Experiences with Virtual Health Care for Children with Chronic Conditions Requiring High Needs for Pediatric Subspecialist Care and their Caregivers

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PREFACE

This thesis comprises two manuscripts primarily authored by the student (Stephanie Musa), with contributions from her supervisor, Dr. Beth K. Potter; her co-supervisor, Dr. Jamie Brehaut; and thesis advisory committee member, Dr. Eyal Cohen. Additional co-authors of the first manuscript include Becky Skidmore, Andrea Chow, Yousif Al-Baldawi, and Wubalem Muchie.

The student is the primary author of both manuscripts and was responsible for reviewing the literature, co-designing the methodology, conducting data analysis, and drafting the manuscript. The supervisor, co-supervisor, thesis advisory committee member, and additional co-authors provided methodological and analytical guidance and input throughout the research and writing process. Ethics approval from relevant Research Ethics Boards was obtained where necessary.

Manuscript 1

Title: Experiences with Virtual Health Care Among Children with Medical Complexity and/or Their Family Caregivers: A Scoping Review

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Manuscript 2

Title: Family Experiences with Health Care for Children with Inherited Metabolic Diseases during the COVID-19 Pandemic: A Secondary Data Analysis

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ABSTRACT

Purpose: This thesis investigated virtual health care experiences among children with high health care needs and their caregivers.

Methods: Using a scoping review, we synthesized studies of virtual care experiences for children with medical complexity. Analyzing cross-sectional survey data from an existing Canadian study, we then examined caregiver-reported experiences with virtual care for children with inherited metabolic diseases (IMDs) during the COVID-19 pandemic.

Results: Of 34 studies included in the scoping review, we identified inconsistencies in the conceptualization and measurement of virtual care, and limited attention to experiences specific to sociodemographic subgroups (5 studies). Seventy-one caregivers participated in the survey; they endorsed benefits of virtual care such as a reduced need to travel and challenges such as communication barriers.

Conclusions: Findings highlight the need for harmonized, equity-focused frameworks for evaluating virtual care. While virtual care presented benefits, some challenges persisted, suggesting areas for improvement in post-pandemic virtual care for pediatric populations.

150/150 words

TABLE OF CONTENTS

Chapter 1- Introduction

1 Background

1.1 Experiences with health care among children with high health care needs

Patient and family experience with care is a key component of health care quality.[1,2] Care experiences are important to the well-being and satisfaction of patients and are associated with patient safety and other quality outcomes. [1,2] Patient experience with care is a multidimensional concept and has been defined in different ways; one definition is " the sum of all interactions that influence patient perceptions across the continuum of care." [3] Health care experiences are often described or measured based on principles of patient-centred care.[4] These principles emphasize that the specific health needs and outcomes that are important to a patient should be the main driving force behind health care quality measurements and decisions.[4,5] For example, the Picker Institute framework defines eight principles of patient-centred care: care coordination, emotional comfort, physical comfort, information sharing/communication, access to care, family involvement, continuity/follow-up and respect for patient/family. [6]

In pediatric health care, parents or other family caregivers are typically actively involved in the patient's care, as they often assume the crucial roles of advocates, care providers, proxy decision-makers and communicators on behalf of the child.[7] Implementing family-centred care is thus essential to high quality care for children. Principles of family-centred care have been defined differently by different organizations, [8–11] but typically include open and unbiased information sharing between families and providers, partnership and collaboration in decision making, coordination between team members, care in the context of family and community, and respect and honoring differences. [8–11] Much of the literature on pediatric health care experiences focuses on children with chronic and complex illness, including those with medical complexity or who otherwise have elevated health care needs. [12–14]These pediatric patients frequently receive

care from several different health care providers in multiple settings (hospital, community and/or home), necessitating care coordination across providers and services. [15] Thus, children with chronic and complex conditions who have high care needs are susceptible to gaps in health services that may negatively impact their experiences with care.

Children with medical complexity represent a particularly high needs group. [16,17] Definitions are inconsistent in the literature, [16,18] but generally children with medical complexity have one or more chronic and severe conditions that involve functional limitations that may be associated with a need for medical devices (such as feeding tubes); and they have high needs for health care that often require multiple pediatric sub-specialties.[16,18,19] As a result of their frequent health care interactions involving multiple health care providers, this group of children and their caregivers are at risk of poor experiences with care coordination and communication.[16]

Children with rare genetic diseases, including inherited metabolic diseases (IMDs), also often have high health care needs. IMDs are a diverse group of genetic conditions involving deficiencies in enzymes needed in metabolic pathways. [20,21] Many, but not all, children with IMDs would also be considered to have medical complexity. Similar to caregivers in the broader population of families of children with medical complexity, caregivers of children with IMDs often find themselves taking on many tasks involving the health care system, including identifying and gaining access to specialists for their children, as well as coordinating medical appointments. [22–24] Some of these families have reported challenging experiences with care, especially when dealing with systems and providers unfamiliar with their children's rare diagnoses. [25,26]

1.2 Virtual Care and its Implications

Virtual care has been defined as "any interaction between patients and/or members of their circle of care, occurring remotely, using any forms of communication or information technologies, with the aim of facilitating or maximizing the quality and effectiveness of patient care." [27] Virtual care has sometimes been referred to as telehealth or telemedicine and includes modalities such as telephone-based visits, video conference-based visits, text-based counselling and communication using patient portals and instant messaging. [28]

The emergence of the COVID-19 pandemic, which was publicly recognised by the World Health Organisation (WHO) in March 2020, prompted a change in the delivery of health care services across the world. [29] While virtual care existed prior to the pandemic, its use increased dramatically as COVID-19 physical distancing measures limited the availability of in-person care.[30–32] In Canada, the utilization of virtual care services saw a significant increase, rising from 10%-20% of all healthcare visits in 2019 to 60% in 2020.[33,34] By March 2022, nearly half of Canadians reported being offered a virtual visit alongside in-person options, and approximately one-third of all patient-reported visits between January 2021 and March 2022 were conducted virtually. [32,35] Although this usage has declined from its peak, it remains higher than pre-pandemic levels. [32,35]

The increased use of virtual care has important implications for children with high health care needs and has potential to both increase and decrease existing inequities in access to care for families who face systemic barriers to high-quality care, depending on the specific context. [36,37] For example, virtual care may improve access to specialist services for children and their families who may reside in rural or remote areas, those with mobility issues, and families with fewer resources, for whom the time and financial costs associated with in-person care are most

difficult. This is important for high users of health care services, including families who describe having to travel frequently in the absence of virtual options. However, caregivers of pediatric patients may face barriers in accessing telehealth care due to various factors such as socioeconomic status, geographic location, or language. [38–40] For example, families with lower income levels may face difficulties in participating in virtual care because they may not have access to technological devices or internet capabilities to allow for high-quality telehealth experiences. [39] Similarly, while virtual care may improve accessibility in some ways for individuals in remote areas, some remote areas may not have accessible high-quality internet access.[36] Newcomers into English-speaking countries who do not speak English as a first language may also face barriers, for example, when using virtual care that has been tailored for English-speaking individuals.[40] This has prompted recommendations for inclusive cultural and linguistic adaptive tools to improve virtual care experiences.[41,42]

1.3 Describing or Evaluating Experiences with Pediatric Virtual Care: Frameworks, Methods, and Tools

Aligned with the increase in delivery of care using virtual modalities, there has been increased interest in recent years in evaluating patient and family experiences with virtual care. Various frameworks and methods have been created to guide evaluations of virtual care.[43,44] For example, the American Academy of Pediatrics (AAP) Section on Telehealth Care's SPROUT (Supporting Pediatric Research on Outcomes and Utilization of Telehealth) developed the SPROUT Telehealth Evaluation & Measurement (STEM). STEM is a synergized framework that combines evaluation strategies developed by the WHO. Researchers have used this framework to analyse virtual care impacts on patients and their caregivers. The STEM framework includes four measurement domains: 1) health outcomes, 2) health delivery (quality and cost), 3) experience

(quality and characteristics of telehealth encounters), and 4) program implementation and key performance indicators.[43,44] The STEM framework has recently been expanded by a group of Canadian pediatric researchers, who have subdivided the 4 domains into 19 subdomains.[44] This includes 7 subdomains within the area of experiences with telehealth, which relate to experiences of health care providers and support staff in addition to those of patients and their families. [44] These seven subdomains include: technology reliability, usefulness, ease of use, interaction quality, satisfaction and future use, workload burden, and patient-centredness. [44]

Various methods have been used to measure child and family virtual care experiences, including satisfaction surveys and qualitative methods. [44,45] Some examples of tools that have been developed and/or used to measure family experiences with virtual pediatric care are the Telehealth Usability Questionnaire, [46,47] Net Promoter Scores, and the Telehealth Satisfaction Questionnaire. [48,49] Other studies have used questionnaires developed by the researchers or other non-validated questionnaires. [46,49,50]

1.4 Virtual Care Experiences for Children with Chronic and Complex Conditions

Several recent studies have evaluated the virtual care experiences of children with chronic and complex conditions and their caregivers. [51,52] For example, studies have examined family/caregiver and provider satisfaction with virtual care, [51] evaluating factors such as communication effectiveness, accessibility, convenience of accessing services, preference for virtual care compared to in-person care, and sensitivity and respect by clinicians. [51–53] A small number of studies, to my knowledge prior to conducting the study reported in Chapter 2, have evaluated virtual health care experiences specifically for children with complex needs and/or those with IMDs. [54–56] There are also currently few pediatric studies of experiences with virtual

care that have focused on children and families who face barriers to care related to language, socioeconomic status, rural residence, or other factors related to health inequity.[38–40,57]

1.5 Summary of Rationale

In summary, child and family experience is an important dimension of health care quality and is particularly impactful for children with high health care needs, including those with medical complexity and those who have rare genetic disease diagnoses, such as IMDs. Considering the frequent interactions these children and their families have with the health care system, and the increase in virtual health care delivery that has occurred in large part due to the pandemic, it is important to understand experiences with virtual care for these high needs groups. This understanding can inform future research and the development of interventions to improve virtual care experiences; and may provide insights generalizable to other areas of virtual pediatric care.

Given the breadth of this topic and the recent attention it has received, an important starting point was to describe the nature of existing evidence on virtual care experiences for a group of children with high health needs and their caregivers. This served to identify key gaps, underexplored domains, and methodological considerations in the existing literature. It also provided a context within which to carry out a secondary analysis of virtual care experiences for a particular group of children with rare diseases and their families, those with IMDs, ascertained during the pandemic.

1.6 Thesis Objectives

The aim of this thesis project was to understand the virtual care experiences of children with chronic and complex conditions who have high needs for care and their family caregivers. The specific study objectives were:

1) Scoping Review Objective: To synthesize and categorize the key themes, methodologies, guiding frameworks, and domains of care experiences explored in existing literature on the virtual care experiences of children with medical complexity and/or their family caregivers, while identifying gaps and areas for future research.
    1.1 As a secondary objective, to describe this evidence specific to the virtual care experiences of children and families who may face systemic barriers to high-quality care related to sociodemographic characteristics, for example, place of residence, race/ethnicity/culture/language, occupation, gender/sex, religion, education, socioeconomic status, age, and social capital. [58,59]
2) Secondary Analysis Objective: To describe caregivers' self-reported ratings of the impact of the COVID-19 pandemic on health and health care for their young child (<12 years of age) with an IMD, including their views on the advantages and drawbacks of virtual care.

1.7 Thesis Structure

This thesis project consists of a scoping review and a secondary analysis of a cross-sectional survey embedded in a cohort study, structured in a thesis-by-manuscript format. The chapters are as follows:

- Chapter 1 has provided an overview of the current literature on health care for pediatric patients with high health needs, the use of virtual care, and existing frameworks for evaluating virtual care experiences. It also introduced the rationale and overarching objectives of this thesis.
- Chapter 2 focuses on the first objective and presents a scoping review manuscript ("Experiences with Virtual Health Care Among Children with Medical Complexity and/or Their Family Caregivers: A Scoping Review"). This chapter provides information on the

settings and modalities of virtual care that have been investigated for children with medical complexities. It also presents the methods and tools that have been used in published studies to evaluate the experiences of children and their caregivers with virtual care. Additionally, this chapter describes the extent which sociodemographic characteristics have been analyzed in relation to virtual care experiences.

- Chapter 3 addresses the second objective through a quantitative secondary analysis of cross-sectional survey responses embedded within a cohort study ("Family Experiences with Health care for Children with Inherited Metabolic Disorders during the COVID-19 Pandemic: A Secondary Data Analysis "). This chapter provides insights into caregiver perspectives on health and health care for their young child ≤12 years of age with an IMD during the COVID-19 pandemic, with data collected from November 2020 to May 2022.
- Finally, Chapter 4 presents an integrative discussion that synthesizes and interprets the studies' findings, highlighting their implications for future research and for the design and delivery of virtual health care for pediatric patients with high health care needs.

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CHAPTER 2: Experiences with Virtual Health Care Among Children with Medical Complexity and/or Their Family Caregivers: A Scoping Review

PREFACE

The manuscript in this chapter presents a scoping review examining primary studies that describe virtual health care experiences among children with medical complexity and their family caregivers. The study was led by Stephanie Musa, who co-conceived the idea, drafted the research proposal, informed the search strategy, screened citations, extracted and analyzed the data, prepared initial drafts of all tables and figures, and prepared the initial draft of the manuscript. Dr. Beth Potter supervised the study, co-conceived the idea, screened citations, verified data extraction, interpreted the findings, and critically reviewed the manuscript. Drs. Jamie Brehaut and Eyal Cohen co-supervised the study, provided methodological and clinical advice as the study progressed, interpreted the findings, and critically reviewed the manuscript. Becky Skidmore developed and implemented the search strategy and critically reviewed the manuscript. Yousif Al-Baldawi and Wubalem Muchie screened citations, verified data extraction, and critically reviewed the manuscript. All authors have read and approved the final version of this manuscript. This manuscript has not yet been submitted for publication but has been formatted in preparation for submission to BMC Pediatrics. Ethics approval was not required for this study.

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ABSTRACT

Background: Virtual delivery of pediatric health care is not new but has increased dramatically in response to the COVID-19 pandemic. Children with medical complexity are often frequent users of health care services and may thus be among those most affected by the benefits (e.g., reduced need for travel) and potential challenges (e.g., concerns about equitable access due to language or technology barriers) of virtual care. In this scoping review, we aimed to synthesize and map the nature and extent of evidence regarding the virtual care experiences of children with medical complexity and/or their family caregivers.

Methods: We developed and implemented a search strategy in multiple electronic databases (Medline, Embase, PsycINFO, CINAHL) to identify relevant primary articles using any study design to evaluate or describe virtual care experiences for children with medical complexity and/or their caregivers. The articles were screened in Covidence by two reviewers working independently. Data from the included studies were then extracted by one researcher and verified by a second researcher. We describe the settings, methods, care modalities, and domains of care assessed.

Results: From 8,591 citations retrieved by the search, 37 articles describing 34 studies met inclusion criteria, including 18 intervention studies (randomized controlled trials and quasi-experimental studies) and 16 observational studies. Virtual care was delivered through a variety of means, including videoconferencing, telephone calls, and messaging platforms. Nine studies used an established framework to guide their assessment of experiences with virtual care. Most studies assessed experiences using quantitative surveys (74%) with 12 studies relying on a previously published survey. From 16 possible domains of experiences with care (14 selected a priori, 2 identified inductively), "Satisfaction and Future Use" was most frequently assessed (30 studies), followed by "Usefulness" and "Access to Care" (assessed in 21 and 16 studies, respectively). Only five studies examined experiences by sociodemographic subgroups.

Conclusion: To advance the field, harmonizing frameworks and tools for evaluating virtual care is crucial for generating consistent, comparable evidence across studies. Examining how sociodemographic factors shape families' experiences is also critical to ensure that expanding virtual care models remain equitable, accessible, and family-centered for children with medical complexity.

Key Words/ Abbreviations: Children with Medical Complexities, Virtual Care, Experiences, e-medicine, telemedicine, videoconferencing, virtual health care.

2.1 BACKGROUND

While delivery of health care for pediatric and other populations using virtual modalities such as telephone visits, video conference-based visits, patient portals, and instant messaging is not new, the COVID-19 pandemic led to a rapid increase in the use of virtual care by prompting urgent changes to policy and practice; these changes were necessary as part of the pandemic response but also helped to overcome longstanding barriers to telehealth.[1–4] As a result, virtual care is likely to continue to be widely offered beyond the pandemic, given its potential benefits with respect to increased accessibility, for example, reducing the need for travel, particularly for populations in remote or rural areas.[5–8] This shift in care delivery may be most impactful for individuals and families who have the highest needs for health care services, including children with medical complexity. [9–11] Generally children with medical complexity have one or more chronic and severe conditions that involve functional limitations and/or neurological impairment and that may be associated with a need for medical devices (such as feeding tubes); and they have high needs for health care that often require multiple sub-specialties.[9,12,13] As a result of their frequent health care interactions involving multiple health care providers, this group of children and their caregivers are at risk of poor experiences with care coordination and communication.[9]

Children with medical complexities and their families may be a particularly important target for virtual modes of health care delivery. Given their frequent interaction with multiple health care providers over many settings, virtual care holds the promise of increasing accessibility and convenience, facilitating visits to clinics and other health care services without the need of travel to specialized centres such as academic hospitals. However, virtual care may also present accessibility challenges for some children and their families, for example, due to

poor access to technology or language barriers.[14] These challenges may exacerbate existing disparities in health care for children and families from underserved populations (e.g., minoritized populations, those with fewer socioeconomic resources) who already face systemic barriers to obtaining high-quality health care for their children. Mapping the current state of the literature regarding the virtual care experiences of children with medical complexity and their families could present areas for future research and development and ultimately inform the creation of more inclusive and accessible care for this high-needs population.

We aimed to synthesize and categorize the key themes, methodologies, guiding frameworks, and domains of care experiences explored in existing literature on the virtual care experiences of children with medical complexity and/or their family caregivers, while identifying gaps and areas for future research. As a secondary objective, we aimed to describe this evidence specific to the virtual care experiences of children and families who may face systemic barriers to high-quality care related to sociodemographic characteristics, for example, place of residence, race/ethnicity/culture/language, occupation, gender/sex, religion, education, socioeconomic status, age, and social capital. [15,16]

2.2 METHODS

We conducted a scoping review, following the recommended methods from JBI[17] and reporting the review following the Preferred Reporting Items for Systematic Review and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR)[18] (checklist, Appendix 1). The protocol for this study was registered on Open Science Framework (osf.io/ds4xq). A scoping review was best suited to explore key themes and identify gaps in the literature on virtual care experiences for children with medical complexities.[17]

2.2.1 Literature Search and Information Sources

An experienced medical information specialist (BS) developed and tested the search strategies through an iterative process in consultation with the review team. Another senior information specialist peer reviewed the MEDLINE strategy prior to execution using the PRESS Checklist.[19] Using the multifile option and deduplication tool available on the Ovid platform, we searched Ovid MEDLINE® ALL, EmbaseClassic+Embase, and APA PsycINFO. We also searched CINAHL (Ebsco platform). All searches were performed on October17, 2023 and included records from the inception of the relevant databases. The strategies utilized a combination of controlled vocabulary (e.g., “Chronic Disease”, “Telemedicine”, “Patient Satisfaction”) and keywords (e.g., “medically complex”, “ehealth”, “experience”). Vocabulary and syntax were adjusted across the databases and where possible, animal-only records and opinion pieces were removed. There were no language or date restrictions. Records were downloaded and deduplicated using EndNote version 9.3.3 (Clarivate Analytics) and uploaded to Covidence (Veritas Health Innovation). The full MEDLINE search strategy can be found in Appendix 2.

2.2.2 Eligibility Criteria and Selection of Sources of Evidence

Eligible studies included primary articles that assessed or evaluated virtual care experiences of children with medical complexity and/or their family caregivers. For clarity, in this paper, the terms "family" and "caregivers" refer to any of the following individuals involved in the child’s care: biological parents, legal guardians, adoptive parents, foster parents, or any primary adult caregivers. Medical complexity was defined to include one or more of the following: [9,20] a) complex chronic conditions that are severe and in multiple systems, b) significant functional limitation(s) that often result in dependence on medical devices such as

tracheostomies or feeding tubes, c) substantial need for caregiver/family support for home and health care coordination, and d) high health care use often requiring services from multiple subspecialities.

Our specific inclusion criteria were (Table 1): i) Study participants were children (0-18 years) or their family caregivers (parents, guardians, siblings, extended family); ii) the article empirically described or evaluated child-reported or family caregiver-reported experiences (e.g., preferences, ratings, satisfaction); iii) the article focused on synchronous and/or asynchronous virtual care; iv) the article must have been a primary research study; and v) study participants were children with a diagnosis that met the criteria for medical complexity. Non-English publications were excluded due to resource constraints.

