

DEVELOPMENT AND INITIAL TESTING OF A CARE COORDINATION MHEALTH
APPLICATION FOR FAMILIES OF CHILDREN WITH DOWN SYNDROME

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ABSTRACT

Bethany Skelton Cosgrove: Development and Initial Testing of a Care Coordination mHealth Application for Families of Children with Down Syndrome
(Under the direction of Marcia Van Riper, PhD, FAAN)

Children with Down syndrome (DS) often require health care from multiple providers and are at increased risk for missed care coordination, potentially leading to gaps in care. Care coordination or the linking together of health information and members of health care teams, is vital to ensure components of health care are not missed. This responsibility, along with maintaining health information, often falls to a family caregiver. A personal health record (PHR) is the health information maintained by caregivers outside of the medical record and helps caregivers manage care coordination. To support caregivers, we needed to better understand their care coordination needs. This dissertation was designed to describe the care coordination needs of caregivers of children with DS and design a mHealth application addressing those needs to be used as a PHR. A scoping review of literature explored prior research on the care coordination needs of families of children with DS and found that communication, information, and utilization of resources were critical components of successful care coordination. A mixed methods study investigated care coordination needs and technology use of caregivers and health care providers. Both caregivers and health care providers reported valuing communication as well as the ability to manage health information and resources, monitor adherence to current health care guidelines for children with DS, and coordinate appointment schedules. These findings were then used to develop a prototype of a mHealth app (321Connect) to serve as a PHR and support family

caregivers' care coordination needs. 321Connect was developed using principles of user centered design with end users, caregivers of children with DS, participating in each phase of the design process. Initial usability testing elicited positive feedback from family caregivers who reported 321Connect had strong potential for supporting caregivers in coordinating care and maintaining a PHR.

To my support team and cheerleaders: my mom, stepdad Miff, and my partner Jeff. Especially to Jeff, whose answer always to “Can I do this?” was “You’re actively doing it!”

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LIST OF ABBREVIATIONS

AAP	American Academy of Pediatrics
App	Application
DS	Down syndrome
EHR	Electronic Health Record
FaMM	Family Management Measure
FCMH	Family centered medical home
FMSF	Family Management Style Framework
HCP	Health care provider
NSCH-D	National Survey of Children's Health- Section D
PHR	Personal Health Record

CHAPTER 1: INTRODUCTION

Background and Significance

Navigating the health care system can be challenging for any family, particularly families raising a child with a genetic condition such as Down syndrome (DS). One way to support families in meeting their health care needs is to provide care coordination. Care coordination involves linking health care providers, community resources, and families to manage the health care needs of a child within the child's primary healthcare practice (McAllister et al., 2007; Agency for Healthcare Research and Quality, 2021). Despite evidence that care coordination improves health outcomes, children with DS are at higher risk for missed care coordination than other children with special health care needs (McGrath et al., 2011; Phelps et al., 2012). This is likely in part due to the fact that children with DS on average have three times as many outpatient healthcare appointments (McGrath et al., 2011; Phelps et al., 2012; Williams et al., 2017). These outpatient appointments are increased because of the higher risk for co-occurring conditions in children with DS, such as congenital heart disease or hypothyroidism, which often require specialty care by multiple providers (Council on Children with Disabilities and Medical Home Implementation Project Advisory Committee, 2014; Skotko et al., 2012; Williams et al., 2017). These specialty appointments are critical to provide screening and treatment of co-occurring conditions (Bull et al., 2011, Williams et al., 2017). Care coordination ensures that children with DS receive the care they need from the appropriate provider (Uppal et al., 2015).

Care coordination is not limited to specialty care needs but also includes managing routine pediatric health care needs such as developmental concerns and immunization updates (Bull et al., 2011). To manage co-occurring conditions as well as health maintenance, the American Academy of Pediatrics (AAP) created age-based guidelines for the care of children with DS (Bull et al., 2011). It is estimated that only 9.8% of children with DS are up to date in all recommended components of care coordination within the guidelines (Skotko et al., 2012). While originally designed for providers, given high demands placed on primary care professionals, care coordination is at risk for being missed, leaving the child's primary caregiver(s) responsible for coordinating the services needed to address the full spectrum of their child's health care needs (Ranade-Kharkar et al., 2017). Primary caregivers are known to carry thick binders of health information to their visits with the various providers involved in their child's care in order to manage care coordination and to ensure they have access to all the information that providers might request (Allshouse et al., 2018). However, little is known about strategies employed by caregivers of children with DS to manage care coordination needs. To reduce risk of missed health care for a child with DS, better methods are needed to initiate and sustain caregiver tracking of care coordination so that it aligns with AAP DS care guidelines and addresses the unique needs of the child with DS and the child's family.

One way that primary caregivers can participate in the planning and tracking of care coordination and health information is through a personal health record (PHR) (Ranade-Kharkar et al., 2017; Gallo et al., 2009). A PHR contains the health information and data managed by a family or caregiver that is not linked to the electronic health record (EHR) (Dameff et al., 2019). EHR's while critical in managing health information, differ in that they are housed within a health care system and maintained by providers (Dixon et al., 2018). While EHR's have

improved information access to providers within the same health system, providers report continued issues from health care systems using multiple EHR platforms and challenges sharing information (Richardson et al., 2015). Additionally, children with DS typically see providers in several different specialty clinics, which may be in different health systems, thus limiting access to all the child's relevant health information in one place (Williams et al., 2017).

PHR's are a way for caregivers to maintain health information for care coordination and to document patient reported outcome data. For example, health data related to sleep, feeding/growth and developmental changes are used to help diagnose and manage co-occurring conditions (Bull et al., 2011; Zhou et al., 2019). One portable way to maintain a PHR is through the use of mHealth application (app) technology (Calderon et al., 2015; Tozzi et al., 2015). PHR information is then readily accessible at clinic appointments, and sharable with providers (Tozzi et al., 2015). A PHR mHealth app gives caregivers the ability to link health care providers, community resources, and family members, enabling them to coordinate the care needs of a child with DS across providers and care systems (Dixon et al., 2018). Studies have shown that primary caregivers are open to the idea of using mHealth apps to support management of their child's chronic condition and recognize that technology may facilitate improved information access and communication (Calderon et al., 2015; Liu et al., 2016; Quigley et al., 2014; Tozzi et al., 2015).

Today, an estimated 85% of Americans own a smartphone, making mHealth apps widely available and disseminated (Ali et al. 2016; Pew Research Center, 2021). mHealth apps also have distinct advantages to be used in care coordination because of the accessibility and portability of health information (Klasnja & Pratt, 2012). mHealth apps have been shown to be a useful tool in the management and care coordination of other chronic conditions such as

asthma or diabetes (Dzubar et al., 2015; Morita et al., 2019; Padman et al., 2013). In addition to improving the management of diabetes care, mHealth app usage positively affected health outcomes. Caregivers using a mHealth app to track blood glucose levels for their children, showed an improvement in the child's HgbA1C levels (Garabedian et al., 2015). A mHealth app tailored for families following a bone marrow transplant, helped to foster self-efficacy and independence by giving families a way to track dietary needs, medications, and lab findings (Maher et al., 2016). Self-efficacy and increased engagement in communication with providers was also found in using mHealth to manage care plans for children with medical complexity (Ming et al., 2018; Opiari-Arrigan et al., 2020). These findings support the potential use of mHealth to manage health needs in a DS population.

While mHealth apps have been shown to have the potential to improve self-management in other chronic conditions, there has been limited development of research based mHealth applications designed for families of children with DS (Bathgate et al., 2017; Choi & Van Riper, 2019; Whitehead & Seaton, 2016). Currently only two mHealth applications were identified for specific use in a DS population, one solely addressing dietary needs of adolescents and young adults with DS and the other using mHealth to engage in therapeutic conversations to address family adaptation (Bathgate et al., 2017; Choi & Van Riper, 2019). Families of children with DS have shown an openness to the use of mHealth apps due to the potential of taking a more active role in health care management (Tozzi et al., 2015). Given complexity and importance of care coordination, caregivers of children with DS would likely benefit from a mHealth app, to support health information tracking and access.

Equally important to identifying the potential need for mHealth apps to help manage care coordination, is the mHealth app design process. One way to increase the likelihood of a

mHealth app being adopted by families is to employ user centered design methods in the development of mHealth technologies (McCurdie et al., 2012). User centered design is an iterative process, endorsed by the World Health Organization task force for mHealth technology, to understand and apply user needs to the design process by having users involved in each step of the design (Kao & Liebovitz, 2017; McCurdie et al., 2012; Nielsen & Mack, 1994; World Health Organization, 2011). User centered design includes concept design, prototype development, and evaluation (McCurdie et al., 2012). User requirements are the features and appearance of the mHealth app that are preferred by the end user and are identified through the steps of user centered design (McCurdie et al., 2012; Nielsen & Mack, 1994). The end users in this case are caregivers of children with DS. However, it is also important to involve others who may interact with the mHealth app, such as health care providers in its design and evaluation of its acceptability and usability (Canter et al., 2018; McCurdie et al., 2012). Engaging end users in the user centered design process increases the likelihood of developing a user friendly, easy to use app (Schnall et al., 2016).

Given the complexity of managing the health care needs of a child with DS and the rapidly evolving use of information and communication technology in health care, this dissertation sought to identify care coordination needs of families and develop and evaluate a prototype mHealth application to support caregivers' care coordination and information management efforts. The aims of this dissertation were to:

Specific Aim 1: Complete a scoping review of published research to assess care coordination needs in families of children with DS.

The following questions were addressed: (1) What are the care coordination needs of primary caregivers of children with DS? (2) What strategies and resources do primary caregivers use to address their care coordination needs?

Specific Aim 2: Examine primary caregivers' and health care providers' perspectives on desirable characteristics and features of mHealth application content and application use outcomes that could be used to develop an algorithm for the design of a mHealth application using a mixed methods analysis.

Specific Aim 3: Using the results of Aims 1 and 2, develop and evaluate a mHealth prototype.

3a: To develop a prototype for a mHealth application based on user centered design findings from Aim 2.

3b: To complete initial usability testing of a mHealth application prototype.

Theoretical Framework

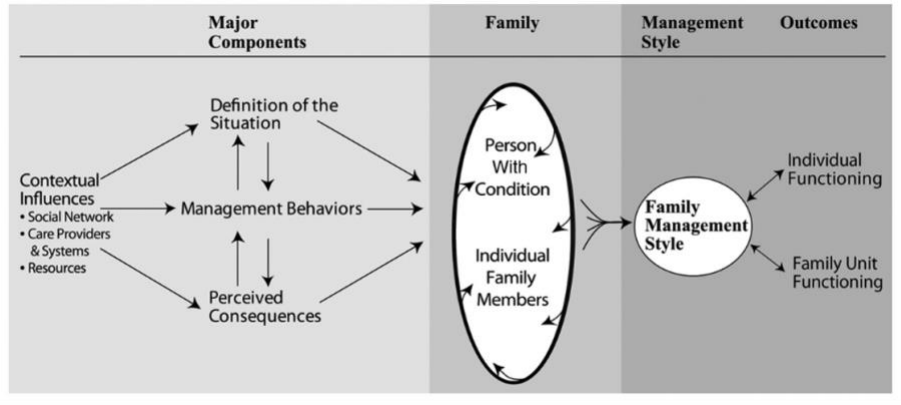
With a focus on family management and theoretically grounded in the Family Management Style Framework (FMSF), a mHealth application designed using user centered design principles could provide a platform for supporting primary caregivers' care coordination efforts and their ability to maintain up-to-date health information for their child in a PHR (Knafl et al., 2012; McCurdie et al., 2012; Nielsen & Mack, 1994; Williams et al., 2017; Zhou et al., 2019). The FMSF has been used previously to identify information management strategies and management behaviors with regards to a genetic diagnosis (Gallo et al., 2009). Although there is evidence of the efficacy of mHealth apps in other chronic conditions, little is known about how caregivers of children with DS may use mHealth apps and factors contributing to their acceptability (Dzubar et al., 2015; Maher et al., 2016; Ming et al., 2018; Morita et al., 2019;

Opipari-Arrigan et al., 2020; Padman et al., 2013). Therefore, this dissertation was grounded in both the FMSF and the principles of user centered design in identifying care coordination and information management needs as well as identifying features related to mHealth app design and use (Knafl et al., 2012; Nielsen & Mack, 1994).

The FMSF identifies the central components of family management of a child's chronic health condition (Figure 1.1) (Knafl et al., 2012). For this dissertation, the focus was on the contextual factors and major components of the FMSF. The contextual factors include the family's social network and access to health care and resources that may contribute to family management (Knafl et al., 2012). These contextual factors as well as individual and family factors were used in the analysis of articles for the scoping review in Aim 1. Contextual factors were also used in the development of questions for the qualitative interviews in Aim 2. Further, the major components of definition of the situation, management behaviors, and perceived consequences were used in the development of interview questions. Informed by the FMSF, three subscales of the Family Management Measure (FaMM) were used to determine if demographic and care coordination needs were predictors of family management ability (Knafl et al., 2011, Knafl et al., 2012). These findings will be used to identify families who may benefit from aspects of care coordination in future studies piloting the use of the mHealth app.

Figure 1.1

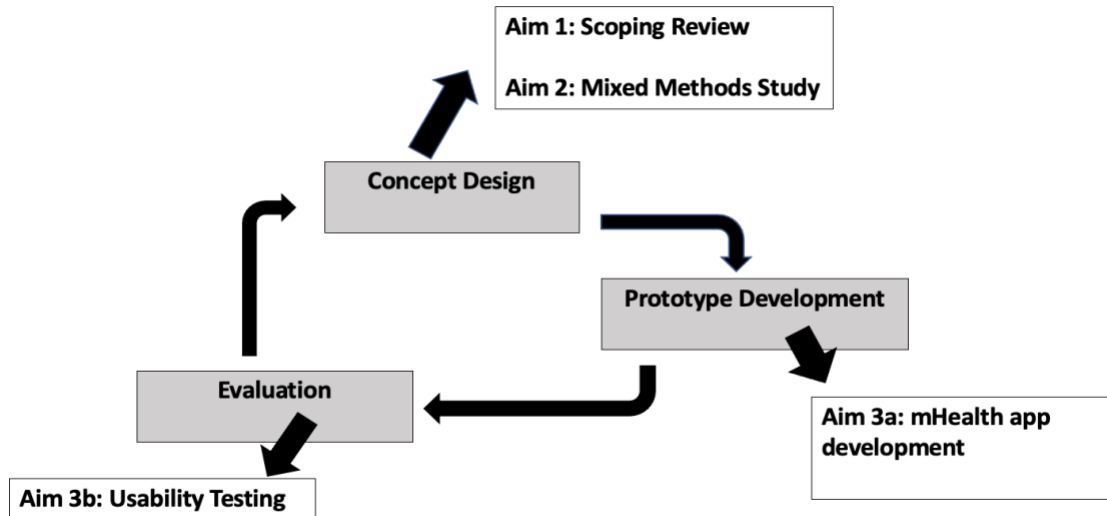
Family Management Style Framework



In conjunction with the FMSF, components of user centered design structure each of the aims in the development of a mHealth app (Figure 1.2) (McCurdie et al., 2012). User centered design consists of three parts: concept design, prototype development and evaluation. Concept design identifies the user requirements and the proof of concept related to mHealth app design (McCurdie et al., 2012; Nielsen & Mack, 1994). Concept design was included as part of study Aims 1 and 2 through the scoping review and the mixed methods analysis. The data collected in Aim 1 and 2 were then used to design the prototype of the mHealth app as part of Aim 3a. Finally, evaluation of the mHealth app prototype addressed Aim 3b.

Figure 1.2

Study Aims addressing User Centered Design



MANUSCRIPTS

This is a three-manuscript dissertation. Chapter 1 is an introduction to care coordination needs of families of children with DS as well as the use of mHealth applications in chronic condition management. Chapter 2 is a scoping review addressing care coordination needs and use of mobile technology to support condition management and care coordination among families of children with DS (Arksey & O'Malley, 2005; Tricco et al., 2018). Chapter 3 presents the results from a sequential mixed-methods study to examine caregiver and health care provider perspectives on care coordination needs and desirable characteristics of a mHealth application. Findings reported in this paper include results from a survey of primary caregivers of children with DS concerning care coordination needs and use of mobile technology to support condition management and qualitative interviews of caregivers of children with DS and health care

providers to elicit further information regarding care coordination management and desirable content for a mHealth app.

Chapter 4 presents findings from the development of the mHealth prototype as well as findings from initial usability and acceptability testing. Participants completing usability and acceptability testing included caregivers of children with DS. As end users, caregivers of children with DS, also completed qualitative interviews addressing the content included in the mHealth app.

Chapter 5 is a synthesis and discussion of findings across all three aims of the dissertation. Also included in Chapter 5 are implications for future research and considerations for future pilot testing of the mHealth app. The titles of the chapters are as follows:

Chapter 2: Care Coordination Needs of Families of Children with Down Syndrome: A Scoping Review to Inform Development of mHealth Applications for Families

Chapter 3: A Mixed Methods Analysis of Care Coordination Needs and Desirable Features of a mHealth Application to Support Families of Children with Down Syndrome

Chapter 4: Initial Usability Testing of a mHealth Application for the Management of Personal Health Record for Families of Children with Down Syndrome

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CHAPTER 2: CARE COORDINATION NEEDS OF FAMILIES OF CHILDREN WITH DOWN SYNDROME: A SCOPING REVIEW TO INFORM DEVELOPMENT OF MHEALTH APPLICATIONS FOR FAMILIES¹

SUMMARY

Care coordination is a critical component of health management aimed at linking care providers and health information-involved care management. Our intent in this scoping review was to identify care coordination needs of families of children with Down syndrome (DS) and the strategies they used to meet those needs, with the goal of contributing to the evidence base for developing interventions using a mHealth application (mHealth apps) for these families. Using established guidelines for scoping reviews, we searched 5 databases, yielding 2,149 articles. Following abstract and full text review, we identified 38 articles meeting inclusion criteria. Studies incorporated varied research designs, samples, measures, and analytic approaches, with only one testing an intervention using mHealth apps. Across studies, data came from 4,882 families. Common aspects of families' care coordination needs included communication and information needs and utilization of healthcare resources. Additional themes were identified related to individual, family, and healthcare contextual factors. Authors also reported families' recommendations for desirable characteristics of mHealth apps that addressed the design of a personal health record, meeting age-specific information needs, and ensuring

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access to up-to-date information. These results will further the development of mHealth apps that are tailored to the needs of families with a child with DS.

INTRODUCTION

Care coordination is critical for the management of care for any child with a chronic condition, but particularly for those with a genetic condition that could span multiple body systems, such as children with Down syndrome (DS). Management of the care provided by members of the health care team from a variety of health care settings is called care coordination (McAllister et al., 2007). Care coordination is a central component in guidelines set forth by the American Academy of Pediatrics (AAP) policy on family centered medical home and has been shown to improve family-centered outcomes (Turchi et al., 2009). Researchers have defined essential components to be included in successful implementation of care coordination in primary care; these include being family-centered and comprehensive in nature to meet both health and psychosocial needs (Schor, 2019). As a part of care coordination, it is important to include all persons who may be involved in the management of care for a child. This includes community service providers such as physical and speech therapists as well as those involved in the management of individual educational plans (IEP) (Moreno, 2019).

Care coordination is critical to ensuring adequate management of co-occurring conditions with DS. The American Academy of Pediatrics (AAP) along with the Down Syndrome Medical Interest Group (DSMIG), led a task force in developing care guidelines for providers to manage the care coordination needs of children with DS (Bull et al., 2011). These guidelines encompass not only co-occurring conditions commonly associated with DS but also well-child care including developmental screenings and immunizations. However, research has shown that provider adherence to these guidelines can vary greatly from zero completion of screenings to completion of more than 75% of the recommendations, with only an estimated 9.8% of children with DS being up to date of recommendations for care (Santoro et al., 2017; Skotko et al., 2012).

Missing these recommended care considerations could lead to complications from co-occurring conditions as well as gaps in care (Skotko et al., 2012; Williams et al., 2017). Further, it has been found that successful implementation of care coordination can increase completion of well-child care (Ramirez et al., 2020). Care coordination can also influence caregivers' perceptions of how well the care provided meets the needs of their families (Lemke et al., 2018; Turchi et al., 2009).

