Understanding Parents' Experiences and Information Needs to Inform a Digital Knowledge Translation Tool about Pediatric Functional Constipation

by

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Abstract

Background: Functional constipation is very common and has devastating effects on the physical, psychological, social, and financial well-being of children and families, while also inflating costs and healthcare resource use. Adequate clinical care for this difficult condition is lacking and treatment outcomes are often insufficient to prevent lifelong constipation. Although parents are key stakeholders in treatment, there are limited resources available and little evidence to pinpoint what information will meet parents needs and accurately reflect their experiences. Therefore, finding innovative ways to support families living with pediatric functional constipation can make a positive contribution towards improving care. Specifically, patient-direct knowledge translation has the potential to increase knowledge, improve experiences, optimize health resource use, and encourage effective health behaviours. In addition, the integration of patient engagement methods in research can enhance relevance and recognize patients as valuable contributors to knowledge development.

Purpose: The overarching purpose of this dissertation is to contribute towards improving care for children and families affected by pediatric functional constipation. The step-wise objectives of the research are to: i) identify and synthesize existing evidence about parents' experiences and information needs related to pediatric functional constipation; ii) explore in-depth parents' experience and information needs caring for a child with functional constipation; iii) collaborate with parents to create a novel knowledge translation tool that integrates best evidence with parental perspectives; iv) assess usability of the knowledge translation tool amongst parents in real-life context

Methods: This dissertation consists of a knowledge translation tool and four related papers: (i) a systematic review of research evidence about parents' experiences and information needs caring for a child with functional constipation; (ii) a qualitative study using interpretive description methodology to explore the experiences and information needs of parents related to pediatric functional constipation; (iii) a study protocol for the evaluation of the patient engagement process in child health research; and (iv) a descriptive study of the development and usability testing of the knowledge translation tool.

Findings: Results from the systematic review demonstrate that research specifically exploring parents' experiences with pediatric functional constipation is scarce and insufficient to meaningfully inform improvements in practice. The qualitative inquiry reveals that parents have significant unmet needs for information and support related to pediatric functional constipation, including validation and recognition of extreme caregiver burden for the condition, physiology of the condition, pathophysiology of soiling, safety and use of medications, and greater understanding of treatment goals and duration. Both the systematic review and qualitative inquiry findings also highlighted the need for improved resources for healthcare providers and consideration of alternative models of care delivery to better meet parents' needs. These results contributed to the development of a knowledge translation tool for parents. Usability testing of the tool with parents in a real-life context was overwhelmingly positive, citing ease of use and clarity of information as strengths.

Conclusions: This dissertation provides a comprehensive understanding of parents' experiences and information needs when caring for a child with functional constipation. In addition to developing knowledge, this research led to the creation of an enduring, patient-direct knowledge translation tool for parents. The development of practice relevant knowledge and a novel resource for parents are substantive contributions to the field of pediatric functional constipation, with implications for patient engagement and knowledge translation science. The research findings, methodological, and practical contributions of this dissertation have the potential to improve health outcomes for patients and families while also creating a foundation for future research.

Preface

This thesis is an original work by Alison Thompson. The research projects that comprise this thesis, received ethics approval from the University of Alberta Research Ethics Board. Project title – "Collaborating with parents to understand and address information needs when caring for a child with functional constipation" #Pro 00087548 approved April 17, 2019 and Project title - "Digital KT tools for parents, on common pediatric conditions in ED visits" #Pro 00062904 was approved on April 19, 2016.

Paper one has been published as Thompson, A. P., Wine, E., MacDonald, S. E., Campbell, A., & Scott, S. D. (2020). Parents' experiences and information needs while caring for a child with functional constipation: A systematic review. *Clinical Pediatrics*, *60*(3), 154–169. https://doi.org/10.1177/0009922820964457. Ms Slater and Ms Kung created and conducted the systematic search. Ms Marta Michas translated one study from Polish to English. I was responsible for the conceptualization, initial search guidance, screening, data extraction, data analysis, writing, and submission of this paper for publication. Ms Campbell assisted with screening, data extraction, and revising of the manuscript. Dr. Scott was the primary supervisor for this doctoral research and provided key guidance on the conceptual development of this review. Drs Scott, Wine, and MacDonald contributed to the conceptualization and intellectual development of this paper. All authors suggested substantive revisions of the paper and approved the final manuscript.

Paper two has been published as Thompson, A. P., MacDonald, S. E., Wine, E., & Scott, S. D. (2021). Understanding parents' experiences when caring for a child with functional constipation: Interpretive description study. *JMIR Pediatrics and Parenting*, *4*(1), e24851. https://doi.org/10.2196/24851. I was responsible for the conceptualization, participant recruitment, data collection, data analysis, writing, and submission of this paper. Dr. Scott was the primary supervisor for this doctoral research and provided key guidance on the conceptual development of this study. Drs Scott, Wine, and MacDonald contributed to the conceptualization and intellectual development of this paper. All authors suggested substantive revisions of the paper and approved the final manuscript.

Paper three has been published as Thompson, A. P., MacDonald, S. E., Wine, E., & Scott, S. D. (2020). An evaluation of parents' experiences of patient engagement in research to develop a digital knowledge translation tool: Protocol for a multi-method study. *JMIR Research Protocols*, *9*(8), e19108. https://doi.org/10.2196/19108. I was responsible for the conceptualization, writing, and submission of this paper. Dr. Scott was the primary supervisor for this doctoral research and provided key guidance on the conceptual development of this study. Drs Scott, MacDonald, and Wine contributed to the conceptualization and intellectual development of this paper. All authors suggested substantive revisions of the paper and approved the final manuscript.

Paper four is being prepared for submission as: Thompson, A. P., Hartling, L., & Scott, S. D. Development and usability of a knowledge translation tool for parents managing pediatric functional constipation. (Target journal: *Journal for Specialists in Pediatric Nursing,* April 2021). Participant recruitment was facilitated by an onsite research liaison. I was responsible for contributing to; the conceptualization of the paper, development and refinement of the tool, data analysis (assisted by Ms Le), writing, and submission of this paper. Dr. Scott was the primary supervisor for this doctoral research and provided key guidance on the conceptual development of this study. Drs Scott and Hartling contributed to the conceptualization and intellectual development of this paper. All authors suggested substantive revisions of the paper and approved the final manuscript.

The research conducted for this thesis forms part of a research collaboration co-led by Dr. Shannon Scott (supervisor) with Dr. Lisa Hartling at the University of Alberta. The methods employed in this dissertation reflect a methodological process to develop a parent knowledge translation tool established by Drs. Scott and Hartling, specifically combining the results of a knowledge synthesis (Paper 1) with the results of a qualitative study (Paper 2) to develop a knowledge translation tool for parents. The knowledge translation tool described in Paper 4 was co-designed by Drs. Scott and Hartling, their research team, and me with feedback received by expert health care professionals and members from a Pediatric Parent Advisory Group that informs Drs Scott and Hartling's research program. The infrastructure required to develop the knowledge translation tool was provided by Dr. Scott's research program and funding awarded to her and Dr. Hartling from the Canadian Institutes of Health Research and the Stollery Science Lab's Distinguished Researcher funding. I was responsible for contributing to the development and refinement of the tool, interfacing with the Pediatric Parent Advisory Group, as well as leading the interpretation of the data and final reporting. Dr. Scott was the supervisory author of this project and was extensively involved in the development and evaluation of the KT tool.