We used Covidence[21] to manage the screening process. Four team members (SM, BP, WM, YA) contributed to screening retrieved citations for eligibility in two stages: title/abstract and full text. Following training led by SM, two independent reviewers screened each citation/study at both stages, following a pilot of 100 citations at the title/abstract stage and 10 full-text articles at the second stage. The reasons for exclusions were recorded at the full-text stage. The studies that were moved to the full-text screening stage were initially screened based on criteria (i-iv). We found it difficult to apply the medical complexity criterion. Therefore, we made a post-hoc decision, at the full-text stage, to rely on the Complex Chronic Condition (CCC) Version 3.0 International Classification of Diseases (ICD) codes[22] as a guide for determining eligibility based on inclusion of child participants with diagnoses that met criteria for medical complexity, ascertained from the group of articles that met all other eligibility criteria. Conflicts between reviewers at both stages of screening were resolved by discussion.

2.2.3 Data Extraction Process and Items

We created a Microsoft Excel data extraction form to extract, enter and manage data from the included studies. The extraction form was constructed with input from all team members. One reviewer (SM) extracted the data from included studies using the extraction form and the abstracted data were verified by a second reviewer (of YA, WM), with discrepancies resolved by discussion, involving a third reviewer (BP) as needed. The data extraction stage was piloted with two articles by all reviewers. The full list of fields that were extracted can be found in Appendix 3. The PROGRESS-Plus (place of residence, race/ethnicity/culture/language, occupation, gender/sex, religion, education, socioeconomic status, social capital, and age) framework was used to identify equity-relevant sociodemographic participant characteristics collected in the included studies.[15]

2.2.4 Synthesis of Results

We descriptively analyzed and narratively synthesized the abstracted results from the included studies. We planned to inductively synthesize the extracted data on the aspects of virtual care experiences that were measured in the reviewed studies, but this was difficult due to high variability in how experiential data were collected and described. Therefore, we made a post-hoc decision at the synthesis phase to rely on two previously published frameworks to identify and apply broad domains of health care experiences, using the definitions of the constructs as outlined in: the Picker principles of patient-centered care,[23–25] and the concepts that informed the development of the Telehealth Usability Questionnaire[26] (Table 3). These two frameworks yielded 14 domains, to which we added two further domains ("Integration into Family Life" and "Information Protection and Privacy") inductively during the synthesis process. To assign information about virtual care experiences from the reviewed studies into these 16 domains, we

relied on the abstracted data, and we consulted the original articles, including: narrative descriptions from the Methods and Results sections; questionnaires (where available) for the survey-based studies; and interview guides (where available) from the qualitative studies. In using Results sections from qualitative studies, we relied on the highest level themes reported as indicators of aspects of experiences that were assessed. SM coded the abstracted data and accompanying surveys/interview guides for each study and BP verified the coding; we met to discuss discrepancies as needed.

2.3 RESULTS

2.3.1 Search and Screening Results

The database search yielded a total of 8,591 citations screened at the title-abstract stage, from which 175 articles were considered at the full-text stage, yielding 37 articles and 15 conference abstracts that met the eligibility criteria (Figure 1). We made a post-hoc decision to exclude the conference abstracts from further analysis because they did not present sufficient information to inform our findings.[27] Of the 37 articles included in the review, we identified three pairs of articles that represented different reports from the same underlying study. We extracted data separately for each of the 37 articles but synthesized findings at the study level (n=34). Where different information was reported in different articles from the same study, we relied on the most comprehensive information available for the synthesis (e.g., larger sample size if a second article was based on a sub-sample, all domains of experiences with care if some domains were reported in one article and others in a second article).

2.3.2 Study Characteristics

The characteristics of the 34 included studies are summarized in Table 2 and Appendices 4A-4B. 18 of the 34 studies used an intervention design (randomized controlled trial or quasi-

experimental study), while 16 were observational (Table 2). Based on the country from which participants were recruited, the studies most commonly originated from the United States (n=17), followed by Canada (n=5) . Half of the 16 observational studies were cross-sectional (n=8). Most of the intervention studies were quasi-experimental (n=14), with four randomized control trials. The included articles were published between 2004 and 2023. Data collection periods covered primarily pre-pandemic years (61.8%), with fewer studies collecting data in 2020 (41.2%), 2021 (20.6%), and 2022 (14.7%). The sample size for children with medical complexity (CMC) ranged from four to 288 with a median of 28. Studies most commonly enrolled children with medical complexity generally (n=12), without identifying specific diagnoses. The remaining studies included children with a range of specific conditions, with cerebral palsy (n=6) and cystic fibrosis (n=3) most frequently reported (Table 2).

2.3.3 Frameworks, Methods, and Domains to Evaluate Virtual Care Experiences

The studies reported the delivery of virtual care to children with medical complexity through several modes, including videoconference (n=23), telephone calls (n=12), messaging (n=8), digital applications (n=4), emails (n=2) and remote monitoring (n=2) (Table 2). Nine studies reported using specific frameworks to guide the evaluation of virtual care experiences for this population, with no studies using the same framework (Box 1). Most studies (n=25) used quantitative surveys to measure experiences with virtual care (Table 2). Of the studies using quantitative studies, eight used surveys of their own design, 12 used or adapted previously published survey tools, and eight did not specify the nature of survey tools used. Among established tools, the Telehealth Usability Questionnaire (TUQ) was used in three studies and the Consumer Assessment of Health Providers and Systems (CAHPS) was used in two studies; all other survey instruments were reported in only one study (Box 1).

From the 16 domains of virtual care, we considered (Table 3), the most commonly assessed domain we identified was Satisfaction and Future Use (measured in n=30 studies), which included assessments of overall satisfaction with virtual care, general perceptions, and children's/caregivers' responses to questions about their plans and/or interest in continuing to use virtual care or adopting it in the future[26] (Figure 2). Studies also frequently assessed participants' perceived Usefulness (n=21) of virtual care and how it impacted their Access to Care (n=16); these domains were overlapping since the TUQ incorporates the concepts of access and convenience into its definition of Usefulness (Table 3). Additional domains considered in at least 10 of the 34 studies included Information, Communication, and Education (n=15), Interaction Quality (n=13), Ease of Use (n=13), Reliability (n=11), and Interface Quality (n=10) (Figure 2).

While Satisfaction and Future Use was the most frequently assessed domain across both study types (88% of intervention studies and 89% of observational studies), Usefulness was more commonly evaluated in interventional studies (69%) than in observational studies (56%) (Figure 2). In contrast, observational studies assessed Access to Care (56%) and Information/Communication (56%) more frequently than intervention studies (44% and 31%, respectively). Observational studies also more often examined Respect for Patients/Families (39% vs. 25%) and Family Involvement (28% vs. 19%).

2.3.4 Virtual Care Experiences across Sociodemographic Subgroups

Across the 34 included studies, only five evaluated children's and/or caregivers' experiences with virtual care separately by sociodemographic characteristics (Appendix 5).[28–32] All of these used quantitative methods to assess experiences with care, and one also included qualitative approaches.[29] Among these five studies, age was the most frequently examined sociodemographic factor (n=3).[29,31,32]

2.4 DISCUSSION

This scoping review provides an overview of more than 30 studies of the virtual care experiences of children with medical complexity and their caregivers, including the settings, care modalities, frameworks, methods and instruments used to assess virtual care experiences; the domains of care assessed; and the extent to which experiences of sociodemographic subgroups were considered. The studies we reviewed were published between 2004 and 2023. We identified multiple modes of virtual care, variability in the frameworks and methods used to measure experiences, and limited attention to sociodemographic factors that may shape these experiences.

We found that the methods studies used to evaluate care were predominantly quantitative, with high variability in the specific survey instruments incorporated. This methodological heterogeneity mirrors findings in a recent mixed-methods environmental scan on the evaluation of pediatric virtual care experiences in Canada, which identified a need for standardized evaluation tools to improve the monitoring, acceptability, and quality of virtual care models for pediatric populations.[33] Establishing a harmonized core set of existing or new standardized tools for pediatric studies or specific to children with medical complexity would facilitate consistency in data collection, enhancing comparability of studies across clinical contexts and institutions and encouraging health systems and researchers to incorporate essential domains of patient- and family-centered care models for this particularly high-needs population. The need to identify the most meaningful aspects of virtual care experiences for measurement in future studies is further highlighted by the limited use of frameworks used by the reviewed studies to collect data on experiences with virtual care. Of the nine studies that referenced specific frameworks, none used the same framework. This lack of consistent or unified conceptual guidance for evaluating virtual care experiences for pediatric populations has been highlighted previously as a challenge[34–36]

and hinders efforts to develop a coherent body of evidence. A promising development in this area is a pediatric virtual care framework proposed by Dulude and colleagues which builds upon existing virtual care evaluation frameworks and literature and was developed using a consensus approach with a multidisciplinary team. [37] The framework was validated by being applied to an existing virtual care program, demonstrating its utility in a real-world care setting, and could be considered or adapted for children with medical complexities.[37]

Only five of the reviewed studies examined virtual health care experiences for children with medical complexity in a manner that considered the influence of sociodemographic factors such as place of residence, race/ethnicity/culture/language, occupation, gender/sex, religion, education, socioeconomic status, social capital and age.[15,16] These factors are known to impact health outcomes for children with medical complexity and their families[38–41] and have been hypothesized to be related to potential barriers to effective virtual care, for example, related to resources for equipment and internet service and to language barriers.[42,43] We thus identify this as a critical gap in the literature.

The most frequently reported domain of care experience across the included studies was "Satisfaction and Future Use"[26]. While satisfaction is an important measure of experience, it represents only one facet of the complex and multifaceted care experiences of children with medical complexity and their families.[44] Children with medical complexities require substantial care coordination and compassionate interactions for positive health care outcomes.[45–47] However, family involvement, a critical component of care, was among the underexplored dimensions (considered in only 8 studies).[48,49] Similarly, other dimensions, such as care coordination (n=6), and emotional support (n=1), were rarely explored despite their importance for children with medical complexity. [45,47,50–52] Family involvement includes incorporating

family members in the care process, ensuring that they have meaningful opportunities to participate in decision-making related to their child's health. [23–25] Studies have shown that involving family members in care, such as through bedside training during hospital admissions, can reduce caregiver distress and enhance home-based care support for this population.[39,40] Understanding how virtual care facilitates or hinders family involvement and emotional support could provide insights into optimizing virtual care delivery models for children with elevated health care needs. A recent review of studies on the experiences of inpatient (in-person) health care services among children with medical complexities similarly highlighted a scarcity of interventional studies including patient and family-oriented outcomes into their evaluations.[53] Thus, this research gap is not specific to virtual care. The distribution of domains across study designs likely reflects both the nature of the domains and the specific goals of the studies. For example, the Information, Education and Communication domain was more commonly addressed in intervention studies due to its focus on communication-focused outcomes, while reliability and interface quality were more commonly assessed in observational studies, given the tools used, such as the Telehealth Usability Questionnaire (TUQ).

A high proportion of the included studies were conducted in the United States, which may reflect the country's emphasis on virtual health care innovation and its early adoption of telehealth systems.[54] However, this geographic concentration raises questions about the generalizability of findings to other health care systems, particularly in low- and middle-income countries where infrastructure, policies, and access to virtual care may differ substantially.[55]

2.4.2 Strengths and Limitations

A strength of our study was our incorporation of pre-pandemic studies of virtual care, providing a more complete summary of this care modality by summarizing research that took

place both before and during the pandemic. We also documented the methods used to evaluate care experiences comprehensively, including synthesizing the domains of experience they considered, which served to highlight key gaps in the existing evidence. Following established guidance for scoping reviews[56] helped to ensure the methodological rigour of our approach, for example, using a systematic search strategy, involving two independent reviewers in selecting evidence sources, and having a second reviewer verify extracted data. Our study also has important limitations. First, despite developing and implementing a robust search strategy across multiple databases, due to variability in how studies of children with medical complexity and of virtual care are indexed, specifically the varied terms used to describe both virtual care and patient experience, it is likely that some relevant studies were not captured in our search. Secondly, as most included studies were conducted in English-speaking countries and we excluded non-English-language articles, our findings may not fully capture care experiences in other cultural or health care system contexts. Finally, synthesizing the domains of experiences with care that were considered in each study was necessarily subjective and we had to rely on the level of detail provided by the study authors. We involved two reviewers in this process (with the second reviewer verifying the coding from the first reviewer) and made use of information available in appended surveys and interview guides but reporting was not always strong, particularly when assessing experiences with virtual care was not a study's primary objective. Finally, this is a rapidly evolving field, as evidenced by the large number of conference abstracts retrieved by the search that would have been eligible had they been completed study reports. We were able to provide a comprehensive picture of studies published by late 2023 in order to identify gaps in a timely way to inform this dynamic field.

2.4.3 CONCLUSION

To advance the field of research on experiences with virtual care for children with medical complexity and their caregivers, we recommend selecting and/or developing harmonized evaluation frameworks, tools or core experiential outcomes. These tools would facilitate consistent data collection and reporting, enable comparisons across studies, and inform the design of equitable, family-centered virtual care models. Understanding how sociodemographic factors influence families' experiences is also essential for addressing potential disparities. Future research should prioritize addressing gaps in understanding the potentially differential impacts of virtual care across varying modes (e.g. phone, videoconference, text), different population subgroups (such as different types of conditions) and consider underexplored domains of care experiences such as care coordination and family involvement. As virtual care continues to expand, these efforts will be critical to ensuring high-quality, accessible, and inclusive care that is also patient- and family-centered for children with medical complexity.

LIST OF ABBREVIATIONS

CMC: Children with Medical Complexity

TUQ: Telehealth Usability Questionnaire

CAHPS: Consumer Assessment of Health Providers and Systems

NSHCN: National Survey of Children with Special Health Care Needs

MPOC-20: Measure of Processes of Care-20

PACIC: Patients Assessment of Chronic Illness Care

TSS: Telehealth Satisfaction Survey

TEI-SF : Treatment Evaluation Inventory-Short Form

TARF-R : Treatment Acceptability Rating Form–Revised

SUS: System Usability Scale

DECLARATIONS

Ethics approval and consent to participate

Not applicable

Consent for publication

Not applicable

Availability of data and materials

All data generated or analysed during this review are included in this manuscript.

Competing interests

The authors declare that they have no competing interests.

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Authors' Contributions

SM co-conceived the study, drafted the research proposal, contributed to the search strategy, screened citations, extracted and analyzed data, and prepared initial drafts of tables, figures, and

the manuscript. BKP supervised the study, co-conceived the idea, screened citations, verified data extraction, interpreted findings, and critically reviewed the manuscript. JB and EC co-supervised the study, provided methodological and clinical guidance and critically reviewed the manuscript. BS developed and implemented the search strategy and critically reviewed the manuscript. YA and WM screened citations, verified data extraction, and critically reviewed the manuscript. All authors have approved the final manuscript.

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Table 1. Article Selection Criteria (inclusion/exclusion criteria)

Inclusion Criteria	Exclusion Criteria
i) The population in the article must include children (0-18 years) or their family caregivers (parents, guardians, siblings, extended family).	
ii)The article must evaluate experiences (e.g., preferences, ratings, satisfaction) with care from the participants. The evaluation must be by self-report from the pediatric patient or caregiver	
iii) The article must focus on virtual care (synchronous or asynchronous) as a component of health care delivery .	
iv)The article must be a primary research study.	Non-primary research studies such as commentaries, letters, editorials, reviews, and guidelines were excluded. Non-English publications were excluded.
v) The population in the article must include children with medical complexities as defined by one or more of the following: a. Complex chronic conditions that are severe and in multiple systems. b. Significant functional limitation/s that often result in dependence on medical devices such as tracheostomies or feeding tubes. c. Substantial need for caregiver/family support for home and health care coordination. d. High health care use often requiring services from multiple subspecialities. Studies where a proportion of the participants met one or more of these criteria for medical complexity were included if at least half of the participants meet the criteria or if results were presented separately for at least one subgroup that meets outlined criteria.	

Figure 1. PRISMA flow diagram

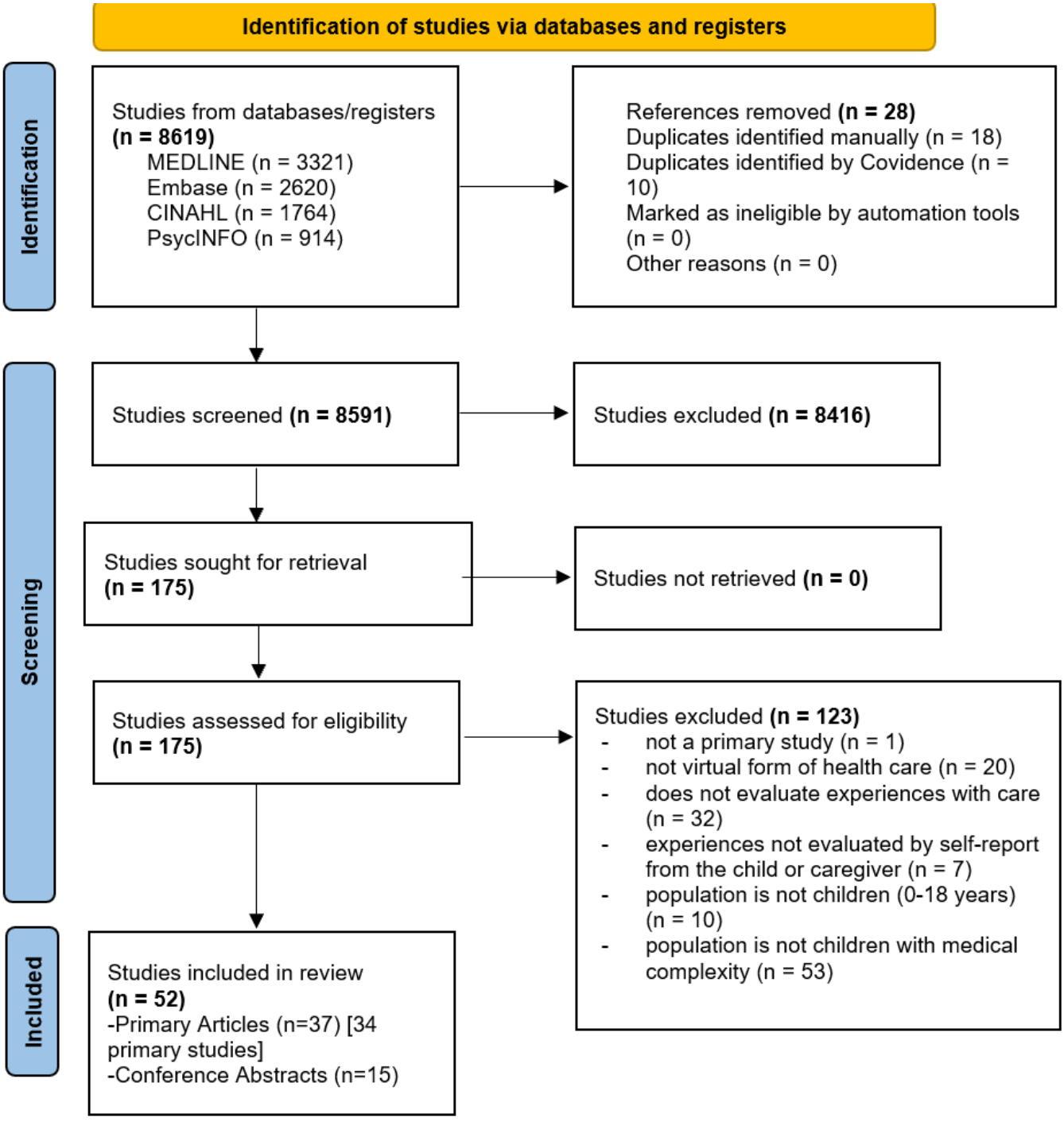


TABLE 2: Study Characteristics of All Included Studies. All data presented as n (%), unless otherwise specified.