When thinking about care coordination, it is also critical to consider who is responsible for managing the care of a child with DS. For children with medical complexities, the burden of management of health information and care coordination often falls to the parent or caregiver (Cady & Belew, 2017). There are also barriers from the health care providers in aiding families in implementing care coordination such as lack of personnel, lack of communication skills and lack of time (Tschudy et al., 2016). While there have been excellent developments in health information management and communication through the use of electronic health records (EHR), because of the possible range of persons involved in care, much of the health information ends up outside of a single electronic health record system. Given these barriers, mHealth apps may be a way to fill this gap by supporting caregivers and families in managing their care coordination and health information management needs.

mHealth is defined by the World Health Organization as any “medical and public health practice supported by mobile devices, such as mobile phones, patient monitoring devices, personal digital assistants, and other wireless devices” (World Health Organization, 2011, p.6). There are many potential uses of mHealth apps including communication needs between individuals and health services, health monitoring, and access to information (World Health Organization, 2011). Studies have shown that primary caregivers are open to the idea of using

mHealth apps to support management of their child’s chronic condition and recognize that technology may facilitate improved information access and communication (Dzubar et al., 2015; Liu et al., 2016; Quigley et al., 2014). To begin the development of an intervention such as a mHealth app, it is first important to gain a full understanding of family and caregiver needs and preferences. This scoping review was designed to examine the extent and nature of research on caregivers’ role in care coordination by addressing the following questions: (1) What are the care coordination needs of primary caregivers of children with DS?; and (2) What strategies and resources do primary caregivers use to address their care coordination needs? These findings will help guide user-centered design of a mHealth app to support families of children with DS.

METHODS

Protocol

The protocol for the search and analysis was developed using guidelines outlined by Arksey and O’Malley (2005) as well as Preferred Reporting Items for Systematic Reviews and Meta-analysis Protocols for Scoping Reviews (Arksey & O’Malley, 2005; Tricco et al., 2018).

Eligibility Criteria

Listed below are the inclusion and exclusion criteria developed for selection of articles to be included in our scoping review (Arksey & O’Malley, 2005).

Inclusion Criteria

- articles appearing in peer reviewed journals
- research implementing any study methodology and design
- published in journals between January 2010 and January 2020
- English language
- study population includes caregivers of children with DS birth to twenty-one years old

- article addresses health management of child with DS, including dental management
- article reports results reflecting perspectives of parents.

Exclusion Criteria

- not research
- Age of child with DS not reported
- focused exclusively on educational management of a child with DS
- did not differentiate children with DS from other possible conditions included in the sample
- focused exclusively on the prenatal period or diagnosis experience
- review articles
- reporting only the providers' perspective

Information Sources

With guidance from a research librarian, a literature search was completed on January 21, 2020. Databases included in the search were CINAHL, Embase, ProQuest Health Management, PsycInfo, and PubMed. These databases were selected to capture journals from a range of healthcare disciplines as well as research on health management. The final search results from each database were exported to Endnote, where duplicates were removed. Results were then exported to Covidence to complete both the title and abstract screening, as well as the full text review (Veritas Health Solutions, Melbourne, Australia).

Search

Search terms were intentionally broad to cast a wide net to find all studies possibly addressing care coordination in the context of DS (Arksey & O'Malley, 2005). While each search was modified slightly to meet the constraints of the search tool, the PubMed (Medline) search string was as follows: [(mother* or father* or parent* or caregiv* or family* or families*

or "family"[mesh] or "pediatric*" or "child"[mesh] or "child, preschool"[mesh] or "adolescent"[mesh] or "teen" or "teenager" or "child*" or "infant*" or "toddler") AND ("care coordination" or "coordination" or "patient-centered care"[mesh] or "case management"[mesh] or "meaningful use"[mesh] or "case managers"[mesh] or "health communication"[mesh] or "needs" or "health care" or "healthcare") AND ("Down syndrome"[mesh] or "down syndrome") and "last 10 years"[PDat]]. In addition to database searches, reference lists of each article included in the review were hand checked to identify studies that were not captured within search strings.

Selection of Sources of Evidence

All studies were screened by two reviewers using Covidence software at both the title and abstract screening as well as the full text review level (Veritas Health Solutions, Melbourne, Australia). The first two authors resolved disagreements for inclusion by discussion to reach final consensus.

As the full text review was completed, it became evident that additional inclusion and exclusion criteria were required to successfully identify studies related to the research questions (Tricco et al., 2018). As part of the inclusion criteria, studies needed to have a focus on health management; however, this definition was further refined to include interaction with the health care system and providers.

Charting the Data

A structured template for charting the data was developed by the first two authors (BC and KK). Data categories included in the template were: study purpose, study design, respondent, conceptual underpinnings of study, definition of care coordination, age range of children in sample, measures, use of technology, and findings related to care coordination and

health information management. Following the development of the extraction template, BC and KK independently charted the data from five studies and then met to discuss the completeness and clarity of the extraction template and make final revisions. Extractions were completed in Microsoft Word. As a check on the extraction process, KK, checked every fifth extraction for accuracy and completeness. Only minor changes were needed providing evidence of the quality of the first authors' extractions.

Collating and Summarizing Data

To collate and summarize study findings, the Marshall definition of care coordination and the Family Management Style Framework (FMSF) structured the analysis (Knafl et al., 2012). Marshall (2019) defined care coordination as “[relying] on communication between primary care providers and specialty care services and access to and facilitation of services and support” and was used to analyze the data (Marshall et al., 2019, p.79). While the purpose statement gave a glimpse into how each study addressed care coordination, within this scoping review it was also necessary to review the studies' findings to answer each of the research questions. While there were 28 articles in which the study purpose included an element of care coordination, all articles were included in this analysis using both the purpose statement and study findings. The constant comparison method was used to develop themes across the purpose statements and results from each study (Corbin & Strauss, 1990). Summaries of each of the themes is included in the results.

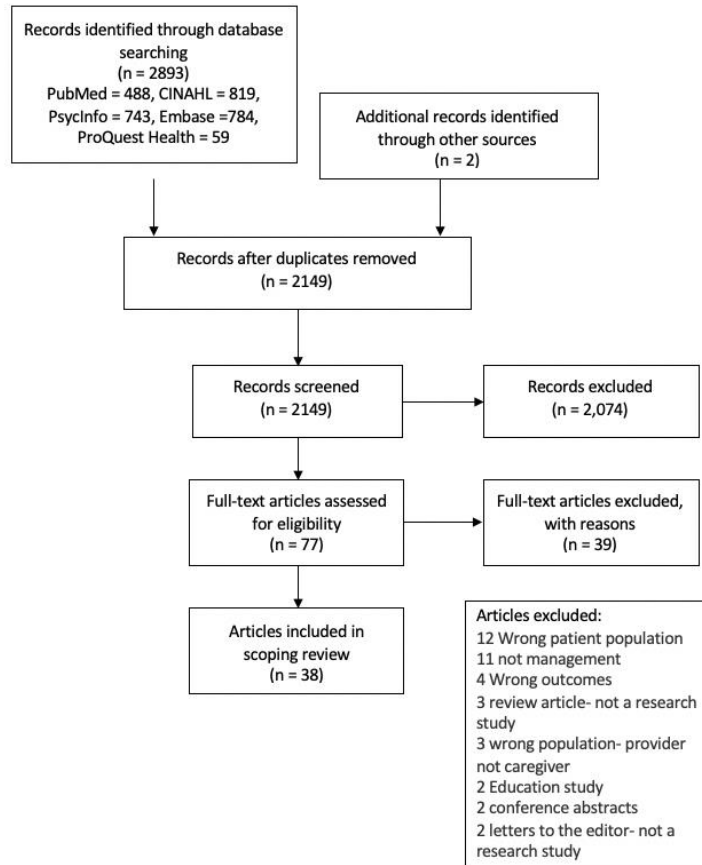
RESULTS

The search of five data bases yielded 2,149 articles for review after duplicates were removed. 2,147 came from database searches and 2 articles came from reference list screening. Following PRISMA guidelines (see Figure 2.1), screening of studies by title and abstract review excluded 2,074, leaving 77 articles for full text review. 38 articles, from 37 studies, were

included in the final sample for extraction. 39 articles were eliminated after the full text review. Reasons for exclusion are listed in the figure below (Figure 2.1).

Figure 2.1

PRISMA Diagram



Characteristics of Sources of Evidence

Data extracted from each study is summarized in Table Appendix 1.1. Findings summarized in extraction included study first author, country where data were collected, study purpose, study design, respondent, age of child with DS, measures, and technology use. These data were extracted to provide a broad overview of the type of studies investigating caregiver interactions with health care with regards to care coordination.

Study Design

The 38 articles in the final sample included multiple study designs. Articles were evenly distributed between qualitative and quantitative study designs. Fourteen studies were qualitative (Amitha et al., 2015; Barros da Silva et al., 2018; Cartwright & Boath, 2018; Farkas et al., 2018; Gibson & Martin, 2019; Huiracocha et al., 2017; Krueger et al., 2019; Marshall et al., 2014; Melvin et al., 2018; Murphy et al., 2017; Nunes & Dupas, 2011; Oliveria et al., 2010; Sheehan & Guerin, 2017; van der Dreissen Mareeuw et al., 2019), eleven were quantitative (Choi & Yoo, 2015; Descamps & Marks, 2015, Hsiao, 2014; Mengoni & Redman, 2018; Nqobobo et al., 2019; Nugent et al., 2018; Parrott et al., 2012; Pikora et al., 2014; Rahim et al., 2014; Santoro et al., 2016; Tozzi et al., 2015), and five were mixed methods (Bertoli et al., 2011; Hall et al., 2012; Hall et al., 2018; Leonard et al., 2016; Marshall et al., 2019;). Investigators in six studies completed secondary analyses, four using data from the National Survey of Children with Special Health Care Needs (NS-CSHCN) (Hubner et al., 2016; Lollar et al., 2012; Phelps et al., 2012, Schieve et al., 2011), one completing a chart review (Skotko et al., 2012), and one using the Intellectual Disability Exploring Answers (IDEA) database (Thomas et al., 2011). Only two studies were longitudinal in design (Crossman et al., 2018; Thomas et al., 2011). Finally, one study employed a pre/post test quasi-experimental design (Choi & Van Riper, 2020).

Measures

There was a lack of consistency across studies in measures used to assess characteristics of care coordination. Outside of the secondary data analyses using the NS-CHCSN survey, only two studies used the same care coordination measure. Caregivers' management of their child's special health care needs with DS was assessed using standardized tools such as the Family Experiences Survey (FES), Family Problem Solving and Communication (FPSC), Family

Management Measure (FaMM), and Parenting Stress Index (PSI). Each of these measures was used in two studies included in the review (Choi & Van Riper, 2020; Choi & Yoo, 2015; Crossman et al., 2018; Hall et al., 2012; Hsiao, 2014, Marshall et al., 2019). Additionally, two studies used the Oral assessment in DS Questionnaire (Descamps & Marks, 2015; Rahim et al., 2014). The Emotionality, Activity, Sociability Survey, Family Adaptability and Cohesion Evaluation Scale, the brief Family Assessment Measure (FAM), Family Support Scale (FSS), Family Coping Index (FCI), Family Environment Scale, Family Index of Regeneratively and Adaptation (FIRA-G), Parent-caregiver Perception Questionnaire (P-CPQ), Perceived Social Support Scale (PSSS), Family Assessment Device (FAD), Beck Depression Inventory (BDI), Index of Social Competence (ISC) and Family APGAR were each used in one study (Choi & Yoo, 2015; Choi & Van Riper, 2020; Crossman et al., 2018; Hsiao, 2014; Nqcoobo et al., 2019; Pikora et al., 2014). Nine studies used measures generated by the investigators to assess caregiver needs (Amitha et al., 2015; Bertoli et al., 2011; Hall et al., 2018; Leonard et al., 2016; Megoni & Redman, 2019; Parrott et al., 2012; Pikora et al., 2014; Santoro et al., 2016; Tozzi et al., 2015).

In the qualitative studies, investigators collected data through interviews or focus groups using interview/discussion guides developed for the study. Twelve qualitative studies collected interview data (Barros da Silva et al., 2018; Farkas et al., 2019; Gibson & Martin, 2018; Hall et al., 2018; Leonard et al., 2016; Marshall et al., 2014; Melvin et al., 2019; Murphy et al., 2017; Nunes & Dupas, 2011; Oliveira et al., 2010; Sheehan & Guerin, 2017; van den Driessen Mareeuw et al., 2019). Investigators in four studies collected data from focus groups (Cartwright & Boath, 2018; Hall et al., 2012; Huiracocha et al., 2017; Marshall et al., 2014).

Analysis

Several different analytic methods were employed to analyze qualitative data. In five studies investigators analyzed interview data using a grounded theory approach (Farkas et al., 2019; Gibson & Martin, 2018; Hall et al., 2018; Murphy et al., 2017; Nunes & Dupas, 2011). Krueger and colleagues (2019) were the only investigators to describe using triangulation for analysis, while van den Driessen Mareeuw (2019) was the only author to report using framework analysis. Finally, investigators in six studies used content analysis (Barros da Silva et al., 2018; Leonard et al., 2016; Marshall et al., 2014; Melvin et al., 2019; Oliveira et al., 2010; Sheehan & Guerin, 2017). Cartwright and Boath (2018) analyzed focus group findings using a phenomenological approach, while the others completed a content analysis (Hall et al., 2012; Huiracocha et al., 2017; Marshall et al., 2014).

Descriptive statistics were reported for all eleven quantitative studies as well as the five mixed methods studies (Bertoli et al., 2011; Choi & Yoo, 2015; Descamps & Marks, 2015; Hall et al., 2012; Hall et al., 2018; Hsiao, 2014; Leonard et al., 2016; Marshall et al., 2019; Mengoni & Redman, 2018; Nqcocho et al., 2019; Nugent et al., 2018; Parrott et al., 2012; Pikora et al., 2014; Rahim et al., 2014; Santoro et al., 2016; Tozzi et al., 2015). Twelve of the studies including quantitative data completed tests of association or correlation (Descamps & Marks, 2015; Hall et al., 2012; Hall et al., 2018; Leonard et al., 2016; Hsiao, 2014; Marshall et al., 2019; Nqcocho et al., 2019; Nugent et al., 2018; Parrott et al., 2012; Pikora et al., 2014; Rahim et al., 2014; Santoro et al., 2016). Frequency counts were employed by five studies as part of the analysis (Bertoli et al., 2011; Marshall et al., 2019; Megoni & Redman, 2019; Rahim et al., 2014; Santoro et al., 2016). Regression modeling was used in six studies including investigations of family adaptation (Choi & Yoo, 2015), functioning and social support (Hsiao, 2014), care

transitions (Nugent et al., 2018), uncertainty and communication (Parrott et al., 2012), social competence (Rahim et al., 2014), and attitudes towards mHealth app usage (Tozzi et al., 2015).

Respondents

3024 families of children with DS were included across 31 primary studies. Eliminated from this total were secondary data analyses and chart reviews. In five secondary analyses, investigators from four studies used data from the National Survey of Children with Special Healthcare Needs years 05-06 (1,128 participants) or 09-10 (504 participants) and in one secondary analysis, the investigator used data from the Health Utilization Survey completed in Western Australia between 1997 and 2004, to which 121 families of children with DS responded (Hubner et al., 2016; Lollar et al., 2012; Phelps et al., 2012; Schieve et al., 2011; Thomas et al., 2011). Lastly, one investigator completed a chart review of 105 medical charts of children with DS (Skotko et al., 2012).

When identified, the primary caregiver in 20 studies, was the mother. Five studies focused exclusively on mothers. Of the 3024 families, 1669 respondents identified as mothers. Only one study exclusively focused on fathers, with 93 respondents. Across studies, 416 respondents identified as fathers. Few studies identified other family member respondents. Bertoli et al., (2011) included 100 siblings of persons with DS, and Parrott et al., (2012) included 18 sibling respondents as well as caregivers. Three studies each had one respondent identifying as a grandmother (Hall et al., 2012; Hall et al., 2018; Melvin et al. 2019). The remaining studies did not specify which parent or caregiver completed the study. Only one study focused exclusively on caregiver dyads (Choi & Van Riper, 2020). Several studies included caregiver dyads within the sample but did not analyze the data as dyads (Decamps & Marks, 2015; Huiracocha et al., 2017; Nunes & Dupas, 2011; Sheehan & Guerin, 2017). One study focused on

caregiver and child dyads however health related quality of life data were only collected from mothers and were linked to child's dental health physical exam findings (Nqcoo et al., 2019). While the focus of this scoping review was families and caregivers of children with DS, several studies also included health care providers, with 72 provider respondents.

Age of Children with DS

Data were extracted regarding the age range of the children with DS. There was great variation in how studies reported age of the child with DS. Since the respondent in each of the studies reviewed was the caregiver and not the child, data on the child was often incomplete or not included. Four studies did not include the age range of the children with DS (Amitha et al., 2015; Hall et al., 2018; Oliveria et al., 2010; Parrott et al., 2012). Twenty-one studies reported the average age of the child with DS (Cartwright & Boath, 2018; Choi & Van Riper, 2019; Choi & Yoo, 2015; Decamps & Marks, 2015; Farkas et al., 2018; Hall et al., 2012; Hsiao, 2014; Krueger et al., 2019; Megoni & Redman, 2019; Melvin et al., 2018; Nqcoo et al., 2019; Nunes & Dupas, 2011; Pikora et al., 2014; Rahim et al., 2014; Sheehan & Guerin, 2018; Skotko et al., 2012; Thomas et al., 2011; Tozzi et al., 2015; van den Driessen Mareeuw et al., 2019). The overall average age across these studies was 9.2 years. Thirteen studies reported only an age range and not an average age of the child with DS (Barros da Silva et al., 2018; Bertoli et al., 2011; Crossman et al., 2018; Gibson & Martin, 2019; Hubner et al., 2016; Huiracocha et al., 2017; Leonard et al., 2016; Lollar et al., 2012; Marshall et al., 2019; Marshall et al., 2014; Murphy et al., 2017; Nugent et al., 2018; Phelps et al., 2012; Santoro et al., 2016; Schieve et al., 2011). While the focus of the scoping review was children and adolescents under the age of 21, some of the studies also included adults with DS. The age ranges reflect this inclusion ranging from birth to 61 years of age. In 6 articles from 5 studies, the sample included caregivers of both

children and adults with DS (Bertoli et al., 2011; Farkas et al., 2018; Gibson & Martin, 2019; Krueger et al., 2019; Leonard et al., 2016; Pikora et al., 2014).

Study Purpose

The purpose of study each article was examined. Although all studies addressed caregivers' interactions and experiences with health care systems and providers for their child with DS, only two studies explicitly used the term "care coordination" in the study purpose (Marshall et al., 2019; Phelps et al., 2012). Additionally, 26 articles contained elements of health care management included in care coordination in their purpose statements. These elements related to care coordination included knowledge of services (Amitha et al., 2015; Decamps & Marks, 2015; Oliveira et al., 2010; Nugent et al., 2018), communication (Melvin et al., 2019; Parrott et al., 2012), access to care (Rahim et al., 2014), information availability (Cartwright & Boath, 2018; Gibson & Martin, 2018; Lollar et al., 2012; Sheehan & Guerin, 2017), engagement and advocacy (Hubner et al., 2016; Krueger et al., 2019; Leonard et al., 2016; van den Driessen Mareeuw et al., 2019), and utilization and availability of services (Bertoli et al., 2011; Choi & Van Riper, 2020; Crossman et al., 2018; Hall et al., 2018; Marshall et al., 2014; Pikora et al., 2014; Thomas et al., 2011; Tozzi et al., 2015). There were three studies that addressed the AAP guidelines for care of a child with DS (Skotko et al., 2012; Mengoni & Redman 2019; Santoro et al., 2016). The other ten studies contained elements in the purpose that could be related to care coordination but was not immediately apparent from the details included in the purpose statement and were found in the study results.

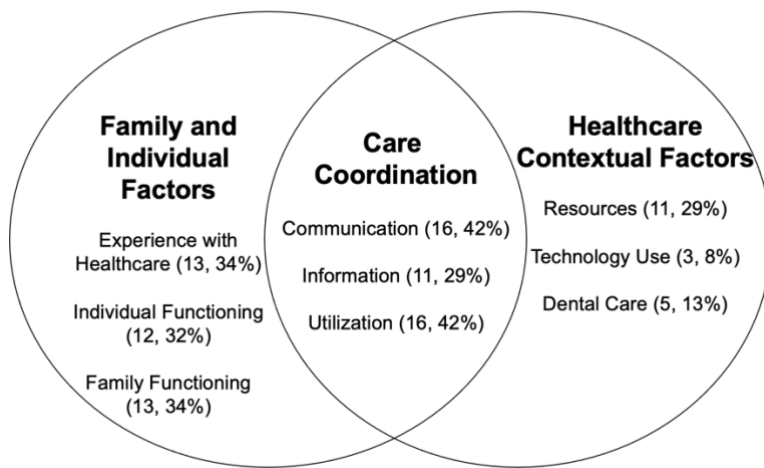
Research Question One

Research question one was designed to address what are the care coordination needs of primary caregivers of children with DS? A model was created from the themes developed from

collating data collected from the purposes and results of each article (Figure 2). All 38 articles were analyzed for factors related to the care coordination needs reported by primary caregivers of children with DS as relates to research question 1. Themes are ordered by the frequency with which they occurred within each topic area.