Acknowledgements

Thank you to all the parents who so graciously participated in my research. It was an honour to hear your stories and learn from your experiences.

To my doctoral supervisor Dr Shannon Scott, words cannot begin to express the profound gratitude I have for you as a supervisor, researcher, and mentor. Thank you for your tireless commitment to creating the best opportunities for your students. You motivated me when I was lagging and made me want to live up to your expectations. You are an inspiration.

To Dr Shannon MacDonald and Dr Eytan Wine, thank you for being an incredible doctoral committee. It was such a pleasure to work with you both. You gave so generously of your time, always offering constructive and thoughtful insights. Thank-you for being mentors who made me feel respected, supported, and challenged by your contributions.

Thank you David. You believed in me with such enthusiasm it filled the confidence gap I had in my own abilities.

To Mackenzie, Reid, and Jillian, thank you for your patience and cheering, especially when it came at a personal cost.

Mom, Dad, Erin, and Susan thank you for a lifetime of unwavering love and support.

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List of Abbreviations

FC = Functional Constipation

ID = Interpretive Description

KT = Knowledge Translation

KTA framework = Knowledge to Action framework

PCG = Parent Collaborator Group

PE = Patient Engagement

P-PAG = Pediatric Parent Advisory Group

PPEET = Public and Patient Engagement Evaluation Tool

PEIR framework = Patient Engagement in Research framework

Chapter 1. Situating the Research

This chapter introduces the reader to the main elements that comprise the research project including the condition of pediatric functional constipation and its clinical management, and the fields of patient-direct knowledge translation, narrative methods, and patient engagement in research. After providing an overview of these key components, details of the research phases, methods, objectives, philosophical and theoretical foundations, and the adaptions that occurred during the research are described. Each paper that forms the dissertation is briefly introduced and its' connection to the full research project is presented.

Functional Constipation

Functional constipation (FC), idiopathic constipation and inorganic constipation are often used interchangeably and refer to a constellation of gastrointestinal symptoms that occur without an underlying anatomical or physiological cause (National Collaborating Centre for Women's and Children's Health, 2010, p. 4). The condition is often marked by a cyclical pathophysiology; such that painful defecation due to large, firm stool prompts stool-withholding, which increases water absorption from stool and causes harder, larger stools, and exacerbates difficult elimination and painful defecation. Physical symptoms of FC include abdominal pain, distention and bloating, decreased appetite, decreased stool frequency, hard stools, painful or difficult defecation, fecal impaction, fecal incontinence, urinary incontinence and increased frequency of urinary tract infections (Rowan-Legg & Canadian Paediatric Society, 2011; van den Berg et al., 2006; Varni, Nurko, et al., 2015). Across all presentations of childhood constipation, FC accounts for more than 95% of cases (Loening-Baucke, 2005). Although reported prevalence rates vary widely (Koppen, Vriesman, Saps, et al., 2018; Mugie, Benninga, et al., 2011; van den Berg et al., 2006), FC is known to be very common. Results from a recent systematic review indicate the condition likely affects at least 1 in 10 children worldwide (Koppen, Vriesman, Saps, et al., 2018) with the true prevalence expected to be greater due to under-recognition and under-reporting (Rajindrajith et al., 2016; Timmerman et al., 2019). Despite perceptions that pediatric FC is merely an uncomfortable condition, studies reveal there are devastating physical, emotional, social, and school-related consequences for children and families (van den Berg et al., 2006; Vriesman et al., 2019; Youssef et al., 2005). For example, some degree of fecal soiling is estimated to occur in up to 80% of children with FC (Rajindrajith et al., 2013), which can hinder school success and peer relationships, further increasing a child's risk of being isolated, stigmatized, or bullied (Bongers et al., 2009; Joinson et al., n.d.; Kaugars et al., 2010; Klages et al., 2016; Kovacic et al., 2015). In addition, high levels of stress and conflict are common within families affected by pediatric FC (Kaugars et al., 2010; Klages et al., 2016; Kovacic et al., 2015; Varni, Bendo, et al., 2015). The cumulative significance of symptoms associated with FC is highlighted by resounding evidence that indicates children with FC experience lower quality of life than healthy peers (Kaugars et al., 2010; Klages et al., 2016; Kovacic et al., 2015; Vriesman et al., 2019; Wang et al., 2013; Youssef et al., 2005) and those with organic gastrointestinal disease (e.g. Crohn's disease) (Varni, Bendo, et al., 2015; Youssef et al., 2005).

Over and above the individual and family-level consequences of FC, healthcare systems are also affected by pediatric FC (Choung et al., 2011; Liem et al., 2009). Financial implications of the condition include immediate and directs costs such as increased emergency department visits, diagnostic testing, inpatient admissions, outpatient clinic visits, and medication use (Choung et al., 2011; Liem et al., 2009). For instance, FC accounts for an estimated 25% of pediatric gastroenterology visits (Jurgens et al., 2011). Given the increased use of healthcare resources, it is not surprising that healthcare costs for children with FC are consistently higher than matched controls from childhood through to early adulthood (Choung et al., 2011). Specifically, mean costs of inpatient and outpatient care were found to be three to four times higher than for children without constipation (Choung et al., 2011; Liem et al., 2009). Indirect costs of pediatric FC are difficult to capture accurately but include a high rate of school absenteeism which can trigger domino-like effects for families such as missed work for parents, loss of income, and obstacles to a child's academic success (Bongers et al., 2009; Choung et al., 2011; Kaugars et al., 2010; Kovacic et al., 2015; Liem et al., 2009). Lastly, long-term costs related to pediatric FC may be attributed to the low rate of treatment and resolution of symptoms. FC rarely improves without intensive interventions (detailed below) and about 40% of children with FC will develop persistent symptoms lasting for more than one year, while ¼ of affected children will become adults with chronic or lifelong FC (Bongers et al., 2010). The cumulative burden and costs associated with pediatric FC are similar to higher profile childhood conditions such as asthma and attention deficit– hyperactivity disorder (Liem et al., 2009). The myriad of physical symptoms, psychological and social effects, and economic sequelae establishes compelling evidence to support continued research and innovation towards improving the care for families affected by childhood FC.

