. LENA is a wearable, digital recording device and software package. The small recording device is worn in the front pocket of specially designed clothing. The recordings are then processed by the LENA software to generate frequency counts of the number of words a child is exposed to, the number of vocalizations the child produces, and the number of conversational turns the child takes with an adult for up to 16 hours. LENA also produces data about the environment, including electronic media, distant speech, background noise, and silence. The LENA-generated feedback can be used to identify patterns in communication that can be discussed during feedback sessions. Total daily counts can be used to monitor progress and set goals much like a pedometer encourages the user to increase their step count. With the clinician, LENA feedback can be used as a basis for discussing ways to improve the language environment. As the

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software algorithm identifies speech sounds, LENA can be used with non–English-speaking families. LENA has been validated in several languages, including Mandarin, Swedish, Spanish, and French [33,39-41]. Like a pedometer, LENA assists parents to see the language environment they provide for their child, understand their role in this, and reinforce positive behavioral change.

LENA has been shown to influence adult language behavior and thus improve a child's home language environment. Intervention, including weekly feedback with LENA results, showed a significant and prolonged increase in caregiver language output [37]. This leads to improvements in a child's language environment. A randomized controlled pilot study for families with low SES using LENA in combination with an intervention curriculum showed significant but short-term increases in both parent language interactions with their children and child language outcomes. These outcomes included increases in daily adult word count, conversational turn count, and number of child vocalizations [42]. Internationally, other parent-led language intervention programs based on LENA in the United States include the Thirty Million Words Initiative [43] and Providence Talks [44] and Talking Matters in New Zealand [45].

Unified Theory of Acceptance and Use of Technology

Successful implementation of LENA technology is determined by its acceptability to parents with children who are developmentally at risk. Unified theory of acceptance and use of technology (UTAUT) is a simplified theory that explains user acceptance and the use of technology. In this model, technology acceptance predicts its use [46]. Behavioral intention or acceptability is predicted by three antecedents: (1) performance expectancy, which is "the degree to which an individual believes that the system helps improve job performance," (2) effort expectancy, which is the system's ease of use, and (3) social influence, which is the "degree to which an individual perceives that important others believe he or she should use the new system" [47]. Various extensions have been made to UTAUT, including the addition of contextual factors [48]. Important contextual factors specific to LENA use by mothers with children who are at risk of poor language development outcomes include perceived risk and parenting self-efficacy. Perceived risk with technology increasingly includes concerns about privacy problems [49]. LENA is a recording device that carries an associated privacy risk. Second, parenting self-efficacy is commonly considered in parenting intervention programs [50]. Parenting self-efficacy refers to parents' assessment of their ability or effectiveness to successfully perform the parenting role and is a social learning theory component [51]. Task-specific parenting self-efficacy refers to a single parenting domain, such as, in this instance, communication [52].

There is no published information on the acceptability of LENA for Australian families whose children are at risk of poor language outcomes. This is in comparison with the United States, where LENA is used more extensively in trials for families living in areas of disadvantage [42,53]. Understanding the factors that influence the acceptability of LENA for children with developmental risks will help improve technology uptake.

Methods

Recruitment

Participants were identified as mothers with a child aged between 1 and 30 months, with at least 1 risk factor for poor child developmental outcomes. These included a history of or current mental health disorders or problems, major stressors in the perinatal period, substance misuse, government benefit, and education less than Year 12 level (Table 1).

Table 1. Risk factor distribution (n=8).

Number of risk factors[a]	Participants, n (%)
1	2 (25)
2	1 (13)
3	3 (38)
4	2 (25)

[a]Risk factors defined as current or history of maternal mental health problems, perinatal major stressors, substance misuse, maternal education less than Year 12, and low household income.

Participants were recruited purposively using maximal variation sampling to ensure a mix of socioeconomic and ethnodemographic backgrounds. See *Participant Characteristics* below that further describes this broad distribution. Mothers were recruited widely from those attending a variety of child and family health services for vulnerable families. These included parenting groups run by drug and alcohol services, child and family nursing home visiting programs, and community pediatrics clinics for children at risk. A participant information sheet describing the study was given to the coordinator of each of these services, outlining the study and eligibility criteria. All mothers were invited through a third person, such as the coordinator of a playgroup, to prevent coercion. All mothers were provided with a participant information sheet that included the requirements and purpose of the study. Participation was voluntary. There was no relationship between the researcher and recruited mothers.

Each participant was asked to make 2 full-day recordings (16 hours each) and to keep a record of the day's activities to support feedback. After each recording, the mothers were provided with an individual LENA linguistic feedback report. The researcher assisted the mothers' understanding of the results and supported goal setting for the second recording. LENA feedback reports

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included graphs of hourly word counts, including adult words, child vocalizations, conversational turns, and representation of the audio environment. A summary of the total daily word counts comparing the first and second recordings was also provided after the second recording. Percentile ranks were not provided, as Australian norms were not available. The discussion of the results formed the basis of the postrecording interviews. A total of 8 mothers were recruited, of which 6 were able to complete 2 recordings. One mother withdrew from the study because of relapse of a mental health condition. Another mother was unable to complete the recording as the clothing was not large enough for her child.