Figure 2.2

Themes Related to Care Coordination



*(number of studies addressing theme included in review, percentage of studies in review)

Communication

Sixteen studies included in the scoping review reported results related to communication between caregivers and providers (Barros da Silva et al., 2018; Cartwright & Boath, 2018; Choi & Yoo, 2015; Gibson & Martin, 2018; Hall et al., 2012; Hall et al., 2018; Hubner et al., 2016; Huiracocha et al., 2017; Leonard et al., 2016; Marshall et al., 2014; Marshall et al., 2019; Melvin et al., 2019; Murphy et al., 2017; Parrott et al., 2012; Phelps et al., 2012; Sheehan & Guerin, 2017). Caregivers’ perceptions of the quality of communication were linked to both positive (Choi & Van Riper, 2020; Choi & Yoo, 2015; Hall et al., 2018; Hubner et al., 2016; Leonard et al., 2016; Marshall et al., 2019) and negative (Barros da Silva et al., 2018; Cartwright & Boath,

2018; Gibson & Martin, 2018; Huiracocha et al., 2017; Marshall et al., 2014; Phelps et al., 2012; Sheehan & Guerin, 2017) outcomes for families and children in both quantitative and qualitative studies. In four studies investigators focused on caregivers' beliefs about the importance of communication rather than their assessment of the quality of their communication with professionals (Hall et al., 2012; Melvin et al., 2019; Murphy et al., 2017; Parrott et al., 2012). These findings, both negative or positive in nature, have the potential to impact the quality of communication and thus care coordination.

Shared decision making was a common theme that caregivers expressed as promoting effective and positive communication (Hall et al., 2018; Hubner et al., 2016; Leonard et al., 2016; Melvin et al., 2019). Communication was also found to be a protective factor for caregivers in developing resilience and family adaptation (Choi & Yoo, 2015). In Marshall et al., (2019), 88% of respondents expressed satisfaction with communication between health care providers as well as satisfaction between providers and other professionals.

However, there were negative findings related to communication between caregivers and providers. Caregivers reported supportive communication to be rare and more often felt that providers did not understand family needs related to DS and frequently evoked negative emotions (Cartwright & Boath, 2018; Krueger et al., 2019). Caregivers expressed that there was the perception of information being withheld and caregivers did not feel supported by health care providers (Barros da Silva et al., 2018; Gibson & Martin, 2018). Almost half of caregivers (44.6%) were less than very satisfied with the communication between different providers involved in their child's care (Phelps et al., 2012). Caregivers also described non-existent or scattered communication between providers when describing care coordination (Marshall et al., 2014).

Four studies detailed how communication was a contributing factor in effective care coordination practices (Hall et al., 2012; Melvin et al., 2019; Murphy et al., 2017; Parrott et al., 2012). Hall et al. (2012) found the perception of open versus closed communication with providers to be influenced by the caregiver's stress level, with stressed families describing more closed communication in focus groups (Hall et al., 2012). Both Murphy et al., (2017) and Melvin et al., (2018) identified communication as a factor influencing caregiver perception of quality of life and self-management ability respectively. Finally Parrot et al., 2012, found that as caregiver uncertainty increased related to medical management, the caregivers' desire for increased communication also increased. These findings are important in understanding how communication may be both influencing and influenced by individual family member well-being.

Results related to communication also addressed caregivers' perceptions of their communication with other parents of children with DS. Caregivers wanted to connect with other families of children with DS and learn from their experiences (Barros da Silva et al., 2018; Choi & Van Riper, 2020; Marshall et al., 2019; Sheehan & Guerin, 2017). Caregivers reported that communication with other caregivers fostered the exchange of information as well as creating a bond between families (Barros da Silva et al., 2018). Caregivers described these experiences as having a positive impact on their ability to coordinate care.

Information

Eleven studies investigated the information needs of caregivers and families (Barros da Silva et al., 2018; Cartwright & Boath, 2018; Choi & Van Riper, 2020; Gibson & Martin, 2018; Leonard et al., 2016; Marshall et al., 2014; Marshall et al., 2019; Melvin et al., 2019; Nunes & Dupas, 2011; Sheehan & Guerin, 2017; Santoro et al., 2016). Included under this theme were

study results addressing the quality and timing of information. Across studies, it was found that caregivers were motivated to acquire information regarding their child and DS (Cartwright & Boath, 2018; Choi & Van Riper, 2020; Leonard et al., 2016; Marshall et al., 2014; Marshall et al., 2019; Melvin et al., 2019; Tozzi et al., 2015). However, there were several factors impacting the desire for information. Families wanted information to be from trustworthy sources (Gibson & Martin, 2018) and were often frustrated by the quality and lack of information provided by health care providers (Phelps et al., 2012). Caregivers also reported feeling responsible for accessing reliable health management information (Nunes & Dupas, 2011). Families were also concerned about providers not being up to date and knowledgeable in the management of health concerns associated with DS (Barros da Silva et al., 2018; Cartwright & Boath, 2018; Melvin et al., 2019; Sheehan & Guerin, 2017). Caregivers were concerned that information presented in printed materials was often out of date and information on available resources was lacking (Marshall et al., 2014).

Age and timing were also considerations in caregiver's information needs. Families expressed a desire to have targeted information at times of transition or at specific age milestones (Leonard et al., 2016; Sheehan & Guerin, 2017). However, families did not want to be overloaded by information (Barros da Silva et al., 2018; Sheehan & Guerin, 2017). In Choi & Van Riper (2020) study of a mHealth app addressing family adaptation and therapeutic communication, researchers reported that families' found information most helpful from birth to age 12 months. Melvin et al. (2018) defined these critical information periods as early development ages (0 to 3 years) and school transitions and also specified that families should be offered information early in the child's life to be able to develop realistic goals and expectations.

However, less than half of families reported receiving information about DS in the birth setting (Marshall et al., 2019).

Utilization

In sixteen studies investigators linked elements of health care utilization to care coordination (Bertoli et al., 2011; Hubner et al., 2016; Leonard et al., 2016; Marshall et al., 2014; Marshall et al., 2019; Mengoni & Redman, 2019; Murphy et al., 2017; Nugent et al., 2018; Phelps et al., 2012; Pikora et al., 2014; Rahim et al., 2014; Santoro et al., 2016; Schieve et al., 2011; Skotko et al., 2012, Thomas et al., 2011; Tozzi et al., 2015). The most prominent feature of utilization was the role of the medical home. The medical home is a model of care defined as “holistic care of children and their families where each family has an ongoing relationship with a health care professional” (National Association of Pediatric Nurse Practitioners Executive Board, 2015, p. 17A). Care coordination is a key component in the implementation of medical home policy (Council on Children with Disabilities and Medical Home Implementation Project Advisory, 2014). Investigators found that children with DS were at significantly more risk for not having a medical home than other children with special health care needs (Hubner et al., 2016). Between 29.7-37.5% of caregivers of children with DS reported having a medical home compared 47.3% of families with other special health care needs reporting having a medical home (Hubner et al., 2016; Phelps et al., 2012). While only about 1/3 of families reported having a medical home, 88% reported seeing a general healthcare provider in the past three years (Pikora et al., 2014). In studies for this review, children without a medical home were at greater risk for missed components of care management and failure to meet health care transitions (Hubner et al., 2016; Nugent et al., 2018; Phelps et al., 2012; Schieve et al., 2011; Skotko et al., 2012). Caregivers of children with DS without a medical home may be less prepared to aid their

child with the transition from pediatric to adult health care because of a lack of support and encouragement from their health care provider (Nugent et al., 2018). Utilization was also investigated by three studies looking at the use of the AAP/DSMIG guidelines for the management of children with DS (Mengoni & Redman, 2019; Santoro et al., 2016; Skotko et al., 2012).

Investigators also examined the amount of time caregivers spent addressing their child's special needs. Caregiver time is also an important consideration when investigating health care utilization. Examples of outcomes related to caregiver time included in these studies were issues such as travel time to appointments and missed days of work. However only one study investigated healthcare utilization related to travel times, reporting that 1/3 of the sample (n=41) took more than an hour to reach a pathology center (Tozzi et al., 2015). Caregivers reported missing on average 7 workdays annually for health care visits (Tozzi et al., 2015). Phelps et al. (2012) reported that 30.2% of caregivers reported dedicating eleven or more hours per week to care coordination for their child with DS.

Experiences with Healthcare

While all of the studies included some connection to caregiver and family experiences with healthcare, there were thirteen articles that highlighted specific details with regards to these interactions (Barros da Silva et al., 2018; Cartwright & Boath, 2018; Choi & Van Riper, 2020; Farkas et al., 2018; Gibson & Martin, 2019; Hall et al., 2018; Huiracocha et al., 2017; Krueger et al., 2019; Marshall et al., 2014; Marshall et al., 2019; Melvin et al., 2018; Phelps et al., 2012; Sheehan & Guerin, 2017). Some of the results of caregiver experiences fell into other categories because they specifically addressed themes such as information, but findings from nine studies reported more general experiences with healthcare or experiences that did not fit into other

categories described above (Barros da Silva et al., 2018; Cartwright & Boath, 2018; Farkas et al., 2018; Huiracocha et al., 2017; Krueger et al., 2019; Marshall et al., 2014; Melvin et al., 2019; Phelps et al., 2012; Sheehan & Guerin, 2017).

These experiences related to caregivers' general perceptions of the interaction with health care providers. Caregivers reported providers were helpful in providing information, however where providers fell short was understanding caregivers' emotional responses to their child's condition and special needs (Sheehan & Guerin, 2017). There were other experiences expressed across studies that were perceived as negative. For example, caregivers in several studies expressed dissatisfaction with health care providers who were perceived as insensitive and lacking support for behaviors such as breastfeeding (Barros da Silva et al., 2018; Farkas et al., 2019; Huiracocha et al., 2017). Caregivers felt there was a lack of distinction in care needs for children with DS versus typically developing children (Cartwright & Boath, 2018). In one study, less than half of families reported feeling like a partner in care with their child's provider (Phelps et al., 2012).

Individual Functioning

A second theme within individual and family factors addressed the individual functioning of a child with DS. Twelve studies included elements of individual functioning of children with DS (Barros da Silva et al., 2018; Bertoli et al., 2011; Cartwright & Boath, 2018; Lollar et al., 2012; Hall et al., 2018; Mengoni & Redman, 2019; Nugent et al., 2018; Pikora et al., 2014; Schieve et al., 2011; Skotko et al., 2012; Thomas et al., 2011). Articles focused on various components of individual functioning including co-occurring conditions commonly associated with DS, impact on activities of daily living and demographic factors influencing functioning (Bertoli et al., 2011; Lollar et al., 2012; Mengoni & Redman, 2019; Nugent et al., 2018; Pikora et

al., 2014; Schieve et al., 2011; Skotko et al., 2012; Thomas et al., 2011). Two studies specifically addressed feeding concerns related to breastfeeding and feeding methods (Barros da Silva et al., 2018; Cartwright & Boath, 2018).

Several studies investigated the impact of functional status on the care received. Researchers reported there was variation in the specialty referrals for individual functional difficulties made by providers, leading to discrepancies in care received (Hall et al., 2018; Schieve et al., 2011). Children with higher functional impairments had lower odds of having a medical home, which studies have shown improves care coordination outcomes (Council on Children with Disabilities and Medical Home Implementation Project Advisory Committee, 2014; Hubner et al., 2016). Children with DS were more likely to have delays in necessary therapies than children with other special health care needs (Schieve et al., 2011).

Family Functioning

While family factors were addressed across thirteen studies (Choi & Van Riper, 2020; Choi & Yoo, 2015; Crossman et al., 2018; Hall et al., 2012; Hsiao, 2014; Huiracocha et al. 2017; Krueger et al., 2019; Leonard et al., 2016; Murphy et al., 2017; Nqobobo et al., 2019; Nunes & Dupas, 2011; Parrott et al., 2012; Sheehan & Geurin, 2017), three factors of family functioning were identified as playing a role in care coordination, resiliency, advocacy, and uncertainty (Hall et al., 2012; Krueger et al., 2019; Parrott et al., 2012). Families described care coordination across providers as one way they were able to advocate for their child, and this advocacy most common occurred in health care and school settings (Krueger et al., 2019; Nunes & Dupas, 2011). Caregivers' perception of family resiliency also was a factor contributing family response to stressors (Hall et al., 2012). Families who displayed more resiliency and less stress reported an increased perception of communication and support in a health care setting, important factors

in care coordination (Hall et al., 2012). Caregivers and families with increased uncertainty also expressed a strong desire to communicate (Parrott et al., 2012).

Resources

Children with DS often require many resources to manage co-occurring conditions as a part of care coordination. These resources may be in the form of services such as therapies, as well as financial resources and transportation. Eleven studies included in our sample reported findings related to resources (Amitha et al., 2015; Bertoli et al., 2011; Choi & Yoo, 2015; Gibson & Martin, 2018; Hall et al., 2018; Huircocha et al., 2017; Marshall et al., 2014; Murphy et al., 2017; Nugent et al., 2018; Nunes & Dupas, 2011; Tozzi et al., 2015). Most commonly financial constraints were reported as a barrier to receiving necessary care (Amitha et al., 2015; Gibson & Martin, 2018; Hall et al., 2018; Huircocha et al., 2017; Nunes & Dupas, 2011). However, the lack of available social services and resources and how to access them was also reported as a concern for caregivers (Bertoli et al., 2011; Hall et al., 2018; Marshall et al., 2014; Marshall et al., 2019; Nugent et al., 2018; Schieve et al., 2011). Investigators in one study reported that availability of community services was a positive indicator in family adaptation (Choi & Yoo, 2015). Caregivers reported frustration with resources from frequently having to change providers to maintain government supported services such as developmental therapies (Murphy et al., 2017). Support also varied greatly on the perceived level of the disability and the availability of the services (Hall et al., 2018).

Technology Use

The ultimate goal of this scoping review is to determine the content of a mHealth app that would best support caregivers of children with DS. Because of this goal, it was important to investigate if any of the studies incorporated the use of technology into management of care

needs and how it may impact care. Choi & Van Riper (2020), studied family adaptation and DS using a mHealth app to engage in therapeutic communication with a nurse, and included a definition of mHealth from the World Health Organization (WHO) as “medical and public health practice supported by mobile devices, such as mobile phones, patient monitoring devices, personal digital assistants, and other wireless devices” (p.2). Investigators in a second study included in this review investigated caregivers’ attitudes and expectations for the use of a mHealth app (Tozzi et al, 2015). Tozzi et al., (2015) found that almost all families connected to the internet either at home or work, and half of those families connected through a smartphone (Tozzi et al, 2015).

While technology was not included in the project aims, Melvin et al. (2018) found that caregivers expressed a desire to have an electronic way to organize information as well as provide age-appropriate and research-based information to support their child’s communication (Melvin et al., 2019). Researchers also reported caregivers’ desire to have trusted information from online sources, particularly at times of transition such as the start of school, and the beginning of puberty (Cartwright & Boath, 2018; Gibson & Martin, 2018). Technology was also described as an important facilitator of communication between caregivers of children with DS through use of online forums as well as platforms like Facebook (Barros da Silva et al., 2018).

Dental Care

An unexpected finding of this scoping review were five studies exclusively addressing dental care (Amitha et al., 2015, Oliveira et al., 2010; Descamps & Marks, 2015; Nqco et al., 2019; Rahim et al., 2014). While the focus was on dental care, many of the themes overlapped in terms of resources, information, and experiences with health care. Studies focused on dental care primarily reported frequency of visits, type of dental provider, use of anesthesia and

knowledge related to the dental care of children with special needs (Amitha et al., 2015; Oliveira et al., 2010; Descamps & Marks, 2015; Nqcoobo et al., 2019; Rahim et al., 2014). Nqcoobo et al. 2019 also reported on the frequency and treatment of dental caries in children with DS. Rahim et al., (2014) found that children with DS received less dental care than typically developing children and none had received orthodontic care.

Research Question Two

A similar thematic analysis of the results was completed to address the second research question: What strategies and resources do primary caregivers use to address their care coordination needs? This question yielded fewer findings than the first question describing caregiver experiences with elements of care coordination. However, there were results that could inform future care coordination recommendations and be used in the development of interventions using mHealth apps. Findings addressing the second research question fell within the themes of care coordination, family, and individual factors.

Care Coordination Factors

Caregivers' recommendations addressing strategies and resources related to care coordination, included communication, information, and utilization. As reported in research question one, parents viewed shared decision-making as a strategy enhanced communication, particularly when initiated in the medical home (Hall et al., 2018; Hubner et al., 2016; Leonard et al., 2016; Melvin et al., 2019; Nugent et al., 2018; Skotko et al., 2012). This was also reflected in the need for care to be family centered (Marshall et al., 2019; Hsiao, 2014; Huiracocha et al., 2017). Additionally, caregivers recommended strategies to maintain an organized, up to date health history as a way to minimize the time focused on updating the health history during appointments with providers (Murphy et al., 2017). This was particularly true for appointments

related to child development (Murphy et al., 2017). Two studies recommended the use of mHealth apps to support time management and caregivers' active participation in their child's health care (Choi & Van Riper, 2020; Tozzi et al., 2015).

Because of the amount of information required for care coordination and health management, it was recommended that information be age specific as well as electronically available to help manage the amount and timing of information (Choi & Van Riper, 2020; Melvin et al., 2019). Information was particularly important in transition planning (Gibson & Martin, 2018; Leonard et al., 2016; Nugent et al., 2018). Also, using knowledge gained on related protective family factors such as communication, resources and support, nurses can develop individual strategies for families related to adaptation and resiliency (Choi & Yoo, 2015). One study, Thomas et al., (2011), made a recommendation related to resource use and utilization. To maximize utilization, it was recommended by caregivers that providers have knowledge of health insurance coverage for specialty referrals (Thomas et al., 2011).

Family and Individual Factors

Family factors also contributed to caregivers' strategies for effective care coordination. Krueger et al., (2019) reported that caregivers recommended being assertive as well as persistent in their communication with healthcare providers to ensure receiving necessary services and information. Families also need to develop good organization skills to manage care coordination (van den Driessen Mareeuw et al., 2019).

DISCUSSION

Findings from this scoping review support the conclusion that effective care coordination has the potential to increase family satisfaction and improve outcomes in managing care for a child with DS. These findings are consistent with study findings from a study of caregivers of

adolescents with chronic conditions (Lemke et al., 2018). Care coordination has also shown the ability to reduce functional difficulties for children with special needs (Litt & McCormick, 2014). However, care coordination needs to be utilized in a manner that best supports individual family and child needs. Using the themes developed from the findings of this scoping review, we considered how these findings could be used to inform the development of an intervention using mHealth apps for families. While fewer studies addressed research question two, there were findings from themes from both research questions that will aid in the development of a mhealth application.

The results of this scoping review point to reliable and up to date information as one of the most important caregivers needs with regards to care coordination for their child with DS. There are several findings that were identified from this scoping review that could be used to manage health information within a mHealth app to improve care coordination efforts. One area in which a mHealth apps could fill a gap is in the development of a personal health record (PHR). A PHR is designed to fill the information gap in the electronic health record by having persons or caregivers manage their own health information (Zhou et al., 2019). We know that children with DS have the potential to see many specialty providers in a variety of health care settings. A PHR may help organize required documentation of health behaviors for referrals (Maher et al., 2016). For example, if there is a concern about obstructive sleep apnea, a PHR could serve as a journal for a caregiver to document sleep history (Maher et al., 2016). mHealth apps can be used to document information such as medications, provider information and lab results as part of a PHR (Maher et al., 2016). Families expressed a desire to be able to keep health information in a way that is easy to bring with them between visits and a mHealth app is one intervention to fill this gap.

In conjunction with a PHR, other technologies are being developed to support caregivers of children with DS. Down Syndrome Clinic to You (DSC2U) is a web-based tool that will help caregivers identify referral needs for their child using the AAP guidelines (Chung et al., 2021). It may be possible for a mHealth app technology to work in conjunction with a website like DSC2U for families to document information pertinent to referral appointments. The linking together of health care technologies increases caregivers' ability to manage health information and care coordination.