Characteristics	All studies (n=34)	Intervention design studies (n=18)	Observational studies (n=16)
Country of Publication			
Brazil	1 (2.9%)	-	1 (6.2%)
Canada	5 (14.7%)	3 (16.7%)	2 (12.5%)
China	1 (2.9%)	1 (5.6%)	-
Ethiopia	1 (2.9%)	-	1 (6.2%)
European countries[a]	8 (23.5%)	2 (11.1%)	6 (37.5%)
Israel	1 (2.9%)	1 (5.6%)	-
USA	17 (50%)	11 (61.1%)	6 (37.5%)
Study Design			
Case study	1 (2.9%)	-	1 (6.2%)
Case-control	2 (5.9%)	-	2 (12.5%)
Cross-sectional	8 (23.5%)	-	8 (50%)
Prospective cohort	3 (8.8%)	-	3 (18.8%)
Retrospective cohort	2 (5.9%)	-	2 (12.5%)
Randomized Control Trial(RCT)	4 (11.8%)	4 (22.2%)	-
Quasi-experimental	14 (41.2%)	14 (77.8%)	-
Timing of primary data collection about virtual care (not mutually exclusive)[b]			
Pre-pandemic (prior to 2020)	21 (61.8%)	15 (83.3%)	6 (37.5%)
2020	14 (41.2%)	7 (38.9%)	7 (43.8%)
2021	7 (20.6%)	1 (5.6%)	6 (37.5%)
2022	5 (14.7%)	1 (5.6%)	4 (25%)
Child Sample Size			
≤10 participants	7 (20.6%)	5 (27.8%)	2 (12.5%)
11–50 participants	16 (47.1%)	9 (50%)	7 (43.8%)
51–200 participants	8 (23.5%)	3 (16.7%)	5 (31.3%)
>200 participants	1 (2.9%)	0 (0%)	1 (6.2%)
Child (Median[range])[c]	28 [4–288]	24 [4–163]	31 [5–388]
Caregiver Sample Size			
≤10 participants	5 (14.7%)	4 (22.2%)	1 (6.2%)
11–50 participants	11 (32.4%)	6 (33.3%)	5 (31.3%)
51–200 participants	6 (17.6%)	3 (16.7%))	3 (18.8%)
201-300 participants	2 (8.8%)	0 (0%)	2 (12.5%)
>300 participants	1 (2.9%)	0 (0%)	1(6.2%)
Caregiver (Median[range])[d]	32 [4–3209]	30 [4–136]	71 [7–3209]

Characteristics	All studies (n=34)	Intervention design studies (n=18)	Observational studies (n=16)
All Participant Diagnoses[e]			
Children with Medical Complexity(CMC)	12 (35.3%)	8 (44.4%)	4 (25%)
Individual Conditions[f]			
Cerebral Palsy	6 (17.6%)	3 (16.7%)	3 (18.8%)
Cystic Fibrosis	3 (8.8%)	1 (5.6%)	2 (12.5%)
Congenital heart disease	2 (5.9%)	-	2 (12.5%)
Congenital Myopathy	2 (5.9%)	-	2 (12.5%)
Epilepsy	2 (5.9%)	1 (5.6%)	1 (6.2%)
Other Diagnoses[g]	15 (44.1%)	-	15 (93.8%)
Other Diagnoses[h]	7 (20.6%)	7 (38.8%)	-
Mode of Virtual Care[i]			
Videoconference	23 (67.6%)	11 (61.1%)	12 (75%)
Phone call	12 (35.3%)	4 (22.2%)	8 (50%)
Messaging	8 (23.5%)	4 (22.2%)	4 (25%)
Application	4 (11.8%)	2 (11.1%)	2 (12.5%)
Emails	2 (5.9%)	1 (5.6%)	1 (6.2%)
Other: remote monitoring	2 (5.9%)	1 (5.6%)	1 (6.2%)
Not Specified	4 (11.8%)	2 (11.1%)	2 (12.5%)
Methods of Experiential Data Collection			
Qualitative	6 (17.6%)	4 (22.2%)	2 (12.5%)
Quantitative	25 (73.5%)	12 (66.7%)	13 (81.2%)
Both	3 (8.8%)	2 (11.1%)	1 (6.2%)
Nature of Quantitative Surveys			
Pre-existing Surveys	12 (35.3%)	8 (44.4%)	4 (25%)
de novo Surveys	8 (23.5%)	4 (22.2%)	4 (25%)
Not Specified[j]	8 (23.5%)	2 (11.1%)	6 (37.5%)

a: European Countries include Germany(n=1), Italy(n=2), Netherlands(n=1), Spain(n=1), Turkey (n=1),[91] UK (n=2).
b, e and i: These characteristics are not mutually exclusive
c :n= 2 articles did not report child sample size.
d :n= 9 articles did not report caregiver sample size.
e: The diagnoses listed are not mutually exclusive, as some articles included combinations of multiple diagnoses.
f: Not all the conditions listed were considered medical complexities (see Table 1 and Methods section).
g: Conditions reported in one study (observational): children born with colorectal malformations, complex surgical infants, diabetes, fragile X syndrome, mitochondrial encephalopathy, movement

Characteristics	All studies (n=34)	Intervention design studies (n=18)	Observational studies (n=16)
disorder, muscular dystrophy, neurodevelopmental disorders, neuropathy, recurrent fever syndrome, rheumatic heart disease, seizure disorder, spinal muscular atrophy, thyroid disorder, and other neurological disorders. **h**: Conditions reported in one study (intervention Study) and include the following: chronic conditions with individual technology use, dysarthria, Rett syndrome, sickle cell disease, tracheostomy dependence, varying cancer diagnoses, and other diagnoses not mentioned. **j**: These articles did not report the nature of the survey used.			

Box1. Pre-existing Surveys(n=11) and Frameworks (n=11) used across included studies.

Names of Pre-existing Surveys[a]	Frameworks
• Telehealth Usability Questionnaire (TUQ)	• AAP Telehealth Support to assess organizational needs, goals, and resources [d]
• Measure of Processes of Care(MPOC-20)	• Telehealth Start-Up Resource Guide[d]
• Consumer Assessment of Health Providers and Systems (CAHPS)[b]	• The behavioural model of health service
• National Survey of Children with Special Health Care Needs(NSHCN) [b]	• RE-AIM implementation research evaluation framework[f]
• CollaboRATE Questionnaire[c]	• A Model of Health care Service Coproduction[e]
• Patients Assessment of Chronic Illness Care (PACIC)[c]	• Model for Improvement Change Framework
• Telehealth Satisfaction Survey (TSS)	• Holistic eHealth framework
• Treatment Evaluation Inventory-Short Form (TEI-SF)	• Performance Appraisal Framework
• Single Ease Question Satisfaction Scale	• An economic evaluation framework for pediatric telehealth
• Treatment Acceptability Rating Form–Revised(TARF-R)	• A conceptual framework developed by the study team
• System Usability Scale (SUS)	• Chronic Care Model[e]

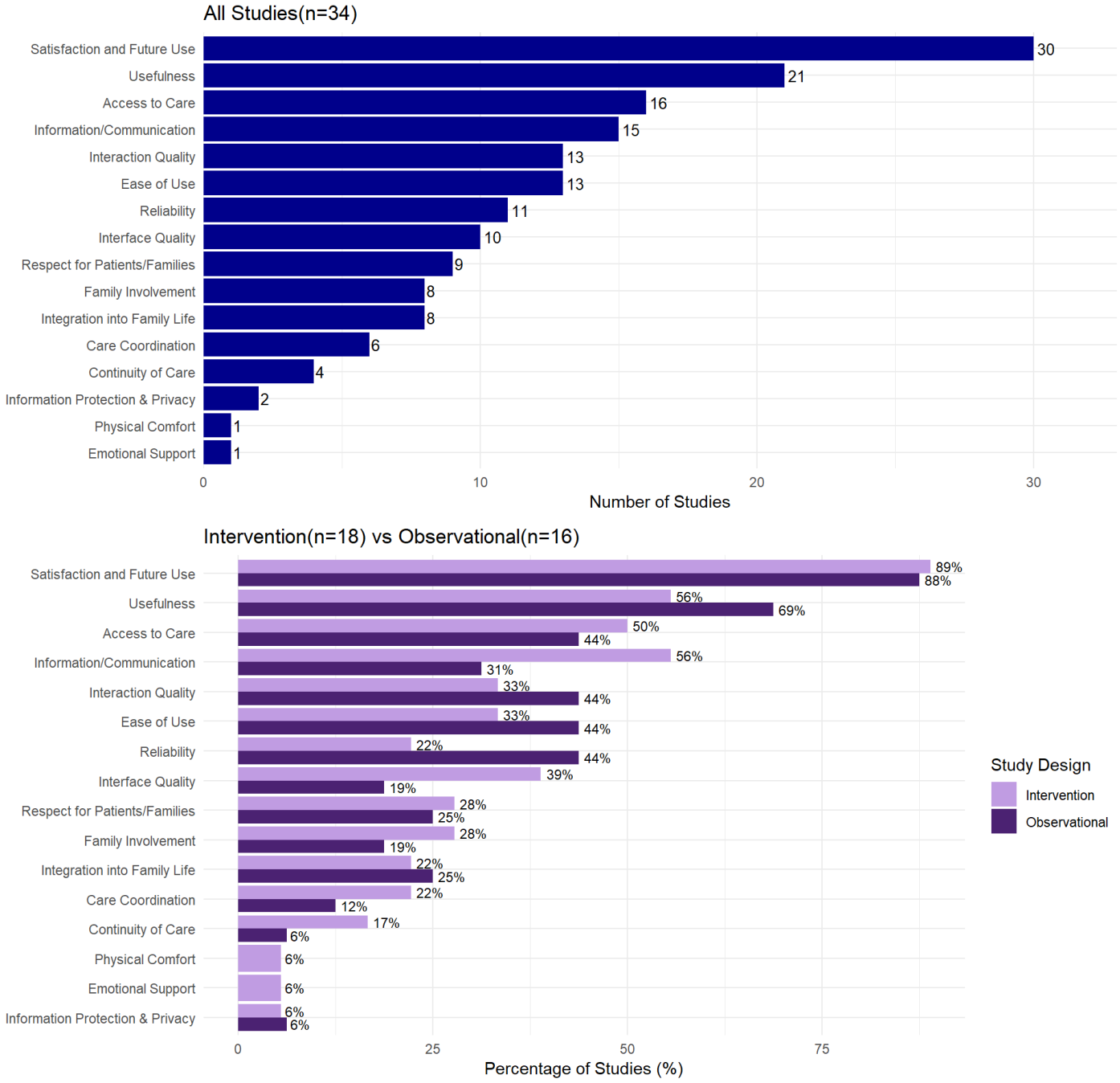
a: All surveys used in only 1 article except the TUQ (used in 3 studies[28,29,57] and the CAHPS used in 2 studies[32,58]
Support to assess organizational needs, goals, and resources frameworks.
b:These were used by the same study
c: These were used by the same study
d: American Academy of Pediatric Telehealth Support to assess organizational needs, goals, and resources. The same study used both the Telehealth Start-Up Resource Guide and the AAP Telehealth
e: These were used by the same study.
f: RE-AIM- (Reach, Effectiveness, Adoption, Implementation, and Maintenance) implementation research evaluation framework.

Table 3. Definitions and Themes used to categorise Domains of Virtual Care Experiences

Themes	Definition
Access to Care[23–25]	Evaluates whether access to health care is timely and reliable. This includes references to travel times, wait times for treatments and referrals, easy and direct access to appropriate professionals, and convenient scheduling
Care Coordination / Integration [23–25]	Evaluates the organization of health care , including effective information exchange among providers and coordination of clinical support
Information, Education and Communication between Providers and Families[23–25]	Assesses whether communication with children/caregivers is clear, transparent and accessible, including the quality of information provided about the patient's status, treatment processes, and prognosis. Encompasses references to whether information is conveyed in an understandable manner at appropriate times
Respect for Patients/Families [23–25]	Evaluates whether care is delivered with respect for patient-centred values, cultural and social backgrounds, and expressed preferences. Includes references to whether providers engage in equitable partnerships, respect patient choices, and support shared decision-making
Emotional Support[23–25]	Evaluates whether effective approaches are used to address psychological distress, fear, and anxiety. Includes references to whether providers offer compassionate care, empathy, and holistic support to meet emotional needs with sensitivity.
Physical Comfort[23–25]	Assesses the provision of a safe, supportive health care environment. Includes effective pain management, assistance with daily activities, and maintaining clean and comfortable surroundings
Family/Friends Involvement[23–25]	Evaluates support for the involvement of family members in the care process, ensuring that they have meaningful opportunities to participate in decision-making related to their child's health.
Follow-up / Continuity of care [23–25]	Evaluates effectiveness of transitions and continuity in care, such as references to whether patients receive clear instructions and consistent information, whether they have ongoing relationships with health care staff, and whether management is well-coordinated post-discharge.
Usefulness [26]	Reflects whether the virtual care system functions and allows care delivery similar to in-person – includes concepts of convenience and access
Ease of Use/Learnability[26]	Addresses how simple and intuitive the system is for children and caregivers to learn and navigate
Interface Quality[26]	Underscores acceptability and user-friendliness of the technical interface, including whether children and caregivers find it enjoyable [26]
Interaction Quality[26]	Evaluates children's and caregivers' views on the quality of virtual interactions with health care providers, including audio and visual communication (as applicable)
Reliability[26]	Focuses on the technology's functionality and the absence of errors, examining how easily technical problems can be resolved
Satisfaction and Future Use[26]	Considers overall satisfaction with virtual care, the likelihood that children and caregivers would use the service again, and general reflections on benefits and challenges, where reported

Integration into Family Life [a] [26]	Focuses on the intersection of virtual care into family life and routines. Considers family dynamics and assesses the potential disruptiveness of virtual care.
Information Protection and Privacy [a] [26]	Concentrates on the protection of patient/family personal or health information.
a: Additional themes included beyond the Picker Principles and Telehealth Usability Questionnaire Subcomponents	

Figure 2. Reported Domains of Experiences with Virtual Care across all Included Studies (n=34) , Intervention (n=18) and Observational (n=16) Studies.



Note: Percentages for the Observational and Intervention plots use a denominator of 16 and 18 respectively.

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APPENDICES

Appendix 1. PRISMA-ScR Checklist - Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) Checklist

SECTION	ITEM	PRISMA-ScR CHECKLIST ITEM	REPORTED ON PAGE #
TITLE			
Title	1	Identify the report as a scoping review.	13
ABSTRACT			
Structured summary	2	Provide a structured summary that includes (as applicable): background, objectives, eligibility criteria, sources of evidence, charting methods, results, and conclusions that relate to the review questions and objectives.	15
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known. Explain why the review questions/objectives lend themselves to a scoping review approach.	16
Objectives	4	Provide an explicit statement of the questions and objectives being addressed with reference to their key elements (e.g., population or participants, concepts, and context) or other relevant key elements used to conceptualize the review questions and/or objectives.	17
METHODS			
Protocol and registration	5	Indicate whether a review protocol exists; state if and where it can be accessed (e.g., a Web address); and if available, provide registration information, including the registration number.	17
Eligibility criteria	6	Specify characteristics of the sources of evidence used as eligibility criteria (e.g., years considered, language, and publication status), and provide a rationale.	18
Information sources*	7	Describe all information sources in the search (e.g., databases with dates of coverage and contact with authors to identify additional sources), as well as the date the most recent search was executed.	18
Search	8	Present the full electronic search strategy for at least 1 database, including any limits used, such that it could be repeated.	50
Selection of sources of evidence†	9	State the process for selecting sources of evidence (i.e., screening and eligibility) included in the scoping review.	21
Data charting process‡	10	Describe the methods of charting data from the included sources of evidence (e.g., calibrated forms or forms that have been tested by the team before their use, and whether data charting was done independently or in duplicate) and any processes for obtaining and confirming data from investigators.	20

Data items	11	List and define all variables for which data were sought and any assumptions and simplifications made.	54
Critical appraisal of individual sources of evidence§	12	If done, provide a rationale for conducting a critical appraisal of included sources of evidence; describe the methods used and how this information was used in any data synthesis (if appropriate).	NA
Synthesis of results	13	Describe the methods of handling and summarizing the data that were charted.	20

SECTION	ITEM	PRISMA-ScR CHECKLIST ITEM	REPORTED ON PAGE #
RESULTS			
Selection of sources of evidence	14	Give numbers of sources of evidence screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally using a flow diagram.	32
Characteristics of sources of evidence	15	For each source of evidence, present characteristics for which data were charted and provide the citations.	59
Critical appraisal within sources of evidence	16	If done, present data on critical appraisal of included sources of evidence (see item 12).	NA
Results of individual sources of evidence	17	For each included source of evidence, present the relevant data that were charted that relate to the review questions and objectives.	59
Synthesis of results	18	Summarize and/or present the charting results as they relate to the review questions and objectives.	21
DISCUSSION			
Summary of evidence	19	Summarize the main results (including an overview of concepts, themes, and types of evidence available), link to the review questions and objectives, and consider the relevance to key groups.	24
Limitations	20	Discuss the limitations of the scoping review process.	26
Conclusions	21	Provide a general interpretation of the results with respect to the review questions and objectives, as well as potential implications and/or next steps.	28
FUNDING			
Funding	22	Describe sources of funding for the included sources of evidence, as well as sources of funding for the scoping review. Describe the role of the funders of the scoping review.	29

JBI = Joanna Briggs Institute; PRISMA-ScR = Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews.

* Where *sources of evidence* (see second footnote) are compiled from, such as bibliographic databases, social media platforms, and Web sites.

† A more inclusive/heterogeneous term used to account for the different types of evidence or data sources (e.g.,quantitative and/or qualitative research, expert opinion, and policy documents) that may be

eligible in a scoping review as opposed to only studies. This is not to be confused with *information sources* (see first footnote).

‡ The frameworks by Arksey and O'Malley (6) and Levac and colleagues (7) and the JBI guidance (4, 5) refer to the process of data extraction in a scoping review as data charting.

Appendix 2. Electronic Search Strategy (MEDLINE)

Database: Ovid MEDLINE(R) ALL <1946 to October 16, 2023>
Search Strategy:
---

1 Adolescent/ (2223481)
2 exp Child/ (2165822)
3 exp Infant/ (1257476)
4 Pediatrics/ (57968)
5 (baby or babies or neonat* or newborn* or child* or infant or infants or infanc* or toddler* or preschool* or pre-school* or adolescen* or teen or teens or teen-age* or teenage* or youth or youths or p?ediatric*).tw,kw,kf. (2736498)
6 or/1-5 [CHILDREN/ADOLESCENTS AGES 0-18] (4777751)
7 Disabled Children/ (7024)
8 (disabled or disabilit* or handicap*).tw,kw,kf. (289070)
9 Chronic Disease/ (282098)
10 (chronic* adj (condition? or disease? or disorder? or health or ill or illness* or sick or sickness*)).tw,kw,kf. (149107)
11 Multiple Chronic Conditions/ (717)
12 ((multiple or multi-organ? or multiorgan? or multi-system? or multisystem?) adj3 (chronic* or condition? or disabl* or disabilit* or disease? or disorder? or illness* or sickness*)).tw,kw,kf. (71494)
13 ((chronic* or condition? or disabl* or disabilit* or disease? or disorder? or health or ill or illness* or sick or sickness* or medical*) adj3 complex*).tw,kw,kf. (86241)
14 ((care or healthcare or health-care or medicine or needs) adj3 complex*).tw,kw,kf. (16760)
15 Functional Status/ (1307)
16 (function* adj3 (depend* or disabl* or disabilit* or limit* or relianc* or reliant* or rely*)).tw,kw,kf. (85186)
17 Enteral Nutrition/ (22041)
18 ((enteral* or force? or gastric of tube?) adj3 (eat* or feed* or food or nutrition* or nourish*)).tw,kw,kf. (22321)
19 Tracheostomy/ (8864)
20 (tracheostom* or (tube? adj3 trachea*)).tw,kw,kf. (21397)
21 ((airway? or breath* or respirat*) adj3 (control* or maintain* or manag* or support*)).tw,kw,kf. (45555)
22 or/7-21 [MEDICAL COMPLEXITY] (982674)
23 6 and 22 [CHILDREN/ADOLESCENTS WITH MEDICALLY COMPLEX DISEASE] (229812)
24 exp Telemedicine/ (45454)
25 (telemed* or tele-med* or telecare or tele-care or teleconsult* or tele-consult* or teleconferenc* or tele-conferenc* or telehealth* or tele-health* or telemonitor* or tele-monitor* or telepractic* or tele-practic* or telerehab* or tele-rehab*).tw,kw,kf. (42625)
26 (ehealth* or e-health* or mhealth* or m-health* or emental health* or e-mental health* or epsychiatr* or e-psychiatr* or epsychol* or e-psychol* or etherap* or e-therap*).tw,kw,kf. (21758)
27 (e-provider? or e-clinician? or e-counsel?or? or e-doctor? or e-nurse? or e-physician? or e-practitioner? or e-therapist? or m-provider? or m-clinician? or m-counsel?or? or m-doctor? or m-nurse? or m-physician? or m-practitioner? or m-therapist?).tw,kw,kf. (109)
28 (emedicine or e-medicine*).tw,kw,kf. (105)
29 (mobile adj3 (care or counsel* or health* or healthcare or health-care or medicine or practice? or psychotherap* or psycho-therap*)).tw,kw,kf. (12648)
30 ((digital* or virtual* or remote*) adj3 (care or counsel* or health* or healthcare or health-care or medicine or practice? or psychotherap* or psycho-therap*)).tw,kw,kf. (24284)