Participant Characteristics

The 8 participating mothers were aged between 24 and 40 years (mean age 33.4 years). Of these 8 mothers, 2 (25%) were of non–English-speaking backgrounds, 1 (13%) had an Aboriginal background, 5 (63%) reported having either nil income or received government benefits, 2 (25%) reported a household income between Aus $1000 (US $734.50) and Aus $2000 (US $1469) per week, and 1 (13%) reported a household income of greater than Aus $2000 (US $1469) per week. Furthermore, 50% (4/8) of the mothers had an education level less than Year 12, 38% (3/8) completed high school or a diploma, 13% (1/8) had a bachelor's degree, and 75% (6/8) of mothers reported a history of or current mental health problems. The number of risk factors for each mother is documented in Table 1. Participating children's ages were relatively evenly distributed in 6-month bands from 0 to 30 months (mean age 14 months). There was an even distribution of child gender. Of the 8 children, 5 (63%) lived with 2 adult parents at home. One child also had grandparents involved in their care, but they did not live in the same household.

Data Collection

A basic qualitative interpretive design was chosen to understand the mothers' perceptions of LENA. In-depth interviews were chosen to explore individual perceptions of the use of the device. The researcher sought to understand how mothers interpret their experience of using LENA and understand the characteristics and patterns of mothers' use of LENA. In particular, we gathered insights from mothers with identified risk factors for developmental vulnerability and poor child language outcomes. Interviews were semistructured and conducted face to face before, during, and after the use of LENA to assess acceptability from different temporal perspectives. An interview guide was also used. The interview guide covers three main areas. These were mothers' impressions of LENA, difficulties encountered with recording, and likelihood of using LENA in the long term. In-depth interviews were conducted by the researcher at the location of the participant's choice. Often, this was at the play group or community health center; sometimes, it was in their home. Each in-depth interview lasted between half an hour and 1 hour, and each mother was interviewed on 4 separate occasions: before the first recording, after the first recording, before the second recording, and after the second recording. As each participant was interviewed multiple times, data saturation for each participant was achieved. Where English was not the participant's main language, interviews were conducted with the help of an interpreter. Field notes were also recorded.

Audio-recorded interviews were transcribed unless the participant disagreed with the recording. One mother disagreed with the interview recordings. Notes were instead handwritten during these interviews. Participants were allocated codes and names deidentified in the transcript to maintain their anonymity. Observational notes were also recorded in a journal.

Data Analysis

Overview

Data were analyzed using thematic analysis and constant comparison. Transcripts were manually coded using an inductive approach. Each transcript was analyzed individually to identify the thematic statements. Data were analyzed by the first author and reviewed by the second author. As themes emerged, these were checked against the original transcripts and previous codes. Emerging themes were explored by the participants during sequential interviews. Successive readings led to the emergence of more focused themes. Thematic statements were clustered and checked against the original transcripts. Each cluster was analyzed to determine its essential meaning. From these clusters, major themes were generated that incorporated themes for all participants. Field notes contained a critical examination of ideas that emerged as the research progressed and the researcher's reflections and insights. A parallel literature review of technology acceptance identified extended models of UTAUT as helpful to understanding factors influencing the likelihood of technology use. Themes generated inductively were mapped to an extended UTAUT model using an abductive approach.

Rigor

Transcripts and their recordings were reviewed to ensure the accuracy of the written text used in the analysis. To examine internal coherence, emergent themes were compared for consistency with the data. Two researchers coded portions of the same transcript to establish intercoder reliability. The differences and similarities in codes were then discussed. Themes were discussed by the first author and second author and reviewed with an expert mentor. An audit trail was maintained, which consisted of a diary to record the researcher's theoretical choices, decision-making, and conclusions drawn.

Reflexivity

The researcher has a medical background with an interest in child development. LENA appears to be a helpful intervention for stimulating child-directed language in the home environment. LENA has been used in populations in the United States, with children who are at risk for poor developmental outcomes with positive short-term outcomes. No such published research has been conducted in Australia. The researcher is interested in identifying the conditions under which mothers with children who are at risk of poor developmental outcomes are likely to adopt LENA. This will help in identifying future caregivers who are willing to draw on the capacities of LENA to improve caregiver language output and enhance their child's language environment.