However, one consideration to keep in mind when developing mhealth technologies is cost. Having mhealth apps be free and publicly available may help to increase parents' access to health information needed to manage their child's care. There is limited but positive evidence that mhealth apps are cost effective (De la Torre-Diez et al., 2015; Rinaldi et al., 2020). Examples of free mhealth apps focusing on the DS community include HealthSwap, focusing on the nutritional needs of persons with DS and mhealth apps targeting learning and communication needs such as the mhealth app Otsimo (GooglePlay, 2021). The DSC2U website mentioned above does carry a cost for use, but this cost can be offset by insurance companies (DSC2U, 2021). However, there currently is no mhealth app addressing the care coordination needs of persons with DS. The global COVID- 19 pandemic shed light on the importance of remote healthcare tools, and this is likely to continue into the future. As such, further research considering the cost and use of mhealth technologies is warranted.

Caregivers expressed the desire to have age specific health information as a part of coordinating care (Leonard et al., 2016; Sheehan & Geurin, 2017). A mHealth app could allow for information to be communicated through pop-up reminders that could be tailored to a child's age and the families' specific needs. Pop-up reminders would be particularly relevant in infant

and preschool years when there is an increase in specialty care visits and developmental changes (Melvin et al., 2019). Caregivers also wanted information at times of transition such as the start of school and the beginning of puberty and pop-ups could be used for this information (Melvin et al., 2019). This would also prevent the concern of receiving too much information all at once (Sheehan & Geurin, 2017). Pop-up information could also be used to create appointment reminders, reducing caregiver concern related to scheduling (Phelps et al., 2012). Finally, pop-up reminders could be used for caregivers to document information related to daily habits requiring documentation for co-occurring conditions as part of a PHR (Maher et al., 2016). Pop-up reminders could serve many functions within a PHR and a mHealth app.

Additional key findings from this scoping review that can be used in development of a mHealth app include the need for up-to-date information and easy to access data (Barros da Silva et al., 2018; Cartwright & Boath, 2018; Choi & Van Riper, 2020; Gibson & Martin, 2018; Marshall et al., 2014; Marshall et al., 2019; Tozzi et al., 2015). These findings with regards to health information align with findings related to user centered design practices for developing mHealth apps. Desai et al., (2020) outlined six design preferences for mHealth apps. Of these six design elements, using a table layout, a problem-based organization system, and linking content between different areas will help address these caregiver information needs (Desai et al., 2020). Information within a mHealth app can be presented in a table format as well as linking pertinent topics together for caregivers. The findings from this scoping review related to care coordination needs as well as these design elements, will help develop a mHealth app that meets the needs of caregivers of children with DS.

Limitations

There were limitations to this scoping review. Articles were restricted to only those in the English language. However, the search terms for this scoping review were purposely broad to cast a wide net, creating great variety in the type of studies identified. There was a lack of continuity across studies in type of data, measures, and study design within the extraction. There was also inconsistent reporting on the age of the child with DS. Additional information on age would allow for additional analysis across studies based on the age of the child. It is possible that relevant articles may have been missed. This risk was minimized by seeking the input of a research librarian to design the search string. This scoping review was also limited by focusing exclusively on caregiver perceptions of care coordination. It was important to include health care providers' input on care coordination and future reviews focusing on healthcare providers' knowledge of care coordination, type of practice, and knowledge and ability to treat children with DS would also advance knowledge of strategies for ensuring optimal care coordination. Despite limitations, this scoping review provides valuable insights into families' care coordination needs for a child with DS.

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CHAPTER 3: A MIXED METHODS ANALYSIS OF CARE COORDINATION NEEDS TO INFORM THE DEVELOPMENT OF A MHEALTH APP FOR CAREGIVERS OF CHILDREN WITH DOWN SYNDROME

SUMMARY

Care coordination is critical for the management of health care needs for children with Down syndrome. Caregivers are often left to manage referrals and health information between providers and visits. This mixed methods study was designed to identify caregiver and health care provider perspectives to inform the development of an mHealth application. Ninety-three caregivers, primarily mothers, completed survey materials addressing DS health care needs for their child, app use, and experiences with care coordination. Eleven caregivers and ten health care providers were interviewed to further understand care coordination needs and desires for mHealth technology. While most caregivers reported having a primary health care provider, caregivers reported wanting increased communication and help managing care coordination. Qualitative data, analyzed using framework analysis, identified eight subthemes related to challenges families experience surrounding care coordination. Subthemes included information management, information sharing, use of health care guidelines, tracking health care data, resources, technology use, previous app use, and coordination of schedules. These themes were then linked to desired features of an mHealth app. These findings will be used to guide the development of a mHealth app for caregivers of children with Down syndrome.

INTRODUCTION

Providing family centered care within a child's medical home, the child's primary health care setting, is critical in achieving optimal health and family centered outcomes (Boudreau et al., 2012, Turchi et al., 2009). This is particularly true when caring for children with special health care needs, such as Down syndrome (DS) (Marshall et al., 2019; Phelps et al., 2012). The National Association of Pediatric Nurse Practitioners (NAPNAP), along with the American Academy of Pediatrics (AAP), outline and endorse guidelines of the family centered medical home (FCMH) which includes components such as a personal care provider, a place for both acute and well care, and a place to receive referrals (Litt & McCormick, 2015; NAPNAP, 2009; AAP, 2014). An additional key feature of the FCMH is comprehensive care coordination (Litt & McCormick, 2015; NAPNAP, 2009). Care coordination is described as delivering family centered care that meets the child's health care needs across many realms of health care including, medical, social, support therapies, and educational (AAP, 2014; Antonelli et al., 2009; Schor, 2019). Additionally care coordination is defined by promoting self-care skills (Kuo et al., 2018). However, research has shown, children with DS are more at risk for missed care coordination than other populations of children with special health care needs because they are more likely to receive care outside of the medical home (Phelps et al., 2012; Williams et al., 2017). Because of this increased risk and need for care coordination, there needs to be a focus on how best to serve families in a primary care setting.

To aid in the management and care coordination of children with DS, the AAP established guidelines for care from birth to twenty-one years of age (Bull et al., 2011). These guidelines help to organize referrals and monitoring for co-occurring conditions that may present in a child with DS (Bull et al., 2011). However, outside of the guidelines, there are still many

care coordination needs that fall to families (Cady & Belew, 2017). These needs include information sharing and management (Cady et al., 2020; Ranade-Kharkar et al., 2017). Results from a review of the literature indicate that communication, up to date information, and utilization of resources were vital to families with regards to care coordination for their child with DS (Skelton et al., 2021).

Because of the ongoing challenges families of children with DS face, there is a need to develop new tools to support families in their care coordination and information management needs. Research has shown that primary caregivers express a desire to use mHealth or mobile applications to maintain a personal health record (PHR) because of portability benefits and the ability to input vital information related to care (Calderon et al., 2015). One new support tool that exists is Down Syndrome Clinic to You (DSC2U), developed to manage the AAP guidelines, but does not include components to maintaining a PHR (Chung et al., 2021). One potential way to maintain a PHR is through a mHealth application.

mHealth apps have the potential to fill the gap of familial management of health information and care coordination. While it is estimated that there are as many as 325,000 mHealth apps on the market, mHealth has been shown to improve self-management of symptom control, as well as health outcomes (AAP, 2016; Research2Research, 2017; Whitehead & Seaton, 2016). This is true for chronic conditions such as diabetes and asthma (Dzubur et al., 2015; Padman et al., 2013). However, there is limited research on the use of mHealth apps by caregivers, especially with regards to care coordination. What is also lacking from many mHealth tools is evidence of their development being guided by user-centered design. There is a need to develop mHealth technologies by engaging the end user in the development process (Slater et al., 2017).

With this in mind, the aim of this study was to examine primary caregivers' and health care providers' (HCPs) perspectives on care coordination needs and desirable characteristics of mHealth application content and application use outcomes. These findings will be used to develop an algorithm for the design of an mHealth application to support care coordination and information management. Participant data were gathered using qualitative and quantitative methods to assess children's current health status, current app use, potential for app use, current care coordination status, and desires for a mHealth app.

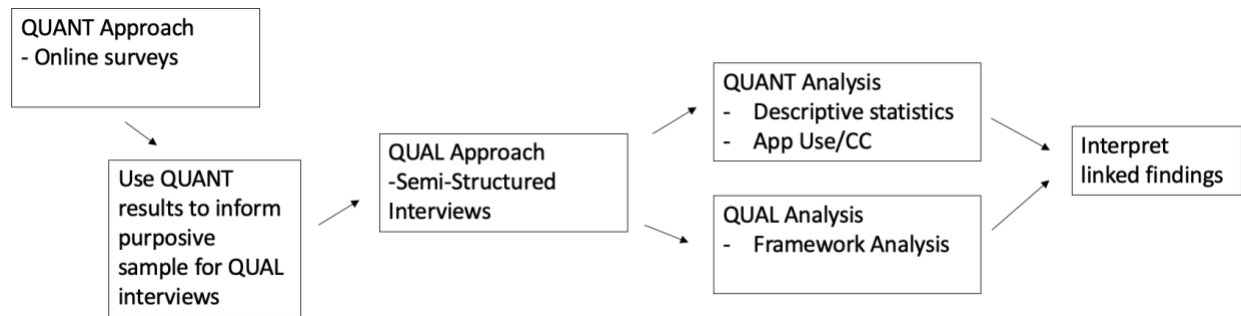
METHODS

Design

A sequential mixed methods design was selected to use both quantitative and qualitative methods and integrate results to gain a more complete understanding of care coordination needs and desired features related to mHealth technologies (Creamer, 2018; Creswell & Plano Clark, 2018; Richie et al. 2014). There was a greater focus on the qualitative findings in the analysis, however the quantitative data played a key role in understanding current mobile technology use, care coordination, and health care needs for children with DS. Additionally, quantitative findings were also used for purposive sampling for qualitative interviews (Creamer, 2018). Quantitative findings from the National Survey of Children's Health Section D (NSCH-D) were linked to key findings from the qualitative interviews. Figure 3.1 outlines the sequential mixed method design. Further quantitative analysis will be presented in future publications.

Figure 3.1

Sequential Mixed Methods Design



Sample

Maximum variation purposive sampling was employed to recruit a diverse sample with regards to geographical location, caregiver role, and age of child with DS for caregiver qualitative interviews with a goal of ten caregivers (Creswell & Plano Clark, 2018; Creswell & Poth, 2017). These variables and sample size were selected to yield a diverse sample related to knowledge of and experience with care coordination (Creswell & Poth, 2017). Purposive sampling was also employed to yield a sample of ten HCPs with varying levels of experience, care setting, and HCP role.

Measures

Primary caregivers completed questionnaires eliciting data on respondents' demographic characteristics, health care usage, care coordination needs and care condition management (e.g., NSCH-D). Questions also assessed respondents' use of mobile devices and desired features related to mHealth.

Demographics Questionnaire. Demographic information about primary caregivers included age, sex, marital status, level of education, child seen at a DS clinic, distance from hospital system with DS clinic, specialty referrals and information about the child (age, sex,

sibling with DS, and co-occurring conditions). Demographic information for healthcare professionals/support staff included healthcare role, years of experience caring for children with DS, and experience with mHealth applications.

National Survey of Children's Health. Questions from the NSCH-D address current care coordination approaches employed in health care management, primary caregivers' perceptions of positive experiences with care coordination, potential gaps in care, and perceptions of areas of needed improvement in care coordination (Health and Human Resources Administration, n.d.).

Interview Guides. Questions for both the caregiver and HCP interviews were grounded conceptually in the Family Management Style Framework (Knafl et al., 2012) and elicited respondents' perspectives on how an mHealth app could contribute to better family management of care coordination. Interview questions were piloted with both a caregiver and a provider and further refined to eliminate redundant questions and clarify the type of information the investigator was seeking. The interview guides can be found in Appendix 3.1.

Procedures

The study underwent UNC-CH IRB review and was deemed exempt in June 2020, with recruitment and enrollment starting in September 2020. Caregivers were recruited from two main sources, North Carolina Down Syndrome Alliance (NCDSA) and DS-Connect. NCDSA is an organization serving families of children with DS across North Carolina. NCDSA listed study information in their monthly newsletter distributed to families. DS-Connect is a national database as part of the National Institute of Child and Human Development (NICHD) as part of the National Institutes of Health (NIH). DS-Connect emailed study enrollment information to 1,959 families of a child ages birth to 21 years with DS within the database. Additionally,

contacts from Down Syndrome Diagnosis Network (DSDN) were posted through their parent groups to further supplement recruitment efforts. Participants emailed PIs expressing interest in the study and they were sent an online link to consent and survey information through Qualtrics (Provo, UT).

Emails for participation in interviews were sent to respondents who expressed willingness to participate in the survey and met the criteria for maximum variation purposive sampling. The PI completed interviews via telephone and the interview was recorded for transcription. At the beginning of each interview, consent information was reviewed, and caregivers were informed that the interview could be stopped at any time if questions caused distress.

To recruit providers, each caregiver who was interviewed was asked to share their primary care provider's contact information. Additional provider recruitment was completed through a known contact at the Children's National Health System (Washington, DC, United States). Invitations to participate were sent to providers and office managers from seven branches of outpatient pediatric primary care centers in the Washington, DC, metro area. This allowed for recruitment not only of primary providers but also recruitment of staff such as office managers and a case manager. Two provider participants were recruited through NCDSA as both were parents of a child with DS as well as HCPs. Interviews were completed via telephone at a time selected by the participant. Consent information was reviewed at the beginning of each interview.

Survey and interview data were stored within a password protected server. All data were deidentified and interview participants were assigned a random identifier. Interviews ranged

from twenty to seventy minutes in length. Upon completion, interviews were transcribed by a professional transcriptionist and checked for accuracy by BC. Audio files were then destroyed.

Analysis

Quantitative Analysis. All quantitative analysis was completed using SPSS Statistics (Version 25). Descriptive statistics were calculated on demographic data including parental age, number of children, birth order, sex, child's age, geographical location, and co-occurring conditions as well as responses related to care coordination and mobile technology use. Descriptive statistics were calculated using means and standard deviations.

Qualitative Analysis. Framework Analysis was selected as the method of analysis for these cross-sectional qualitative findings (Richie et al., 2014; Ward et al., 2013). Framework analysis is completed in four phases: familiarization, identifying themes, indexing, and sorting data, and summarization (Richie et al., 2014; Ward et al., 2013). Familiarization involved a full immersion into the data by reading and rereading each interview transcript and a thorough review of field notes. Familiarization was primarily conducted by BC and co-author KK reviewed thirty percent of interview transcripts. Data were charted into a matrix based on themes reflected in the FMSF, and themes identified in a scoping review (Knafl et al., 2012; Skelton et al., 2021; Ward et al., 2013). BC charted the data and then worked in collaboration with KK in organization of themes and subthemes. The matrix display supported comparison of themes across respondents. In the final stage of framework analysis, the charted data were summarized to present a narrative description of findings.

RESULTS

Sample

A sample of 90 caregivers completed greater than seventy-five percent of the survey materials. However, of the 90 participants, only 84 completed the total survey materials. Missing data were handled by excluding cases in a pairwise manner (Pallant, 2020).

Demographic Characteristics

Overall, the sample was homogeneous in nature, with most participants self-identifying as a mother (93.3%), from a 2-parent household (93.3%), who are highly educated (84.9% completion of college or graduate education). Demographic characteristics are listed in Table 3.1.

Table 3.1

Demographic Characteristics of Participants

Characteristic	n	%
Sex		
Male	5	5.6
Female	85	91.4
Caregiver Role		
Parent	89	98.9
Relative (Aunt)	1	1.1
Family Type		
2 parent household	84	93.3
1 parent household	5	5.6
other	1	1.1
Child Sex		
Male	49	52.7
Female	40	43
Education		
High school	3	3.2
Associates degree	2	2.2
Partial completion of college	6	6.5
Completion of college	36	38.7
Graduate degree	43	46.2
Live		
Urban	14	15.6
Suburban	55	61.1
Rural	21	23.3
Distance to Ds Care		
< 19 miles	49	55.7
20-49 miles	22	25
>50 miles	17	19.3
	Mean (years)	SD
Age		
Caregiver	44.23	+/-7.93
Child with DS	7.36	+/-6.09

Qualitative Purposive Sample

Email invitations were sent to 20 caregivers, with 11 completing interviews. The final sample included 9 mothers, 1 father and 1 aunt. The age range of the child with DS ranged from 6 months to 14 years old. Seven participants self-described living in a suburban environment, while 2 described an urban living environment and 2 a rural living environment. In all but one

family, the child with DS had siblings, with 2 families identifying the first child as a child with DS.

Qualitative Sample Health Care Providers

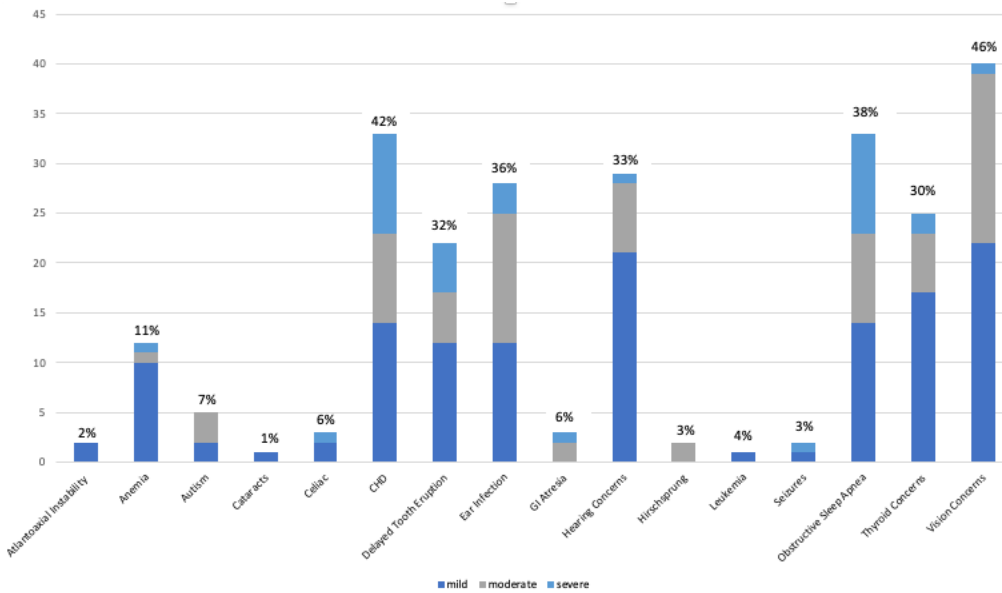
Interviews were completed with ten HCPs, including two nurse practitioners, three registered nurses (RN), four medical doctors (MD), and one respiratory therapist. The RNs served in several capacities including office manager, in-patient transition care nursery and as a case manager. Four major health care systems were represented within this sample, with only two providers coming from an independent practice.

Health Management for Child with Down Syndrome

To better understand the mHealth needs of caregivers of children with DS, it was also important to understand the health care management for their child. This included collecting data describing co-occurring conditions and severity, as represented in Figure 3.2. These values are similar to national averages of co-occurring conditions except for atlantoaxial instability (National Down Syndrome Society, 2021). The discrepancy seen in the height of the bar graph versus the percentage listed, is the percentage calculated from the total number of participants selecting that co-occurring condition, versus the number in the bar graph reporting the symptoms as mild, moderate, or severe.

Figure 3.2

Caregiver Report of Severity Co-occurring Conditions



Most caregivers reported seeing a specialist for the management of some aspect of care (89.9%), ranging from one to twelve specialists involved in managing care. Approximately two thirds of participants reported having been seen at a DS clinic (65.6%), varying in frequency from one lifetime visit to annually. While less than the 25% of families drove greater than ninety minutes for DS care in previous studies, these findings identified 19.3% of participants who reported traveling greater than fifty miles for DS care (Joslyn et al., 2020).