Clinical Management

The first stage for families with a child affected by FC is to recognize a problem exists and access healthcare services. Families living with pediatric FC can be affected by systemic barriers (e.g. determinants of health, discrimination, etc) which hinder access to appropriate healthcare (Christian, 2017) and may compound the stigma of a defecation-related disorder. Although research in the field is sparse, a variety of factors may influence parents' capacity and willingness to discuss their child's symptoms with a clinician (Bernardbonnin et al., 1993;

Brennan-Parsons, 2000; Farrell et al., 2003; Timmerman et al., 2019). Beyond existing systemic barriers to accessing healthcare, it can be difficult for children and parents to identify constipation as a serious medical condition (Timmerman et al., 2019; van Tilburg et al., 2012). For instance, social constructions typically portray constipation as a simple lack of dietary fibre and/or water, fostering the myth of an uncomfortable but benign episode (Müller-Lissner et al., 2005; Staller & Cash, 2020). This type of misconception may form a backdrop that interferes with parents' capacity to recognize that a serious problem exists until the condition has continued for a prolonged time or has progressed in severity (Klages et al., 2016; Kovacic et al., 2015; Malowitz et al., 2016). In addition, parents may be encouraged by family and friends to defer medical care with hopes the symptoms reflect normal variability and will be outgrown over time. Furthermore, because many children with FC begin to show symptoms during periods of transition such as toilet training or school entry (Malowitz et al., 2016), the changes related to defecation may be hidden or easily attributable to the growth and development process of the child. Lastly, FC can be a difficult topic of conversation for parents who feel embarrassed or ashamed to talk about topics perceived to be private or confined to the bathroom (Kaugars et al., 2010). For instance, some parents are reluctant to discuss defecation and soiling, whereas other parents may feel guilty they have not been able to independently solve what they consider to be a simple problem (Farrell et al., 2003; Kaugars et al., 2010).

Unfortunately, the challenges parents face in the early stages of FC may be compounded by non-existent or negative interactions with professionals in the healthcare system. Specifically, pediatric FC is underrecognized and undertreated by clinicians (Borowitz et al., 2005; Sood et al., 2018; Timmerman et al., 2019; Yang & Punati, 2015). In a recent survey of pediatricians and pediatric gastroenterologists, more than 38% of U.S. clinicians were not familiar with an international guideline for the treatment of FC in children (Koppen, Vriesman, Tabbers, et al., 2018). Similarly, an earlier study found 84% of clinicians described themselves as ‘unfamiliar’ or only ‘slightly familiar’ with the guideline (Yang & Punati, 2015). In addition to lack of knowledge, clinicians may underestimate the severity of the condition. For example, despite evidence that up to 80% children with FC may experience fecal incontinence (Rajindrajith et al., 2013), the majority of clinicians indicated that fecal incontinence was present in only 0-10% of their patients (Yang & Punati, 2015). It is unclear from the research whether clinicians are not asking about soiling or parents and children are not disclosing this information. The mismatch between expected prevalence and what pediatricians and pediatric gastroenterologists estimated from their practice is problematic because the presence of soiling episodes typically suggests some degree of impaction, which is a primary decision-point in the treatment algorithm (Tabbers et al., 2014). Thus, failure to recognize or adequately assess for impaction is likely to exacerbate the undertreatment of pediatric FC in clinical practice. Although there are numerous factors that contribute to a child’s health outcomes, evidence indicates that up to 50% of children with FC still have symptoms after 5 years of treatment (Mugie, Di Lorenzo, et al., 2011). In addition, delayed treatment initiation has been linked to prolonged and more severe symptom presentations, meaning that expedient treatment is important to minimize chronicity and disease burden (Mousa et al., 2020; Yang & Punati, 2015). Management of pediatric FC usually includes behavioural interventions (regular toilet time, positive reinforcement etc.) and daily oral medication therapy using osmotic laxatives such as polyethylene glycol 3350, with additional agents according to individual needs (Brazzelli et al., 2011; Gordon et al., 2016; National Collaborating Centre for Women’s and Children’s Health, 2010; Rowan-Legg & Canadian Paediatric Society, 2011; Tabbers et al., 2014). Overall, despite a variety of therapeutic options (Brazzelli et al., 2011; Gordon et al., 2016; Mousa et al., 2020; van Engelenburg-van Lonkhuyzen et al., 2017), care for children and families with pediatric FC is typically not meetings families' needs nor is it aligning with best practice evidence (Borowitz et al., 2005; Farrell et al., 2003; Sood et al., 2018; Timmerman et al., 2019; Yang & Punati, 2015).

Although there are many avenues to target for improvement, clinical practice guidelines for childhood FC emphasize the importance of family education as a central component of treatment (National Collaborating Centre for Women's and Children's Health, 2010; Rowan-Legg & Canadian Paediatric Society, 2011; Tabbers et al., 2014). Parents are responsible for implementing and monitoring treatments that may be required for many months, meaning they are key stakeholders that merit the attention of clinicians and researchers. Unfortunately, there is a very little evidence exploring how families live with pediatric FC and existing research indicates parents' experiences with FC are frequently misunderstood by healthcare professionals (Farrell et al., 2003). Not surprisingly, parents have expressed the need for relevant, accessible, and reliable information sources to support their child-health decision-making (Jackson et al., 2008). Consequently, there is a significant knowledge gap that exists at the intersection of parents' experiences and information needs related to caring for a child with FC. Therefore, relevant resources to support and empower families caring for a child with FC have the potential to make a meaningful contribution to improve care.

Patient-Direct Knowledge Translation and Narrative Methods

Knowledge translation (KT) is a complex process to decrease gaps between research and practice (Straus et al., 2013), thus many KT projects are focused on healthcare providers; however, connecting parents and families to research evidence is an important component of KT with significant potential to improve outcomes (Stacey & Hill, 2013). Specifically, patient-direct KT positions patients as the intended audience and the approach has the potential to increase knowledge, improve experiences, optimize health resource use, and encourage effective health behaviours (Stacey & Hill, 2013). Research about parents' use of information resources found that material that was clear, relevant, and involved parents in the development process was most likely to be effective (Neill et al., 2015). Additionally, evidence demonstrated that patient-direct KT using creative methodologies are well-suited to pediatric practice (Hartling et al., 2013; Scott et al., 2012).

Storytelling has a long history of social use as means to share information and influence beliefs, with narrative-based approaches to patient and family education becoming increasingly common in healthcare contexts. In particular, narrative-based methods such as stories have been recognized for their ability to make information relatable in health education initiatives (Archibald et al., 2018; Cunningham & Boom, 2013; Lee et al., 2018; Njeru et al., 2015). Specifically, emotional engagement in narratives is helpful to increase personal relevance and acceptance of the information presented (Green & Brock, 2000). In addition, the widespread traditional use of stories makes narrative-based approaches familiar and acceptable for many cultures (Goding, 2013; Houston et al., 2011; Lee et al., 2016; Restrepo & Davis, 2003). Stories have been used successfully to facilitate education and support behaviour change in complex contexts such as smoking cessation, vaccine education, and diabetes management (Cunningham & Boom, 2013; Fix et al., 2012; Houston et al., 2011; Lee et al., 2016; Njeru et al., 2015). Therefore, integrating narrative-based approaches with patient-direct KT may have great potential to improve health resources and care for families in general, and for this research specifically—those affected by pediatric FC.