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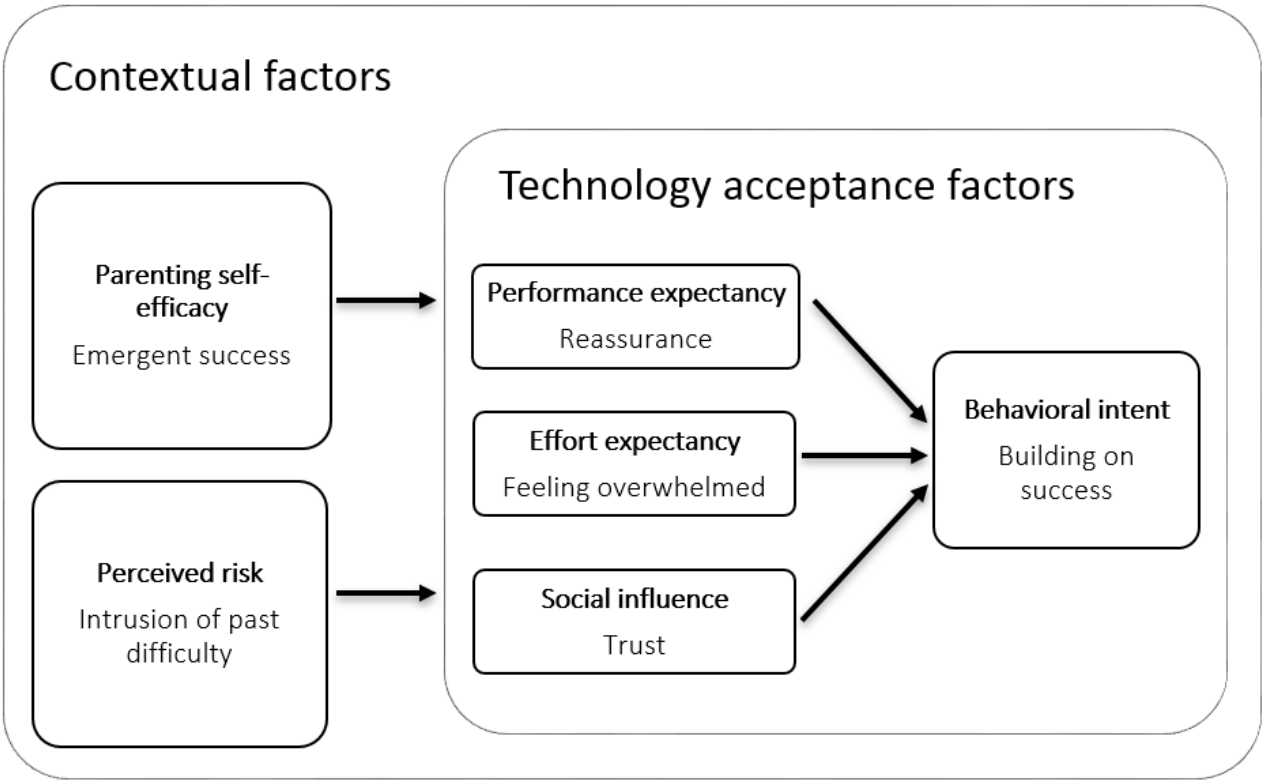
Results

Overview

Thematic analysis revealed technology-related and context-related themes that impact mothers' acceptance of LENA. Figure 1 shows these themes within an extended UTAUT model. Within technology acceptance factors, themes of reassurance, feeling overwhelmed, and trust mapped to performance expectancy, effort expectancy, and social influence. Within contextual factors, themes of emergent success and intrusion of past difficulty were mapped to parenting self-efficacy and perceived risk. The theme of building on success best described the behavioral intention or acceptability of LENA.

Figure 1. An extended unified theory of acceptance and use of technology model for Language Environment Analysis acceptability.



Contextual Factor Themes

Emergent Success and Parenting Self-Efficacy

The theme of emergent success was mapped to parenting self-efficacy. Parenting self-efficacy describes a parent's assessment of their ability or effectiveness in successfully performing the parenting role. Those mothers who demonstrated emerging confidence in their parenting were interested in exploring how LENA could improve their communication with their child. They had a sense of positivity and demonstrated building upon previous success:

> *I think when you are feeling good about yourself, you, you manage to sort of come from rock bottom. You are not as vulnerable. Basically, you've got your confidence back, you feel confident that you know you've already done some good things in getting, you know, communication with the child back and you are building upon that. You are feeling positive and you've got that momentum going forward in the direction that you want. You've turned it around and now you feel like, "Okay well now I'm doing so well, I can, let's capitalise on this and do it [LENA]."* [P5]

Mothers with a background of mental health issues, particularly anxiety and depression, experienced periods of low parenting self-efficacy. Being recorded was associated with the impression of being judged. Some mothers expressed anticipatory concern that feedback could be critical and reveal flaws in parent-child interactions:

> *Because in my headspace I thought I was doing everything wrong and I [would have] thought, "Oh, this is just another way for someone to find out what I'm doing wrong."* [P5]

One mother expressed confidence in her parenting self-efficacy based on previous experience. She felt that she was able to assess the development of her child better without the need for technology:

> *My child's condition, I can see it. He is making his own progress. I don't know if it [LENA] helps. I have two other children and from previous experience I*

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never needed it [LENA]...I don't need the information to monitor his progress. [P3]

Intrusion of Past Difficulty and Perceived Risk

The theme of intrusion of past difficulty was mapped to perceived risk. Perceived risk with technology commonly includes concerns about privacy problems [49]. Past difficulties impacted mothers' experience of using LENA. Some mothers described issues involving child protection agencies, incarceration, mental health difficulties, and drug rehabilitation. There was a perceived risk associated with the recording.