31 ((digital* or virtual* or remote*) adj3 (appointment* or coach* or conferenc* or consult* or diagnos* or interven* or manag* or meet* or monitor* or palliat* or rehab* or support* or surger* or surgic* or therap* or treatment? or visit*)).tw,kf. (41356)
32 Monitoring, Ambulatory/ (8657)
33 ((outpatient* or out-patient* or ambulator* or home? or homebased or home-based) adj3 (appointment* or coach* or conferenc* or consult* or diagnos* or interven* or manag* or meet* or monitor* or palliat* or rehab* or support* or surger* or surgic* or therap* or treatment? or visit*)).tw,kw,kf. (128417)
34 Patient Portals/ (754)
35 ((messag* or patient or patients? or web or webbased or web-based) adj3 portal?).tw,kw,kf. (9880)
36 (secure* adj3 (communicat* or messag* or platform* or portal*)).tw,kw,kf. (1928)
37 exp Therapy, Computer-Assisted/ (45933)
38 ((internet* or app or apps or computer* or cyber* or e-application? or e-mail* or email* or electronic mail* or Google* or iphone? or i-phone? or Microsoft team? or MS team? or (mobile adj2 application?) or mobile-based or mobile phone? or online or smarthome* or smart-home* or smartphone* or smart phone* or technolog* or video* or web or webbased or web-based or webdeliver* or web-deliver* or "web 2.0" or Zoom) adj3 (care or health care or healthcare or health-care or interven* or appointment* or coach* or communicat* or conferenc* or consult* or counsel* or diagnos* or interven* or manag* or meet* or monitor* or palliat* or psychotherap* or psycho-therap* or rehab* or support* or surger* or surgic* or therap* or treatment? or visit*)).tw,kw,kf. (168338)
39 or/24-38 [VIRTUAL CARE PT 1] (448111)
40 exp *Delivery of Health Care/ (733580)
41 exp Cell Phone/ (22699)
42 "Cell Phone Use"/ (387)
43 exp Computers/ (86867)
44 Digital Technology/ (760)
45 Electronic Mail/ (2985)
46 Internet/ (81781)
47 Internet Access/ (154)
48 "Internet Use"/ (549)
49 Telecommunications/ (5055)
50 Telephone/ (13425)
51 Videoconferencing/ (2337)
52 (internet* or app or apps or computer* or cyber* or digital* or e-application? or e-mail* or email* or electronic mail* or Google* or iphone? or i-phone? or Microsoft team? or MS team? or (mobile adj2 application?) or mobile-based or mobile phone? or online or smarthome* or smart-home* or smartphone* or smart phone* or technolog* or telephon* or textmessag* or text-messag* or video* or web or webbased or web-based or webdeliver* or web-deliver* or "web 2.0" or Zoom).ti,kw,kf. (483030)
53 40 and (41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49 or 50 or 51 or 52) [VIRTUAL CARE PT 2] (38203)
54 39 or 53 [VIRTUAL CARE PTs 1 & 2] (463811)
55 23 and 54 [CHILDREN/ADOLESCENTS WITH MEDICALLY COMPLEX DISEASE - VIRTUAL CARE] (6673)
56 Patient Participation/ (29575)
57 ((adolescen* or child* or client* or patient? or person$2 or personally) adj3 (activat* or engag* or involvement? or participat*)).tw,kw,kf. (124344)
58 ((caregiver* or care giver* or carer or carers or family* or families or guardian* or parent* or user or users?) adj3 (activat* or engag* or involvement? or participat*)).tw,kw,kf. (40786)
59 ((adolescen* or child* or client* or patient? or person$2 or personally) adj3 (action* or collaborat* or consult* or contribut* or dialog* or partner*)).tw,kw,kf. (74526)

60 ((caregiver* or carer? or family or families or guardian* or parent* or user or users) adj2 (action* or collaborat* or consult* or contribut* or dialog* or partner*)).tw,kw,kf. (14176)
61 Patient Advocacy/ (24234)
62 ((adolescen* or child* or client* or patient? or person$2 or personally) adj3 (advoca* or input* or ombuds* or represent*)).tw,kw,kf. (47579)
63 ((caregiver* or carer? or family or families or guardian* or parent* or user or users) adj3 (advoca* or input* or ombuds* or represent*)).tw,kw,kf. (16398)
64 (clinical adj3 (advoca* or ombuds* or representative?)).tw,kw,kf. (2425)
65 Patient Acceptance of Health Care/ (55117)
66 exp Patient Satisfaction/ (99857)
67 ((adolescen* or child* or client* or patient? or person$2 or personally) adj3 behav*).tw,kw,kf. (88584)
68 ((caregiver* or care giver* or carer or carers or family* or families or guardian* or parent* or user or users?) adj3 behav*).tw,kw,kf. (32091)
69 Adaptation, Psychological/ (104702)
70 Attitude/ (54191)
71 Attitude to Computers/ (4892)
72 exp Attitude to Health/ (472221)
73 Optimism/ (1077)
74 Pessimism/ (392)
75 (optimism or optimistic* or pessimis*).tw,kw,kf. (22133)
76 Social Perception/ (25047)
77 Perceived Discrimination/ (130)
78 Caregivers/px [psychology] (26900)
79 exp Patients/px [psychology] (18543)
80 exp Parents/px [psychology] (58614)
81 ((accept* or attitud* or dissatisf* or dis-satisf* or emotion* or expect* or experienc* or feeling* or knowledge* or opinion* or perceiv* or perspective? or "point-of-view" or "points-of-view" or prefer* or priorit* or reflect* or respons* or satisf* or understand* or understood or unsatisf* or value or valued or values or valuing or view or views or viewpoint? or voice? or voicing) adj3 (adolescen* or child* or caregiver* or care giver* or carer or carers or client* or family* or families or guardian* or parent* or patient? or person$2 or personally or user or users?)).tw,kw,kf. (872544)
82 (accept* or attitud* or dissatisf* or dis-satisf* or emotion* or expect* or experienc* or feeling* or knowledge* or opinion* or perceiv* or perspective? or "point-of-view" or "points-of-view" or prefer* or priorit* or reflect* or respons* or satisf* or understand* or understood or unsatisf* or value or valued or values or valuing or view or views or viewpoint? or voice? or voicing).ti,kw,kf. (2209140)
83 "Power (Psychology)"/ (13320)
84 empower*.tw,kw,kf. (39703)
85 (experienc* or experiential*).ti,kw,kf. (327919)
86 Diaries as Topic/ (178)
87 Focus Groups/ (36160)
88 Interview.pt. (30779)
89 Interviews as Topic/ (66840)
90 "Surveys and Questionnaires"/ (566446)
91 exp Health Care Surveys/ (49214)
92 Self Report/ (43699)
93 (diary or diaries or focus group? or interview* or questionnaire? or survey*).ti,kw,kf. (311055)
94 exp Qualitative Research/ (83608)
95 qualitative*.tw,kw,kf. (387840)
96 or/56-95 [EXPERIENCE] (4330460)

97 55 and 96 [CHILDREN/ADOLESCENTS WITH MEDICALLY COMPLEX DISEASE - VIRTUAL CARE - EXPERIENCE] (3343)
98 exp Animals/ not Humans/ (5161325)
99 97 not 98 [ANIMAL-ONLY REMOVED] (3343)
100 (comment or editorial or letter or news or newspaper article).pt. (2418781)
101 99 not 100 [OPINION PIECES REMOVED] (3332)

Appendix 3. List of all Items Extracted

Domain to extract	Definitions and Details	Example extraction or choices
First Author &Year	Last Name and Year of Publication	Cohen K. 2013
Country of Publication	Country of study publication or country of the corresponding author	
Main Study Objective	Overall objective of the study	
Study objective specific to the scoping review	The study objective that is specific to the scoping review's objective i.e. investigating experiences with virtual care for children with a medical complexity (refer to Methods Section)	
Years of Data Collection	Years data on virtual care experiences were collected in the study based on the following : Pre-pandemic, 2020, 2021, 2022, and 2023	• Yes • No • Not Specified
Study Design	Whether the study had an interventional or non-interventional design. Further information on the type of Interventional and Non-Interventional Designs.	• Interventional Design -RCT (Pilot RCT, Feasibility RCT, or Full-size RCT). -Quasi-Experimental • Non-Interventional Design - Case-Study - Cross-sectional - Retrospective Cohort - Prospective Cohort - Case control
Study Centers	Indication of whether the study was conducted at a single center or multi-center in one country or multi-center in multiple countries	• Single Center • Multi-center one country • Multi-center International
Care Setting Origin	Indication of whether the care setting the study was carried out was hospital or community based.	• Hospital • Community-based • Other: Specify • Not Specified
Reported Child Age Range	Reported age range of all child participants recruited	
Reported Child Participant Age Range	Reported age range of all children included in the study investigating experiences with virtual care	
How Child Age was Reported	Indication of how child age was reported in the study based on the following : Mean/SD, Median/IQR, Categories, Range, and Other :specify	• Yes • No • Other: specify
All Diagnoses	Diagnoses of all participants in the study	

Medical Complexity Diagnoses	Diagnoses that met the medical complexity criteria (refer to Table 2)	
Caregiver/Child Sample Size Sample Size	The number of caregiver and child participants	
Collected Sociodemographic Characteristics of Caregiver and/or Child	Whether socio demographic characteristics of child and/or caregiver were collected in the study guided by the PROGRESS-Plus recommendations (refer to methods section). Yes/No were selected as appropriate for the following options: • Age • Race/Ethnicity • Language • Place of Residence (Rural/Urban residence) • Gender/Sex • Education • Occupation • Individual/Household Income • Immigrant/Citizenship status • Social Capital • Religion • Other: Specify	• Yes • No • NA
Framework/Theory to guide virtual care experience evaluation presented?	Indication of whether a framework or theory was presented in the study to guide virtual care experience evaluation. Including open text field for the specific framework presented	• Yes • No
Method of data collection	The method used in the study to collect virtual care experiences	• Quantitative • Qualitative • Both
How were the experiences collected	Tools or specific methods the virtual care experiences were collected. The following fields to indicate choices were: • Self-complete survey • Interviewer-administered survey • Qualitative Interview • Qualitative Focus Groups • Other: specify	• Yes • No • NA-Quantitative only • NA-Qualitative only
Name of survey tool	If study indicated use of quantitative methods to measure experience, the	

	specific survey tool was indicated here	
If survey name specified, is this a pre-existing survey or denovo?	Whether the survey used in the study was new and created by the study investigators or a survey that was existing	• Pre-existing • Denovo • NA
If an interview, how was this carried out?	Specification of how the qualitative interviews to collect virtual care experiences in the studies were executed	• Video conference • Phone Interview • In-person • Online • Both (two or more of any of the above options) • NA Survey only • Not Specified • Other: Specify
If survey selected, how was it administered ?	Specification of the means through which the survey examining experiences was administered	• Online survey • Paper survey • Phone survey • Other: specify • Not Specified • NA- interview only
-Is the survey tool available as part of the publication? -If yes, where ?	Specification of where the survey tool was presented in the article	• Yes or No • Where? -Table or figure - In the text -Supplement/ appendix -Bibliography -External link -Available upon request
-Virtual Care platform -If denovo, specify the name	Indication of if the virtual care platform used to deliver care was a standard/commonly used software or denovo	• Denovo • Standard • Not Specified
Nature of Virtual Care	Indication of the mode used to deliver virtual care	• Videoconference • Phone call • Application • Emails • Messaging • Other:specify
Specific virtual platform /software	Specification of the virtual care software used in the study	• Zoom • Microsoft Teams • Skype • SpeakHealth • Connecting2gether

		• Vidyo • KioApp • Not Specified
Summary of main findings/conclusion with respect to virtual care experiences	Study's results with respect to virtual care experiences	
Were these main finding reported separately based on sociodemographic subgroups?	Indication of whether or not the study's results on experiences with virtual care were reported separately based on sociodemographic subgroups	• Yes • No
What were these sociodemographic subgroups?	The sociodemographic subgroup options were guided by the PROGRESS-Plus recommendations (refer to methods section): • Age • Race/Ethnicity • Language • Place of Residence (Rural/Urban residence) • Gender/Sex • Education • Occupation • Individual/Household Income • Immigrant/Citizenship status • Social Capital • Religion • Other: Specify	• Yes • No • NA
Summary of findings for these subgroups	Study's results with respect to findings for these subgroups	
Aspects of experience collected as described in the study	The domains of experience that were evaluated in the study were based on the following (refer to Table 3): • PP11: Access to Care • PP22: Care coordination/Integration • PP3: Information, education and communication between providers and families. • PP4: Respect for patients/families • PP5: Emotional support • PP6: Physical Comfort	• Yes • No

	• PP7:Family/Friends Involvement • PP8: Follow up/continuity of care • TUQ1: Usefulness • TUQ2: Ease of use/learnability • TUQ3: Interface Quality • TUQ4: Interaction Quality • TUQ5: Reliability • TUQ6: Satisfaction and Future Use • IPP1: Information Protection and Privacy • FF1: Integration of virtual care into family life	

Appendix 4A. Detailed Characteristics of Observational Design Articles (n=16)

		YEARS OF DATA COLLECTION								
First Author & Publication Year	**Country**	**Pre-pandemic**	**2020**	**2021**	**2022**	**2023**	**Study Design**	**All diagnoses**	**Medical Complexity Diagnoses***	**Caregiver sample size**	**Child sample size**
Abegaz A. 2023[28]	Ethiopia	no	no	yes	no	no	Cross-sectional	Congenital Heart disease , Diabetes Mellitus , Seizure Disorder ; Other neurologic disorder Thyroid disorder, Cerebral Palsy, Rheumatic heart disease	Seizure disorder, Congenital Heart Disease, Cerebral Palsy	n=288	n=288
Báez-Suárez A. 2022[67]	Spain	no	yes	no	no	no	Prospective cohort	Cerebral palsy , Neurodevelopmental disorders and Movement disorders	Cerebral palsy	n=113	n=113
Curfman A. 2023[62]	USA	yes	yes	no	no	no	Retrospective cohort	Children with medical complexities	Children with medical complexities	Not Specified	n=80
Davis J. 2021[59]	USA	no	yes	no	no	no	Cross-sectional	Cystic Fibrosis	Cystic Fibrosis	n=141	n=141
Fong.V 2023[61]	Canada	no	no	yes	yes	no	Cross-sectional	Children with medical complexities	Children with medical complexities	n=30	n=31
Hall S. 2022[66]	USA	no	no	yes	yes	no	Case study	Fragile X syndrome	Fragile X syndrome	n=11	n=11

Kayaalp G. 2023[30]	Turkey	no	yes	yes	no	no	Cross-sectional	Recurrent fever syndromes	Recurrent fever syndromes	n=245	Not Specified
Lopez J. 2021[64]	USA	yes	yes	no	no	no	Retrospective cohort	Children born with colorectal malformations	children born with colorectal malformations	Not Specified	n=17
McCrossan B. 2008[65]	UK	yes	no	no	no	no	Prospective cohort	Congenital Heart Disease (single-ventricle pathology)	Congenital Heart Disease (single-ventricle pathology)	Not Specified	n=5
Nageswaran 2023 [29]	USA	no	no	no	yes	no	Cross-sectional	Children with Medical Complexities	Children with Medical Complexities	n=23	n=23
Pitch N. 2023[63]	Canada	no	no	yes	yes	no	Cross-sectional	Children with medical complexities	Children with medical complexities	n=12	n=12
Rudolf I. 2019[68]	Germany	yes	no	no	no	no	Case control	Cystic Fibrosis	Cystic Fibrosis	Not Specified	n=25
Sudati I. 2023[70]	Brazil	no	yes	yes	no	no	Cross-sectional	Cerebral Palsy, congenital myopathy	Cerebral Palsy, congenital myopathy	n=7	n=7
Trivisano M. 2020[60]	Italy	no	yes	no	no	no	Cross-sectional	Epilepsy	Epilepsy	n=3209	n=112

Trucco F. 2019[69]	Italy	yes	no	no	no	no	Case control	Duchenne muscular dystrophy (DMD), spinal muscular atrophy type 2 (SMA 2), congenital myopathy (CM), spinal muscular atrophy type 1 (SMA 1),limb girdle muscular dystrophy (LGMD), congenital muscular dystrophy (CMD), neuropathy and mitochondrial encephalopathy	Duchenne muscular dystrophy (DMD), spinal muscular atrophy type 2 (SMA 2), congenital myopathy (CM), spinal muscular atrophy type 1 (SMA 1),limb girdle muscular dystrophy (LGMD), congenital muscular dystrophy (CMD), neuropathy and mitochondrial encephalopathy	n=48	n=48
Williard A. 2018[71]	USA	yes	no	no	no	no	Prospective cohort	Complex surgical infants	Complex surgical infants	n=93	n=93

Appendix 4B. Detailed Characteristics of Intervention Design Articles (n=21).

		YEARS OF DATA COLLECTION								Intervention Group(s)		Comparator Group(s)	
First Author& Publication Year	Country	Pre-pandemic	2020	2021	2022	2023	Study Design *	Type of RCT, where applicable*	All Diagnoses	Medical Complexity Diagnoses	Caregiver sample size	Child sample size	Caregiver sample size	Child sample size
Bird M. 2022[88]	Canada	no	yes	no	no	no	Quasi experimental	NA	Children with medical complexities	Children with medical complexities	n=6	n=6	NA	NA
Bourque M. 2023[81]	USA	yes	yes	no	no	no	Quasi experimental	NA	Level IV NICU infants with a medical complexity	Level IV NICU infants with a medical complexity	Not Specified	n=38	NA	NA
[a]Cady R. 2015[75]	USA	yes	no	no	no	no	RCT	Full-size RCT	Children with medical complexity	Children with medical complexity	n=48(telephone),n=51 (video+telephone)	n=54[phone intervention], n=54 [video & phone intervention]	n=37	n=55
Connolly M. 2023[84]	USA	no	yes	yes	yes	no	Quasi experimental	NA	Sickle Cell Disease (SCD)	Sickle Cell Disease (SCD)	n=20	n=20	NA	NA
Cormack C. 2016[74]	USA	yes	no	no	no	no	Quasi experimental	NA	Children with medical complexity	Children with medical complexity	n=32	n=32(same participants)	n=32	n=32(same participants)

Davidson L. 2022[79]	USA	yes	yes	no	no	no	RCT	Full-size RCT	Cerebral Palsy and other diagnoses not mentioned	Cerebral Palsy	n=53	n=53	n=68	n=68
Dick P. 2004[31]	Canada	yes	no	no	no	no	Quasi experimental	NA	Chronic conditions with individual technology use	Chronic conditions with individual technology use	n=10	n=10	NA	NA
[b]Frush J. 2023[89]	USA	yes	yes	no	no	no	Quasi experimental	NA	Children with medical complexities	Children with medical complexities	n=12	n=12	NA	NA
Gali 2022[82]	USA	yes	no	no	no	no	Quasi experimental	NA	Epilepsy	Epilepsy	Not Specified	n=61	NA	NA
[c]Gulmans J. 2010[77]	Netherlands	yes	no	no	no	no	Quasi experimental	NA	Cerebral Palsy	Cerebral Palsy	n=21	n=21	n=9	n=9
[c]Gulmans J. 2012[76]	Netherlands	yes	no	no	no	no	Quasi experimental	NA	Cerebral Palsy	Cerebral Palsy	n=21	n=21	n=9	n=9
[a]Looman W. 2015[58]	USA	yes	no	no	no	no	RCT	Full-size RCT	Children with medical complexity	Children with medical complexity	n=50(telephone), n=51(telephone +video)	n=50(telephone, n=51(telephone +video)	n=47	n=47
Lotan M.2021[83]	Israel	Not Specified	Not Spe	no	no	no	Quasi experimental	NA	Rett Syndrome	Rett Syndrome	Not Specified	n=5	NA	NA

		cifie d												
Ming D. 2019[87]	USA	yes	no	no	no	no	Quasi experimental	NA	Children with Medical complexities	Children with Medical complexities	Not Specified	n=50	NA	NA
[b]Ming D. 2021[57]	USA	yes	yes	no	no	no	Quasi experimental	NA	Children with medical complexity	Children with medical complexity	n=28	n=28	n=20	n=20
Moreno L. (2020)[80]	USA	yes	no	no	no	no	Quasi experimental	NA	Tracheostomy dependence	Tracheostomy dependence	n=2	n=2	n=2	n=2
Notario P. 2019[73]	USA	yes	no	no	no	no	RCT	Feasibility	Children with medical complexity	Children with medical complexity	Not Specified	n=15	Not Specified	n=9
Oates G. 2023[78]	USA	yes	yes	no	no	no	Quasi experimental	NA	Cystic Fibrosis	Cystic Fibrosis	n=29	n=9	n=29	n=9
Parpia C. 2023[86]	Canada	yes	yes	no	no	no	Quasi experimental	NA	Children with medical complexities	Children with medical complexities	n=36	Not Specified	NA	NA
Pennington L. 2018[72]	UK	yes	no	no	no	no	RCT	Pilot	Cerebral palsy and dysarthria	Cerebral palsy	n=11	n=11	n=11	n=11

Wang J.2017[85]	China	yes	no	no	no	no	Quasi experimental	NA	Varying cancer diagnoses(Osteosarcoma, Non-Hodgkin lymphoma, Acute lymphoblastic, leukemia, Acute myeloid leukemia, Lymphoma.)	Varying cancer diagnoses(Osteosarcoma, Non-Hodgkin lymphoma, Acute lymphoblastic, leukemia, Acute myeloid leukemia, Lymphoma.)	n=5	n=10	NA	NA

*RCT: randomized control trial
a: These two articles are part of the same study
b: These two articles are part of the same study
c: These two articles are part of the same study

Appendix 5. Results Reported Separately by Sociodemographic Characteristics (n=5)

	Sociodemographic subgroups										
First Author (Year)	**Age**	**Race/Ethnicity/ Culture**	**Language**	**Place of residence**	**Gender/Sex**	**Education**	**Occupation**	**Socioeconomic status (SES)**	**Immigrant /citizenship status**	**Social Capital**	**Religion**
Abegaz A. (2023)[28]	No	No	No	No	Yes	No	No	No	No	Yes	No
Davidson L. (2022)[79]	Yes	Yes	No	No	No	No	No	No	No	No	No
Dick P. (2004)[31]	Yes	No	Yes	No	Yes	Yes	Yes	Yes	No	Yes	No
Kayaalp G. (2023)[30]	No	No	No	Yes	No	Yes	No	Yes	No	No	No
Nageswaran (2023)[29]	Yes	Yes	Yes	Yes	No	No	No	No	No	No	No

Appendix 6 . Detailed Reported Methods of Data Collection for Virtual Care Experiences (n=37).