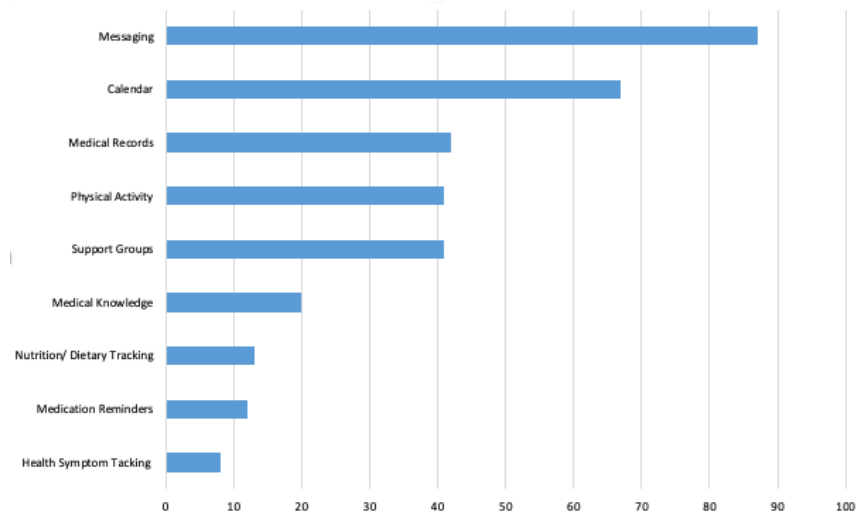
Mobile Technology Use

Respondents also were asked about current mobile device use. Most participants reported using a smartphone as their primary mobile device (97.7%), with only two participants reporting using their tablet. Most participants also reported using iOS (72.4%) as the operating system for their mobile device, with 26.4% reporting using an Android device, and one participant (.01%) using the Windows platform. Figure 3 describes current weekly app use as

reported by caregivers. As reflected in Figure 3, most caregivers used their mobile device for messaging, with over 40% accessing health records through a mobile device.

Figure 3.3

Caregiver Report of Weekly Use of Apps or Websites Accessed from Mobile Device



Qualitative Findings

Two themes were identified from the categorizing and indexing of the data and subthemes as part of phase three of the framework analysis. Initially subthemes were organized using the FMSF and themes related to care coordination such as communication, organization, and utilization (Knafl et al., 2012; Skelton et al., 2021). Extracted data and direct quotations were organized in a table under each subtheme. Subthemes were refined and final subthemes are listed in Table 3.2. As subthemes were indexed two themes emerged, (1) the challenges related to care coordination and (2) the desired content for a mHealth app expressed by caregivers' and HCPs. Caregivers' and HCPs comments about the care coordination challenges were found to be mirrored by their recommendations for app content (Table 3.2).

Table 3.2

Themes and Subthemes from Framework Analysis of Qualitative Findings

Theme	Challenges	Desired Content
Subthemes	Management of Health Information	Management of Health Information
	Information Sharing	Information Sharing
	Use of Guidelines	Use of Guidelines
	Tracking Health Data	Tracking Health Data
	Resources	Resources
	Technology Use Concerns	Design Features
	Previous App Use- negative examples	Previous App Use- positive examples
	Coordination of schedule	Coordination of Schedule

Theme One: Challenges

Each subtheme related to ‘challenges’ is described below, with exemplar quotes and number of times subtheme mentioned being provided in Table 3.3.

Table 3.3

Exemplar Quotes of Challenges and Number of Participants Expressing Each Subtheme

Subtheme	Caregiver	# Caregivers	Health care provider (HCP)	# HCPs
Management of Health Information	“But then in the hospital down here doesn’t do the MyChart, it’s hard to transfer the files from what I have on my phone to them and once I go and find them and it out or email them to 6 different people, because they don’t participate in the same chart system or whatever” (CCC003)	11	“So, it is just a pain from every aspect. Whether you’re a provider or a parent, I feel like it is just a lot because here, when we try to log in, when we have patient, even though we’re all on Epic, it’s really hard. We can’t see what’s going on at other places.” (CCHC006)	8
Information Sharing	“There was no communication between groups there, ... I don’t even think he sent a referral to the pediatrician, because she didn’t know anything about it when I met with her last week, so I just told her the findings. And it’s fine since I’m on top of everything, but for parents who are more intimidated or just not maybe as organized, this’d just be a nightmare” (Caregiver, CCC006)	9	“Because I do feel badly. Sometimes those things just fall through the cracks. They might’ve been seen by different people, or the provider, I don’t know.” (CCHC003)	7

Use of Guidelines	“And some things that you may mention (guidelines), I love our pediatrician here, but some things that you may mention to a pediatrician, and they just don't have the experience or exposure to really put it in context for themselves or for me.” (CCC011)	5	“And I pull up that article, I guess it's the AAP guidelines, some probably old, but for me, it's confusing because I'm trying to look at that. I'm like, "Did you do this?" And so, I think it was because she went to genetics and kind of got everything checked off that they were pretty on top of things, on schedule” (CCHC004)	5
Tracking Health Data	“at least just save the questions that you want to remember to ask when you are sitting down with them, which that's one thing that I think would be hugely important because even with MyChart, you don't have those kinds of options”. (CCC007)	8	“I think one of the things that would probably be helpful for families is to ask them what they need. Sometimes as care providers, we may make assumptions that a person needs XYZ, but it may not be what they need. They may need something else, but to ask them what they need, to make sure that they're involved in their own care and try to support them as best as we can, but also allow them to have the autonomy to make sure that they are keeping abreast of what needs to be done.” (CCHC009)	6
Resources	“When a child is born with Down syndrome, how do we reach those families? Because I Googled like everyone else has, I'm sure. And you know what you see if you Google. Not great stuff.” (CCC008)	4	“We may or may not be thinking about that, and then the family is not even realizing that there are these different services and different sort of supports that they can get.” (CCHC003)	5
Technology Use Concerns	“And I don't like the thought that my information will be stored in the Cloud and anybody that is tech savvy can hack in there and find out my schedule” (CCC003)	2	“The biggest concern, of course, is confidentiality and security type thing. That's always the main thing with health records.” (CCHC007)	4
Previous App Use- negative examples	“And the baby's feed app also didn't have a lot of details for feeding that would be related to more special needs kiddos, because it just said you can put in a meal and then, "Was it good or bad?", basically. Whereas we needed to track how many ounces of puree he'd eat, how many ounces of milk did he have? Did he try any self-feed solid foods? We needed to be a bit more detailed.” (CCC004)	8	“Like the app for that newborn is probably great for parents for whatever reason, but not so helpful for me, on my side.” (CCHC004)	3
Coordination of schedule	“We had a lot of doctors' appointments, and it took a lot of coordination to get everybody where they needed to be, when they needed to be” (CCC005)	8	“There's no closed loop. And a lot of it's counting on the mom. So I can order reoccurring lab, monthly for four months or something if I need to but I can't order it to be checked twice yearly or something like that. So if the family doesn't remember and I don't have a cue, then I don't remember there's 1200 patients in my panel.” (CCHC010)	2

Management of Health Information. Health information management was a central subtheme of challenges described by both caregivers and HCPs. Participants reported health information being siloed within different provider offices and not having a central place to store and access health information. To meet this challenge, caregivers described the need to keep hard copies of health information either in a binder, electronic filing system such as Google documents or the calendar within their smartphone. Caregivers reported feeling solely responsible for keeping a complete health record with one mom calling herself her daughter's "primary care manager." Several caregivers identified the primary care office as the "keeper of health information," attempting to have all specialty health information sent to the primary care office.

There were additional factors related to information management that caregivers and HCPs found challenging. Caregivers described information management as time consuming, with two caregivers describing themselves as "lazy" because they had not completed filing of health information. Another challenge detailed by both caregivers and HCPs was managing information when the family relocated. One HCP who worked within the armed services reported the clinic having support services to help manage information during moves, but due to the outdated technology, families still needed to rely on hard copies of charts. This was similar to a caregiver describing an overseas move.

Information sharing. Caregivers and HCPs both expressed challenges related to care coordination with regards to information sharing. When describing information sharing with the health care team, caregivers described themselves as the "middleman," the "bridge," and responsible for "keeping everyone on the same page." This was often due to familial caregivers being the sole person who can access health information across all providers. Caregivers

reported feeling responsible for the sharing of health information between primary and specialty care, with one mother reporting that the primary care provider relied on her to provide information about other health visits. One mother described that all her providers use MyChart, but each had different levels of access, and had difficulty accessing information outside of their specialty. Another caregiver reported that even with specialists housed within the same hospital system, they often could not see information from other health care teams and relied on the caregiver to communicate health information. A caregiver described frustrations with having to repeat the child's health history at each visit.

HCPs expressed similar challenges related to the fragmentation of information and their inability to share information with other providers. HCPs also wanted a more efficient way for primary care and specialty providers to exchange information. One HCP gave the example of providing a note for the family to share with the specialty provider at the next visit. However, by the time the specialty visit had occurred the family had misplaced the note and the provider needed to fax the information emergently at the time of the visit.

Use of guidelines. The use of the AAP guidelines for care management of a child with DS, was expressed as a challenge by both caregivers and HCPs. While all participants who addressed the guidelines felt it was a vital tool for management, challenges surrounded the usability and accessibility of the guidelines. One HCP stated being unsure of who should have "ownership of the guidelines," the provider or the parent, leading to confusion over who was monitoring adherence to guidelines. Both caregivers and healthcare providers described challenges with accessibility of the guidelines both in having the document available at the time of the visit and in the readability of the table. Two HCPs reported feeling overwhelmed by the recommendations and feeling fearful of missing a component of care management. Even with

the detailed nature of the guidelines, one provider expressed feeling unsure of the timing of the referrals. Possibly because of these concerns from HCPs, both caregivers and HCPs felt the need for children to be seen in a DS specific clinic. However, families described encountering long wait lists for care. One caregiver even reported receiving pushback from their HCP on the necessity of adhering to the guidelines.

Tracking Health Data. Many co-occurring conditions with DS (e.g., congenital heart conditions, atypical development, obstructive sleep apnea) required monitoring, with caregivers responsible for tracking health data to report to care providers (Pikora et al., 2014). Both caregivers and HCPs described challenges with documenting growth on DS specific growth charts. Caregivers and HCPs also described challenges related to tracking information related to medications and labs, while caregivers also went on to describe needing to track dietary needs. Caregivers described various strategies for tracking data, including calendars, check lists, and installing white boards to keep track of completion of “homework” related to different therapies and activities aimed at promoting the child’s optimal development. Caregivers also discussed challenges surrounding tracking therapies and children’s development such as remembering the age/date of certain developmental goals were met, vital information when setting individualized education plans (IEP) with school staff.

Previous App Use. In describing previous app use, caregivers reported challenges they encountered when they tried to use growth and development mHealth apps. As with tracking health data, challenges arose from not being able to find a Down syndrome specific growth curve within mHealth apps. Other caregivers encountered difficulties when they attempted to document concerns about possibly atypical development. One caregiver described abandoning

use of an mHealth app that allowed for some individualization of developmental tracking because it was not DS specific.

HCPs also described different challenges with mHealth app use. One HCP stated that an mHealth app would not make efficient use of visit time because of the time needed to screen all the stored data for relevant information. Providers also described poor previous experiences with app use and the poor quality of existing mHealth apps tied to EHR systems. One HCP highlighted the poor quality of the EHR mHealth app as “garbage in, garbage out.” As with caregivers, HCPs described already using apps and thinking information was already stored across too many apps. Additionally, an HCP expressed concern about the accuracy of the information if the app was reliant on caregiver input of health information.

Resources. Caregivers and providers identified multiple challenges related to resources and access to educational resources. Caregivers reported challenges in finding accurate information on co-occurring conditions, common concerns, anticipatory guidance, and clinical trials. Caregivers also wanted a way to access information on support groups. Lastly, caregivers reported challenges related to the presentation of information, with one mother reporting that she was offended by the language used in some of the information sources (e.g., birth defects).

HCPs reported challenges related to maintaining an inventory of resources for families, with one provider saying that all her resources are kept in her head. Another provider described challenges encountered when trying to explain to caregivers the necessity of the multiple components of health management some children required, noting that resources to support those conversations were needed. Providers described the challenges of finding appropriate support groups to recommend to caregivers and not knowing how to advise families about accessing

information they needed. HCPs reported using web searches and the insurance companies to find information for families.

Technology Use Concerns. Trust was the most common challenge expressed by caregivers and HCPs related to concerns about technology use. Trust concerns included those related to the privacy of health information, technology use, and data entry. One caregiver expressed a hesitancy to use an mHealth app because of having lost data from her mobile device when it was accidentally deleted by her child. HCPs also stated concerns about health information privacy and security, with one HCP expressing families' concerns of having one provider seeing all health information. Lastly, challenges with usability factors were described by both caregivers and HCPs within the subtheme of technology use concerns. These included comments related to lack of technology knowledge, time consumption, and the complicated nature of mHealth apps.

Coordination of Schedule. Data for this subtheme came predominantly from caregivers. Only two HCPs mentioned scheduling as a challenge in relation to scheduling multiple specialty appointments and monitoring AAP guidelines. In contrast, eight caregivers described multiple scheduling challenges. Caregivers described challenges in remembering multiple appointments and efforts to maximize efficiency, by scheduling multiple appointments at one facility on the same day. Caregivers also described challenges with scheduling with regards to other responsibilities related to the child's care such as medication administration, meals, and lab studies. As was with the information management subtheme, caregivers described strategies for overcoming the scheduling challenges such as creating family calendars, having calendars in every room of the house, and using technology such as Google calendar.

Theme: Desired Content

A second theme was identified within the data as ‘desired content for an mHealth app’. As data were analyzed in phase three it became clear that the subthemes for desired content were linked to the subthemes identified for challenges. Findings from each of the subthemes are described below.

Management of Health Information. Caregivers and HCPs described several desirable features of an mHealth app relating to the management of health information. Mirroring the challenge expressed by both caregivers and HCPs that information was siloed, respondents recommended development of an app that would support relevant information being organized in one location. HCPs stated that organization of information in one location could contribute to more efficient use of appointment time. Caregivers expressed the need for an app that would allow them to create a one-page summary of health history to give to providers. Two caregivers noted a desired feature in the ability to upload PDFs to a single location or folder. Another feature of information management reported as a desirable app feature by caregivers and HCPs was the capacity to store contact information for different providers involved in care. The storage of provider contact information was also described as a desirable app feature by a HCP, noting that without a set place to keep contact information, they would often use the child’s problem list within the EHR.

Information sharing. A second subtheme of information sharing provided important insights into desirable features of a mHealth technology. Three caregivers mentioned that a key desired feature of an mHealth app would allow partnered families to share and access information with one another and with others involved in childcare. Two caregivers expressed the desire to share feeding and nutrition data with specialty providers prior to visit.

Caregivers and HCPs described several other features of information sharing and communication. Caregivers and HCPs wanted to be able to share lab data across specialists and the primary care medical home so all providers could be up to date on current lab findings. Caregivers also expressed the utility in having a chat feature with providers included in a mHealth technology.

Guidelines. While use of the AAP guidelines could overlap with information management or tracking of health information, there were enough unique responses related to the guidelines to justify it being a separate subtheme. Both caregivers and HCPs expressed the desire to use the guidelines as a checklist within a mHealth app modality. Embedding the AAP guidelines was particularly desirable for the ability to tailor recommendations based on age as well as determine the timing and need of referrals and monitoring. Further, an HCP described a desire to use the guidelines within a mHealth app to prioritize care needs for families and reorder items based on an individual child's needs. One caregiver expressed wanting to use the guidelines to organize anticipatory guidance information for future care needs.

Tracking Health Data. The option for tracking data was a desirable app feature expressed by both caregivers and HCPs, but this was particularly evident in interviews with caregivers. Areas in which caregivers and HCPs wanted to track health data included feeding, monitoring growth on growth charts, medications, and lab findings. Caregivers discussed how they would like an app that would support tracking data related to the amounts and types of food their child was eating, monitoring for food sensitivities or allergies, and medically complex feeding needs such as tube feedings. Caregivers also mentioned tracking health data within a mHealth app by logging data about output, both for stooling concerns and toilet training. One

caregiver described a desire to track sleep data to monitor for co-occurring conditions such as obstructive sleep apnea.

Other suggestions for tracking health data related to developmental monitoring. Both caregivers and HCPs discussed the tailoring of developmental tracking to meet the child's individual needs. Lacking in current EHR apps such as MyChart, caregivers envisioned using an mHealth app to track developmental goals and “homework” working on with various therapies. This information could then be communicated to providers in an organized manner.

Both caregivers and HCPs described the importance of developing an app that would support caregivers' ability to keep notes. One mother described this as a way to protect against “mommy brain” or feeling scattered and forgetful. Participants included examples of notes such as documenting questions for the providers, or behaviors/symptoms to discuss at next visit. Two caregivers described a desirable feature of notetaking as being able to embed videos or pictures of concerns to share with provider.

Resources. There was considerable overlap between caregivers and HCPs in desired content related to resources. Caregivers and HCPs emphasized it was important to ensure educational resources came from reputable sources, that were DS specific. HCPs stated it was also important that health information conveys the rationale underlying referrals and monitoring studies. As with the AAP guidelines, both caregivers and providers suggested that health information be presented in an age specific manner and tailored to the families' desired amount of information. In addition to being age specific, HCPs wanted health information to be available regarding other transitions of care such as sexual development, vocational training, and community services. Caregivers specified resources include information on dietary and dental needs as well as common co-occurring conditions. One provider also stated it would be helpful

to include Early Intervention resources since families are able to self-refer to those without a referral from a provider. Both caregivers and HCPs specified informational needs regarding advocacy and clinical trials.

The desire for including resources to support caregivers in the app was reported by both caregivers and HCPs. Caregivers described not only the overall need for parental support but also the need for subspecialty groups related to specific concerns. Interviews included data on caregivers' desire to want to learn from other's experiences.

Design. Caregivers and HCPs provided valuable insights into design elements that would enhance the potential use of a mHealth app. Desirable features related to design include clear icons for data entry such as labs, body systems and provider information, as well as a simple, clean format, specifically designed for mobile technology. Other design elements mentioned included a Q&A section for parents, the capacity to auto-populate provider information, and to tailor input to the families' information needs. One caregiver suggested that the capacity to turn off certain app features or limit the amount of information required when a co-occurring condition was no longer being monitored would be a desirable app feature.

HCP noted the importance of language choice within the mHealth app. Two providers encouraged the use of language that promoted neurodiversity, and moved away from terms such as developmental milestones, into more inclusive language, such as developmental goals. Another HCP recommended positive and supportive language use throughout the app.

Previous App Use. Data reflecting this subtheme addressed positive past experiences that caregivers reported making them more likely to use an mHealth app, with most data addressing accessibility. Three caregivers and two HCPs described experiences using mobile technology to store and access information. One provider commented on the potential for

mobile technology to reach and provide information to families in more rural communities. Two caregivers related potential app use to positive experiences tracking their child's early dietary habits, and one provider discussed success of a health app using a checklist format to organize immunizations. Caregivers and providers also expressed enthusiasm about the potential for a mHealth app related specifically to DS health information. Lastly, one caregiver and health provider, discussed the potential of a mHealth app to support their ability to complete timely, efficient reviews of health information.

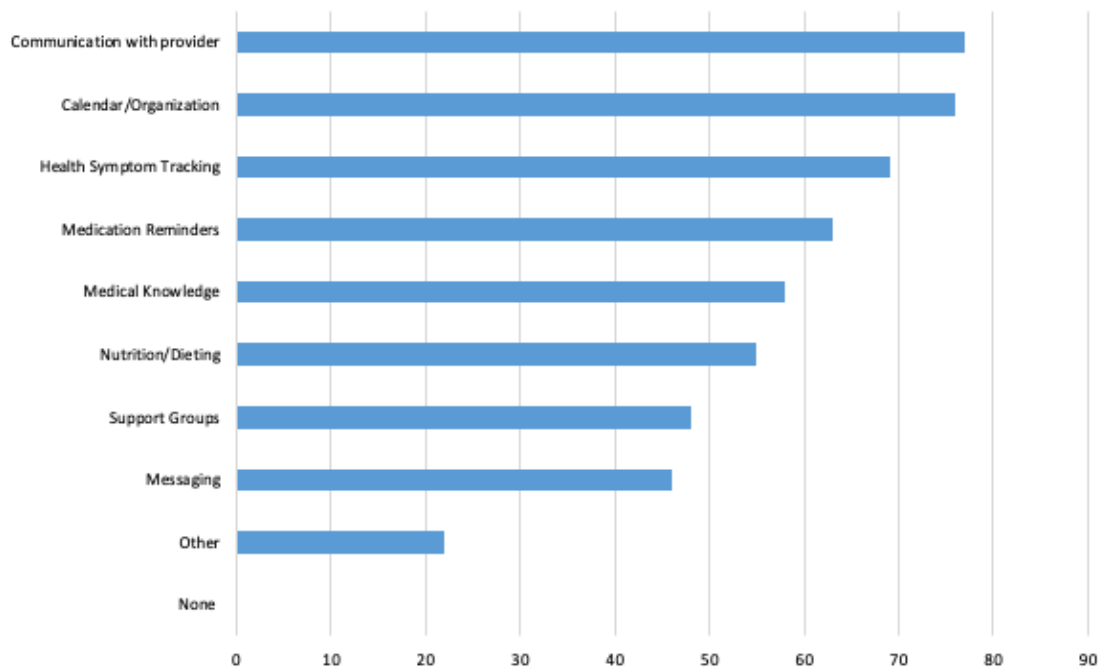
Coordination of Schedule. A majority of caregivers interviewed, discussed the benefits of using a mHealth app to coordinate the schedule of their child's upcoming care needs. This included the ability to maintain a calendar of scheduled appointments and receive reminders for appointments and upcoming care needs, including lab monitoring, medication administration, and behavioral monitoring of such things as toilet training and developmental therapies. The two providers who commented on schedule coordination mirrored caregivers' desire for reminders for upcoming care needs. Caregivers expressed the desire for reminders to be delivered in the form of pop-up reminders.