For 2 mothers, the recording device carried a stigma of being spied on, and concerns were expressed about the loss of privacy and tampering with recorded information. The recording device was sometimes considered suspicious by others. Concerns relating to child protection and domestic violence were also raised:

But – "Oh, watch out! She's wired!" – muck around...I tried to point it [LENA] out...but none of them would listen, so I just thought pfft. If you's are so weird about me recording her or her recording me and her, and then yeah. And they didn't answer, so I just walked out. [P2]

Technology Acceptance Factors

Reassurance and Performance Expectancy

The theme of reassurance was mapped to performance expectancy. Performance expectancy describes the perceived usefulness of technology [47]. All mothers, except one, expressed anxiety about their communication ability. Commonly, these mothers were interested in receiving feedback on their interaction with their child and sought reassurance about the efficacy of their communication with their child:

Um, I'm just hoping to see how much I can improve to help [child's name], like I'd like to know, cause I can't really, sounds silly, but I can't really hear myself, I don't know. You know, I want to know how much I'm putting into making him speak, so whatever I could improve to help [child's name], that's what I want to do. [P7]

Was I talking enough? [P2]

Mothers were often concerned about their child's development and perceived their child to be at risk of poor language development. This could be in the context of the child's pre-existing medical problems, such as neonatal withdrawal, behavioral difficulties in the child, or a family history of developmental issues such as language delay. These mothers felt that LENA was useful in improving their child's communication. However, for 1 family, the experience of LENA represented a deficit model, which was incompatible with their strong focus on positive health and well-being. The LENA system was seen to focus on potential issues:

I don't want to predict [language difficulties]. I don't want that trouble...My husband asked me, 'Why worry? His problems have all gone now and the doctor said he will be normal.' He does not think he needs the recording, he is not concerned and believes he will pick up [language] as he grows up. [P3]

Feeling Overwhelmed and Effort Expectancy

The theme of feeling overwhelmed was mapped to effort expectancy. Effort expectancy describes the ease of use of technology [47]. Not all mothers were in a position to be able to take on the additional demands required to participate. For example, 1 mother experienced a relapse and withdrew from the study. Others expressed interest but declined because of other commitments. Rescheduling appointments was common where recordings were not completed before the next review.

On the recording day, some mothers found it difficult to balance competing demands. These demands were not always avoidable or predictable. These included teething, illness, and unsettled behavior in children. Sometimes, the needs of a sibling led to reduced interaction with the child being recorded: "We weren't having a good day, he was in such a state and I know, there's no point putting it [LENA] on" [P1].

Trust and Social Influence

The theme of trust was mapped to social influence. Social influence is the "degree to which an individual perceives that important others believe he or she should use the new system" [48]. Mothers experienced various difficulties, including mental health issues, addiction, child medical health problems, and relational breakdown. Some mothers reflected on parenting issues such as being unavailable to their children both physically and emotionally, uncertainty in how to parent, and not identifying medical and developmental issues early enough.

Despite these difficulties, several mothers described a continuing process of coming out of these difficulties, often through the help of a caseworker or health professional. Building upon a trusted professional relationship was important to 4 of the mothers. The caseworker or trusted professional was seen as a bridge to the introduction of LENA:

Because I know that umm [caseworker's] interested in [child's] progress and my progress, umm and would only recommend something that was beneficial to both of us. Umm whereas if anyone else sort of, you'd be like, "Oh." And because she knew by being in the home...what was going on specifically, so she thought that potentially that [LENA] could be umm helpful, but yeah with someone else, you'd just be like, "Oh okay." [P5]

Participants were more likely to trust and use the technology when they were positively influenced by a trusted professional:

If it wasn't safe my caseworker wouldn't let me do it, that's why I give everything to her, run it by her first...she knows what's happening and if it's good for me and baby. [P2]

Building on Success and Behavioral Intention

The theme of building on success was mapped to behavioral intention. Behavioral intention has been defined as "the degree to which a person has formulated conscious plans regarding whether to perform a specified future behavior" [49]. Positive

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results were comforting for some mothers who regarded their results with pride and satisfaction. Several mothers valued results as a validation of their self-efficacy:

> *I like the fact that I know that she's communicating back and I'm communicating to her and it just reassures me that I'm doing the right thing, yeah, so it's good to know...It makes me feel good and yeah, happy that I'm doing something right.* [P4]

Success in achieving a good word count motivated some mothers to do more with their children: "So, seeing the results would probably make me want to do it more and more to see if I can get it up to that level" [P2]. However, not all parents found the goal of a word count as motivational:

> *See, I'm still bogged down in all of that sort of stuff rather than, "Oh yay, I haven't got my word count." I couldn't care less if I got the ** word count in.* [P5]

Discussion

Principal Findings

This is the first Australian study to describe the factors that influence the acceptance of LENA for mothers with children at risk of experiencing delayed language development. The results of this study are relevant to understanding the acceptability of the LENA system for developmentally at-risk children in Australia. The findings may be used to develop interventions and foster conditions that aim to improve early language development. The findings of this study can help increase the likelihood that mothers with at-risk children will use LENA to improve their language output and enhance their children's language environment.

The UTAUT model is a well-established, statistically validated model that describes the conditions that influence behavioral intention and use of technology [48]. This study developed an extended UTAUT model to represent technology-specific and context-specific factors that influence the acceptance of LENA by mothers with children at risk of poor developmental outcomes. Contextual factors were as important as specific technology factors for acceptance of LENA. A mother's experience of using LENA was highly influenced by her individual circumstances. These contextual factors were represented by themes of emerging success and the intrusion of past difficulty. Emerging success was considered to be encompassed by the condition of parenting self-efficacy. Parenting self-efficacy was interwoven with the acceptability of LENA and influenced the appraisal of the technology. Mothers with emerging success found satisfaction with their results. These mothers had some anxiety about their communication self-efficacy and expected LENA to help them improve their communication with their children. However, very low or high parenting self-efficacy appeared to negatively influence the performance expectation of the technology and reduced the likelihood of LENA acceptance. For one mother, the condition of low parenting self-efficacy led to the impression that LENA was an additional burden. High parenting self-efficacy was also negatively correlated with acceptability. For 1 mother who felt confident in her parenting ability, LENA was not expected to provide any beneficial new information, and she expressed low interest in ongoing use.