			How were these experiences collected?					
First Author (Publication Year)	**Framework presented as a guide to collect virtual experiences**	**Method of data collection for virtual care experiences**	**Self-complete survey**	**Interview-Administered Survey**	**Qualitative Interview**	**Qualitative Focus Groups**	**Name of survey tool**	**Is this a pre-existing survey or denovo?**
Abegaz A. (2023)[28]	No framework	Quantitative	No	Yes	NA	NA	Telehealth Usability Questionnaire(TUQ)	preexisting
Báez-Suárez A. (2022)[67]	No framework	Quantitative	Yes	No	NA	NA	Not Specified	denovo
Bird M. (2022)[88]	Holistic eHealth framework	Both	Yes	No	Yes	No	Single Ease Question satisfaction scale	preexisting
Bourque M. (2023)[81]	No framework	Quantitative	Yes	No	NA	NA	Not Specified	denovo
[a]Cady R. (2015)[75]	No framework	Quantitative	Yes	No	NA	NA	National Survey of Children with Special Health Care Needs(NSHCN)	preexisting
Connolly M. (2023)[84]	No framework	Quantitative	Yes	No	NA	NA	Treatment Evaluation Inventory-Short Form (TEI-SF)	preexisting
Cormack C. (2016)[74]	No framework	Quantitative	Yes	No	NA	NA	Measure of Processes of Care(MPOC-20)	preexisting
Curfman A. (2023)[62]	An economic evaluation framework for pediatric telehealth	Quantitative	Yes	No	NA	NA	Not Specified	Not Specified

Davidson L. (2022)[90]	No framework	Quantitative	Yes	No	NA	NA	Consumer Assessment of Health Providers and Systems (CAHPS)	preexisting
Davis J. (2021)[59]	No framework	Quantitative	Yes	No	NA	NA	Cystic Fibrosis Telemedicine Questionnaire	denovo
Dick P. (2004)[31]	Performance Appraisal Framework	Quantitative	Yes	No	NA	NA	Not Specified	denovo
Fong V. (2023)[61]	No framework	Qualitative	NA	NA	Yes	No	NA	NA
[b]Frush J. (2023)[89]	No framework	Qualitative	NA	NA	Yes	No	NA	NA
Gali K. (2022)[82]	No framework	Quantitative	Yes	No	NA	NA	Not Specified	Not Specified
[c]Gulmans J. (2010)[77]	A model of health care service coproduction	Quantitative	Yes	No	NA	NA	Not Specified	Not Specified
[c]Gulmans J. (2012)[76]	Chronic Care Model	Quantitative	Yes	No	NA	NA	T1 and T0 Questionnaires	denovo
Hall S. (2022)[66]	No framework	Quantitative	Yes	No	NA	NA	Treatment Acceptability Rating Form–Revised (TARF-R)	preexisting
Kayaalp G. (2023)[30]	No framework	Quantitative	Yes	No	NA	NA	Not Specified	Not Specified
[a]Looman W. (2015)[58]	The behavioural model of health service	Quantitative	Yes	No	NA	NA	Consumer Assessment of Health Providers and Systems (CAHPS)	preexisting
Lopez J. (2021)[64]	No framework	Quantitative	Yes	No	NA	NA	Not Specified	Not Specified

Lotan M.(2021)[83]	No framework	Quantitative	Yes	No	NA	NA	Not Specified	denovo
McCrossan B. (2008)[65]	No framework	Quantitative	No	Yes	NA	NA	Not Specified	Not Specified
[b]Ming D. (2019)[87]	No framework	Qualitative	NA	NA	Yes	No	NA	NA
[b]Ming D. (2021)[57]	RE-AIM (Reach, Effectiveness, Adoption, Implementation, and Maintenance) implementation research evaluation framework	Quantitative	Yes	No	NA	NA	Telehealth Usability Questionnaire (TUQ)	preexisting
Moreno L. (2020)[80]	AAAP* Telehealth Support to assess organizational needs, goals, and resources, Telehealth Start-Up Resource Guide	Quantitative	Yes	No	NA	NA	Telehealth Satisfaction Survey (TSS)	preexisting
Nageswaran S. (2023) [29]	A conceptual framework developed by the study team	Both	Yes	No	Yes	No	Telehealth Usability Questionnaire(TUQ)	preexisting
Notario P. (2019)[73]	No framework	Quantitative	Yes	Yes	NA	NA	Not Specified	Not Specified
Oates G. (2023)[78]	No framework	Quantitative	Yes	No	NA	NA	CollaboRATE, Patients Assessment of Chronic Illness Care (PACIC)	preexisting
Parpia C. (2023)[86]	No framework	Qualitative	NA	NA	Yes	No	NA	NA
Pennington L. (2018)[72]	No framework	Qualitative	NA	NA	Yes	No	NA	NA

Pitch N. (2023)[63]	No framework	Qualitative	NA	NA	Yes	No	NA	NA
Rudolf I. (2019)[68]	No framework	Quantitative	Yes	No	NA	NA	System Usability Scale (SUS)	preexisting
Sudati I. (2023)[70]	No framework	Quantitative	Yes	No	NA	NA	Not Specified	denovo
Trivisano M. (2020)[60]	No framework	Quantitative	Yes	No	NA	NA	Not Specified	denovo
Trucco F. (2019)[69]	No framework	Quantitative	Yes	No	NA	NA	Not Specified	Not Specified
Wang J.(2017)[85]	No framework	Qualitative	NA	NA	Yes	No	NA	NA
Williard A. (2018)[71]	Model for Improvement Change Framework	Quantitative	Yes	No	NA	NA	Not Specified	Not Specified

*: AAP- American Academy of Pediatric Telehealth Support to assess organizational needs, goals, and resources.
a: These two articles are part of the same study
b: These two articles are part of the same study
c: These two articles are part of the same study

CHAPTER 3: Family Experiences with Health care for Children with Inherited Metabolic Diseases during the COVID-19 Pandemic: A Secondary Data Analysis

PREFACE

The manuscript in this chapter presents results from a secondary data analysis of a cross-sectional survey embedded within a cohort study of caregivers/parents to children (<=12 years) diagnosed with an IMD and receiving care in Canada. The objectives of this analysis were to examine caregivers' ratings of overall health care quality for their child with an IMD during the pandemic, including their views on pandemic-induced changes to their child's health care and associated impacts. We also described caregivers' evaluations of virtual appointments compared to their recollections of similar in-person appointments prior to the pandemic. Ethics approval was obtained from the University of Ottawa Research Ethics Board, Children's Hospital of Eastern Ontario's Research Ethics Board, and research ethics boards at all participating centres.

The original cohort study from which we obtained the data for this secondary analysis was a large multi-centre study. This version of the manuscript has been prepared for Stephanie Musa's MSc Epidemiology thesis defence and the co-authors listed below have contributed substantively to the manuscript in the manner listed. Prior to submitting for publication, additional investigators on the larger study will be given an opportunity to critically review the manuscript and to meet co-authorship criteria as appropriate.

Stephanie Musa (SM, MSc student) was responsible for conducting the analysis of the survey data, interpreting the findings, and drafting the manuscript. Beth Potter (SM thesis supervisor) supervised the design and analysis of this study and the survey analysis, contributed to the interpretation of the findings, and critically reviewed the manuscript. Jamie Brehaut (SM

thesis co-supervisor) and Eyal Cohen (SM thesis advisory committee member) contributed to supervising the design of this study, interpretation of the findings, and critically reviewed the manuscript. Andrea Chow (study coordinator) advised the survey analysis, provided the data cut used for the analysis, and critically reviewed the manuscript. All authors listed below have read and approved the final version of this manuscript. This manuscript has not yet been submitted for publication. It has been formatted for Orphanet Journal of Rare Diseases.

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ABSTRACT

Background: Children with inherited metabolic diseases (IMDs) often require frequent health care visits, specialized treatments, and coordinated services, all of which may be disrupted by public health crises. This study aimed to describe caregivers' perceptions of how the COVID-19 pandemic affected their child's health care quality, access, and well-being, focusing on the advantages and drawbacks of virtual care.

Methods: We performed a secondary analysis of cross-sectional baseline survey data embedded in a broader cohort study of family caregivers of children (≤12 years) with IMDs in Canada. Caregivers were recruited from November 2020 to March 2022 across 11 participating pediatric tertiary-care metabolic centres. The questionnaire included items on demographic characteristics, pandemic-related changes to health care (e.g., appointment cancellations, shifts to virtual care), perceived impacts on child and family well-being, and overall ratings of virtual care compared to pre-pandemic in-person encounters. We analyzed data descriptively.

Results: Seventy-one caregivers participated in the study. Over half of participants (57%) indicated that they believed the overall quality of their child's health care remained unchanged during the pandemic, yet most (90%) reported pandemic-driven changes in health care *delivery*, including a shift from in-person to virtual appointments (79%). While 51% of caregivers who noted this shift reported that they perceived no difference in the quality of virtual pandemic care relative to pre-pandemic in-person care, 32% found virtual care worse and 17% found it better. Frequent benefits of virtual care endorsed by participants included convenience, reduced travel time, and lower costs. However, 27% described communication with providers as more difficult virtually. Some caregivers also reported a reduction in child health monitoring that they attributed to the pandemic (30%) and noted increased caregiving challenges, financial strain, and concerns about the child contracting COVID-19.

Conclusion: Caregivers of children with IMDs described substantial disruptions to health care during the pandemic. Although virtual care emerged as a valuable alternative in some respects, offering convenience and cost savings, some participants reported challenges in communication and maintaining rapport with providers. Our findings highlight the importance of addressing not only the logistical aspects of remote care, but also the relational and contextual factors that influence its effectiveness for this population.

Key Words/Abbreviations : virtual care, pediatrics, Inherited Metabolic Diseases (IMDs), experiences with care, caregiving, pandemic.

3.1 BACKGROUND

Inherited metabolic diseases (IMDs) encompass a diverse group of single-gene diseases involving deficiencies in enzymes needed in metabolic pathways.[1,2] These rare conditions are commonly diagnosed during childhood.[3,4] Many children with IMDs have complex health care needs which may include specialized medications, dietary needs, coordinated multidisciplinary care, and recurrent health care encounters to mitigate negative health outcomes and preserve patient quality of life. [5–7]

Family caregivers, including parents, are integral to pediatric health care, especially for children with rare diseases like IMDs, as they play a central role in the patient's care. They often assume critical roles as advocates, care providers, and communicators on the child's behalf. [8] Caregivers of children with IMDs often take on multiple responsibilities within the health care system, including identifying and access to specialists for their children, and coordinating medical appointments.[9–11] These families have reported challenging experiences with care, especially when dealing with systems, care coordination and providers unfamiliar with their children's rare diagnoses.[4,12]

The emergence of the COVID-19 pandemic has impacted pediatric and other health care delivery, including the accessibility of many health care services.[13,14] One of the important ways that the pandemic changed health care was its mode of delivery, specifically by accelerating the use of virtual care modalities. For instance, in Canada, the percentage of the population utilizing virtual care services rose from 10%-20% in 2019 to 60% in 2020. [15,16] By March 2022, nearly half of Canadians indicated that they had been offered a virtual visit in addition to other in-person options, and approximately one-third of all patient-reported visits from January 2021 to

March 2022 were conducted virtually.[15,16] Although use of virtual care has decreased from its peak, it remains substantially higher than pre-pandemic levels.[15,16]

Added to pre-existing health care challenges faced by families of children with rare diseases such as IMDs, some studies have reported that the pandemic further exacerbated difficulties in accessing aspects of health care, such as timely treatment and access to medications.[17–19] Given their frequent interactions with the health care system, it is particularly important to understand the experiences of children with IMDs and their family caregivers regarding pandemic-related changes to care, including virtual care, to inform future health care delivery. Using a quantitative cross-sectional survey, this study aimed to describe caregivers' self-reported ratings of the impact of the COVID-19 pandemic on health and health care for their young child (≤12 years of age) with an IMD, including their views on the advantages and drawbacks of virtual care. Our objectives were to:

i. Describe self-reported ratings of overall health care quality for their child with an IMD during a specific time period within the COVID-19 pandemic (November 2020 to May 2022);
ii. Describe caregivers' perspectives on changes to their child's health care induced by the pandemic;
iii. Describe the impacts of the pandemic and associated changes to care on the family and on the child's health care and status, from caregiver perspectives;
iv. Report caregivers' pandemic-related concerns about their child's health care and wellbeing (e.g., avoidance of in-person care, concerns about difficulty in accessing medications); and

v. Report caregivers' ratings of their general experiences with the child's virtual appointments relative to their recollections of similar (pre-pandemic) in-person appointments.

3.2 METHODS

3.2.1 Study Design and Setting

We analyzed quantitative data from a cross-sectional survey embedded within a cohort study of caregivers/parents to children (≤12 years) diagnosed with an IMD and receiving care in Canada.[20] Caregivers were recruited to the cohort study from November 2020 to March 2022; those who contributed data to a cross-sectional web-based baseline survey were included in the present analysis. The cohort study focused broadly on participants' experiences with receiving health care for their child with an IMD. The cohort study's launch was delayed due to measures associated with managing the COVID-19 pandemic; this provided an opportunity for the team to add questions to the baseline survey about virtual care and perceptions of the impact of the pandemic. This set of questions formed the basis of the present study.

3.2.2 Eligibility Criteria and Recruitment

Eligibility criteria for the cohort study were: being a family caregiver (parent or other guardian) to a child who was receiving care at one of 11 participating metabolic centres, based at pediatric tertiary care hospitals in Canada (in Vancouver, Edmonton, Calgary, Winnipeg, Ottawa, Kingston, Toronto, Hamilton, London, Montreal, and Halifax); the child had a diagnosis of one of 31 IMDs that were included in a previous study conducted by the same team[21] or a different IMD diagnosis if that diagnosis was characterized by a multi-system and progressive clinical course, adjudicated by a clinician at each metabolic centre (as the 31 original diagnoses included few complex progressive conditions; Appendix 1); the child was aged 12 years or younger at the time of recruitment; the child and participating caregiver were currently residing in Canada; the

participating caregiver was able to participate in the online study in English (as study questionnaires were only available in English); and the child was expected by the caregiver to have a minimum of one health care encounter per month during a four-month prospective component of the larger cohort study.

The cohort study team used a purposive maximum variation sampling approach to pursue a heterogenous sample[20] across six selection variables that were expected to impact experiences with health care: travel time from home to the participating metabolic centre, the child's sex assigned at birth, the child's age (years), the participating metabolic centre in Canada where the child received care, and the "IMD category", which was a team-developed classification of the included IMD diagnoses.[20] Specifically, diagnoses were grouped according to their common clinical course, based on the a priori judgement of clinician investigators, into three mutually exclusive categories: acute and episodic, chronic and generally non-progressive, or multi-system and progressive (Appendix 1). If a caregiver had multiple children with IMDs, we invited the caregiver to participate with the child whose characteristics across the selection variables were judged by the research team to best fit the goal of a heterogenous sample.

Potential participants were invited (in person or by telephone or email) by team members at participating metabolic centres to provide an email address indicating an interest in receiving more information about the study. Team members either entered email addresses into the database, or potential participants entered themselves. Duplicates were removed from the database, and potential participants received an email and a link to a website with additional information, including an opportunity to complete a screening questionnaire. The study sample was frequently examined for diversity on the study selection variables listed above and

participants were recruited accordingly based on information from the screening questionnaire. All participants provided informed consent.

3.2.3 Data Collection

The baseline questionnaire that was the source of data for this study included questions addressing demographic characteristics of the participant, child, and household; child health status; family management of the IMD at home; use of community services and supports; and, added as an amendment prior to the start of recruitment, caregiver-reported impacts of the COVID-19 pandemic on the child's health care and health status as well as comparisons of virtual versus in-person care. The questions about COVID-19 and virtual care in the baseline questionnaire were developed by the team (Appendix 2). The questionnaire included a structured response format, specifically, check one option at a time and check all that apply formats.[20] Pilot testing of the questionnaire was conducted with seven patient advisors, who were either parents of children with an IMD or adults living with an IMD. Advisors reviewed the questionnaire and provided feedback, which helped the team to refine the final version.[20]

The baseline questionnaire was administered and managed via a secure web application, Research Electronic Data Capture (REDCap), hosted at the Children's Hospital of Eastern Ontario (CHEO). Participants were enrolled in the cohort study on a rolling basis between November 2020 and March 2022 and the baseline questionnaires were completed from December 2020 through May 2022.

3.2.4 Research Ethics

The study received Research Ethics Board approval from the University of Ottawa Research Ethics Board, Children's Hospital of Eastern Ontario's Research Ethics Board, and the research ethics board at all participating centres. To maintain confidentiality, participants

received a survey link via email and needed their email address to log in. If they did not complete the survey in one sitting, they could save the information and return to the survey at a later time. Survey data were stored securely within the REDCap system, with access restricted to authorized personnel only.

3.2.5 Statistical analysis

All statistical analyses were conducted in SAS version 9.4, R studio version 4.3.2, and Microsoft Excel.[22,23] All data were analyzed descriptively. All variables were categorical, and we have presented counts and proportions, overall and in some cases, stratified by child age (0-4 years and 5-13 years) and IMD category (chronic and generally non-progressive diseases, acute and episodic diseases, and progressive multisystem diseases). While we did not have a priori hypotheses about differences in reported experiences and views across subgroups by age or IMD category, and thus did not conduct formal statistical hypothesis tests, for variables stratified by age and IMD category, we reported effect sizes (Cramer's V, an indication of the strength of association between two categorical variables with more than two categories)[24,25] to aid interpreting findings. When stratifying by age and IMD category, it was necessary to describe the results in subgroups defined by both variables at the same time as they were related (e.g., younger children in the sample were somewhat more likely to have a chronic and non-progressive diagnosis, while older children were more likely to have a multi-system and progressive diagnosis). Results were reported using frequency tables and, for some ordinal variables, stacked bar charts.

3.3 RESULTS

3.3.1 Caregiver, Child, and Family/Household characteristics

Caregiver and Household Characteristics

Although the research team originally aimed to recruit 100 families for the cohort study, 82 caregivers who consented to participate were invited after completing the screening process, of whom 71 started and 70 completed the baseline questionnaire, including the questions about the COVID-19 pandemic and virtual care. Most participating caregivers (80%) reported having at least a university or college degree and a majority (62%) were employed full- or part-time (Table 1). All participants were parents and 90% were mothers to the child with the IMD. A minority (12%) of caregivers identified as recent (<10 years) immigrants to Canada. Just over half (51%) reported a household income of ≥ $100,000 per annum in the past year. Slightly more than half (55%) reported that it took them one hour or less to commute to the metabolic clinic where their child received disease-specific care, while 14% spent over four hours commuting.