Patient Engagement

Patient engagement (PE) is defined as “meaningful and active collaboration in governance, priority setting, conducting research and knowledge translation” (Canadian Institutes of Health Research, 2014). Typically, *patient* is used as a broad rather than specific term, to include individuals, groups, communities, caregivers, friends, and families who have personal experience and knowledge of a health issue[34]. Although terminology varies globally, in Canadian health research, the terms *patient-oriented research* and *patient engagement* are commonly used and align with guidance from the Canadian Institutes of Health Research. Whereas research has traditionally been the exclusive domain of scientists and academics, the outputs are often intended to benefit patients and the public. Research processes and the resulting outcomes have increasingly been criticized for failing to acknowledge or integrate patients’ knowledge and experience (Snyder & Engström, 2016). The shift towards patient inclusion stems from multiple underlying motivations, including moral/ethical, socio-political, methodological, and outcomes-related (Canadian Institutes of Health Research, 2014; Domecq et al., 2014). Regardless of the original impetus, the movement is in full motion and is supported by requirements for patient or public involvement by many funders and publishers (Frank et al., 2015). Despite the momentum for PE in research, there remain many unknowns that are impeding optimized practice and outcomes in the field. In particular, comparing and contrasting best-practices for PE in research has been difficult due to the wide range of approaches that are categorized and operationalized as PE (Manafo et al., 2018; Supple et al., 2015). In addition, evaluation of the processes and outcomes of PE in research has lagged, resulting in a meagre evidence base for PE in heath research (Brett et al., 2014b; Domecq et al., 2014; Esmail et al., 2015; Lavallee et al., 2012; Staley, 2015; Staniszewska & Denegri, 2013). Furthermore, parents are a unique subgroup of the PE population that deserves attention because of their dual roles representing both themselves as caregivers and their children as patients (Amirav et al., 2017; Curran et al., 2018; Pérez Jolles et al., 2017). These knowledge gaps make it difficult to foster capacity and sustainability for patients and researchers alike. Evaluations can help identify strengths, barriers, challenges, and outcomes of PE, to inform subsequent projects with aims to maximize the value for contributors and the effectiveness of the outputs.

Existing evidence and recommendations identify key elements needed to strengthen the field of PE in research including; explicit PE methods, planned evaluations (process and outcomes), detailed reporting of PE approach, inclusion of multiple stakeholders, use of theories or frameworks, and use of validated tools (Boivin et al., 2018; Brett et al., 2014a; Esmail et al., 2015; Hamilton et al., 2017; Lavallee et al., 2012; Manafo et al., 2018; Staniszewska et al., 2011; Staniszewska & Denegri, 2013). It is important to clarify that although measuring and evaluating PE in research are both important contributions, they differ significantly. Measuring PE is aimed at quantification of methods and outcomes (Goodman et al., 2019; Soobiah et al., 2019), whereas evaluation is primarily focused on understanding processes. Together, measurement and evaluation can be used to improve PE methods and ultimately to improve outcomes. To this end, evaluating parents' experiences participating in child-health research is a meaningful step in building the science of PE.

Philosophical and Theoretical Foundations

Ontological and epistemological diversity are characteristic of the nursing discipline (Chinn & Kramer, 2015; Tarlier, 2005). This diversity allows a broad range of research questions, methods, and philosophical stances that contribute to nursing knowledge. Similarly, the numerous viewpoints, methodologies, and approaches used in research are helpful to build knowledge that is suitable for the complexities of nursing practice (Risjord, 2010; Tarlier, 2005). In my research, I have adopted a pragmatic epistemological position, supporting the integration of approaches, methods, and tools that may be helpful to improve nursing practice (G. Doane & Varcoe, 2005; McCready, 2010). From this perspective, knowledge gaps or deficits in practice offer direction for research and theoretical development (Risjord, 2010). In this research, the existing gap in care for families affected by pediatric FC provides the motivation for my studies, while the underpinnings of relational practice in nursing frame the methods chosen (G. H. Doane & Varcoe, 2007). The use of Interpretive Description (ID) methodology is appropriate for researchers wishing to explore phenomena through co-creation of meaning with participants and is explicitly aimed at generating clinically useful knowledge to improve practice (Thorne, 2016; Thorne et al., 1997). Similarly, PE in research often stems from the impetus to decrease the know-do gap by better integrating research and clinical practice, in particular the experience and wisdom of patients (Brett et al., 2014a; Canadian Institutes of Health Research, 2014; Domecq et al., 2014; Evans et al., 2014; Selby et al., 2012; Ward et al., 2010). For example, PE is hypothesized to facilitate translation, dissemination, and uptake of results while improving research applicability (Esmail et al., 2015). While both ID and PE approaches in research may be undertaken from a range of epistemological positions, establishing an intentional connection between patients and researchers reflects a key relational aspect of knowledge generation that aligns with my nursing practice, research aims, and philosophical position.

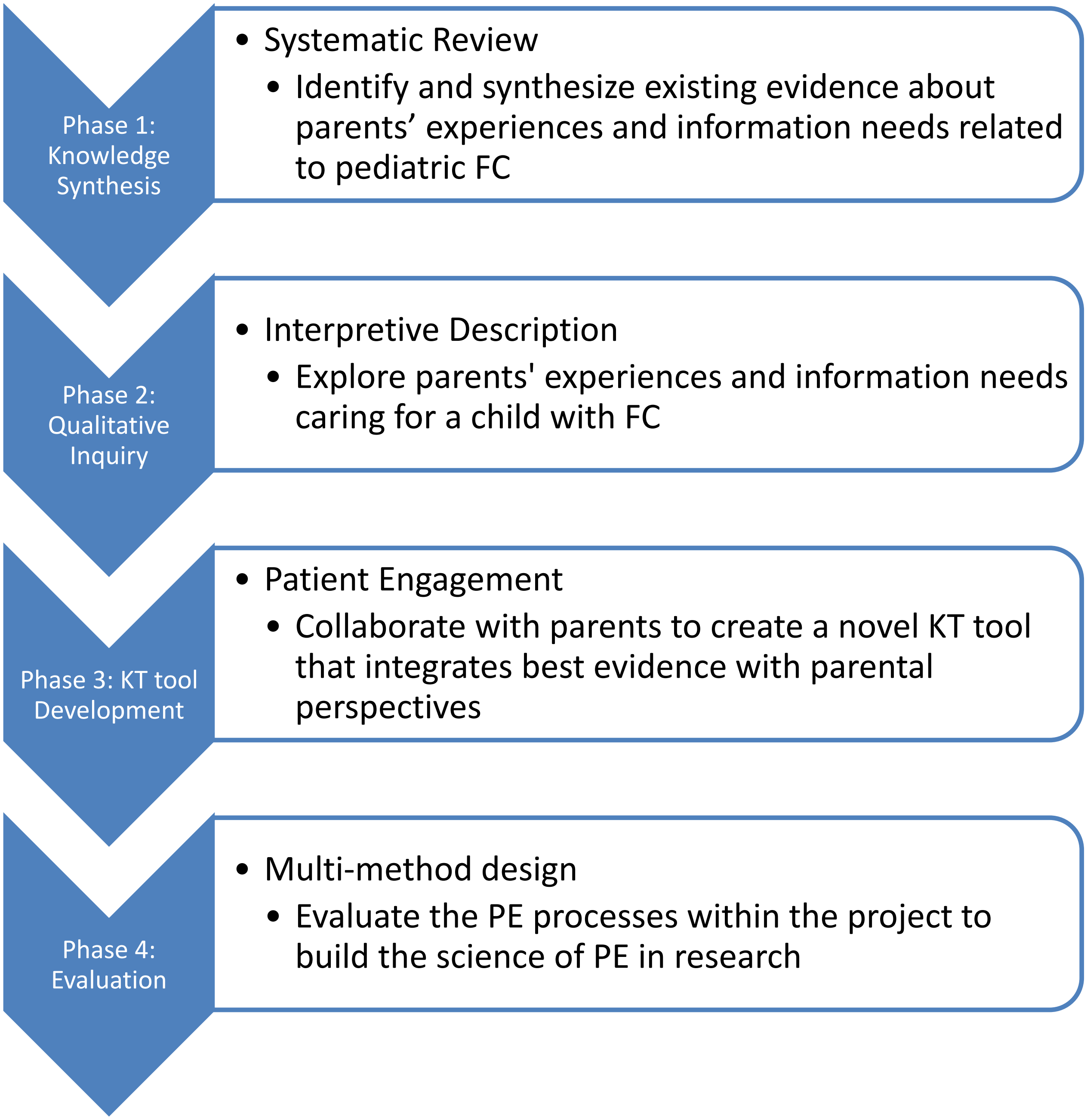
In addition to acknowledging the philosophical traditions of this research, I also used theoretical sources to provide foundational direction for design and planning. Specifically, the Knowledge to Action (KTA) framework is a seminal source in the field of KT and was used to ensure the phases of this project aligned with existing theoretical knowledge (Straus et al., 2013).