This study also revealed other contextual factors that hindered acceptability. The intrusion of past difficulty was mapped to perceived risk. These were seen as negative past experiences for mothers, which led to the perception that LENA was being used to monitor the mother rather than help the child. This is consistent with the research by Allen et al [54], that identified issues of intrusion and recording privacy.

Technology acceptance factors helped explain the conditions that influence the acceptability of LENA. These factors include perceived usefulness, ease of use, and social influence. The theme of reassurance was mapped to performance expectancy. Mothers showed increased interest in LENA when they perceived that the technology provided useful feedback on their communication. However, some aspects of LENA results were not reassuring and may sometimes place a parent at risk of exposing their child's language difficulties.

The theme of feeling overwhelmed described effort expectancy. Overall, mothers described LENA as easy to use and clothing as appealing. They felt that the recording process was natural, and most mothers forgot that they were being recorded. However, contextual difficulties negatively influence the effort expectancy. Increased demands, often in the context of high levels of stress, impacted usability with mothers waiting for ideal recording conditions.

Social influence, as represented by the theme of trust, is an important acceptance factor. Building on a trusted professional relationship improved the acceptance of LENA. This is in agreement with a UK study looking at the acceptability of LENA by parents of young deaf children, which reported the importance of establishing trust through a known professional [54].

Experiencing cumulative success improves the acceptability of LENA. LENA provides short-term goals, such as word count goals, and assists mothers' motivation and confidence as they experience small successes. This may be especially important for mothers to improve their communication self-efficacy. Feedback received from LENA can support parents to persist in improving their child's language environment. Bandura described the powerful influence of the ability to master a task [51]. Mothers who saw evidence of reciprocal conversation felt motivated to increase this. Satisfaction with reports and experiencing cumulative success improves the acceptability of LENA. Conversely, there is a risk of repeated nonmastery demotivating parents and becoming another way to demonstrate what they are *doing wrong*.

LENA was positively perceived by most mothers, with 83% (5/6) of mothers who were able to complete a trial of LENA expressing an interest in continued use. Mothers felt that LENA was beneficial for their children and found feedback useful in increasing awareness of the home language environment, improving confidence in their interactions with their child, and providing reassurance about their child's language development.

Although mothers valued feedback, the researcher observed the need to interpret the results. Presentation of graphical

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information in bar charts with word counts throughout the day was not always readily accessible to participants. It failed to answer the question, *Was I talking enough?* This finding is consistent with observations by Allen et al [54], which emphasized the need for clinician interpretation of results. Other pilot studies have incorporated individual feedback provided by research assistants to help caregivers understand their specific results [37,55]. Clinicians need to add value to the interpretation of results.

Limitations

Emphasis in this study was placed on understanding the experience of an individual mother's use of LENA with children at risk of poor developmental outcomes in central and inner west Sydney. The study was able to sample from a wide ethnodemographic population, as the software can be used with linguistically diverse families. The results are informative but not necessarily generalizable because of the small sample size.

The nature of the research was explorative, and the trial duration was not long enough to assess any improvements in child and adult language measures. These results are informative for the development of future interventions. These should include regular outcome measures, including pre-, post-, and follow-up intervention measures for children and adults to demonstrate changes over time.

A full-day recording was chosen to reduce the *Hawthorne effect*, where individuals may modify their behavior in response to knowingly being observed. However, for some mothers, this likely contributed to mothers' feeling overwhelmed by requiring recordings to be completed in a specific 16-hour period.

Fathers in 2-parent households were underrepresented in this study. Their voices were expressed through their partners, but they were not directly interviewed. Assessment of their perspective is important, as their attitudes toward LENA also influence the recording and acceptability of language intervention. Allen et al [54] identified the importance of the involvement of both parents, particularly in the beginning, to establish understanding and consent.

Implications for Future Research

LENA is a linguistic technology that provides parents and associated professionals with information about a child's home language environment. There are now published Australian norms of daily adult word count and child vocalizations [56]. The growth of expressive communication in the first 3.5 years is very similar to that in American children [57]. The positive correlation identified in American research between adult word count and child vocalization is presumed to be held in the Australian context and is the basis of quantitative feedback. On an individual level, absolute word counts can be used to track progress. There is a need for a simplified reporting system so that parents can determine whether they are on target or not. Having normed Australian data integrated into the LENA system to show an individual level may help parents to better assess their progress.

Further research is needed to determine how community-based interventions can incorporate the use of LENA. Future research needs to measure pre- and postintervention language scores to assess the effectiveness of LENA as a strategy for reversing developmental risk in populations at risk for poor child developmental outcomes. Trials will benefit from incorporating learning from this study, including strategies for supporting parents. This includes partnering with parents through a trusting clinician relationship and helping mothers build on their parenting self-efficacy. Gauging mothers' acceptability of LENA through trial use will help establish which mothers are both ready and interested in using LENA long term. LENA is best placed as an adjunct to a parent-delivered language program and has been described as a strategy to increase the effectiveness of behavioral interventions [37].