Child Characteristics

Children with IMDs, as reported by their caregivers, were distributed across the eligible age range, with 39% aged ≤1 to 3 years, 28% aged 4-6 years, and 32% aged 7 to 13 years at the time of baseline questionnaire completion (Table 1). Just over half of the children of participants (56%) were female. The clinical course of the children's IMD diagnoses varied across the pre-specified categories, with 31% having diagnoses of chronic and generally non-progressive diseases, 41% acute and episodic diseases, and 28% progressive multisystem diseases. Nearly one quarter (24%) of children were reported to have other chronic illnesses aside from the IMD. Participants were recruited to the study from across the participating metabolic clinics, with IWK

Health Centre in Halifax (17%) and McMaster Children's Hospital in Hamilton (14%) having the highest enrollments.

3.3.2 Objective (i): Caregivers' self-reported ratings of health care quality

The majority of participants (57%) reported that they believed the overall quality of health care for their child with an IMD had remained unchanged since the beginning of the pandemic, while 39% reported it was worse or much worse, only 4% reported it was better, and no participants reported that the quality of care was much better (Figure 1a). We observed moderate evidence of potential differences across subgroups when we stratified by age and IMD category (Cramer's V 0.42, 95% CI 0.21-0.60), although subgroup sample sizes were small.[24–26] Specifically, among caregivers of younger children (0-4 years): 3 of 8 caregivers to children in this age group with diagnoses considered multi-system and progressive (37% of this small group) reported that their child's care was much worse since the beginning of the pandemic while no caregivers to young children in either of the other two IMD category groups reported that care was much worse (Figure 1b). Among caregivers to older children (ages 5 years and older): over 70% of caregivers to children in this age group whose children had diagnoses considered acute and episodic or chronic and non-progressive reported no change in the quality of their child's health care since the pandemic, while responses were more variable for caregivers to children with conditions considered progressive and multi-system (Figure 1c).

3.3.3 Objective (ii): Caregiver perspectives on changes to care induced by the pandemic

The vast majority of caregivers (90%) endorsed experiencing one or more types of changes to their child's health care that the caregiver attributed to the pandemic (Table 2). Most caregivers (79%) reported that one or more of their child's health care appointments were changed from in -person to virtual. Over half of the respondents reported experiencing restrictions to the number of caregivers allowed to accompany children to appointments

(60%) and appointment delays (54%), while 29% reported appointment cancellations and 20% reported that they were unable to access a lab, test centre, or pharmacy due to reductions in opening hours at these sites. There was some variation in reported changes across child age group and IMD category, although denominators were small (Appendix 3, Table A3.1). Among these, restrictions on caregivers accompanying children to appointments had the highest subgroup effect size (Cramer's V 0.41, 95% CI, 0.20-0.59) (Appendix 3, Table A3.1). Specifically, among caregivers to older children (ages 5 years and older): 40% and 14% of those whose children were considered to have acute and episodic or chronic and non-progressive diagnoses, respectively, reported restrictions to caregivers being allowed to accompany children to appointments, while this was reported by 73% of caregivers to children with diagnoses considered progressive and multi-system. Among caregivers to younger children (0-4 years), over 70% reported these restrictions to accompanying the child across all three IMD categories.

Of the 20 respondents who reported cancelled services or appointments that they attributed to the pandemic (Table 2), the majority (16/20, 80%) reported cancelled appointments (virtual or clinic-based) with providers who were already a part of the child's care team, while 4 participants or fewer (20% or fewer) reported cancellations for each of the following: appointments with new providers, appointments for therapeutic treatment at hospital settings, laboratory/imaging tests, home-based services, health assistance at school, or other types of care (Appendix 3, Table 3.2). Similarly, of the 38 respondents who indicated delays in accessing health care services and therapies attributable to the pandemic (Table 2), a majority reported delays to appointments (virtual or clinic-based) with providers already part of the child's care team (76%), with 26% reporting delayed appointments with new providers, 32% reporting

delayed access to laboratory/imaging tests, and smaller proportions reporting other delayed services (Appendix 3, Table 3.2).

3.3.4 Objective (iii): Impacts of the pandemic and changes to care on the family and child

The 63 participants who indicated that they experienced one or more changes to health care for their child with an IMD that they attributed to the pandemic were asked about the impacts of the pandemic and of these changes. A majority of these participants (62%) reported impacts for the family, including decreased household income (32%), reduced work hours or job loss among primary caregivers (30%), and increased responsibilities related to time spent managing their child's therapies (25%), diet (29%), and other aspects of their health (24%) at home (Table 2). Participants also reported 'other' impacts and elaborated on these in an open-ended question; responses described impacts related to moving, online or home schooling, issues related to opportunities for child socialization, and lower access to care providers.

Over half of participants who indicated experiencing changes to their child's care due to the pandemic reported an associated change to the child's well-being or to monitoring of the child's health (54%); among these, 30% reported less regular monitoring of their child's health, while 24% reported setbacks in child social development, 16% noted a deterioration in mental health, and 6% reported a deterioration in physical health (Table 2).

3.3.5 Objective (iv): Caregivers' pandemic-related concerns about their child's health care and wellbeing

The majority of caregivers (71%) expressed agreement or strong agreement that they worried about their child contracting COVID-19 during the pandemic and a majority (54%) also strongly agreed or agreed that their child had a higher risk of COVID-19 complications due to their IMD (Figure 2a). While only 15% of caregivers in the study avoided seeking in-person

medical appointments for their child with an IMD during the pandemic, a much larger proportion (41%) avoided bringing their child to the emergency department or other parts of the hospital, and a similar proportion (38%) endorsed being reluctant to bring their child to primary care providers (Figure 2b).

A majority of caregivers in the study (66%) somewhat or strongly disagreed that managing their child's IMD at home became more difficult during the pandemic while 19% agreed or strongly agreed with this statement (Figure 2b). Among the 25 caregivers who answered a question about experiencing stress or anxiety when using public transportation or shared car services for their child's in-person medical appointments (45 respondents found this question "not applicable"), a majority disagreed or strongly disagreed that they had experienced such stress (72%). Over one third (39%) of 57 caregivers responding to a statement about experiencing stress or anxiety when accessing other health care-related needs (e.g. supplies, medication) for their child during the pandemic agreed or strongly agreed with this statement. Finally, 65% of respondents disagreed or strongly disagreed that they faced difficulty obtaining the child's medication/medical products.

3.3.6 Objective (v): Caregiver ratings of virtual relative to in-person appointments

Of the 55 respondents who indicated a change to some of their child's health care delivery from in-person to virtual during the pandemic (Table 2), the majority (51%) of those responding to questions about virtual care rated virtual appointments as being 'no different' overall compared with similar (pre-pandemic) in-person appointments, 17% of caregivers found virtual appointments a little or a lot better, and 32% found virtual appointments a little or

a lot worse (Figure 3).

Scheduling, access, length, convenience, and cost of virtual versus in-person care

Also, among the 55 caregivers who indicated a change from in-person to virtual care for some appointments, almost half (48%) of participants reported that their child's virtual appointments were shorter and 50% reported them as the same length compared to similar in-person appointments before the pandemic (Figure 4a). Sixty-one percent of this group endorsed that provider wait times for virtual appointments were shorter and 37% reported the same wait times, relative to in-person care. Over half (63%) of caregivers indicated that scheduling virtual appointments was accomplished with similar ease as pre-pandemic in-person appointments, while 8% reported that scheduling was harder (Figure 4b). Nearly all respondents (94%) strongly agreed or somewhat agreed that avoiding travel for virtual appointments compared to in-person appointments was convenient, and most (86%) also strongly or somewhat agreed that virtual appointments cost less compared to similar in-person appointments (Figure 4c).

Experiences with providers and treatment during and after virtual versus in-person care

Among caregivers who reported that some of their child's appointments had changed from in-person to virtual care (n=55), 67% rated the ease of communication with providers as the same during virtual appointments compared with in-person appointments pre-pandemic and 27% rated it as "harder", while only 6% reported that communication was easier (Figure 5a). By contrast, 35% reported that it was harder to keep their child comfortable during virtual appointments compared to in-person appointments, 39% rated it the same, and 6% reported that it was easier (Figure 5a). With respect to ease of comprehension of follow-up steps following their child's appointments, 4% of caregivers viewed comprehension to be easier for virtual relative to in-person appointments, 73% found it to be the same, and 23% found it harder (Figure

5a). Most caregivers noted no difference in privacy during virtual versus in-person appointments for their child (88%) (Figure 5b) and no difference in the caregiver's level of involvement in decision-making about their child's health (82%) (Figure 5c). One fifth (20%) of participants agreed or strongly agreed that the treatment received by the child during virtual appointments was less effective compared to similar in-person appointments while 44% disagreed or strongly disagreed with this statement. Finally, 42% agreed or strongly agreed that they were able to talk to more than one provider to a greater degree during virtual appointments compared to similar in-person appointments (Figure 5d).

3.4 DISCUSSION

Our study reveals insights into the health care experiences and challenges faced by families of children with IMDs across Canada during the COVID-19 pandemic, including the transition to and perception of virtual care. Caregivers in our study reported that the overall quality of their child's health care was unchanged (57%) or worse (39%) during the pandemic; no caregivers reported that care was "much better". Changes to some aspects of their child's health care delivery were reported by most participants, with a shift to virtual care and restricted accompaniment to appointments being most common. A majority of caregivers also reported financial strain and increased time spent on their child's care that they attributed to the pandemic, as well as impacts on child health and well-being, particularly social health. Some caregivers expressed concerns about in-person care during the pandemic and most were concerned about their child contracting COVID-19. About half of study participants indicated no difference overall in the quality of virtual appointments relative to their recollections of similar pre-pandemic in-person appointments for their child. Many caregivers found virtual appointments to be shorter, more convenient with respect to less travel, less costly relative to in-person care and offered the benefit of talking to more than one provider. However, over one quarter of

participants noted that they found communication with providers more difficult during virtual appointments.

Our finding that the overall quality of health care during the pandemic was viewed as similar to or worse than pre-pandemic care by caregivers to children with IMDs aligns with recent research in Turkey, where a majority of mothers reported unmet health care needs for their children with IMDs during the pandemic; [19] and with the findings of a European survey conducted early in the pandemic whereby the inability for patients to access the same quality of care as before the pandemic was a primary concern for IMD patient organizations.[27] Beyond IMDs, a study investigating caregivers' perspectives on the pandemic's impact for children with medical complexities highlighted significant disruptions in access to essential health care services. These disruptions affected primary and specialty care, various therapies, and preventive services, resulting in reduced access to medical specialists.[28] With respect to specific impacts attributed by caregivers to the pandemic, our finding that a third of participants indicated that they received less or no monitoring of their child's health during the pandemic is important to highlight given that continuous monitoring and frequent adjustments to both therapeutic regimens and dietary plans are crucial for IMD patients, particularly those with IMD diagnoses where metabolic instability can occur during events such as infections or fever. [29] Some studies have highlighted the broader mental health and psychosocial impacts of the pandemic on children with complex health conditions, further supporting our findings of deterioration in the child's mental health and setbacks in social development reported by caregivers.[45,46]

Most participants in our study reported impacts of the pandemic on the family, including financial strain and increased caregiving responsibilities for their child with an IMD. These findings align with other research showing that the pandemic disproportionately affected the

finances of families of children with complex health care needs, leading to job losses, reduced household income and restricted work hours. [18,30,31]

While our relatively small sample size precluded an in-depth analysis of variation in caregiver experiences by child age and IMD category, there was some evidence in our study that caregivers to children with diagnoses considered multi-system and progressive had more variable and negative experiences than caregivers to children with other IMD diagnoses, with respect to the overall quality of their children's health care during the pandemic, and some evidence that caregivers to younger children may have been more likely to report that care had worsened. These exploratory findings highlight a potentially uneven impact of the pandemic on health care access and quality across age groups and across different conditions. Children with progressive and multi-system conditions are likely to have higher care needs,[32,33] in common with children considered medically complex[34] and their families may thus have experienced the greatest impact associated with changes to care related to pandemic restrictions. In addition, children's health and care needs evolve over time, regardless of changes in care delivery, which may affect how caregivers perceive and experience care, particularly in progressive and multi-system conditions.

The shift from in-person to virtual appointments reported by study participants during the pandemic was expected given broader trends in health care systems in Canada and around the world, where telehealth became a critical tool in maintaining continuity of care while adhering to social distancing guidelines.[35–37] Similarly, the benefits of virtual care we identified with respect to reduced travel time and costs have been established in other studies, corroborating the assertation that virtual care can make health care more accessible and affordable for many families, particularly those who live in rural areas.[38–40] This may be particularly important to

consider given that close to half of participants in our study reported a travel time of more than one hour to the metabolic clinic. Our finding that over a quarter of participants found communication with providers more difficult in virtual compared with in-person appointments may be related to technical issues and/or difficulty for providers in building rapport with the child or family, as seen in other studies examining telehealth experiences for pediatric patients and their caregivers.[41–44]

3.4.1 Study Strengths and Limitations

A strength of this study was its focus on the experiences of a pediatric population with high needs for health care, providing insights into experiences with care during the pandemic among caregivers who were well-positioned to report on the changes they observed and their impact. Furthermore, our diverse sample of participants with varying IMD diagnoses across various provinces in Canada is also a strength. An important limitation of our study was the eligibility requirement in the original cohort study requiring all study participants to be able to participate in English. This could have excluded a population of families and children that may require access to interpretation and translation services, which may influence experiences with virtual care in particular. Additionally, the skewing of participant demographics towards higher-income households and more educated parents may have influenced their access to care and the ability to manage care at home. Our sample size was small, precluding us from drawing strong conclusions about subgroups of children with IMDs. Another potential limitation of our study is recall error, particularly concerning participants' ability to accurately compare virtual care vs pre-pandemic in-person care. Finally, it is difficult to disentangle participants' experiences with

virtual care itself, versus virtual care in the context of a global pandemic that had far-reaching impacts on health and health care more generally.

3.4.2 CONCLUSIONS

Our findings indicate that many caregivers to children with IMDs perceived a decline in some aspects of health care during the pandemic, with accompanying impacts on child and family well-being. These results highlight the need for comprehensive support to assist families to recover from the pandemic's effects and emphasize the importance of establishing strategies to ensure families of children with high health care needs receive necessary health care and support in future pandemics in areas such as health monitoring. Families reported both strengths and drawbacks with virtual care; their priorities and experiences should be prioritized in the future evolution of virtual modalities, which will continue to be integral to pediatric health care delivery .

Further research is needed to better understand how experiences with health care during the pandemic for children with IMDs and their caregivers may vary, for example, depending on patient and family demographics, IMD diagnoses, and types of health care needs. Prospective studies are also important to better understand patient and family experiences with virtual care. Our cross-sectional analysis of caregivers' overall perspectives provides a foundation that will complement and help to contextualize such prospective experiential data, including forthcoming analyses from the larger cohort study in which this study is embedded.

LIST OF ABBREVIATIONS

IMD: Inherited Metabolic Diseases

DECLARATIONS

Ethics approval and consent to participate

Ethics approval was obtained from the University of Ottawa Research Ethics Board, Children's Hospital of Eastern Ontario's Research Ethics Board, and research ethics boards at all participating centres.

Consent for publication

Not applicable

Availability of data and materials

All data generated during this analysis are included in this manuscript.

Competing interests

The authors declare that they have no competing interests.

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Authors' Contributions

Stephanie Musa (SM, MSc student) was responsible for conducting the analysis of the survey data, interpreting the findings, and drafting the manuscript. Beth Potter (SM thesis supervisor) supervised the design and analysis of this study and the survey analysis, contributed to the interpretation of the findings, and critically reviewed the manuscript. Jamie Brehaut (SM thesis

co-supervisor) and Eyal Cohen (SM thesis advisory committee member) contributed to supervising the design of this study, interpretation of the findings, and critically reviewed the manuscript. Andrea Chow (study coordinator) advised the survey analysis, provided the data cut used for the analysis, and critically reviewed the manuscript**.**

TABLE 1: Study Participant Characteristics (n=71)

Caregiver and Household Characteristics	n (%[a])
Education Level	
High school/GED or some college/university (no degree/diploma)	14 (20)
College	17 (24)
University	31 (44)
Graduate/Post-graduate school	9 (13)
Working status, full or part-time (vs any other status)	50 (70)
Relationship to child	
Father (biological, legal, adoptive, step)	7 (10)
Mother (biological, legal, adoptive, step)	64 (90)
Immigrated to Canada recently (within 10 years prior to questionnaire completion)[b]	8 (12)
Household income[c]	
≤ $40,000	7 (11)
$40,001 - $60,000	7 (11)
$60,001 - $80,000	6 (10)
$80,001 - $100,000	11 (17)
$100,001 – $120,000	8 (13)
≥ 120, 001	24 (38)
Travel time from home to metabolic clinic	
≤ 1 hour	39 (55)
>1 hour to 2 hours	12 (17)
>2 hours to 3 hours	5 (7)
>3 hours to 4 hours	5 (7)
>4 hours	10 (14)
Child Characteristics	**n (%[a])**
Sex assigned at birth, female (all other children were reported to be male)	40 (56)
Age in years at the time of survey completion	
≤1 – 3	28 (39)
4 – 6	20 (28)
7 – 13	23 (32)
Child's inherited metabolic disease (IMD) diagnosis category	
Chronic and generally non-progressive disease	22 (31)
Acute and episodic disease	29 (41)
Progressive multi-system disease	20 (28)
Child has another chronic illness (beyond the IMD), Yes[d]	16 (24)
Metabolic Clinic	
Alberta Children's Hospital, Calgary	7 (10)
London Health Sciences Centre	8 (11)
McMaster Children's Hospital, Hamilton	10 (14)
Hospital for Sick Children, Toronto	6 (8)
Kingston General Hospital	8 (11)
Children's Hospital of Eastern Ontario (CHEO), Ottawa	9 (13)
IWK Health Centre, Halifax	12 (17)
Stollery Children's Hospital, Edmonton	5 (7)
Other (BC Children's Hospital, Winnipeg Children's Hospital, Montreal Children's Hospital)	6 (8)

[a] percentages may not add to 100% for each variable due to rounding, denominator N = 71 responses except where indicated
[b] N=69 responses to question about immigration; [c] N=63 responses to question about household income;
[d] N=66 responses to question about other chronic illnesses

TABLE 2: Caregiver perspectives on how the COVID-19 pandemic affected health care for their child with an IMD and associated impacts on the family and the child (n=70)

	n (%)
Changes to health care attributed to the pandemic (check all that apply, n=70)	
Any of these types of changes to care:	63(90)
One or more health care appointments cancelled	20(29)
One or more health care appointments delayed	38(54)
One or more health care appointments changed from in-person to virtual	55(79)
Lab / test centre / pharmacy not accessible*	14(20)
Restricted accompaniment during health care appointments	42(60)
Perceived impact of changes induced by pandemic on the family (check all that apply, n=63 respondents who indicated at least one change to health care attributed to the pandemic)	
Any of these impacts on the family:	39 (62)
Household income decreased	20(32)
≥ 1 primary caregiver(s) had work hours reduced or lost job	19(30)
More time spent managing child's therapies at home	16(25)
More time spent managing child's diet at home	18(29)
More time spent managing other parts of child's health at home	15(24)
Other †	5(8)
Perceived impact of changes attributed to the pandemic on child's wellbeing. (check all that apply, n=63 respondents who indicated at least one change to health care attributed to the pandemic)	
Child's well-being or monitoring of health affected in any of these ways:	34 (54)
Child's physical health has gotten worse	4(6)
Child's mental health has gotten worse	10(16)
Child's social development has been set back	15(24)
No monitoring of child's health	2(3)
Less regular monitoring of child's health	19(30)
Other	4(6)

* Lab / test centre / pharmacy not accessible due to opening hours being reduced

† Other : e.g., moved cities, online or home school, concerns about socialization, lower access to health care providers

Figure 1. Caregiver Ratings of the Overall Quality of Health Care for their Child with an IMD since the Start of the Pandemic, Overall and by the Category of the Child's Diagnosis within Age Groups (n=70)