Linked Findings. Questions from NSCH-D were included to gain an understanding of respondents' current care coordination management and compare to finding of the qualitative analysis. Caregivers for the most part felt their providers provided necessary information, spent enough time, and showed compassionate care. However, less than half of participants received help in coordinating care (36%), when 63% of caregivers felt they would have benefitted from care coordination. Caregivers also reported that only 35% were very satisfied with communication with providers with the rest of caregivers reporting being somewhat satisfied or dissatisfied with communication.

This was also highlighted in interview data with caregivers focusing on needs related to communication and information sharing. Other findings from survey data also support the caregiver needs for communication and organization when considering potential mHealth app use (Figure 3.4). Also reflected in the survey data were the desire for an mHealth to support storage of medical records, provide links to resources, and reminders for upcoming care. This mirrored subthemes from both caregivers and HCP providers related to tracking of health data, resources, and information sharing. Resources were specified as tools for care, health information, and information on clinical trials in both survey data and interviews.

Figure 3.4

Caregiver Report of Potential Use of DS mHealth App



DISCUSSION

Findings from this study of caregivers and HCPs point to a desire to have a tool to improve the management and storage of health information related to care coordination.

Providers also saw potential in an organization tool to manage information from multiple care

teams. The portability and accessibility of mHealth apps make this a viable technology to use to manage this health information. Using the findings from the study, as well as what is known about mHealth apps, the results will help aid in the development of an mHealth app.

Development of an mHealth App. Given the descriptions provided by caregivers of communication status with providers, there is a need to continue to find ways to support familial organization of health information and communication tools, an mHealth app has the potential to fill that void. Results point to a need to design an app that would be compatible with both an iOS and Android platform. Interview data in particular elicited responses that directly related to mHealth app features. Subthemes were able to be linked to potential features in the design of the mHealth app. These features include storing provider visit summaries, contact information, and caregiver notes, as well as tracking health data such as growth charts, medications, lab findings, and development. An mHealth app also has the potential to provide age-specific health information and anticipatory guidance. Given current technology capabilities, several components described in interview data would be considered in future iterations of app development such as interfacing with the EHR and direct communication with providers and peers.

Potential for mHealth App Usage. While it is clear from the data that caregivers are playing an active role in care coordination, a mHealth app has the potential to create a more formalized pathway for a shared decision making and care coordination model (Whitehead & Seaton, 2016). This would help practices to meet the goals of establishing a FCMH, which many families with a child with DS are lacking (McGrath et al., 2011). Technology use also has the potential to expand to different languages, leading to increased PHR access by a wider range of families (Goldsmith et al., 2017). While previous studies looking at the relationship between a

desire for technology usage and distance to care providers have been mixed, access to DS clinics may not be available to all families, and mHealth may be able to fill that information gap (Cady & Belew, 2017; Joslyn et al., 2020; Tozzi et al., 2015). In the future, it is possible to consider linking a care coordination mHealth app with a technology such as DSC2U to integrate care coordination and data management with the use of the PHR (Chung et al., 2021).

Limitations. There were several limitations with this study. First, recruitment was challenging in the context of the COVID-19 pandemic. There were also challenges with the planned method of recruitment of HCPs from caregiver information. Since caregivers often communicated with providers through portals, challenges arose when trying to reach providers through the main office number. The overwhelming majority of respondents were mothers, with a lack of representation of other caregivers in the sample. With a focus on primary care in the medical home, the perspective of specialty providers was missed. Specialty providers may have had insight into specific co-occurring condition management tracking and information needs.

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CHAPTER 4: INITIAL USABILITY TESTING OF A MHEALTH APPLICATION FOR THE MANAGEMENT OF PERSONAL HEALTH RECORDS FOR FAMILIES OF CHILDREN WITH DOWN SYNDROME

SUMMARY

mHealth has been shown to have a positive impact on chronic condition management, however few studies have investigated the use of mHealth to serve as a personal health record (PHR) to aid caregivers in care coordination. PHR's include health information, managed by a family or caregiver, to aid in the management of chronic conditions with the health care team. PHR's are particularly important for a chronic condition like Down syndrome (DS) which may require care from multiple health care providers. This article describes the development of a mHealth app using a user centered design approach to serve as a PHR for caregivers of children with DS. The development of a prototype app, 321Connect, was informed by findings from a scoping review and a mixed methods analysis of caregivers of children with DS. Initial usability testing of 321Connect was undertaken by six caregivers by completing usability exercises, a semi-structured interview and completion of the system usability scale survey (SUS). Overall caregivers found 321Connect to be a highly usable tool, with an SUS score of 90.8, that meets their needs as a PHR. Caregivers provided suggestions to increase usability. While initial usability testing is promising, further usability and pilot testing is warranted.

INTRODUCTION

Technology is rapidly changing how we manage our health information. Patient portals to electronic health records (EHR) have expanded families' access to health information and communication, however information often remains siloed between different health care providers (Allshouse et al., 2018). This is particularly evident for families of children with a genetic condition, such as Down syndrome (DS) where multiple providers and health systems may be involved in care (Williams et al., 2017). Thus, the need to maintain a personal health record (PHR) has emerged. A PHR is unique from a patient portal in a EHR because it is not tied to a particular health system or provider and serves as a family's way to maintain and organize health information across health systems (Dameff et al., 2019). However, there is no well-established mechanism for families to maintain a PHR. The portability and accessibility of mHealth apps make them a viable system for maintenance of a PHR (Zhou et al., 2019).

Potential for App Use

There is great potential to use a mHealth app to maintain a PHR as well as to support families' care coordination needs (Tozzi et al., 2015). mHealth apps have proven successful as a PHR for families of children following bone marrow and organ transplants (Lerret et al., 2021; Maher et al., 2016). Additionally, mHealth apps have improved collaboration with providers positively impacting care management in chronic conditions such as cystic fibrosis and inflammatory bowel diseases (Opipari-Arrigan et al., 2020). mHealth apps have aided families to take a more active role in health care and using the platform to engage in therapeutic conversations with health care professionals has improved family adaptation (Choi & Van Riper, 2020; Tozzi et al., 2015).

In addition to use as a PHR, mHealth apps have the potential to help families determine whether the care their child is receiving is in accordance with established specific health care guidelines. mHealth has successfully been used to monitor asthma care in accordance with health care guidelines (Morita et al., 2019). The American Academy of Pediatrics and the Down Syndrome Medical Interest Group created guidelines for the care of a child with DS (Bull et al., 2011). Embedding these guidelines in a EHR has been shown to improve providers' adherence to recommended guidelines (Santoro et al., 2018). While these examples show successful use of mHealth in chronic conditions, there is not currently a mHealth app to support caregivers' efforts to manage the health care needs and PHR of children with DS.

Development Considerations

When approaching the development of a mHealth app, it is critical to use a methodology such as user centered design to ensure the app meets end-user needs (McCurdie et al., 2012). User centered design is defined as “an evidence-based approach informed by the needs and understanding of a specific end-user group” (McCurdie et al., 2012, p. 49). While the end users are the most important group to consider in development, it is also critical to include all stakeholders in the development of a mHealth application (Kao & Liebovitz, 2017). This includes health care providers (HCPs) as well as other individuals with whom caregivers may interact across health care settings. Design considerations also include ease of use as well as the ability to tailor the app for families' specific needs (Konig et al., 2021; Schnall et al., 2016).

Employing user-centered design makes it possible to identify end users' input of desirable characteristics of the app. For example, caregivers found appointment reminders the most beneficial component for asthma self-management, while providers found the mHealth app most important for information sharing, education, and monitoring of symptoms (Geryk et al.,

2016; Slater et al., 2017). These are similar findings to desirable features related to content and information sharing of adolescents and HCPs managing chronic juvenile arthritis (Waite-Jones et al., 2018). Desirable features included caregivers wanting an app that made it possible to track symptoms that could be shared in the form of notes and pictures with an HCP (Nkoy et al., 2019). Characteristics identified in user-centered design studies included components such as medication logs and reminders, pharmacy information, and desire for communication with providers and peers (Schnall et al., 2015). Findings such as these can contribute to the inclusion of desired features in mHealth applications.

The purpose of this study was to develop a mHealth app prototype and complete preliminary usability testing guided by the principles of user centered design (McCurdie et al., 2012; Dopp et al., 2018). User centered design occurs in 3 phases: (1) concept design, (2) prototype development, and (3) evaluation (McCurdie et al., 2012). These phases are defined in Table 4.1.

Table 4.1

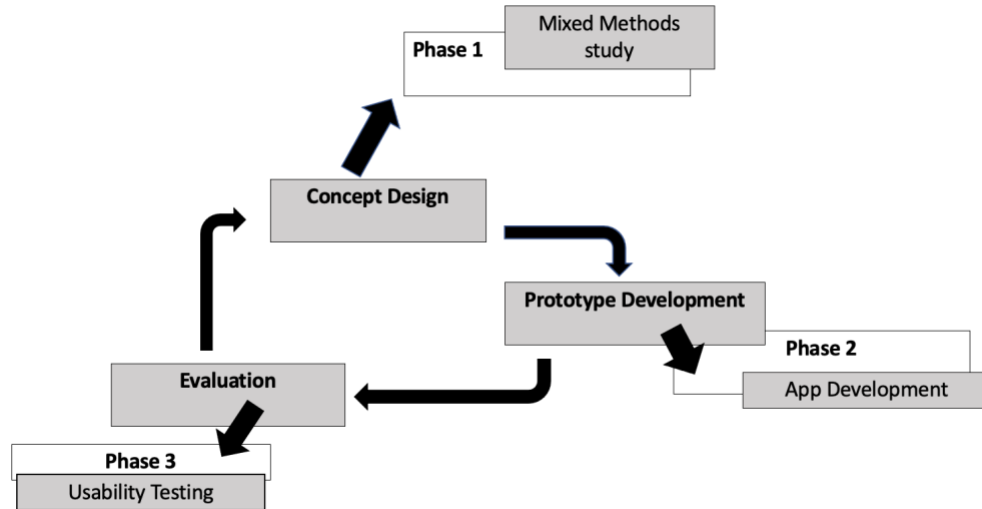
Three Phases of User Centered Design

Concept Design	Understanding the intended use of mHealth application and user requirements in design
Prototype Development	Mockup of features and design elements into app technology
Evaluation	Walkthrough and usability testing to determine if app is meeting intended design goals

Findings from Phase 1, concept design, were reported in Paper 2. These findings served as the proof of concept to aid in the development of the app prototype. This paper describes the prototype development and usability testing as part of Phase 2 and 3 of user centered design (McCurdie et al., 2012). Figure 4.1 diagrams the three phases of user centered design as used in this project.

Figure 4.1

Phases and Products of User Centered Design



PHASE 2: PROTOTYPE DEVELOPMENT.

Design

Building on the characteristics of a desirable app identified during the mixed methods study of Phase 1, BC worked with Dr. Elizabeth Baker (EWB) and her research lab at Virginia Commonwealth University (VCU), to develop the features of the prototype mHealth app. Table 4.2 summarizes desirable app content and features from Phase 1.

Table 4.2

Desired Content and Features for mHealth App Development

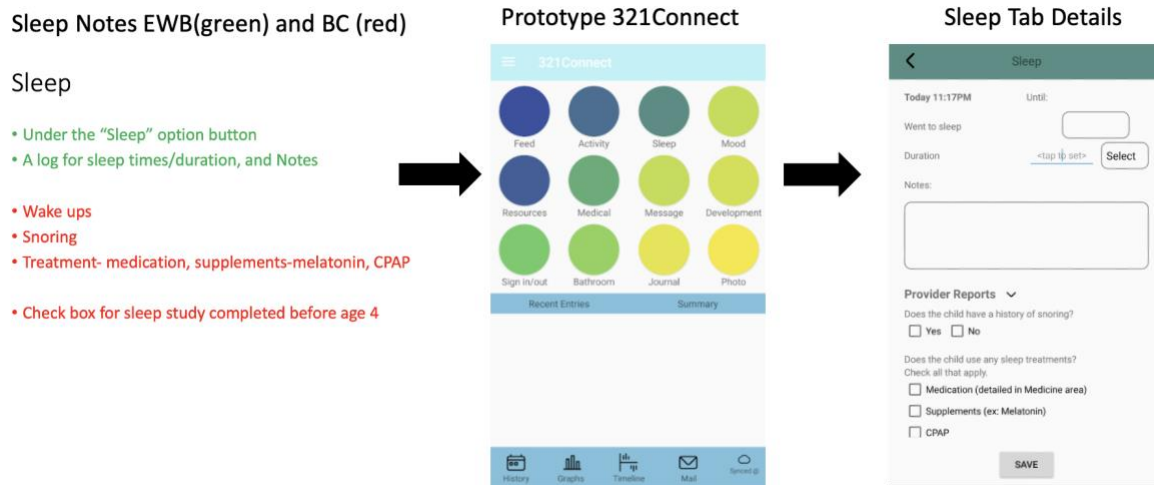
Desired Content	App Features
Management of Health Information	Storing provider notes Contact Information
Information Sharing	Note Taking
Use of Guidelines	Embedding AAP Guidelines
Tracking of Health data	DS Specific Growth Charts Medication Tracking Lab Results Developmental Tracking
Resources	Age-Specific Health Information

Design Features	Inclusive Positive Language
Coordination of Schedule	Calendar/Schedule

Because we were working remotely, we preserved our work on the content of the various sections of the mHealth application using PowerPoint. EWB and BC worked collaboratively integrating the Phase 1 results into an app that incorporated EWB’s knowledge of app development to design the layout and color schemes. We also incorporated other findings from Phase 1 such as the importance of inclusive and positive language use. We conferred regularly through email and Zoom meetings. As part of the design process, we developed a name for the app, 321Connect. 321 is a number commonly associated with DS because approximately 95% of children with Down syndrome have Trisomy 21 or three copies of chromosome 21, which is one more copy chromosome 21 than most children have (Bull, 2020). Figure 4.2 gives an example of one of the PowerPoint layouts as content for 321Connect was shaped and developed. Each section went through multiple iterations of design based on findings from Phase 1 and communication between EWB and BC throughout the design process. Initial development of 321Connect occurred for the Android platform. The iOS version is currently under development and will undergo usability testing when completed in early 2022.

Figure 4.2

Examples of mHealth Content Development



PHASE 3: EVALUATION

The final stage of the user centered design process is evaluation (McCurdie et al., 2012). The evaluation process was designed to assess initial usability as well as the acceptability of design elements. Evaluation entailed testing of users’ ability to navigate the app, completion of the usability scale, and interviewing users to elicit their views of positive and negative aspects of the app, and recommended changes that would strengthen usability (Broekhuis et al., 2019). As a part of the cyclical nature of user centered design, these findings will be used to refine 321Connect for future usability and pilot testing.

Sample

The purpose of Phase 3 Evaluation was to complete usability testing with caregivers of children with DS as the end users. The sample consisted of caregivers of a child with DS who had expressed willingness to participate in Phase 3 when enrolling in Phase 1 and through recruitment of personal contacts. Inclusion criteria for Phase 3 included being a caregiver of a child with DS owning an Android phone or local access to use a lab-owned Android device.

Exclusion criteria were defined as not meeting eligibility requirements. Usability testing recommends five to eight participants to detect usability concerns (Nielsen & Landauer, 1993). This sample size has been found to uncover the majority of usability errors while maintaining cost-effectiveness (Nielsen, 2012). Emails were sent to twelve participants from Phase 1 and six contacts within the North Carolina Down Syndrome Alliance (NCDSA). Six caregivers expressed continued interest in participating in Phase 3 usability testing and completed consent forms.

Measures

Usability Exercise. To assess user ability, each participant completed a series of standardized data entry tasks (Appendix 4.1). Tasks are structured to guide participants in creating an account and adding caregiver and primary provider profiles as initial steps of app usage. Following account creation and developing a profile, participants were asked to create a child profile and to log the child's daily activities such as sleeping, eating and output. The last task was to document a health care provider visit.

Semi-Structured Interview Guide. Following completion of the usability exercise, each participant undertook a brief semi-structured interview. Four questions were asked: "what were your initial impressions of 321Connect?," "what did you like most about 321Connect?," "were there things you didn't like about 321Connect?" and "what would you change about the app?" BC took detailed field notes of participants' responses as well as behavioral observations while participant interacted with various components of 321Connect.

System Usability Scale. The System Usability Scale (SUS) is the gold standard for testing learnability and usability of a mHealth application (Brooke, 1996; Healthcare Information and Management Systems Society, 2012). This 10-item questionnaire addresses effectiveness

and efficiency as a measure of usability (Appendix 4.1) (Lewis, 2018). The SUS has been validated and had strong reliability in previous studies ($\alpha = .70$ to $.91$) (Lewis & Sauro, 2009).

Procedure

Participants who indicated willingness to participate as a part of Phase 1, received an email containing a description of Phase 3 and consent information. Once an interview time was established either in person or via Zoom, a second email containing directions to download the app from the Google Play store onto an Android device, as well as a written copy of the usability exercise instructions (Appendix 4.1) was sent to the participant. This email was sent immediately preceding the interview time to reduce access to the app prior to usability testing and observation. When the interview was occurring in-person, BC brought a lab-issued Android phone with the app downloaded onto the device, but without a profile created within the app. In-person testing occurred at an in-person outdoor setting (precaution due to COVID) with internet access. Because usability testing is about assessing how the app may be used in real life, no attempts were made to control the environment for either the in-person or Zoom interviews (McCurdie et al., 2012).

At the start of each interview, BC reviewed the consent information and encouraged the participant to ask questions. Participants started with usability exercises as described above and in Appendix 4.1. While the participant was completing each task, BC took notes on the time engaging in each section, any navigation issues, and any verbal responses from the participant. Participants were then asked to navigate the app, exploring different features of interest. BC highlighted several features related to the medical visit documentation and the use of the AAP guidelines to elicit feedback. While participants were freely exploring the app, semi-structured interview questions were asked of the participants to gain additional feedback regarding likes

and dislikes of features of the app. Immediately following the usability exercises and interviews, participants completed the SUS survey either by following a link to the survey sent via email or by paper and pencil if in-person. Interviews were completed by asking if there were any final thoughts or questions and participants were thanked for their participation.

Analysis

Interview. Consistent with guidelines proposed by McCurdie et al., (2012), framework analysis was used to analyze responses to the usability exercise and semi-structured interview as a part of Phase 3 and the evaluation process (Richie et al., 2014). The framework analysis was guided by the ten categories of heuristic design for usability testing developed by Jakob Nielsen (Nielsen, 2020). These subthemes include viability, match of app and the real-world usage, user control and freedom, consistency, error prevention, recognition, flexibility, aesthetic, help, and documentation (Nielsen, 2020).

SUS. The 10 items of the SUS are scored together to create a single adjusted score on a scale from 0 to 100 with 0 being the lowest score and 100 the highest score as a measure of usability. There is considerable literature related to the SUS and norming of scores. Norming for this study followed the Sauro-Lewis Curved grading scale (CGS) (Lewis, 2018; Sauro & Lewis, 2016). The Sauro-Lewis CGS has an average score of 68, with benchmark score of 80 or above for usability (Lewis, 2018). A score of 80 equals a percentile rank of 90-95% (Lewis, 2018).

RESULTS

Interviews of six caregivers were completed for Phase 3. An additional partner of a caregiver who was present during one interview was consented to participate and provided feedback related to the app but did not complete the usability exercise or the SUS. Participants

included four mothers, one aunt, and two fathers. Caregivers reported ages for their child with DS from three to nine years old. Four interviews were completed in person using a lab supplied Android device, and two interviews were completed with native Android users via Zoom.

Guided by the principles of framework analysis, data were indexed (i.e., coded) with the ten categories of heuristic design described by Nielsen (2020) (Gale et al., 2013). Six of the ten categories were present in the data from the usability tests and interviews. Following indexing, data were charted into a matrix summarizing data based on each heuristic category for each respondent (Gale et al., 2013). The final step of the analysis was to review the summarized data for each category and draw conclusions about the strengths and limitations of the app and areas in need of further development.