Conclusions

An extended UTAUT model was created specifically for the use of LENA for children at risk of poor language development. This model illustrates the underlying factors contributing to the acceptability of LENA, including technology-specific and contextual factors. Key findings included an increased likelihood of taking up LENA when (1) LENA is introduced by a trusted clinician, (2) LENA results are perceived as useful and motivating to mothers, (3) mothers feel confident in achieving communication goals, and (4) clinicians need to be mindful of the perceived risk associated with recording technology. Language enrichment interventions incorporating LENA may increase parenting self-efficacy, and partnering with families will likely improve technology acceptance and intervention success.

Conflicts of Interest

None declared.

References

1. Sheridan SM, Knoche LL, Kupzyk KA, Edwards CP, Marvin CA. A randomized trial examining the effects of parent engagement on early language and literacy: the Getting Ready intervention. J Sch Psychol 2011 Jun;49(3):361-383 [FREE Full text] [doi: 10.1016/j.jsp.2011.03.001] [Medline: 21640249]
2. Roulstone S, Law J, Rush R, Clegg J, Peters T. The role of language in children's early educational outcomes. Research Brief. 2011. URL: https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment_data/file/181549/DFE-RR134.pdf [accessed 2017-05-07]
3. Hoff E. The specificity of environmental influence: socioeconomic status affects early vocabulary development via maternal speech. Child Dev 2003;74(5):1368-1378. [doi: 10.1111/1467-8624.00612] [Medline: 14552403]

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4. Huttenlocher J, Vasilyeva M, Cymerman E, Levine S. Language input and child syntax. Cogn Psychol 2002 Nov;45(3):337-374. [doi: 10.1016/s0010-0285(02)00500-5] [Medline: 12480478]
5. Connell CM, Prinz RJ. The impact of childcare and parent–child interactions on school readiness and social skills development for low-income African American children. J School Psychol 2002 Mar;40(2):177-193. [doi: 10.1016/s0022-4405(02)00090-0]
6. Shoghi A, Willersdorf E, Braganza L, McDonald M. Let's read literature review. Victoria: The Royal Children's Hospital Centre for Community Child Health, Murdoch Childrens Research Institute; 2013. URL: http://www.letsread.com.au/getmedia/a5313369-d89b-4e11-931f-6cbe7588903e/2013-Let-s-Read-Literature-Review.pdf.aspx [accessed 2017-04-06]
7. Walker D, Greenwood C, Hart B, Carta J. Prediction of school outcomes based on early language production and socioeconomic factors. Child Dev 1994 Apr;65(2 Spec No):606-621. [Medline: 8013242]
8. Shonkoff J, Phillips D. From Neurons to Neighborhoods: The Science of Early Childhood Development. Washington: National Academies Press; 2000.
9. Heckman J, Masterov D. The productivity argument for investing in young children. Appl Econ Persp Policy 2007;29(3):446-493. [doi: 10.3386/w13016]
10. Kingston D, Tough S, Whitfield H. Prenatal and postpartum maternal psychological distress and infant development: a systematic review. Child Psychiatry Hum Dev 2012 Oct;43(5):683-714. [doi: 10.1007/s10578-012-0291-4] [Medline: 22407278]
11. Reilly S, Wake M, Ukoumunne OC, Bavin E, Prior M, Cini E, et al. Predicting language outcomes at 4 years of age: findings from Early Language in Victoria Study. Pediatrics 2010 Dec;126(6):1530-1537. [doi: 10.1542/peds.2010-0254] [Medline: 21059719]
12. Topping K, Dekhinet R, Zeedyk S. Hindrances for parents in enhancing child language. Educ Psychol Rev 2011 Jun 3;23(3):413-455. [doi: 10.1007/s10648-011-9169-4]
13. Pan BA, Rowe ML, Singer JD, Snow CE. Maternal correlates of growth in toddler vocabulary production in low-income families. Child Dev 2005;76(4):763-782. [doi: 10.1111/j.1467-8624.2005.00876.x] [Medline: 16026495]
14. Landry SH, Smith KE, Swank PR, Guttentag C. A responsive parenting intervention: the optimal timing across early childhood for impacting maternal behaviors and child outcomes. Dev Psychol 2008 Sep;44(5):1335-1353 [FREE Full text] [doi: 10.1037/a0013030] [Medline: 18793067]
15. Skurtveit S, Selmer R, Roth C, Hernandez-Diaz S, Handal M. Prenatal exposure to antidepressants and language competence at age three: results from a large population-based pregnancy cohort in Norway. Br J Obstet Gy 2014 Dec;121(13):1621-1631. [doi: 10.1111/1471-0528.12821] [Medline: 24726047]
16. Quevedo LA, Silva RA, Godoy R, Jansen K, Matos MB, Pinheiro KA, et al. The impact of maternal post-partum depression on the language development of children at 12 months. Child Care Health Dev 2012 May;38(3):420-424. [doi: 10.1111/j.1365-2214.2011.01251.x] [Medline: 21651606]
17. Ornoy A, Michailevskaya V, Lukashov I, Bar-Hamburger R, Harel S. The developmental outcome of children born to heroin-dependent mothers, raised at home or adopted. Child Abuse Negl 1996 May;20(5):385-396. [doi: 10.1016/0145-2134(96)00014-2] [Medline: 8735375]
18. Lester BM, Lagasse LL. Children of addicted women. J Addict Dis 2010 Apr;29(2):259-276 [FREE Full text] [doi: 10.1080/10550881003684921] [Medline: 20407981]
19. Hoff-Ginsberg E. The relation of birth order and socioeconomic status to children's language experience and language development. Appl Psycholing 2008 Nov 28;19(4):603-629. [doi: 10.1017/s0142716400010389]
20. Bornstein MH, Hendricks C, Haynes OM, Painter KM. Maternal Sensitivity and Child Responsiveness: Associations with social context, maternal characteristics, and child characteristics in a multivariate analysis. Infancy 2007 Sep;12(2):189-223. [doi: 10.1111/j.1532-7078.2007.tb00240.x] [Medline: 33412745]
21. Halle T, Forry N, Hair E, Perper K, Wandner L, Wessel J. Disparities in Early Learning and Development: Lessons From the Early Childhood Longitudinal Study? Birth Cohort (ECLS-B). Washington, DC: Child Trends; 2009. URL: https://www.childtrends.org/publications/disparities-in-early-learning-and-development-lessons-from-the-early-childhood-longitudinal-study-birth-cohort-ecls-b [accessed 2017-07-15]
22. Hart B, Risley TR. American parenting of language-learning children: Persisting differences in family-child interactions observed in natural home environments. Develop Psychol 1992;28(6):1096-1105. [doi: 10.1037/0012-1649.28.6.1096]
23. Stipek DJ, Ryan RH. Economically disadvantaged preschoolers: ready to learn but further to go. Dev Psychol 1997 Jul;33(4):711-723. [doi: 10.1037//0012-1649.33.4.711] [Medline: 9232386]
24. Forget-Dubois N, Dionne G, Lemelin J, Pérusse D, Tremblay RE, Boivin M. Early child language mediates the relation between home environment and school readiness. Child Dev 2009;80(3):736-749. [doi: 10.1111/j.1467-8624.2009.01294.x] [Medline: 19489900]
25. Durham RE, Farkas G, Hammer CS, Tomblin JB, Catts HW. Kindergarten oral language skill: A key variable in the intergenerational transmission of socioeconomic status. Res Soc Strat Mobility 2007 Dec;25(4):294-305. [doi: 10.1016/j.rssm.2007.03.001]
26. Stein A, Malmberg L, Sylva K, Barnes J, Leach P. The influence of maternal depression, caregiving, and socioeconomic status in the post-natal year on children's language development. Child Care Health Dev 2008 Sep;34(5):603-612. [doi: 10.1111/j.1365-2214.2008.00837.x] [Medline: 18549438]