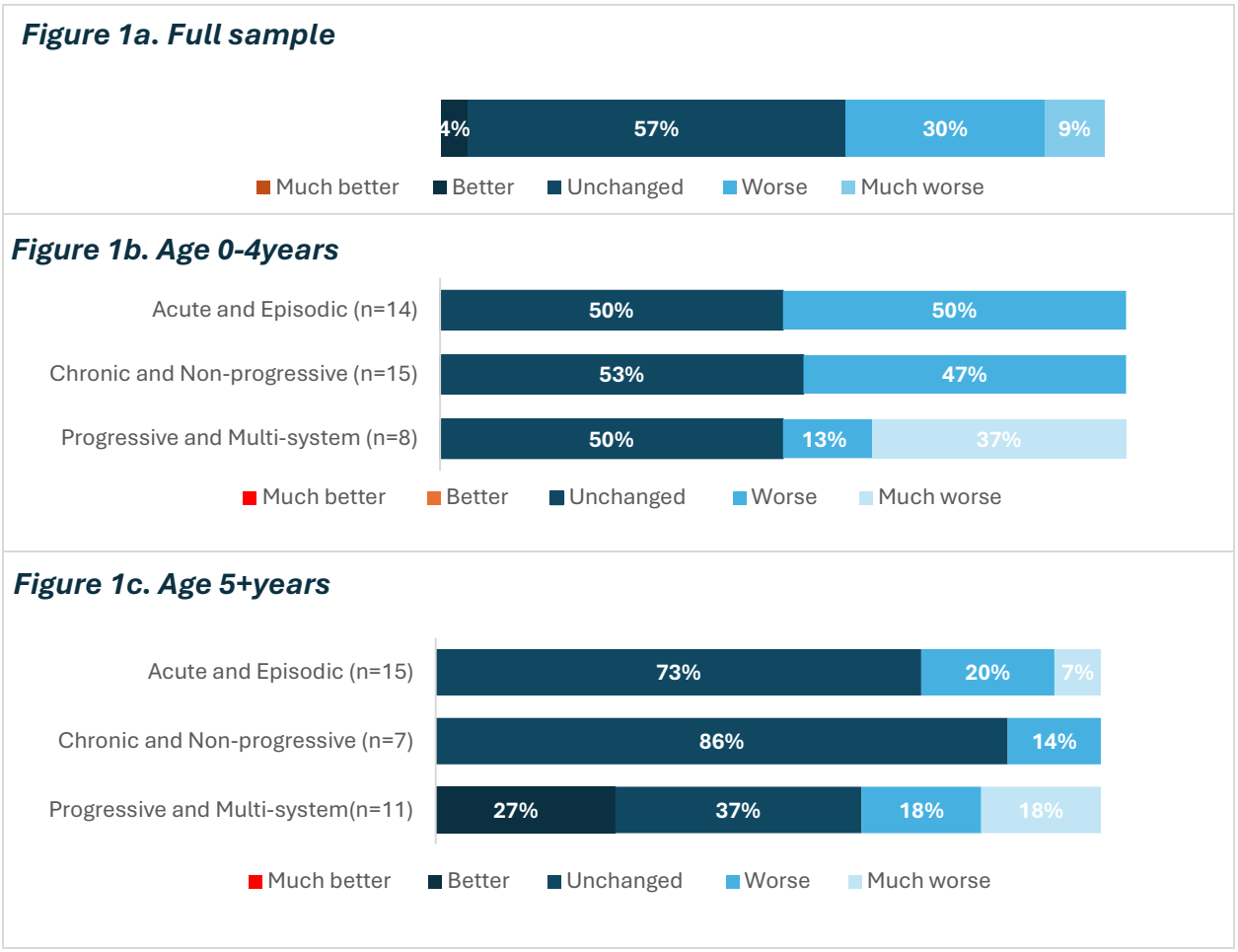


Figure 2. Caregiver Ratings of Concerns Regarding Aspects of Child Health Care during the Pandemic (n=70)

Figure 2a: Caregiver concerns regarding child contracting COVID-19 during the pandemic

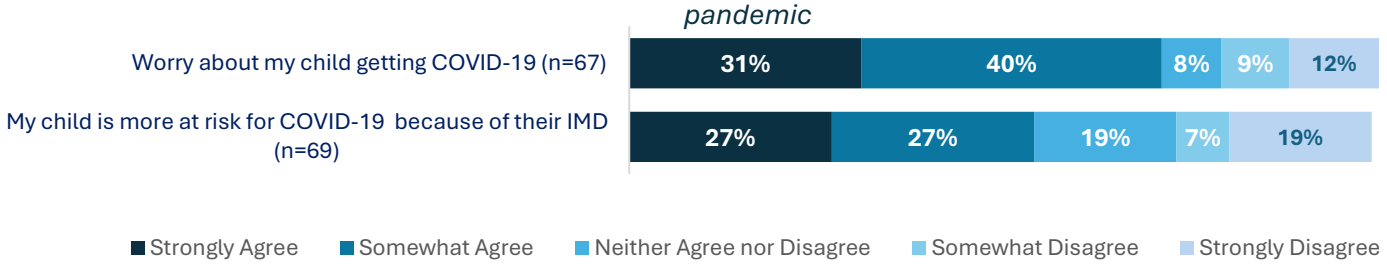


Figure 2b: Caregiver concerns regarding in-person care for their child with IMD

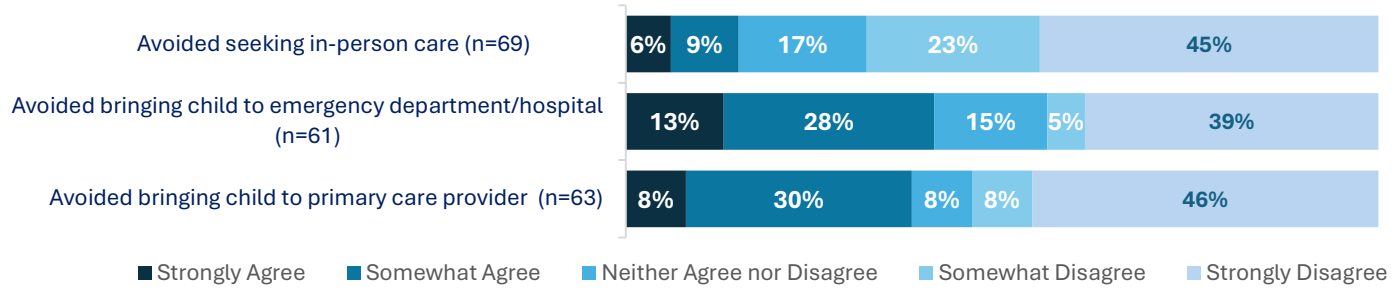


Figure 2c: Impacts of addressing/managing child's IMD health needs on caregivers

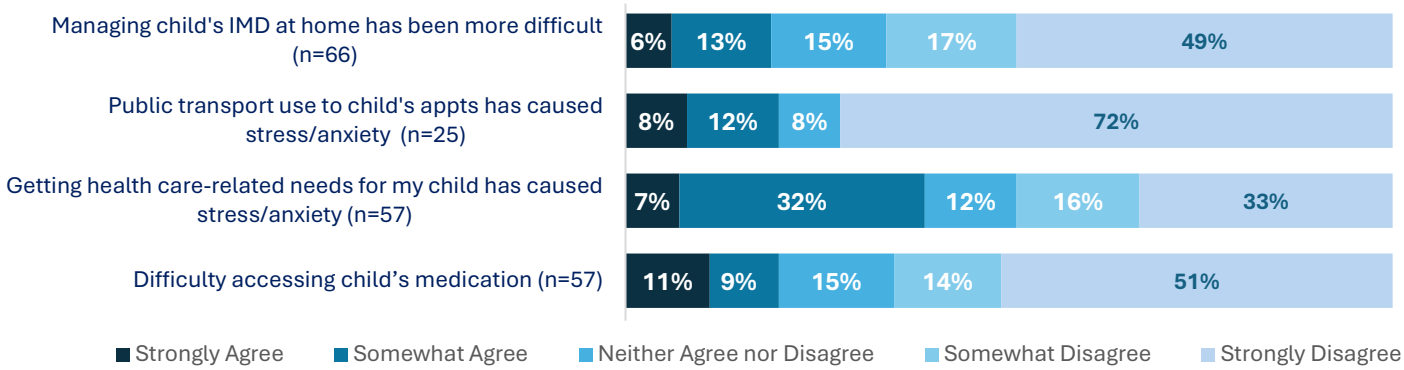


Figure 3. Caregivers' Overall Ratings of their Child's Virtual Appointments vs In-Person Appointments (n=55 asked this question, n=53 responded)

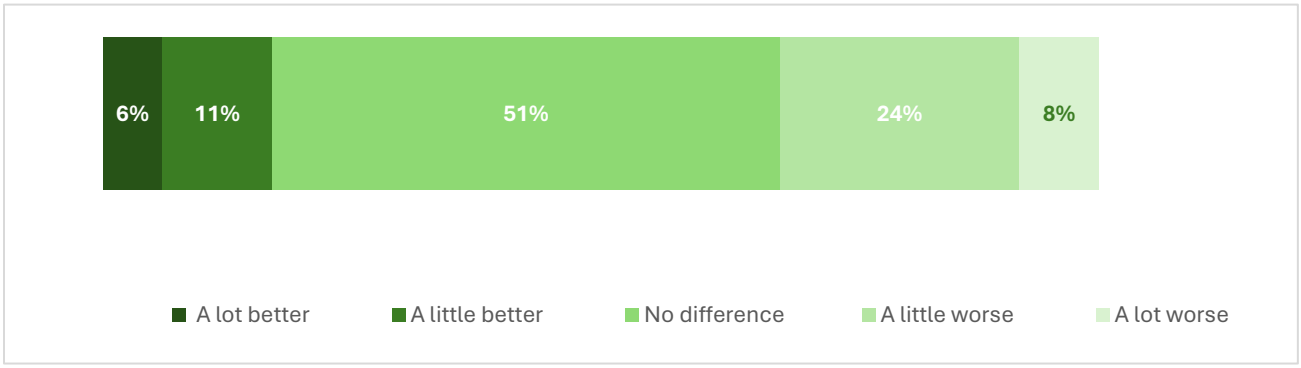


Figure 4. Caregiver Ratings of Virtual Appointments vs Similar (pre-pandemic) In-person Appointments for their Child with an IMD (n=55)

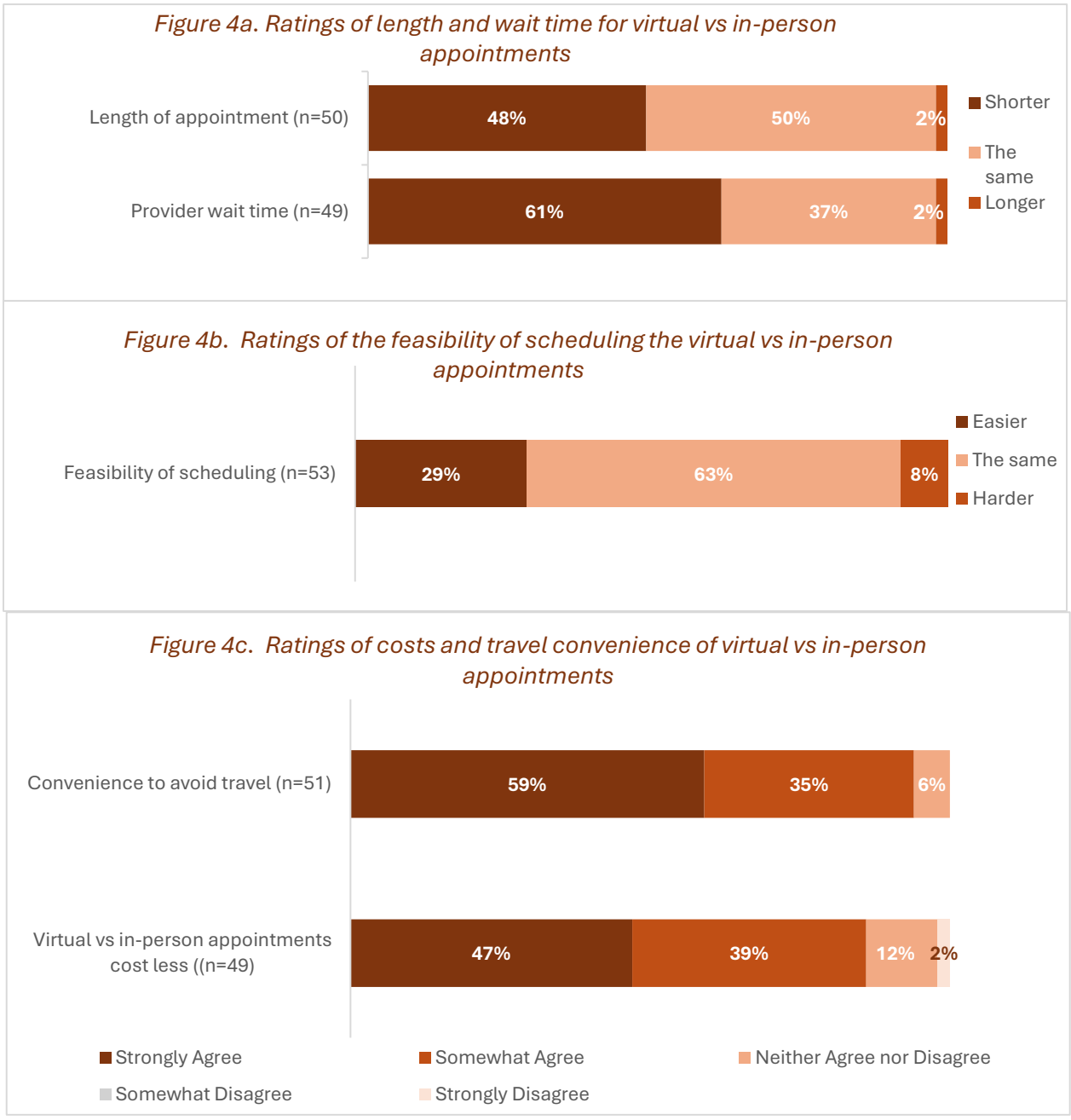


Figure 5 : Caregiver Ratings of Virtual Appointments vs Similar (Pre-pandemic) In-person Appointments for their Child with an IMD (n=55)

Figure 5a.Ratings of follow-up comprehension, chid comfortability and communication with provider during virtual vs in-person appointments

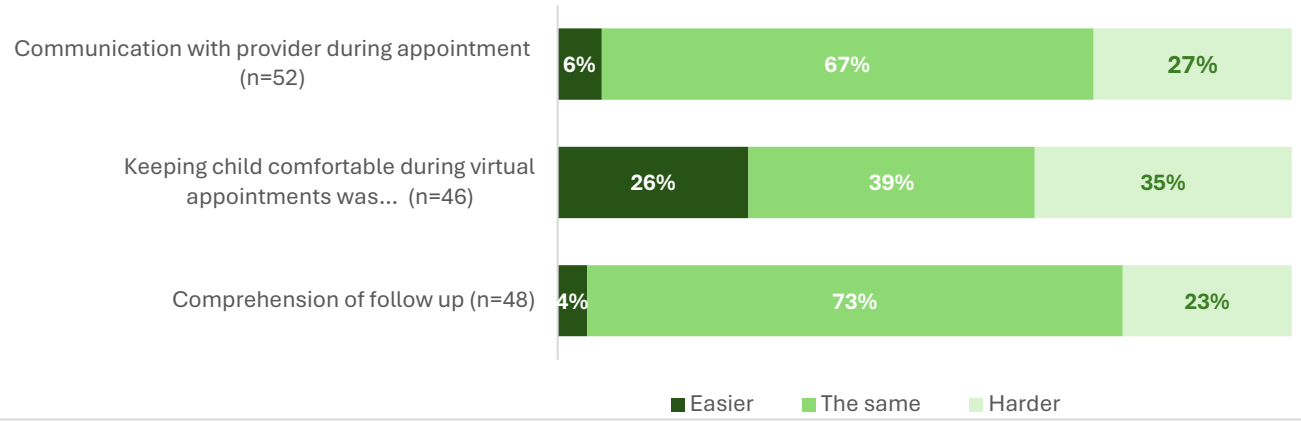


Figure 5b. Ratings of privacy during virtual vs in-person appointments

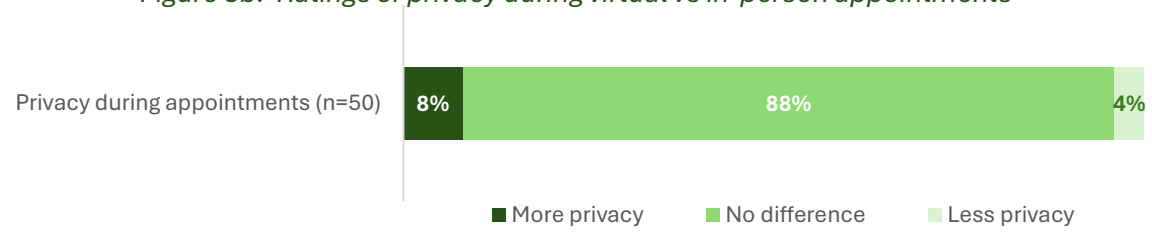


Figure 5c : Ratings of level of involvement during virtual vs in-person appointments

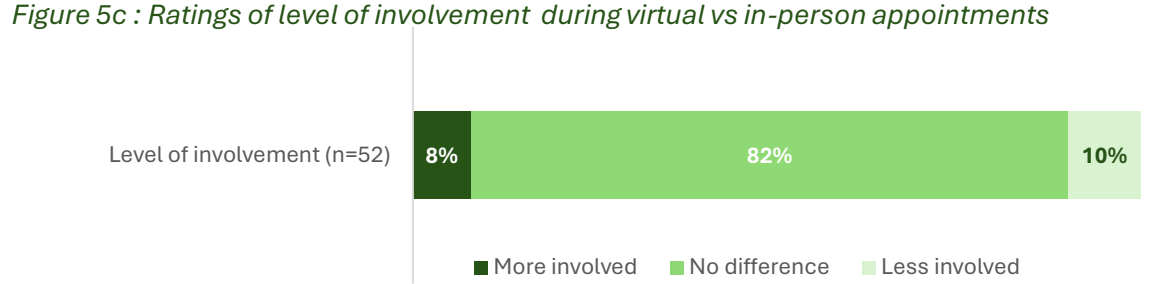
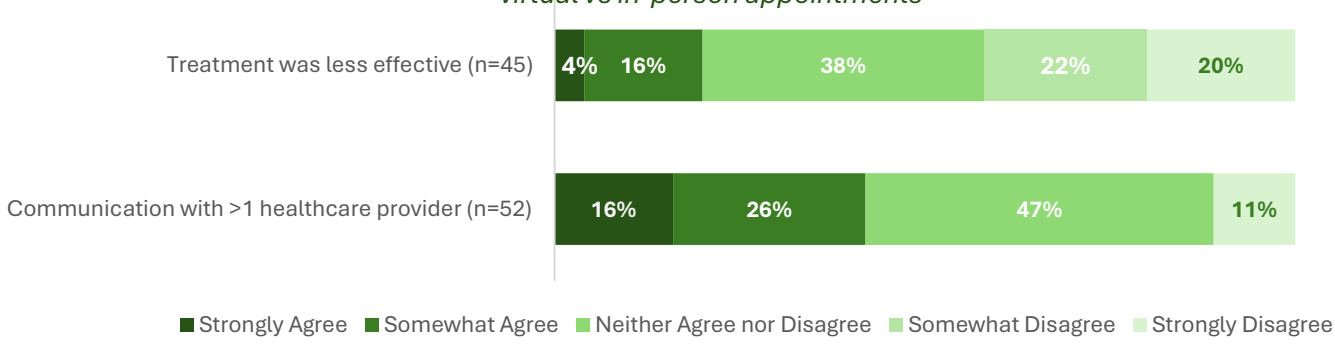


Figure 5d: Ratings of treatment effectiveness and communication with >1 provider during virtual vs in-person appointments



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Appendices

Appendix 1: Eligible IMD diagnoses and their classification for the cohort study

Clinical Course	Chronic and generally non-progressive (Group A)	Acute episodes of severe illness with or without accompanying chronic multisystem sequelae (Group B)	Progressive multisystem disease (Group C)
	Amino acid disorders: - pyridoxine dependent epilepsy - guanidinoacetate methyltransferase deficiency; galactosemia - Phenylalanine hydroxylase deficiency - homocystinuria; tyrosinemia type I	**Organic acid disorders:** ß-Ketothiolase deficiency; Glutaric acidemia type I; HMGCoA lyase Deficiency; Isovaleric acidemia; Methylmalonic acidemias; Propionic acidemia **Fatty acid oxidation disorders**: MCAD deficiency; VLCAD deficiency; Carnitine uptake defect; Long-chain 3-hydroxyacyl-CoA dehydrogenase deficiency; Trifunctional protein deficiency **Amino acid disorders**: Maple syrup urine disease **Urea cycle disorders**: Arginase deficiency; Argininosuccinic acidemia; Carbamoyl phosphate synthetase deficiency; Citrin deficiency; Citrullinemia; Hyperornithinemia-Hyperammonemia-Homocitrullinuria syndrome; Nacetylglutamate synthetase deficiency; Ornithine transcarbamylase deficiency **Other disorders:** - Multiple carboxylase/ biotinidase deficiency - Glycogen storage disease type 1	Other disorders: MPS Type I, Farber disease + Additional diagnoses adjudicated during the recruitment process (e.g., additional MPSs, …)

Appendix 2: Questions from the Baseline Questionnaire by Study Objective

Objective i: Caregivers' self-reported ratings of overall healthcare quality for their child with an IMD since the pandemic

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COVID-19 has changed the way that healthcare is provided. In general, how do you feel about the quality of your child's healthcare since the start of the pandemic (i.e., March 2020)?