For example, the central process of the KTA framework depicts *tailoring knowledge*, which includes knowledge inquiry, synthesis, and tools (Straus et al., 2013). In addition, tailoring knowledge both stems from and feeds into the know-do gap—in this case, the care of families living with pediatric FC. Lastly, co-development of the KT tool through PE in research builds the foundation for *adapting knowledge to the local context*. That is, the collaborative mechanisms of PE of this project leverage parental perspectives to advance adaptation of research evidence for use by parent stakeholders (Banner et al., 2019).

Dissertation Phases, Methods, and Objectives

This paper-based dissertation represents the output of my doctoral research program with the overarching purpose to improve care for children and families affected by pediatric FC. Towards this goal, I established a four-phase research project with each step building upon knowledge from the one previous. The phases, methods, and objective are presented in Figures 1.1 and 1.2.

Figure 1.1 Dissertation Phases, Methods and Objectives



- Systematic Review
    - Identify and synthesize existing evidence about parents’ experiences and information needs related to pediatric FC

- Interpretive Description
    - Explore parents' experiences and information needs caring for a child with FC

- Patient Engagement
    - Collaborate with parents to create a novel KT tool that integrates best evidence with parental perspectives

- Multi-method design
    - Evaluate the PE processes within the project to build the science of PE in research

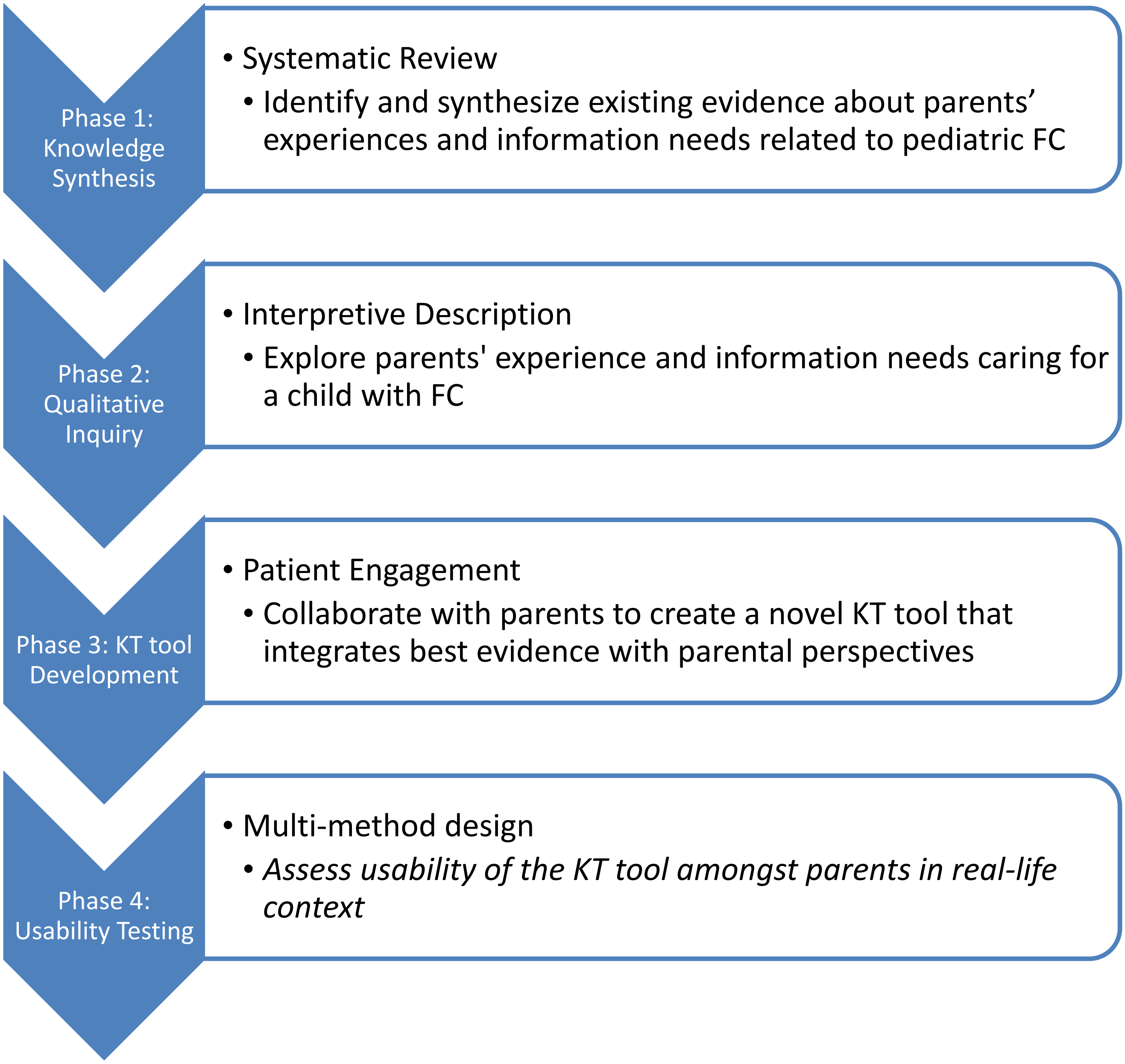
Dissertation Adaptations

In the spring of 2020, while I was analyzing qualitative data and finalizing revisions of the systematic review, our world changed dramatically with the emergence of the Covid-19 global pandemic. In Canada, schools were suddenly closed across the country and provinces enacted emergency lockdown measures. Many parents, arguably fortunate to still have a job, were nevertheless exceedingly strained by trying to learn to work from home while simultaneously caring for young children and supporting online learning. Although the deadly toll of the virus continues to be studied, the magnitude and scope of the concomitant devastation (i.e., economic, mental health, etc.) are not yet fully understood.