XSL•FO
RenderX

27. Petterson SM, Albers AB. Effects of poverty and maternal depression on early child development. Child Dev 2001;72(6):1794-1813. [doi: 10.1111/1467-8624.00379] [Medline: 11768146]
28. Leiferman J. The effect of maternal depressive symptomatology on maternal behaviors associated with child health. Health Educ Behav 2002 Oct;29(5):596-607. [doi: 10.1177/109019802237027] [Medline: 12238703]
29. Murray L, Hipwell A, Hooper R, Stein A, Cooper P. The cognitive development of 5-year-old children of postnatally depressed mothers. J Child Psychol Psychiatry 1996 Nov;37(8):927-935. [doi: 10.1111/j.1469-7610.1996.tb01490.x] [Medline: 9119940]
30. Paulson JF, Keefe HA, Leiferman JA. Early parental depression and child language development. J Child Psychol Psychiatry 2009 Mar;50(3):254-262. [doi: 10.1111/j.1469-7610.2008.01973.x] [Medline: 19175819]
31. Schuler ME, Nair P, Black MM. Ongoing maternal drug use, parenting attitudes, and a home intervention: effects on mother-child interaction at 18 months. J Dev Behav Pediatr 2002 Apr;23(2):87-94 [FREE Full text] [doi: 10.1097/00004703-200204000-00004] [Medline: 11943970]
32. Hawley TL, Halle TG, Drasin RE, Thomas NG. Children of addicted mothers: effects of the 'crack epidemic' on the caregiving environment and the development of preschoolers. Am J Orthopsychiatry 1995 Jul;65(3):364-379. [doi: 10.1037/h0079693] [Medline: 7485422]
33. Weisleder A, Fernald A. Talking to children matters: early language experience strengthens processing and builds vocabulary. Psychol Sci 2013 Nov 01;24(11):2143-2152 [FREE Full text] [doi: 10.1177/0956797613488145] [Medline: 24022649]
34. Rowe ML. Child-directed speech: relation to socioeconomic status, knowledge of child development and child vocabulary skill. J Child Lang 2008 Feb;35(1):185-205. [doi: 10.1017/s0305000907008343] [Medline: 18300434]
35. Zimmerman FJ, Christakis DA, Meltzoff AN. Associations between media viewing and language development in children under age 2 years. J Pediatr 2007 Oct;151(4):364-368. [doi: 10.1016/j.jpeds.2007.04.071] [Medline: 17889070]
36. Roberts MY, Kaiser AP. The effectiveness of parent-implemented language interventions: a meta-analysis. Am J Speech Lang Pathol 2011 Aug;20(3):180-199. [doi: 10.1044/1058-0360(2011/10-0055)] [Medline: 21478280]
37. Suskind D, Leffel KR, Hernandez MW, Sapolich SG, Suskind E, Kirkham E, et al. An exploratory study of “Quantitative Linguistic Feedback”. Commun Disord Q 2013 Feb 01;34(4):199-209. [doi: 10.1177/1525740112473146]
38. Pae S, Yoon H, Seol A, Gilkerson J, Richards JA, Ma L, et al. Effects of feedback on parent–child language with infants and toddlers in Korea. First Lang 2016 Jul 25;36(6):549-569. [doi: 10.1177/0142723716649273]
39. Gilkerson J, Zhang Y, Xu D, Richards JA, Xu X, Jiang F, et al. Evaluating language environment analysis system performance for Chinese: a pilot study in Shanghai. J Speech Lang Hear Res 2015 Apr;58(2):445-452. [doi: 10.1044/2015_JSLHR-L-14-0014] [Medline: 25614978]
40. Schwarz I, Botros N, Lord A, Marcusson A, Tidelius H, Marklund E. The LENA™ system applied to Swedish: reliability of the adult word count estimate. Interspeech 2017:2017-2092. [doi: 10.21437/interspeech.2017-1287]
41. Canault M, Le Normand M, Foudil S, Loundon N, Thai-Van H. Reliability of the Language ENvironment Analysis system (LENA™) in European French. Behav Res Methods 2016 Sep;48(3):1109-1124. [doi: 10.3758/s13428-015-0634-8] [Medline: 26174716]