O It's gotten much better
O It's gotten a little better

O It has not changed

O It's gotten a little worse
O It's gotten much worse

Objective ii: Caregiver perspectives on changes to the child's care induced by the pandemic

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COVID-19 has changed the way that healthcare is provided. In general, how do you feel about the quality of your child's healthcare since the start of the pandemic (i.e., March 2020)?

Over the past 6 months, how have the changes to health care and other services due to the pandemic affected your child's health care?

Because of the pandemic...

Check ALL that apply.

- ☐ One or more of my child's healthcare appointments or services were cancelled
- ☐ One or more of my child's healthcare appointments or services were delayed
- ☐ One or more of my child's healthcare appointments were changed from in-person to virtual (e.g., phone, video)
- ☐ I could not get to the lab, test centre, or pharmacy because their opening hours were reduced
- ☐ Only one primary caregiver was allowed to go with my child to a healthcare encounter
- ☐ None of the above

What services or therapies were cancelled?

Check ALL that apply.

- ☐ Appointment (at clinic or virtual) with a provider who was already part of child's care team
- ☐ Appointment (at clinic or virtual) with a new provider (new referral)
- ☐ Appointment at hospital for therapeutic treatment (e.g., enzyme replacement therapy)
- ☐ Appointment for laboratory tests or diagnostic imaging
- ☐ Home-based healthcare services
- ☐ Health assistance at school
- ☐ Other

Other - please specify: _____

What services or therapies were delayed?

Check ALL that apply.

- ☐ Appointment (at clinic or virtual) with a provider who was already part of child's care team
- ☐ Appointment (at clinic or virtual) with a new provider (new referral)
- ☐ Appointment at hospital for therapeutic treatment (e.g., enzyme replacement therapy)
- ☐ Appointment for laboratory tests or diagnostic imaging
- ☐ Home-based healthcare services
- ☐ Health assistance at school
- ☐ Other

Other - please specify: _____

Objective iii: Impacts of the pandemic and associated changes to care on the family and on the child's healthcare and status, from caregiver perspectives

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Since the start of the pandemic (i.e., March 2020), how has the pandemic affected your family?

Check ALL that apply.

- ☐ One or more primary caregivers has lost a job or had work hours reduced

- ☐ Our household income has decreased
- ☐ We spend more time managing my child's therapies at home
- ☐ We spend more time managing my child's diet at home
- ☐ We spend more time managing other parts of my child's health at home
- ☐ Other
- ☐ None of the above

Other - please specify:

Over the past 6 months, how have these changes to healthcare services due to the pandemic affected your child's CURRENT health or well-being?

Because of the pandemic....

Check ALL that apply.

- ☐ My child's physical health has gotten worse
- ☐ My child's mental health has gotten worse
- ☐ My child's social development has been set back
- ☐ We don't get any monitoring of my child's health
- ☐ We get less regular monitoring of my child's health Other
- ☐ None of the above - My child's health has not been affected

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Objective iv: Caregivers' pandemic-related concerns about their child's healthcare and wellbeing (e.g., avoidance of in-person care, difficulty, or concerns about difficulty in accessing medications)

How much do you agree/disagree with each statement for your child?

Because of the pandemic...

	Strongly agree	Somewhat agree	Neither agree nor disagree	Somewhat disagree	Strongly disagree	Not Applicable
Because of the pandemic, I do not want my child to have in-person medical appointments	○	○	○	○	○	○

During the pandemic, managing my child's IMD at home has been more difficult	○	○	○	○	○	○
During the pandemic, getting other healthcare-related needs for my child (e.g., supplies, medication) has caused me stress or anxiety	○	○	○	○	○	○
During the pandemic, I have taken public transportation or shared car services to take my child to in-person medical appointments. This has caused me stress or anxiety	○	○	○	○	○	○
I worry about my child getting covid-19	○	○	○	○	○	○
Compared to other children, my child is more at risk for COVID-19 complications because of their IMD	○	○	○	○	○	○

Objective v : Caregiver ratings of virtual appointments relative to similar (pre-pandemic) in-person.

Compared to similar in-person encounters before the pandemic (i.e., March 2020), how much do you agree with the following statements?

Overall, how did the virtual appointment(s) compare to similar in-person appointments before the pandemic (i.e., March 2020)?

O They were a lot better

O They were a little better

O No difference

O They were a little worse

O They were a lot worse

Compared to similar in-person appointments before the pandemic (i.e., March 2020)...

	Shorter	The same	Longer	I do not know
..the virtual appointment(s) were [bl_pandemic_length].	○	○	○	○
...on the day of the virtual appointment(s), the wait for the provider was usually [bl pandemic_wait].	○	○	○	○

	Easier	The same	Harder	I do not know	Not applicable
...communicating with the provider during the virtual appointment(s) was [bl_pandemic_comm].	○	○	○	○	○
...scheduling the virtual appointment(s) was [bl_pandemic_sched].	○	○	○	○	○
...understanding what steps would take place after the virtual appointment(s) was [bl_pandemic_follow].	○	○	○	○	○

Compared to similar in-person encounters before the pandemic (i.e., March 2020), how much do you agree with the following statements?

	Strongly agree	Somewhat agree	Neither agree/disagree	Somewhat disagree	Strongly disagree	Not Applicable
It was convenient to avoid travelling.	○	○	○	○	○	○
It cost us less(out-of-pocket costs).	○	○	○	○	○	○
The treatment was less effective .	○	○	○	○	○	○
We were able talk to more than 1 provider at the same time.	○	○	○	○	○	○

How was your privacy during the virtual appointment(s), compared to similar in-person appointments before the pandemic (i.e., March 2020)?

O Had more privacy

O No difference

O Had less privacy

O I don't know

Did you feel more or less involved in decision-making about your child's health during the virtual appointment(s), compared to similar in-person appointments before the pandemic (i.e., March 2020)?

O More involved

O No difference

O Less involved

O I don't know

If the virtual appointment(s) were different in other ways compared to in-person appointments that took place before the pandemic (i.e., March 2020), please describe in the space below.

Appendix 3: Additional results

TABLE A3.1. Caregiver Perspectives on the Impact of the COVID-19 Pandemic on Health Care for their Child with an IMD by Age and IMD category (n=70)

Age		Age 0-4yrs			Age 5-13yrs			
IMD category	TOTAL	[a]Acute (n=14)	[b]Chronic (n=15)	[c]Progressive (n=8)	[a]Acute (n=15)	[b]Chronic (n=7)	[c]Progressive (n=11)	Cramer's V [Lower 95% CI, Upper 95%CI]
Changes to health care attributed to the pandemic (check all that apply, n=70)								
Any changes (from options below)	**63 (90)**	13 (93)	14 (93)	8 (100)	12 (80)	5 (71)	11 (100)	0.31[0.08, 0.51]
≥1 HCA† cancelled / delayed or Lab, test centre, pharm inaccessible*	**46(66)**	9(64)	12(80)	5(63)	8(53)	3(43)	9(81)	0.28 [0.05, 0.48]
≥1 HCA† changed to virtual	**55(79)**	10(71)	12(80)	7(88)	10(67)	5(71)	11(100)	0.28 [0.04, 0.48]
Restricted accompaniment during HCA†	**42(60)**	10(71)	11(73)	6(75)	6(40)	1(14)	8(73)	0.41 [0.20, 0.59]

a: Acute and episodic disease
b: chronic: Chronic and generally non-progressive disease
c: progressive: Progressive multi-system disease
†HCA: Healthcare Appointment
* Lab / test centre / pharmacy not accessible due to opening hours being reduced

TABLE A3.2: Caregiver Ratings on the Nature of Cancelled and Delayed Services due to the Pandemic.

Caregiver reported cancelled services and therapies due to the pandemic (check all that apply, n=20)	
Appointment (at clinic or virtual) with provider already part of child's care team	16(80)
Appointment (at clinic or virtual) with a new provider (new referral)	3(15)
Therapeutic treatment appointments at the hospital	1(5)
Laboratory tests or diagnostic imaging	4(20)
Home-based health care services	4(20)
Health assistance at school	2(10)
Other	2(10)
Caregiver reported delayed services and therapies due to the pandemic. (check all that apply, n=38)	
Appointment (at clinic or virtual) with provider already part of child's care team	29(76)
Appointment (at clinic or virtual) with a new provider (new referral)	10(26)
Therapeutic treatment appointments at the hospital	2(5)
Laboratory tests or diagnostic imaging	12(32)
Home-based health care services	6(16)
Health assistance at school	4(11)
Other	5(13)

CHAPTER 4: INTEGRATED DISCUSSION CHAPTER

4.1 Summary of Findings

This thesis project aimed to understand available evidence about experiences with virtual care among children with high care needs and their caregivers. We focused on children with medical complexities and those with inherited metabolic diseases (IMDs), through two studies, respectively: a scoping review of published studies about virtual care for children with medical complexities; and a secondary data analysis of a cross-sectional survey of caregivers of children ≤12 years of age with IMDs in Canada.

Scoping Review

In our scoping review, we synthesised and mapped existing literature on virtual health care experiences among children with medical complexity and their caregivers (Chapter 2). We found considerable heterogeneity in the methods, frameworks and instruments used to study virtual care experiences. A notable gap was the limited examination of how sociodemographic factors may influence virtual care experiences, considered by the authors in only 5 of the 34 studies we reviewed. We also found that several domains considered to be important aspects of patient- and family-centred care, such as family involvement, emotional support, and care coordination, have received little attention in this body of literature despite their potential importance for children with medical complexity, relative to overall satisfaction with virtual care and its impact on care accessibility, which were considered more frequently. We concluded that the lack of consistency in use of frameworks and tools, and the gaps in consideration of some important areas of care experiences, highlights the need for conceptual guidance to promote more harmonized and meaningful evaluations of virtual care experiences for children with medical complexity and their caregivers.

Secondary Data Analysis

Complementing the mapping of existing evidence from the scoping review, in the secondary analysis we examined caregiver-reported experiences with virtual care for children aged ≤12 years with IMDs in Canada (Chapter 3). Using cross-sectional data from a baseline survey within a Canadian cohort study of 71 children with IMDs,[1] we described caregivers' perceptions of the quality of health care received for their children during the COVID-19 pandemic and their experiences with a transition to virtual care. While many participants reported that virtual care provided benefits such as improved ability to talk to more than one provider, reduced travel time and costs, several participants also indicated concerns related to communication with providers and maintaining child comfort, and they also noted a reduction in monitoring of the health of their child.. These findings revealed the real-world complexities of health care for a child with elevated health needs in an environment characterized by both a transition to virtual care and associated pandemic-related disruptions and impacts.

4.2 Interpretation in the Context of Related Literature

The COVID-19 pandemic necessitated a rapid shift to virtual modalities of health care delivery.[2] The studies we synthesised in our scoping review spanned the pre-pandemic and pandemic time periods, with 21 of the reviewed 34 studies collecting at least some of their data prior to 2020, capturing a time in which virtual care was present but not yet ubiquitous. By contrast, our secondary analysis was conducted during the pandemic period. Some of the survey questions were about virtual care during the pandemic specifically, caregivers who participated were reporting on their child's virtual care in the context of the many disruptions and impacts brought about by COVID-19 and associated public health measures that altered routine care and necessitated remote care measures. This distinction reflects a pattern seen in other pandemic-

related studies, which similarly document that while many families benefitted from telehealth, other concerns such as communication quality, and child comfort, were apparent.[3–6]

These two studies present complementary perspectives: the scoping review identified general patterns in the existing literature on virtual care experiences for children with high health care needs and their caregivers, and the secondary analysis illustrated these concepts in the specific context of a public health crisis (COVID-19 pandemic). Collectively, our findings corroborate and extend previous studies describing the potential benefits and challenges of virtual care for children with high health care needs.[7–9] Some of the benefits that have been often reported are lowered costs, reduction in travel burdens and improved access.[7–9] These were also noted advantages of virtual care endorsed by caregivers in our secondary analysis. With respect to challenges, one challenge described in the literature is the digital divide, which refers to inequities in access to internet and technology, potentially associated with education and socioeconomic status as well as rurality. [10–16] A recent systematic review investigated sociodemographic factors influencing the use of virtual care in adults and children with chronic diseases.[16] That review highlighted that higher socioeconomic status, younger age, and higher levels of education were associated with greater engagement in virtual care, suggesting the role of sociodemographic factors in shaping virtual care experiences. These findings align with prior research on pediatric virtual care that emphasised the need to address how these sociodemographic factors pose potential disparities and barriers to virtual care access[17–19]. For instance, a recent review of pediatric telehealth use during the COVID-19 pandemic among children with limited English proficiency found a dearth of studies that investigated the impact of

language barriers in virtual care[18]. Our scoping review similarly revealed that few studies have focused on sociodemographic factors associated with virtual care experiences.

Our findings contribute to this emerging literature about health inequities by illustrating how virtual care can address specific challenges faced by families of children with high health care needs. For example, nearly half of the participants in our secondary analysis reported traveling over an hour to the metabolic clinic for in-person care, highlighting potential difficulties associated with accessing care due to the need for frequent travel. A reduced need to travel was valued by caregivers, suggesting virtual care's potential to alleviate such difficulties and promote more equitable access to specialty care. From the scoping review, while few studies focused on sociodemographic factors, access to care itself was considered in 16 of the 34 studies, illustrating a recognition of its importance among researchers. These findings echo existing research showing that virtual care is particularly impactful for families in rural or remote settings, where long distances to specialty clinics pose significant barriers to timely and accessible care.[20,21]

While there is broad consensus on the potential for virtual care to enhance access and reduce costs,[22,23] there remains a clear need to ensure patient- and family-centred approaches are carefully integrated into these care models for children with high health needs.[24–26] Our scoping review emphasised that few studies captured family-centred domains such as emotional support, family involvement and care coordination, which are known to be priorities for high-needs groups and their families. [27–33] This reflects findings observed in the telehealth literature and in

health services research more broadly, in which satisfaction and usability measures predominate, often eclipsing deeper explorations of how families experience remote care. [34–38]

In our scoping review, we concluded that the lack of consistency in the use of frameworks and tools highlights the need for conceptual guidance to promote more harmonized evaluations of virtual care experiences. In our review, 12 of 34 studies (35%) collected data exclusively during the pandemic; some of these evaluations may have been implemented rapidly as a part of an emergent response to the COVID-19 pandemic, rather as pre-planned or systematically developed models of care.[39,40] This context may have contributed to the variability we observed in evaluation approaches. However, the majority of studies in our review collected data preceding the pandemic, highlighting ongoing challenges in consistent conceptualization and evaluation of virtual care experiences even outside a crisis context. As virtual care continues to evolve into a more permanent and structured component of pediatric health care, synthesising the current landscape of literature is timely, and provides a foundation for future evaluations that are conceptually guided.

4.3 Strengths and Limitations

This thesis research project examined the virtual care experiences of children with high health care needs and their families, synthesizing and mapping the literature through a scoping review while also offering specific insights through a secondary data analysis of a cross-sectional survey conducted within a recent Canadian cohort study. The scoping review provided a wide-ranging view of virtual care research for children with high health care needs both before and during the COVID-19 pandemic, thereby capturing changing practice patterns and identifying methodological gaps. Meanwhile, the secondary data analysis offered real-world insights into families' experiences during the pandemic, highlighting how rapid shifts to virtual care affected

children with IMDs. The diverse sample of caregivers from various Canadian provinces, representing children with a range of IMD diagnoses, added to that study's relevance and generalizability to similar pediatric populations. Despite these strengths, some limitations should be noted. The scoping review included only English language publications, possibly excluding important perspectives from non-English speaking contexts. For the secondary data analysis, the sample was skewed toward participants with higher household incomes and more highly educated caregivers and was restricted to those who could respond to surveys in English, also limiting generalizability to more diverse populations. For instance, families who may rely on translation services or interpretation during virtual care encounters were excluded. Furthermore, virtual care is a heterogeneous construct, and the studies reported here did not delve into the different modes of virtual care and how they are experienced. There is also heterogeneity in the needs of different populations of children with medical complexity and those with IMDs. It is possible that there are important differences in experiences with care within these groups but our small sample size in the secondary analysis limited our ability to fully describe variations by child age or IMD diagnoses. Additionally, the pandemic's impact on acute exacerbations of medical conditions could have influenced caregiver satisfaction with virtual care. While fewer infections and acute health issues may have reduced the need for healthcare interactions, increasing acute issues post-pandemic may affect caregiver satisfaction or perceptions of virtual care, which is important to consider in future studies. Together, these limitations highlight the need for future studies to incorporate more diverse samples, include non-English publications and

participants, and apply standardized metrics that can systematically evaluate family-centred outcomes.

4.4 Implications for Future Research and Health Care Provision

This thesis produced several insights of relevance to researchers, clinicians and policymakers seeking to optimize virtual care for pediatric populations with elevated health care needs. From a research perspective, we identified a need for harmonized approaches to evaluating virtual care experiences, including frameworks and questionnaires. The selection of frameworks and tools should incorporate insights about what is most meaningful from the perspectives of patients and caregivers, health care providers, and health system decision-makers, including domains that have been identified as important to patient- and family-centred delivery of pediatric care, such as emotional support, involvement of family members, and care coordination. [27–33] For example, the survey we analyzed in Chapter 3 incorporated questions guided by the Picker Institute's Principles of Patient-Centred Care, which did not address technological aspects of virtual care, such as reliability, interface quality, and ease of use. These aspects emerged in the scoping review and are covered by other survey tools like the Telehealth Usability Questionnaire (TUQ), which are important for understanding the user experience with the technology itself.[41] Another promising pediatric virtual care framework in this area is one proposed by Dulude and colleagues, which expands on existing virtual care evaluation models.[42] Harmonizing approaches to evaluating virtual pediatric care with engagement of knowledge users including children and caregivers would enable researchers and clinicians to compare and synthesize findings across studies and to identify performance benchmarks linked to metrics meaningful to children and their caregivers.

The scoping review also highlighted that few studies have considered how sociodemographic factors such as place of residence, race/ethnicity/culture/language, occupation,

gender/sex, religion, education, socioeconomic status, age and social capital [43,44] shape virtual care experiences for children with medical complexity and their caregivers. As noted, the participating caregivers of children with IMDs in the secondary analysis were not representative of a diverse population, particularly with respect to language barriers that could be important to virtual care experiences. Future research should prioritize these considerations and seek to understand the needs of diverse families.

The findings of the secondary analysis reported in Chapter 3 revealed both strengths (e.g., convenience, reduced travel costs) and challenges (e.g., communication barriers, reduced health monitoring) associated with virtual care during the pandemic. As virtual care continues in the context of removal of pandemic restrictions, it will be essential to learn from these experiences. For example, health system administrators and clinical providers could consider the importance of continuing to offer virtual care options to the benefit of children with high health care needs whose caregivers have intensive home care responsibilities, particularly those who must travel longer distances to care; and also, the importance of identifying potential communication and access challenges. Future research could seek to disentangle challenges families face that are related to virtual care generally versus those that are specific to virtual care in the context of the pandemic, to ensure that ongoing development and delivery of virtual care is inclusive and effective.

4.5 Final Conclusions

Children with high health care needs, including those with IMDs and medical complexities, navigate a health care landscape that demands substantial coordination, sustained family engagement and extensive, ongoing care. First, the scoping review comprehensively synthesised the nature of existing evidence and identified gaps in measurement approaches,

guiding frameworks, and sociodemographic analyses. Second, the secondary data analysis described the virtual care experiences of a specific group of children and caregivers in the context of the COVID-19 pandemic, highlighting benefits and challenges with virtual care . Integrating these two studies highlights the need to evaluate underexplored domains of virtual care experiences pertinent to this population and proposes the use of harmonized and equity-focused evaluation frameworks and tools that comprehensively capture the full breadth of virtual care experiences for children with high health care needs.

Ultimately, this work can inform clinicians, researchers, and policymakers striving to improve health care experiences and outcomes for children with high health care needs and their caregivers. Centring the voices of children and families and working toward cohesive approaches for evaluating virtual care will contribute to equitable, inclusive, high-quality, and responsive health care systems that support the well-being of these vulnerable pediatric populations.

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