Consequently, I submitted an amendment to my research ethics application to change the proposed Parent Collaboration Group (PCG) meetings to an online format, in order to prevent potential transmission of the virus. In addition, the funding originally allocated for childcare and refreshments for the meetings was shifted towards providing parents with a 30$ gift card to honor their contribution. In the summer of 2020, I began my attempts to recruit parents to form the planned PCG. At the time of the interviews, all sixteen parents expressed interest in further participating in the project and consented to being contacted by email. In addition, after qualitative data collection had concluded, I received interest from an additional three parents who were keen to be contacted about forming the PCG. Therefore, nineteen parents were contacted by personalized email to explain the project and invite them to attend an online meeting. None of the email messages were returned due to inactive addresses. Three parents replied they were unable to participate due to current life stressors. Four parents replied and confirmed their attendance for the first planned meeting date, with one additional parent expressing interest but being unable to attend on the scheduled date. The meeting was to be recorded, so the parent with the schedule conflict was offered a video recording of the meeting. For the first meeting, only one parent joined the online platform. Together we decided to reschedule the meeting after soliciting input from the other interested parents about preferred dates/times. I received feedback from two parents about the best days and times to attend and rescheduled the meeting accordingly. An email reminder was sent to five parents 48 hours prior to the planned meeting date. On the rescheduled meeting date, the same one parent joined the online platform. Although it was clear that forming the PCG was challenging, the one parent asked to view the prototype KT tool (video) and provided feedback. After the second failed meeting, personalized emails were resent to the four parents who had previously expressed interest. One parent responded that she was unable to attend meetings but shared her feedback about the KT tool via email. No responses were received from the other three parents after one additional follow-up email. I thought it would be impolite to continue sending invitations via email and unfortunately did not have other contact information for the parents. Therefore, the formation of a PCG and subsequent evaluation of this PE strategy was deemed unsuccessful and was cancelled.

Fortunately, within Drs Scott and Hartling's research program there was an existing Pediatric Parent Advisory Group (P-PAG) with ongoing meetings that was interested in providing feedback about the KT tool. The P-PAG was formed in 2016 and had recently collaborated in a PE evaluation with the primary investigators who created the group; therefore, shifting my evaluation protocol to the P-PAG was not suitable. Although contributing to the field through an evaluation of PE was no longer feasible, the ability of the project to retain a commitment to PE throughout the development process of the KT tool was important. Unlike the originally proposed PCG, the parents of the P-PAG were a general audience, rather than those with condition specific experience related to pediatric FC; however, the P-PAG members ongoing commitment to PE in child health research was an asset to help refine and revise the KT tool. At this point, the focus of the final phase of my dissertation research shifted from evaluating parent engagement towards assessing usability of the KT tool, while maintaining an emphasis on patient engagement as a key element of the KT development process. Therefore, my research plan was amended as follows (Figure 1.2).

Figure 1.2 Amended Dissertation Phases, Methods and Objectives



Phase 1: Knowledge Synthesis

- Systematic Review
  - Identify and synthesize existing evidence about parents’ experiences and information needs related to pediatric FC

Phase 2: Qualitative Inquiry

- Interpretive Description
  - Explore parents' experience and information needs caring for a child with FC

Phase 3: KT tool Development

- Patient Engagement
  - Collaborate with parents to create a novel KT tool that integrates best evidence with parental perspectives

Phase 4: Usability Testing

- Multi-method design
  - *Assess usability of the KT tool amongst parents in real-life context*

Dissertation Overview

The four distinct but related papers that form this dissertation focus on the development of knowledge to improve care for patients and families affected by pediatric FC. The papers have been formatted to the specifications of the journals to which they have been published or submitted.

Chapter two details the foundational knowledge synthesis that was used to ensure a comprehensive assessment and understanding of existing literature. Chapter three describes the qualitative inquiry that generated an in-depth exploration of parents' experiences and information needs when caring for a child with FC. Chapter four is a research protocol for the evaluation of PE in research that was published with the intention to enhance transparency and rigour of the methods and outcomes. Chapter 5 describes the development process and usability testing results for the KT tool. In the following paragraphs, I offer a brief summary of each of the four papers and highlight the connections between them. Chapter 6, the conclusion, summarizes the research and presents implications of the findings for future research and practice.

Paper 1

Description: This paper addressed the gap that although family education is a key component of successful treatment, there was little research exploring what information families need and how to best support them when caring for a child with FC. The aim of this systematic review was to synthesize current evidence on the experiences and information needs of parents caring for a child with FC. Following an *a priori* protocol to enhance transparency, we systematically searched published research and completed screening against our inclusion criteria. Thirteen studies (n = 10 quantitative, n = 3 qualitative) were included. We found 2 main themes, *precarious footing and profound and pervasive effects*. Heavy caregiving burdens fueled doubts, misinformation, relationship breakdown, and treatment deviation.

Connection: Paper one formed the foundation for subsequent research phases by systematically identifying and synthesizing the current evidence on the topic of parents experiences caring for a child with FC (Thompson, Wine, et al., 2020). In light of the practice recommendations for pediatric FC, the findings from our SR underscored the disconnect between parents' needs and clinical care provision for the condition. In addition, we concluded that based on the included evidence, it is likely that both parents and healthcare providers would benefit from resources and interventions to improve care related to pediatric FC.

Paper one has been published as: Thompson, A. P., Wine, E., MacDonald, S. E., Campbell, A., & Scott, S. D. (2020). Parents' experiences and information needs while caring for a child with functional constipation: A systematic review. *Clinical Pediatrics*, *60*(3), 154–169. https://doi.org/10.1177/0009922820964457

Paper 2

Description: Exploring parents' experiences and needs related to pediatric FC may offer a critical perspective towards improving clinical care. The purpose of this study was to understand and give voice to parents' experiences and information needs when caring for a child with FC(Thompson et al., 2021). This qualitative design used ID methodology to generate findings aimed at improving clinical care. One-on-one, in-depth interviews were completed either in person or through web-based teleconferencing to explore parents' perspectives. Data collection and analysis occurred concurrently. Analysis of 16 interviews generated 4 major themes: *living in the shadows*; *not taken seriously*, with a subtheme of *persevering and* *advocating*; *missing information and misinformation*; and *self-doubt and strained relationships*. One minor theme of *affirmative influences that foster resilience and hope* was identified.

Connection: Our conclusion that parents have unmet needs for support and information related to pediatric FC underscored the need to find ways to better support parents caring for a child with FC. The results from this study detailed the specific elements that parents identified as priorities for their caregiving support and information needs, which in turn contributed to the evidence base to develop our proposed KT tool.

Paper two has been published as: Thompson, A. P., MacDonald, S. E., Wine, E., & Scott, S. D. (2021). Understanding parents' experiences when caring for a child with functional constipation: Interpretive description study. *JMIR Pediatrics and Parenting*, *4*(1), e24851. https://doi.org/10.2196/24851.

Paper 3

Description: Despite the growing use of PE methods in research, evaluation of the processes has lagged behind, which means that it is difficult to know how to ensure capacity and sustainability for patients and researchers. This paper outlined the aim to use PE methods to establish a research collaboration with parents to co-create a digital KT tool for parents caring for a child with FC (Thompson, MacDonald, et al., 2020). The research protocol detailed plans to formally evaluate the PE processes within this project using a multi-method study design. Use of the validated Public and Patient Engagement Evaluation Tool (PPEET) patient questionnaire was planned to gather data. In addition to descriptive statistics of questionnaire responses, the design included qualitative analysis of open-ended question responses. Directed content analysis would be used to assess themes of the Patient Engagement in Research (PEIR) Framework with a combination of deductive and inductive analyses.