Match of 321Connect to the Real World

Caregivers found many useful connections of 321Connect to how they have previously or would like to manage health information. Two caregivers described 321Connect as “filling a void” and “love the concept” of creating a mHealth app to be used as a PHR. Across the six interviews, at least one participant commented on how much they liked each of the different tabs created for data. These tabs include sleep, bathroom, medical, and development. Four caregivers commented on the importance of managing data related to feeding particularly with regards to different feeding types including use of bottles and tube feedings. Other sections that elicited multiple positive comments included development and sleep.

Caregivers also commented on challenging aspects of navigating the 321Connect and made suggestions for edits that would increase the usefulness of 321Connect as a PHR in matching real-world data. Caregivers had difficulty documenting blood type. While caregivers were able to document the blood type letter, they were unable to document whether blood type

was positive or negative. It was suggested to add a field for positive/negative blood type to the profile page. Another documentation field that presented challenges for caregivers was the height field. Depending on an inpatient or outpatient setting, height values are reported using different units of measurement such as centimeters versus inches. Although caregivers were given a height value in feet and inches during the usability exercise, the data entry field did not specify measurement unit and only provided one field for data entry. Three participants attempted to calculate feet to inches, while another abandoned documenting height.

Other suggestions made by caregivers related to real world use of the PHR and included the ability to add more details related to tabs with regards to feeding, bathroom, sleep, and health tracking. These suggestions included the ability to document adding thickener to feeds, titration of medication for constipation within the bathroom tab and naps under sleep tab. Children with co-occurring conditions such as congenital heart conditions may be eligible for the RSV vaccination Synagis, and it was recommended by one caregiver to add Synagis administration under the medical tab (Committee on Infectious Disease and Bronchiolitis Guidelines Committee, 2014). Additional desired documentation features from caregivers included the ability to document care for other children in the family to keep all health information in one location to give multiple caregivers access information from a single profile. Lastly, one caregiver expressed a desire for the question “have you been connected to your local support organization?” to be added to the medical tab. Incorporating these suggestions through the addition of new fields within 321Connect will help to ensure complete PHR documentation for caregivers.

Aesthetic and Design

Caregivers expressed many positives related to the design including statements like “this is so well-thought out” and “even in this initial stage, it is such a great resource.” A majority of caregivers felt that the current tabs cover all information they would want to track related to co-occurring conditions. Two caregivers suggested two additional components of the medical tab to include a place to document neonatal intensive care unit experience and the possibility of adding separate specialist visits for common co-occurring conditions. Two caregivers suggested that medications could be a separate tab on the home screen for easy documentation of dosing and administration. For sleep, two participants recommended refining the design to support calculation of a 24-hour sleep total from the sleep data entry fields. Additionally, two caregivers wanted to have the ability to add their own references under the reference tab to keep a resource library all in one place.

Caregivers expressed positive comments and suggestions related to the visual aspects of 321Connect. Caregivers were drawn to the ability to visually represent data by using the DS growth charts and timelines of documentation. While two participants liked the color scheme, three wondered if instead of circles, tabs could be identified using commonly used icons. One example given was using “Zzz’s” for the sleep tab. There were also two caregivers who suggested clarification on word choice such as fluid vs. liquid diet, duration, “mode” of eating, and mood vs. behavior tabs.

User Control

All participants interviewed suggested modifications related to user control of data entry. Caregivers expressed support for the ability to add pictures, having a dedicated place to documents questions for providers, and being able to access information without a “million

clicks.” One area of user control in which caregivers expressed differing feedback was the use of check boxes versus space to free text. Caregivers suggested additional checkboxes would be useful in tabs for sleep, medications, and allergies, but also suggested free text related to documentation of naps, sleep quality, and type of multivitamin would be useful. Another consideration related to user control that caregivers found challenging was forced entry boxes. Participants reported that forced entry boxes meant that participants had to document in certain fields to be able to save and close tabs. One forced entry tab in which caregivers were divided was the need to document pregnancy due date regardless of age of child when initiating use of app. Caregivers were also divided on need to document height and weight with each visit. Caregivers who wanted to keep the forced entry feature reported it would help caregivers remember to add these data to growth charts. Caregivers who wanted these data removed stated that not all health care visits (e.g., speech therapy) require height and weight to be documented. One caregiver suggested user control could be improved if it was possible to set up a profile via an online website to shift initial data entry to a keyboard instead of the mobile platform.

Recognition

Overall, caregivers demonstrated an ability to recognize and navigate easily between tabs during the usability exercises. There were two areas in which caregivers expressed challenges in navigation within the app. The first was in attempting to return to profile screens after leaving one profile to move onto the next profile. The second was in adding a provider visit under the medical tab. Since the usability exercise asked caregivers to enter a “physical therapy appointment,” two caregivers attempted to go directly to the physical therapy tab instead of the “add provider visit” tab. Additionally on two occurrences, participants attempted to enter free text data related to diet prior to realizing check boxes existed for diet components. To help

alleviate these navigation errors, one caregiver suggested moving check boxes above the free note section. Another caregiver also suggested that adding an additional notes section after each section of the development tab would help caregivers use the free text option to document additional details related to child development.

Consistency

While caregivers overall found components of 321Connect to be identifiable and consistent, they also described app features they found challenging related to data consistency. These included data entry for the month of the healthcare visit. Caregivers liked being able to select the date by month name but found the switch to a month number in other parts of the app confusing. For example, caregivers would select September 10, 2021, but their entry appears as 8/10/21 in another portion of the app. Caregivers also found the “select visit” tab under provider visit confusing because it was grayed out for all visits except for pediatrician visits and only became an active tab for a pediatrician visit to document the different scheduled age specific routine health care visits. When BC clarified with participants the purpose of the tab, they stated it was a beneficial feature. Additionally, three participants mistakenly entered city and then state, following typical data entry, whereas the tabs are listed state and then city. The final consistency issue arose from the parent profile tab in which the “+” arrow did not work as it did with the child and provider tab, resulting in caregivers skipping that data entry task within the usability exercise.

Errors and Error Prevention

Caregivers completing usability exercises identified several data entry errors, but also successfully avoided errors when receiving error messages. Participants reported that pop-up error messages alerted them to missing data entry fields in creating profiles and prompted them

to find the missing field and complete data entry. Error prevention was also successful when one participant accidentally closed out of a tab without saving but the app automatically saved so none of the data were lost.

The usability exercises also uncovered several correctable errors in the 321Connect prototype. Some of these errors were addressed within other themes above but also revealed free text fields within allergies and feeding, where the participant's cursor moved, they were unable to see typed text. Participants also reported data in profiles showing up within other profile fields. For example, one participant reported that information entered for the child's profile showed up under the provider profile. Other errors may have been related to device errors, for example the app closing out unexpectedly or freezing in certain fields. These errors did not happen consistently across users. Lastly the keyboard on the screen blocked the "save" button on several fields but this error only occurred with non-native Android users.

SUS

Six participants completed the SUS following the usability exercise and interview. The range of scores from the six participants was 80 to 100 with an average score of 90.8. The standard deviation was 8.2. Using the norming benchmark score of 80 from the Sauro and Lewis CGS, the average of 90.8 exceeds expectations and has percentile rank of 90-95% (Lewis, 2018; Sauro & Lewis, 2016). Percentile ranks have been equated to user adjective rating and 90.8 ranks in the excellent range (Bangor et al., 2008). The small sample size for this initial usability testing limited any additional demographic variables being analyzed.

DISCUSSION

The findings from this initial usability testing point to a highly desired tool for caregivers of children with DS to manage a PHR. While caregivers described edits to increase usability

above, the overall concept, structure, and design point to a tool that could fill the gap of maintaining a PHR for families. Caregivers expressed the desire to start using 321Connect for current management and the wish that this tool existed in infant years.

There are several avenues for continued usability and pilot testing for 321 Connect. First, 321Connect is in development for an iOS platform. Once complete, the iOS version will undergo the same usability testing described here. This is a critical step because a majority of caregivers participating in Phase 1 reported using an iOS platform. Following iOS usability testing, the next step is to complete pilot testing of 321Connect. Because this is a tool to gather information and complete documentation as part of a PHR, testing needs to include use of 321Connect in daily life for information management. A three-month pilot study would gain understanding of daily and weekly use of the app. Other pilot studies have shown positive findings related to self-management and satisfaction outcomes with app use but data of adherence to mHealth app use is still mixed (Hamine et al., 2015). Additionally, 321Connect should be tested using a randomized controlled trial to investigate health outcomes for chronic illness management and family self-management behaviors.

There were several features that were described in the mixed methods study as part of Phase 1, concept design, that were not feasible to include in this early prototype but should be considered in future iterations of the 321Connect. These features include the ability of 321Connect to connect directly with the EHR and the patient portal. With current licensing, EHR's tend to be stand alone, however as medical information technology expands there may be an opportunity for systems to share information. There is also the possibility to link 321Connect with other management tools in development such as Down syndrome Clinic to You (DSC2U) a web-based platform to identify care needs for a child with DS (Chung et al., 2021). Caregivers

also described a mHealth app as a way to connect with peer support groups and exchange resources. Lastly caregivers described a desire for appointment reminders and times that may be stored within the app to be linked to programs such as Google Calendar to help streamline management.

Another consideration in the continued development and maintenance of 321Connect is cost. While the plan is for 321Connect to be free for user downloads, there is a cost in maintaining apps over time and this needs to be addressed in the continued development of 321Connect. There are plans from the Down Syndrome Medical Interest Group to update the clinical guidelines from the American Academy of Pediatrics, which will require an update to guidelines within app when released. However, previous updates to the guidelines have only made minor changes to recommendations. Cost also needs to be considered in a possible data security audit for data safety. While reporting and data of cost effectiveness of mHealth varies greatly, the studies that do analyze cost, show that they may be cost effective (Rinaldi et al., 2020). Additional funding for development and maintenance of 321Connect needs to be considered for future testing.

321Connect and the use of mHealth for the maintenance of an PHR to aid in care coordination have many possibilities for the future. The DSMIG developed care guidelines for adults with DS, to continue care from a pediatric population. Since persons with DS often need care until adulthood, future iterations of 321Connect could also include these guidelines (Tsou et al., 2020). Other considerations for future iterations of 321Connect include being able to translate app into Spanish. Finally, future versions of 321Connect could include data entry for adolescents and adults with DS to promote self-management and engagement in care.

Limitations

There were several limitations for this study. First, there were challenges with regards to recruitment. Initial development of 321Connect was completed for Android, however, it was found in Phase 1 that approximately seventy-five percent of participants used an iOS platform. Fortunately, we were able to recruit locally and have participants use a lab-issued Android phone, but we were then asking non-native Android users to complete usability testing. This may have contributed to some of the usability issues reported in findings. Recruitment issues arose from concerns with the COVID pandemic. Interviews were completed in outdoor or open space environments to protect participant safety. We were fortunate enough to be able to complete interviews via Zoom as well as in person. While the 321Connect is designed to address health care management for children ages birth to 21, our sample lacked age representation, with participants caregivers of children ages three to nine. It would be beneficial in future usability testing to include caregivers of infants and adolescents.

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CHAPTER 5: DISCUSSION

This dissertation was designed to identify the care coordination needs of caregivers of children with Down syndrome (DS). These findings were then used to develop a mHealth app to create a personal health record (PHR) to meet these care coordination needs. Chapter 2 reported findings from a scoping review of the published literature investigating the care coordination needs and strategies employed by family caregivers to manage the health care needs for their child with DS. An analysis of the purposes and results section of each article identified three themes related to care coordination, these are: communication, information, and utilization. Chapter 3 presented findings from a mixed methods analysis of the care coordination needs and recommendations for design of a mHealth app from the perspective of caregivers and health care providers. The results of these two chapters, were used to inform the design of a mHealth app prototype, 321Connect. Chapter 4 described the development of 321Connect and initial usability testing as part of user centered design. This is the first known study to design a mHealth app to meet the care coordination needs through a PHR for caregivers of children with DS.

Communication

The scoping review reported in Chapter 2 revealed that communication between caregivers and healthcare providers was a key element of care coordination. Effective caregiver-provider communication has been linked to reduced caregiver stress, improved caregiver quality of life and ability to manage the challenges associated with DS (Diffin et al., 2019; Hall et al., 2012; Melvin et al., 2018; Murphy et al., 2017). These findings echo the recommendations for

family centered care, known to improve health outcomes for children with special health care needs, by encouraging open communication and shared decision making (Leonard et al., 2016; Hall et al., 2018; Hubner et al., 2016; Melvin et al., 2014; Schor, 2019; Turchi et al., 2009).

However, investigators have reported that we have a limited understanding of caregivers' communication needs related to care coordination, with those needs often going unmet (Cartwright & Boath, 2018; Huiracocha et al., 2017). Caregivers of children with DS have described personal communication with providers as scattered or inconsistent, with a similar description of communication between health care providers involved in care (Marshall et al., 2014; Phelps et al., 2012). As reported in Chapter 3, both quantitative survey data and qualitative interview data indicated that caregivers believed communication with providers related to care coordination was inadequate.

mHealth has been shown to improve communication with both health care providers and other persons involved in care (Floch et al., 2018; Opipari-Arrigan et al., 2020). 321Connect includes features with the potential to improve communication. Consistent with findings from this study and from other studies addressing communication and mHealth, design features should include treatment/symptom tracking, note taking and the ability to summarize health information to share with health care providers (Aston et al., 2021; Raval et al., 2017). Articles from the scoping review found mHealth has the potential to serve as a communication platform through messaging with health care providers, sharing information between familial caregivers and connecting with other families (Barros da Silva et al., 2018; Choi & Van Riper, 2020; Marshall et al., 2018; Sheehan & Guerin, 2017). In addition to mirroring the above findings, caregivers completing usability testing described a desire to use 321Connect to share information with school personnel and other non-family caregivers. Additionally, usability testing participants

reported 321Connect has the potential to aid in communication by coordination of scheduling appointments with pop-up reminders. Findings related to the importance of effective communication between caregivers and providers were consistent across the three chapters comprising this dissertation and provide evidence of potential of 321Connect and support effective communication using mHealth.

Information

Findings reported in Chapter 2 and 3 highlight the importance of health information management and access to information resources. Caregivers described feeling responsible for the management health information and reported that health information was fragmented across healthcare providers and specialties, making timely access to health information challenging (Quigley et al., 2014). According to the findings in Chapter 3, caregivers developed strategies for managing information, including utilizing binders and folders to organize paper copies of health care records for tracking health information across providers. During interviews for this study caregivers reported feeling overwhelmed by the volume and types of information they need to track to meet their child's healthcare needs, such as provider contact information, medications, laboratory test results, and plans of care related to developmental therapies. Other investigators have noted that information tracking is a desirable feature of a mHealth app (Maher et al., 2016; Schnall et al., 2015). To address this need, we included spaces within 321Connect for the multiple types of information caregivers needed to track. Results from the caregiver surveys and interviews as well as usability testing and previous research, provided evidence that caregivers wanted flexible information tracking options, including limited use of forced data entry drop-down menus and free text entry boxes (Zhou et al., 2019).

In addition to features related to information management, the scoping review (Chapter 2) and survey and interview data (Chapter 3) highlighted caregivers' desire to access reputable health information resources (Gibson & Martin, 2018; Marshall et al., 2012). To meet this need, we included links to three national, well recognized DS associations that provide information written for caregivers of all literacy levels under the resources tab of 321Connect. We limited links to national resources because caregivers reported concerns that information received from health care offices was sometimes assumed to be out of date (Barros da Silva et al., 2018; Cartwright & Boath, 2018; Melvin et al., 2019; Sheehan & Guerin, 2017). By linking to existing resources we ensured caregivers can search for pertinent, up-to-date information related to their child's specific needs. Including links to existing resources also allowed caregivers to access information relevant to specific age groups and health care needs as described in findings from scoping review (Chapter 2) and qualitative findings (Chapter 3) (Leonard et al., 2016; Sheehan & Guerin, 2017). Caregivers participating in the usability testing of Chapter 4, expressed a desire to add and maintain their own information resources within 321Connect. Building on these findings we increase the ability of 321Connect to meet caregivers information needs.

Utilization

For a mHealth tool to be adopted by caregivers, it needs to be perceived as a usable tool to meet care coordination and information management needs. The intent is for utilization of 321Connect to reduce time dedicated to care coordination, of which caregivers of children with DS spend a significant amount of time (Phelps et al., 2012; Tozzi et al., 2015). Tozzi et al. (2015) found caregivers of children with DS expressed an openness to use mHealth for care management. This is echoed in the findings from this current study, both the mixed methods aspect of the study (Chapter 3) as well as findings from the usability testing (Chapter 4).

Caregivers reported that 321Connect had potential to support caregiver development and management of a PHR, with two caregivers expressing the desire to immediately incorporate 321Connect into healthcare management.

One component critical to the utilization and potential of 321Connect to improve health outcomes is to embed the AAP guidelines into the app. Adding the AAP guidelines to 321Connect, would allow caregivers to be active participants in care management and prompt providers in the use of these health maintenance guidelines (Bull et al., 2011). Embedding the AAP guidelines into EHR, demonstrated improved compliance by providers, of which a similar benefit may be achieved by caregivers by use of a PHR (Santoro et al., 2018). Caregivers also recommended AAP guidelines as well as other features of the app to be tailorable to their child's individual needs. Being able to individualize care planning has been found to increase utilization of apps (Konig et al., 2021).

mHealth apps have demonstrated the potential to improve health information and management, however findings are mixed about sustained utilization of the mHealth apps, with use often fading over time (Agarwal et al., 2019; Hamine et al., 2015; Morita et al., 2019; Whitehead and & Seaton, 2016). Steps need to be taken in designing mHealth to meet caregiver needs and preferences thereby increase the likelihood of sustained caregiver use. For example, previous research into mHealth app use has shown the app design needs to be simple and intuitive (Bendixen et al., 2017). Participants in usability testing in this current study reported similar findings. While caregivers found 321Connect a highly usable tool, they also made suggestions to simplify the design. Suggestions included replacing circles with recognizable icons and adding additional commonly used features to the home screen. Because of the large amount of initial data entry, caregivers completing usability testing recommended simplifying

sign up for 321Connect. One study recommended using a web-based data entry support tool, which could be considered in the future design elements of 321Connect (Marchak et al., 2020).

Data safety also is an important consideration for utilization of 321Connect. Privacy of health information was a concern raised by both caregivers and health care providers in the mixed methods study, with one caregiver suggesting use of an outside data safety consultant during usability testing (Chapter 3 and 4). However, similar to findings by Byambasuren et al., (2020), other participants were less concerned about privacy issues because they would be personally controlling data input as opposed to health data entered by a provider into an EHR. By addressing these care coordination needs in the design of 321Connect, we have taken the first steps in creating a highly usable tool to maintain a PHR for caregivers of children with DS.

Ongoing Analysis

For the future development and pilot testing of the 321Connect app, a regression analysis is planned to examine demographic factors and care coordination needs as predictors of the family management using three subscales of the Family Management Measure (FaMM): condition management ability, management effort, and condition impact. Data for the regression analysis were collected as part of the mixed methods analysis reported in Chapter 3. Respondents completed a demographic survey, the National Survey of Children's Health-Section D addressing care coordination needs, and the three FaMM subscales listed above. These findings, reported in a future manuscript, can then be used to determine if there are caregivers who may benefit from certain aspects of care coordination included in the 321Connect app.

Future Implications for Research and Ongoing Development of 321Connect

In addition to the usability findings reported in Chapter 4, there is a need for continued usability and pilot testing for 321Connect. Usability testing for the iOS version of 321Connect is planned for early 2022. It is also important to complete pilot testing to evaluate use over time since it is known that long term use of mHealth apps is mixed with studies showing use lessening over time (Hamine et al., 2018; Steele Gray et al., 2016). Pilot testing will help assess engagement with the 321Connect as well as to continue to explore components of usability. In addition to pilot testing, mHealth apps allow for translation in other languages. By translating into other languages, 321Connect has the potential to fill an information gap caused by language barriers (Goldsmith et al., 2017). This should be an aim of future usability and pilot testing.