42. Suskind DL, Leffel KR, Graf E, Hernandez MW, Gunderson EA, Sapolich SG, et al. A parent-directed language intervention for children of low socioeconomic status: a randomized controlled pilot study. J Child Lang 2016 Mar;43(2):366-406. [doi: 10.1017/S0305000915000033] [Medline: 26041013]
43. Thirty Million Word Initiative. The University of Chicago. URL: https://tmwcenter.uchicago.edu/ [accessed 2017-05-28]
44. Providence Talks. 2015. URL: http://www.providencetalks.org/ [accessed 2017-05-28]
45. Talking matters to Tamaki. New Zealand Ministry of Education. 2020. URL: https://perma.cc/PJT3-8S9E [accessed 2021-01-07]
46. Brusso R. Employee behavioral intention and technology use: mediating processes and individual difference moderators. Theses & Dissertations: Old Dominion University. 2015. URL: https://digitalcommons.odu.edu/cgi/viewcontent.cgi?article=1013&context=psychology_etds [accessed 2021-05-23]
47. Venkatesh V, Morris MG, Davis GB, Davis FD. User acceptance of information technology: toward a unified view. MIS Q 2003;27(3):425. [doi: 10.2307/30036540]
48. Venkatesh V, Thong J, Xu X. Unified theory of acceptance and use of technology: a synthesis and the road ahead. J Asso Info Syst 2016 May;17(5):328-376. [doi: 10.17705/1jais.00428]
49. Chao C. Factors determining the behavioral intention to use mobile learning: an application and extension of the UTAUT model. Front Psychol 2019 Jul 16;10:1652 [FREE Full text] [doi: 10.3389/fpsyg.2019.01652] [Medline: 31379679]
50. Whittaker K, Cowley S. A survey of parental self-efficacy experiences: maximising potential through health visiting and universal parenting support. J Clin Nurs 2012 Nov;21(21-22):3276-3286. [doi: 10.1111/j.1365-2702.2012.04074.x] [Medline: 23083395]
51. Bandura A. The explanatory and predictive scope of self-efficacy theory. J Soc Clin Psychol 1986 Sep;4(3):359-373. [doi: 10.1521/jscp.1986.4.3.359]
52. Boruszak-Kiziukiewicz J, Kmita G. Parenting self-efficacy in immigrant families-a systematic review. Front Psychol 2020 May 26;11:985 [FREE Full text] [doi: 10.3389/fpsyg.2020.00985] [Medline: 32528373]
53. Pilot findings and next steps. Providence Talks. 2015. URL: https://providencetalks.org/ [accessed 2017-06-15]

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54. Allen S, Crawford P, Mulla I. Exploring the acceptability of innovative technology: a pilot study using LENA with parents of young deaf children in the UK. Child Lang Teach Ther 2016 Nov 16;33(2):117-128. [doi: 10.1177/0265659016671168]
55. Choo D, Dettman S, Dowell R, Cowan R. Talking to toddlers: Drawing on mothers' perceptions of using wearable and mobile technology in the home. Stud Health Technol Inform 2017;239:21-27. [Medline: 28756432]
56. Brushe ME, Lynch J, Reilly S, Melhuish E, Mittinty MN, Brinkman SA. The education word gap emerges by 18 months: findings from an Australian prospective study. BMC Pediatr 2021 May 21;21(1):247 [FREE Full text] [doi: 10.1186/s12887-021-02712-1] [Medline: 34020609]
57. Buzhardt J, Greenwood CR, Hackworth NJ, Jia F, Bennetts SK, Walker D, et al. Cross-cultural exploration of growth in expressive communication of english-speaking infants and toddlers. Early Child Res Q 2019 Jul 01;48:284-294. [doi: 10.1016/j.ecresq.2019.04.002]

Abbreviations

LENA: Language Environment Analysis
SES: socioeconomic status
UTAUT: unified theory of acceptance and use of technology

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