Connection: The protocol and proposed study align with recommendations for how to strengthen the science of PE in research. The results of this proposed work were intended to provide valuable information about parents' experiences participating in child-health research and make a clear contribution to understand how effective patient collaborations in research are built, maintained, and improved.

Paper three has been published as: Thompson, A. P., MacDonald, S. E., Wine, E. & Scott, S. D. An evaluation of parents' experiences of patient engagement in research to develop a digital knowledge translation tool: protocol for a multi-method study. *JMIR Research Protocols* 9, e19108 (2020).

Paper 4

Description: Providing parents with relevant, accessible, and understandable resources to understand pediatric FC and its treatment is an important contribution to child health. This descriptive, multi-method research detailed our PE strategies used to develop the KT tool for parents caring for a child with FC. The paper also reported on the usability testing for the KT tool. The usability results align with previous research findings and indicate parents appreciated the resource for being easy to use and containing information that was easy to understand. Furthermore, the narrative-based format may have enhanced parents' perceptions of relevance of the information.

Connection: This paper outlines the cumulative outputs from this research program; specifically, the use of PE strategies to guide development of a KT tool that has potential to be a meaningful and highly usable resource for parents caring for a child with FC. The paper establishes a connection to future research directions such as rigorous evaluation of knowledge and user-experience outcomes to determine the effectiveness in clinical practice and inform further revisions of the KT tool as needed.

Paper four is being prepared for submission as: Thompson, A. P., Hartling, L., & Scott, S. D. Development and usability of a knowledge translation tool for parents managing pediatric functional constipation. (target journal: *Journal for Specialists in Pediatric Nursing,* planned submission May 2021).

References

Amirav, I., Vandall-Walker, V., Rasiah, J., & Saunders, L. (2017). Patient and researcher engagement in health research: A parent's perspective. *Pediatrics*, *140*(3), e20164127. https://doi.org/10.1542/peds.2016-4127

Archibald, M. M., Hartling, L., Ali, S., Caine, V., & Scott, S. D. (2018). Developing "My Asthma Diary": A process exemplar of a patient-driven arts-based knowledge translation tool. *BMC Pediatrics*, *18*(1), 186. https://doi.org/10.1186/s12887-018-1155-2

Banner, D., Bains, M., Carroll, S., Kandola, D. K., Rolfe, D. E., Wong, C., & Graham, I. D. (2019). Patient and public engagement in integrated knowledge translation research: Are we there yet? *Research Involvement and Engagement*, *5*(1), 8. https://doi.org/10.1186/s40900-019-0139-1

Bernardbonnin, A. C., Haley, N., Belanger, S., & Nadeau, D. (1993). Parental and patient perceptions about encopresis and its treatment. *Journal of Developmental and Behavioral Pediatrics*, *14*(6), 397–400.

Boivin, A., L'Espérance, A., Gauvin, F.-P., Dumez, V., Macaulay, A. C., Lehoux, P., & Abelson, J. (2018). Patient and public engagement in research and health system decision making: A systematic review of evaluation tools. *Health Expectations*, *21*(6), 1075–1084. https://doi.org/10.1111/hex.12804

Bongers, M. E. J., van Dijk, M., Benninga, M. A., & Grootenhuis, M. A. (2009). Health related quality of life in children with constipation-associated fecal incontinence. *The Journal of Pediatrics*, *154*(5), 749-753.e1. https://doi.org/10.1016/j.jpeds.2008.11.029

Bongers, M. E. J., van Wijk, M. P., Reitsma, J. B., & Benninga, M. A. (2010). Long-term prognosis for childhood constipation: Clinical outcomes in adulthood. *Pediatrics*, *126*(1), e156–e162. https://doi.org/10.1542/peds.2009-1009

Borowitz, S. M., Cox, D. J., Kovatchev, B., Ritterband, L. M., Sheen, J., & Sutphen, J. (2005). Treatment of childhood constipation by primary care physicians: Efficacy and predictors of outcome. *Pediatrics*, *115*(4), 873–877. https://doi.org/10.1542/peds.2004-0537

Brazzelli, M., Griffiths, P. V., Cody, J. D., & Tappin, D. (2011). Behavioural and cognitive interventions with or without other treatments for the management of faecal incontinence in children. *Cochrane Database of Systematic Reviews*, *12*. https://doi.org/10.1002/14651858.CD002240.pub4

Brennan-Parsons, S. (2000). *Mothers' experiences in living with a child with encopresis- A phenomenological study.pdf*. Memorial University of Newfoundland.

Brett, J., Staniszewska, S., Mockford, C., Herron-Marx, S., Hughes, J., Tysall, C., & Suleman, R. (2014a). Mapping the impact of patient and public involvement on health and social care research: A systematic review. *Health Expectations*, *17*(5), 637–650. https://doi.org/10.1111/j.1369-7625.2012.00795.x

Brett, J., Staniszewska, S., Mockford, C., Herron-Marx, S., Hughes, J., Tysall, C., & Suleman, R. (2014b). A systematic review of the impact of patient and public involvement on service users, researchers and communities. *The Patient - Patient-Centered Outcomes Research*, *7*(4), 387–395. https://doi.org/10.1007/s40271-014-0065-0

Canadian Institutes of Health Research. (2014). *Strategy for patient-oriented research—Patient engagement framework*. http://www.cihr-irsc.gc.ca/e/48413.html

Chinn, P. L., & Kramer, M. K. (2015). *Knowledge development in nursing: Theory and process* (9th ed.). Elsevier Mosby.

Choung, R. S., Shah, N. D., Chitkara, D., Branda, M. E., Van Tilburg, M. A., Whitehead, W. E., Katusic, S. K., Locke, G. R. I., & Talley, N. J. (2011). Direct medical costs of constipation from childhood to early adulthood: A population-based birth cohort study. *Journal of Pediatric Gastroenterology and Nutrition*, *52*(1), 47–54. https://doi.org/10.1097/MPG.0b013e3181e67058

Christian, B. J. (2017). Translational research—The intersection between sociocultural and environmental factors and the health of children and families. *Journal of Pediatric Nursing: Nursing Care of Children and Families*, *37*, 127–131. https://doi.org/10.1016/j.pedn.2017.09.013

Cunningham, R. M., & Boom, J. A. (2013). Telling stories of vaccine-preventable diseases: Why it works. *South Dakota Medicine*, 21–26.

Curran, J. A., Bishop, A., Chorney, J., MacEachern, L., & Mackay, R. (2018). Partnering with parents to advance child health research. *Healthcare Management Forum*, *31*(2), 45–50. https://doi.org/10.1177/0840470417744568

Doane, G. H., & Varcoe, C. (2007). Relational practice and nursing obligations. *Advances in Nursing Science*, *30*(3), 192–205. https://doi.org/10.1097/01.ANS.0000286619.31398.fc

Doane, G., & Varcoe, C. (2005). Toward compassionate action: Pragmatism and the inseparability of theory/practice. *Advances in Nursing Science*, *28*(1), 81–90.