With this research and the importance of keeping a PHR for families of children with DS, there is the potential to extrapolate these findings to other chronic conditions requiring health care management for multiple health care specialties. mHealth apps have proven their utility in improving health outcomes for conditions such as asthma and diabetes, but there are future implications to explore in the use of mHealth as a PHR (Garabedian et al., 2015; Geryk et al., 2016; Quinn et al., 2018). Because of the immense uses and portability of mobile devices, we will continue to see a push for technology developed for these platforms (Bendixen et al., 2017; Klasnja & Pratt, 2012). EHR's now often include portal access for mobile devices but caregivers are still required to document data for care management that is not included in an EHR. Also, while there is a push to develop mHealth technologies, it is critical to continue to develop mHealth guided by the principles of user centered design (McCurdie et al., 2012). This will help to ensure that mHealth apps may best meet caregiver needs. 321Connect is the first tool of its kind to use mHealth to create a PHR to help improve care coordination for caregivers of children

with DS. There is continued testing to be completed but initial usability testing points to a highly usable tool for the maintenance of a PHR.

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APPENDIX 2.1

Overview of Scoping Review Article Data Extraction

Article First Author, Date, Country of Origin	Purpose (direct quote)	Design	Caregiver*	Age range of children	Measures/Analysis	Use of technology
Amitha (2015) India	Hence the study was done to determine the parental views, attitudes and perceptions of oral health care and treatment requirements among children with disabilities attending various special schools in Mangalore.	Cross sectional survey design	50 families	Does not specify	Researcher generated survey	
Barros da Silva (2018) Portugal	The aim of this study was to understand the breastfeeding experiences of mothers of children with Down syndrome, and their specific practices.	Qualitative interviews	10 mothers	2 months to 10 years old	Interview guide	Several mothers reported using the internet to search for information regarding DS (esp. if health care providers were negative) Also reported using internet forums and Facebook groups for support
Bertoli (2011) Italy	The aim of the present study is to explore the needs and challenges in health, social integration, and	Mixed Methods Cross-sectional,	501 families 286 mothers 155 fathers	0 to 64 years old 181 were under the age of 19,	Researcher generated questionnaire- National Institute of Statistics (ISTAT)	

	daily life, of people with DS living in Rome.	census-based survey	15 other caregivers	with 25 did not disclose age	household survey used as reference	
Cartwright (2018)	The aim of this study was to explore the experiences of mothers of IDS regarding feeding, and to provide information to better inform health professionals caring for new mothers and their babies.	Qualitative focus groups	8 mothers	< 5 years old	Focus groups	Report using the internet to find information and expressed frustration that there was little support in providing information from health professionals
United Kingdom					Interpretative Phenomenology	
Choi (2019)	The aim of this study was twofold: 1) to explore the feasibility of using the mHealth Family Adaptation Intervention (FamilyAdapt-DS) with families of young children with DS and 2) to address the effect of participating in this mHealth intervention on family adaptation.	One group pre-test, post-test design-survey data	8 families 16 parents	Birth to 3 years old	FIRA-G, Family Management Measure (FaMM), and Family Problem Solving Communication (FPSC),	mHealth app entitled FamilyAdapt-DS combined information on mHealth apps with the use of therapeutic conversations with both parents and nurse
South Korea					focus groups	
Choi (2015)	This study examined the factors related to resilience of families of children with DS in Korea.	Cross sectional survey design	126 families 117 mothers 9 fathers	Average age 5.4 years old +/- 3.88	Questionnaire on resource and stress (QRS)	Beck depression inventory (BDI) Emotionality, Activity, Sociability survey (EAS) Family Adaptability and Cohesion
South Korea						

					Evaluation Scale (III), Family Problem Solving and Communication Scale (FPSC) Family APGAR Stigma and Discrimination
Crossman (2018)	The following 3 aims were designed to address gaps in the existing literature: 1.To determine the trajectory of parental competence for fathers of children with DD from age 3 to age 15. 2.Controlling for child and family characteristics, determine the main effects of the family environment, EI service and informal support on paternal competence when their child with a developmental disability was 3. 3.To determine whether there were lasting effects of the family environment, EI services and informal support on differences in paternal competence.	Quantitative, longitudinal analysis of secondary data	93 fathers	3 - 15 years old	Parenting Stress Index- sense of competence subscale Family Environment Scale Family Support Scale
United States					
Descamps (2015)	We were interested in the responses to 3 major questions: 1) what do parents know about dental care for their child with	Cross sectional survey design	100 families 86 mothers 14 fathers	2-21 years old	Adapted Oral Assessment DS questionnaire

Belgium	DS? 2) Have they ever been to a dentist? 3) How would they describe their experience of this dental visit?				Researcher generated survey	
Farkas (2018)	The purpose of the present study was to extend the current literature on parents' experiences by offering a balanced view of both the positive and negative sides of parenting a child with DS, specifically through analysis of parent perspectives.	Qualitative Interviews	435 families	1 to 55 years old mean 9.43 years old	Grounded theory using thematic coding	
United States	The following questions were addressed: How do participants describe their information practices and information seeking experiences?	Qualitative Interview	361 mothers 74 fathers			
Gibson (2019)	What contextual factors contribute to defensive information behaviors and knowledge practices described by participants?		24 mothers	2-26 years old	Grounded theory combining thematic analysis and constant comparative method	Trust was a major component to information seeking from online sources and the trust in maintenance of confidentiality among group participants
United States						
Hall (2018)	The objective of this study are, for children with DS, OME, and hearing loss, to: 1.) Scope the range of current service provision across England; 2.) Explore professional decision making; and 3.) Explore patient, parent	Mixed methods design Survey Interview Focus Groups	13 families 12 mothers 1 grandmother	Not specified	Interview guide Researcher developed survey	
United Kingdom						

	and public views on the direction of future research.				
Hall (2012)	The purpose of this study is to follow up on findings from the focus group interviews to explore parental stress in families of children with disabilities in the context of the Resiliency Model of Stress and Adjustment.	Mixed Methods	25 families 23 mothers 3 fathers 1 grandmother	Mean age of child 9.96 (stressed family group) and 5.58 (non-stressed group)	Parenting Stress Index Short Form Content analysis for qualitative findings
United States		Survey Focus Groups			Family Stressors Index Family Management Measure- condition management effort and family life difficulty scale Perceived Social Support Scale General Family Functioning- Family Assessment device
Hsiao (2014)	The purpose of this study was to examine how family demographics, family demands, and social support relate to family functioning as well as the potential mediating effect of social support on the relationship between family demands and family functioning in Taiwanese families of children with DS.	Cross Sectional Survey Design	83 families 80 mothers 75 fathers	4 to 17 years old	
Taiwan					
Hubner (2016)	This study uses a population based national sample to assess differences in both presence and degree of SDM in ASD and 2 other complex neurodevelopmental disorders- CP and Down syndrome- with	Secondary analysis	353 families	2-17 years old	2009-2010 National Survey of Children with Special Health care needs
United States					

	adjustment for factors (child, family and health system) shown to impact SDM.				
Huiracocha (2017)	This article focuses specifically on (i) how families received a diagnosis of DS and (ii) how the families subsequently adapted and coped.	Qualitative focus groups interviews	4 families 4 mothers 1 father 1 dyad	< 2 to 16 years old	Descriptive coding
Ecuador					
Krueger (2019)	The purpose of this study was to gain information regarding parents' advocacy behaviors and strategies they used for their child with DS.	Qualitative Survey design	435 families 371 mothers 64 fathers	< 1 to 55 years old	Triangulation
United States					
Leonard (2016)	The aim of this study was to further understand the impact of these factors, as well as to describe the current state of parental and student engagement in transition planning in Australia	Mixed methods- Qualitative and Quantitative survey data	190 families	15-30 years old	Researcher generated survey Open ended questions as part of DS "needs opinions wishes" study
Australia					
Lollar (2012)	To describe the functional difficulties of children with special health needs and to demonstrate the shared and unique contributions in predicting health outcomes and informing therapeutic	Secondary data analysis	395 families	Birth to 17 years old	Content analysis 2005-2006 National Survey of Children with Special Health care needs Multiple regression modeling
United States					

	interventions, policies, and research by using data from the 2005-2006- National Survey of Children with Special Health Care Needs					
Marshall (2019)	The purpose of our study was to examine the experiences of parents or caregivers of children with Down syndrome related to prenatal care, the birth setting, primary and specialty care and care coordination	Mixed Methods	101 families 93 mothers 6 fathers 2 other caregiver	0 to 18 years old	Family Experience Survey	
United States		Cross-sectional survey design				
Marshall (2014)	Our purpose was to better understand services and supports most needed and accessed by families of children birth to age 3 who have DS, identify gaps and barriers to accessing these services.	Qualitative-interviews focus groups	13 families 10 mothers 3 fathers	0-3 years old	Interviews Focus group A priori coded transcripts	
United States						
Melvin (2018)	This studied aimed to (1) explore families' experiences of communication information provision and (2) identify families' preferences regarding when, what and how they would like to receive information about communication.	Qualitative-interviews	9 families 5 mothers 3 fathers 1 grandmother	0 to 15 years old	Researcher generated interview guide Thematic analysis	Parents felt that electronic resources would have saved them having to sort piles of paper which were easily lost Wanted access to a website to access information and keep track of key messages.
Australia						

						Also wanted interactive workshops- with written and video information.
Mengoni (2019)	The aim of this study was to investigate whether children with Down syndrome had received recommended health checks at birth and during early childhood and whether known health conditions were monitored.	Cross sectional survey design	24 families	0-5 years old	Researcher generated survey	
United Kingdom						
Murphy (2017)	This current study explored parent observations of QOL important for a sample of children with Down syndrome including children who cannot speak for themselves.	qualitative interviews-	17 mothers	6-18 years old	Grounded theory approach	
Australia						
Nqco (2019)	The aim of this study was therefore to assess the OHRQoL outcomes because of dental caries rate among CSNs.	Cross-sectional survey design and physical exam	62 mother/child dyads	Average age 8.72 years of age	Parent-Caregiver Perception Questionnaire (P-CPQ)	
South Africa						
Nugent (2018)	The goals of this study were to compare the prevalence of successful healthcare transition planning in adolescents with Down syndrome and adolescents with OSHCN and to examine the effect of different social and	Cross-sectional survey design	151 families	12-17 years old	2009-2010 National Survey of Children with Special Health Care Needs	logistical regression
United States						

	demographic factors on transition preparation.				
Nunes (2011)					
Brazil	The objective of this study was to develop a Theoretical Model representative of the experience of families of children with DS.	Qualitative interviews	8 families	5-10 years old	grounded theory and symbolic interactionism
Oliveira (2010)					
Brazil	The aim of this qualitative study was to investigate the perceptions of a group of mothers of children and adolescents with Down syndrome (DS concerning the overall health and oral health of their children.	Qualitative interviews	19 mothers	Did not specify	content thematic analysis
Parrot (2012)					
United States	RQ1: Does diagnostic status within families relate to illness uncertainty, uncertainty management, or communication preference in families affected by NF, or Marfan or DS? RQ2: Do genetic relativism and diagnostic status interact to predict illness uncertainty, negative feelings about a condition, uncertainty management, or communication preferences in families affected by NF, or Marfan or DS?	Cross sectional survey design	106 families	Did not specify	Researcher generated survey
Phelps (2012)	Specifically, we sought to determine how children with DS	Secondary data analysis	395 families	0 to 17 years old	2005-2006 National Survey of Children

United States	<p>compare with other CYSHCN with respect to national performance measures for CYSHCN measures in the NS-CSHCN, including:</p> <ol style="list-style-type: none"> 1.Receipt of health care services with a medical home 2.parental perception of the quality of communication with the physician. 3.Communication of their child’s physician with other physicians and with educators and other professionals. 4.Receipt of needed care coordination 5. Rates of unmet health care needs. <p>Finally, we also sought to determine the relative burden on families of children with DS and ID when compared to other CYSHCN.</p>	with Special Health care Needs.
Pikora (2014)	<p>The aims of this paper were: to examine the prevalence of medical conditions and health service use among adolescents and young adults with Down syndrome; to describe the impact of these conditions on the young person’s daily life; and to explore the relationship between</p>	Index of Social Competence (ISC)
Australia	<p>Cross-sectional 197 families 15-30 years old survey design-</p>	Researcher Generated Survey

the presence of medical conditions and level of functioning daily life.

Rahim (2014)	The main objective of this study was to assess the legal representatives' perceptions on dental care access for DS and non-DS siblings in Malaysia.	Cross-sectional survey design	130 families	Mean age 26.4	Oral Assessment in Down Syndrome Questionnaire
Santoro (2016)	The purpose of this article was to describe current screening practices in patients with mDS, comparing these practices with current AAP screening practices with perceptions of physician adherence to the recommended AAP health supervision guidelines for DS.	Cross sectional survey design	91 families	0-12 years old	Researcher generated survey
Schieve (2011)	In the current study, we conducted an in-depth population-based assessment of the health and functional status of children with DS, using data from the 2005-2006 National Survey of Children with Special Health Care Needs (NS-CSHCN)	Secondary Data Analysis	338 families	3-18 years old	2005-2006 National Survey of children with special health care needs (NS-CSHCN)
Sheehan (2018)	This study aimed to explore the range of emotions experienced by parents in the early years of managing their child's disability, to assess the relevance of the Dual Process Model in	Qualitative interviews	6 families 5 mothers 1 father	5-7 years old	thematic analysis

	understanding these emotions within an adjustment process and to identify the types of support and information perceived by parents as helpful during this time.					
Skotko (2012) United States	What new diagnoses, if any, were identified in our patients with DS as a result of their visit to our clinic? How many of our new patients were up-to-date on the recommended DS healthcare screenings? And, ultimately, what value does a DS specialty clinic bring to people with DS and their families?	secondary data analysis	105 families	>3 years old	Chart Review	
Thomas (2011) Australia	To compare the prevalence of parent reported medical conditions and rates of health service utilization in school-age children with Down syndrome in Western Australia in 1997 and 2004.	Longitudinal data	272 families	5-17 years old	Intellectual Disability Exploring Answers database (IDEA) (formerly referred to as Disability Services Commission)-	
Tozzi (2015) Italy	We therefore performed a study in a population of families of patients with Down's syndrome, William's syndrome and 22q11 deletion syndrome to measure their attitude and expectations regarding specific m-health solutions	Cross sectional survey design	121 families	Average 17 years old	Researcher Generated Survey	Interested in mobile technology for time management and increased involvement in disease management Consultation with physicians

Connected to internet
both at home and at
work

van den Driessen Our aim is therefore to provide
Mareeuw (2019) insight into the perspectives of
the Netherlands PDS, parents and support staff
regarding quality of health care
for PDS in the Netherlands.

Qualitative
Interviews

15 families > 12 years old

Interview guides
Framework analysis

*Only included caregivers of children with Down syndrome (DS)

APPENDIX 3.1

Caregiver Interview Guide

I would like to thank you again for participating in our study and consenting to this interview. You already have provided us with a great deal of useful information about (child's name) and what your family does to manage care coordination. In this interview, we are especially interested in learning your perceptions of health management related to (child's name); how you managed your child's health information; what helped or hindered your management; and what role mobile technology could play in your child's health management.

The questions I'm about to ask are not meant to upset you in any way. We are genuinely interested in understanding your experiences and perspectives. But if at any time you would like to stop, please don't hesitate to let me know. Or, if you think of something else you would like me to know that I haven't asked please feel free to tell me.

To start, tell me a little bit about your child.

Are there health concerns that are managed for your child?

How do you manage visits to different providers or services for your child?

I'm particularly interested in care coordination. This is the linking together of all the providers (both primary and specialty clinics) and services to manage health care needs. What are the visits and services that you have for your child? Does your primary care practice have any tools to help you with coordinating the needs of your child?

How does your provider manage and communicate health information to both you and other health care providers who care for you child?

What has been the "fall out" for you or your child when there is inadequate health information or care coordination?

Do you use any guidelines for care management for a child with DS? Did your provider provide you the guidelines for managing your child with DS? (American Academy of Pediatrics guidelines)

Where do you look for information when you have health questions about your child?

What do you currently do to organize and manage health information for your child with DS?

What community resources do you use to manage the health care needs for your child with DS?

Some families find support groups helpful, others do not. Do you participate in any support groups? If so, what do find helpful about support groups?

How do you think you might use a mobile health application to manage health information?

What features would you like included in the app?

What other health care apps have you used?

What do you find helpful or not helpful in other health care apps you have used?

Is there anything you would like to add?

Are there any questions that you have for me?

Thank you so much for your time and participation.

Healthcare Provider and Staff Interview Guide

I would like to thank you again for participating in our study and consenting to this interview. The purpose of this study is to examine parents' and care providers' perspectives on desirable characteristics and features of mHealth application content and application use outcomes that will be used to develop an algorithm for the design of an mHealth application. In this interview, we are especially interested in learning your perceptions of health management for a child with Down syndrome; how you managed health information in your office; what has helped or hindered management; and what role mobile technology could play in health management.

Rural, suburban, or urban practice? Size of practice? Number of providers? Support staff within the practice- maybe some like a case manager?

To start, tell me about your experience working with children with DS. If you do not have experience specifically with DS, are there other patients that come to mind that require care coordination. (Maybe a good or bad example)

Have you had any experiences linked to lapses in care, missed care coordination, or missed communication?

What do you currently do to organize and manage health information for children with DS in your office? For example, do you have a care manager.

Do you use any technologies to help manage health information?

What have you seen families do to manage care coordination and/or health information for their child with DS?

How do you communicate health information to families and other health care providers who may care for a child with DS?

Where do you look for resources when you have health questions about management of a child with DS?

To what extent do you think families use health care apps in your practice?

- What apps or websites, if any, do you recommend to families?
- How do you think parents could use or benefit from an mHealth to support care coordination?

What do you think would be beneficial features of a mHealth app for families of children with DS? If needed- For example- tracking milestones? Food? Etc?

Is there anything you would like to add?

Are there any questions that you have for me?

Thank you so much for your time and participation.

APPENDIX 4.1

Usability Testing

This exercise is not to test your knowledge of caregiving for Down's Syndrome, but the utility of the app itself. You do not need to be a parent or caregiver to answer these questions. Please answer the following questions using the 321Connect app that has been loaded on your phone.

Question 1 – Getting started with Profiles

To begin using the application, add the three profiles with the information given below. To access the profiles area, click on the three white, horizontal lines at the top left of the screen and choose "Profiles" from the drop-down list. The Child Profile area will appear to fill in this information first.

1) Add a child to the profile

Use the following information to add a child:

Alexander Grant, Male
July 25, 2012, birthdate
August 31, 2012, due date
AB blood type
No allergies or medications

2) Add a caregiver to the profile

Use your personal information to add to the caregiver profile as the parent of Alexander.

3) Add a doctor/provider to the profile

Use the following information to add a provider:

Arabella Euston, Richmond Pediatric Doctors
PT is her specialty
555-123-4444 is her phone; 555-123-4445 is her fax
Her email is aeuston@richpeddoc.com, with her practice website at www.richpeddoc.com
The address of her practice is 123 Main Street; Suite 2; Richmond, VA 23220

Question 2 – Logging the child's daily activities

Now that the profiles have been completed, from the main page, you can start logging activities that the child has completed, similar to the way a caregiver would use the app.

1) Log a sleep episode

Alexander took a nap from 1pm to 4pm today. He was restless but had no fever. Log this in the Sleep area of the app.

2) Log a bathroom episode

Alexander went pee and poop on the potty that was not an accident and lasted 10 minutes. Log this in the Bathroom area of the app.

3) Add a fluid feed

Alexander drank 4oz of milk from a bottle with no vitamins or supplements at noon.

Question 3 – Logging the child’s medical activities

Now from the main page, you can start logging medical activities that the child has completed, similar to the way a caregiver would use the app.

1) Add a doctor visit

Alexander visited Arabella Euston, the PT, yesterday. He was measured at 4 foot 5 and 80 pounds. His head circumference is 24 inches. His temperature is 98.7 F. Log this in the medical area of the app.

Question 4: Semi-structured Interview

What did you like about the app? What would you change about the app? Were there things that you found challenging when working with the app? Anything you would like to add?

Question Set 5 – System Usability Survey: Assessing the usability of the 321Connect application

Strong Disagree

Strongly Agree

1. I think that I would like to use this system frequently

1	2	3	4	5

2. I found the system unnecessarily complex

1	2	3	4	5

3. I thought the system was easy to use

1	2	3	4	5

4. I think that I would need the support of a technical person to be able to use this system

1	2	3	4	5

5. I found the various functions in this system were well integrated

1	2	3	4	5

6. I thought there was too much inconsistency in this system

1	2	3	4	5

7. I would imagine that most people would learn to use this system very quickly

1	2	3	4	5

8. I found the system very cumbersome to use

1	2	3	4	5

9. I felt very confident using the system

1	2	3	4	5

10. I needed to learn a lot of things before I could get going with this system

1	2	3	4	5