Domecq, J. P., Prutsky, G., Elraiyah, T., Wang, Z., Nabhan, M., Shippee, N., Brito, J. P., Boehmer, K., Hasan, R., Firwana, B., Erwin, P., Eton, D., Sloan, J., Montori, V., Asi, N., Abu Dabrh, A. M., & Murad, M. H. (2014). Patient engagement in research: A systematic review. *BMC Health Services Research*, *14*, 89. https://doi.org/10.1186/1472-6963-14-89

Esmail, L., Moore, E., & Rein, A. (2015). Evaluating patient and stakeholder engagement in research: Moving from theory to practice. *Journal of Comparative Effectiveness Research*, *4*(2), 133–145. https://doi.org/10.2217/cer.14.79

Evans, D., Coad, J., Cottrell, K., Dalrymple, J., Davies, R., Donald, C., Laterza, V., Long, A., Longley, A., Moule, P., Pollard, K., Powell, J., Puddicombe, A., Rice, C., & Sayers, R. (2014). *Public involvement in research: Assessing impact through a realist evaluation*. http://www.ncbi.nlm.nih.gov/books/NBK260168/

Farrell, M., Holmes, G., Coldicutt, P., & Peak, M. (2003). Management of childhood constipation: Parents' experiences. *Journal of Advanced Nursing*, *44*(5), 479–489. https://doi.org/10.1046/j.0309-2402.2003.02831.x

Fix, G. M., Houston, T. K., Barker, A. M., Wexler, L., Cook, N., Volkman, J. E., & Bokhour, B. G. (2012). A novel process for integrating patient stories into patient education interventions: Incorporating lessons from theater arts. *Patient Education and Counseling*, *88*(3), 455–459. https://doi.org/10.1016/j.pec.2012.06.012

Frank, L., Forsythe, L., Ellis, L., Schrandt, S., Sheridan, S., Gerson, J., Konopka, K., & Daugherty, S. (2015). Conceptual and practical foundations of patient engagement in research at the patient-centered outcomes research institute. *Quality of Life Research*, *24*(5), 1033–1041. https://doi.org/10.1007/s11136-014-0893-3

Goding, A. (2013). *Storytelling: Reflecting on oral narratives and cultures*. Cognella.

Goodman, M. S., Ackermann, N., Bowen, D. J., & Thompson, V. (2019). Content validation of a quantitative stakeholder engagement measure. *Journal of Community Psychology*, *47*(8), 1937–1951. https://doi.org/10.1002/jcop.22239

Gordon, M., MacDonald, J. K., Parker, C. E., Akobeng, A. K., & Thomas, A. G. (2016). Osmotic and stimulant laxatives for the management of childhood constipation. *Cochrane Database of Systematic Reviews*, *8*. https://doi.org/10.1002/14651858.CD009118.pub3

Green, M. C., & Brock, T. C. (2000). The role of transportation in the persuasiveness of public narratives. *Journal of Personality and Social Psychology*, *79*(5), 701–721. https://doi.org/10.1037/0022-3514.79.5.701

Hamilton, C. B., Leese, J. C., Hoens, A. M., & Li, L. C. (2017). Framework for advancing the reporting of patient engagement in rheumatology research projects. *Current Rheumatology Reports*, *19*(7), 38. https://doi.org/10.1007/s11926-017-0666-4

Hartling, L., Scott, S. D., Johnson, D. W., Bishop, T., & Klassen, T. P. (2013). A randomized controlled trial of storytelling as a communication tool. *PLOS ONE*, *8*(10), e77800. https://doi.org/10.1371/journal.pone.0077800

Houston, T. K., Cherrington, A., Coley, H. L., Robinson, K. M., Trobaugh, J. A., Williams, J. H., Foster, P. H., Ford, D. E., Gerber, B. S., Shewchuk, R. M., & Allison, J. J. (2011). The art and science of patient storytelling-harnessing narrative communication for behavioral interventions: The ACCE project. *Journal of Health Communication*, *16*(7), 686–697. https://doi.org/10.1080/10810730.2011.551997

Jackson, C., Cheater, F. M., & Reid, I. (2008). A systematic review of decision support needs of parents making child health decisions. *Health Expectations*, *11*(3), 232–251. https://doi.org/10.1111/j.1369-7625.2008.00496.x

Joinson, C., Heron, J., Buter, U., von Gontard, A., & Avon Longitudinal Study of Parents and Children Study Team. (n.d.). Psychological differences between children with and without soiling problems. *Pediatrics*, *117*(5), 1575–1584.

Jurgens, H., Oster, C., & Fereday, J. (2011). Management of chronic functional constipation in children: A review of the literature. *Neonatal, Paediatric & Child Health Nursing*, *14*(2), 23–28.

Kaugars, A. S., Silverman, A., Kinservik, M., Heinze, S., Reinemann, L., Sander, M., Schneider, B., & Sood, M. (2010). Families' perspectives on the effect of constipation and fecal incontinence on quality of life. *Journal of Pediatric Gastroenterology and Nutrition*, *51*(6), 747–752. https://doi.org/10.1097/MPG.0b013e3181de0651

Klages, K. L., Berlin, K. S., Silverman, A. H., Mugie, S., Di Lorenzo, C., Nurko, S., Ponnambalam, A., Sanghavi, R., & Sood, M. R. (2016). Empirically derived patterns of pain, stooling, and incontinence and their relations to health-related quality of life among youth with chronic constipation. *Journal of Pediatric Psychology*, jsw068. https://doi.org/10.1093/jpepsy/jsw068

Koppen, I. J. N., Vriesman, M. H., Saps, M., Rajindrajith, S., Shi, X., van Etten-Jamaludin, F. S., Di Lorenzo, C., Benninga, M. A., & Tabbers, M. M. (2018). Prevalence of functional defecation disorders in children: A systematic review and meta-analysis. *The Journal of Pediatrics*, *198*, 121-130.e6. https://doi.org/10.1016/j.jpeds.2018.02.029

Koppen, I. J. N., Vriesman, M. H., Tabbers, M. M., Di Lorenzo, C., & Benninga, M. A. (2018). Awareness and implementation of the 2014 ESPGHAN/NASPGHAN guideline for childhood functional constipation. *Journal of Pediatric Gastroenterology and Nutrition*, *66*(5), 732–737. https://doi.org/10.1097/MPG.0000000000001786

Kovacic, K., Sood, M. R., Mugie, S., Di Lorenzo, C., Nurko, S., Heinz, N., Ponnambalam, A., Beesley, C., Sanghavi, R., & Silverman, A. H. (2015). A multicenter study on childhood constipation and fecal incontinence: Effects on quality of life. *The Journal of Pediatrics*, *166*(6), 1482-1487.e1. https://doi.org/10.1016/j.jpeds.2015.03.016

Lavallee, D. C., Williams, C. J., Tambor, E. S., & Deverka, P. A. (2012). Stakeholder engagement in comparative effectiveness research: How will we measure success? *Journal of Comparative Effectiveness Research*, *1*(5), 397–407.

Lee, H., Fawcett, J., & DeMarco, R. (2016). Storytelling/narrative theory to address health communication with minority populations. *Applied Nursing Research*, *30*, 58–60. https://doi.org/10.1016/j.apnr.2015.09.004

Lee, H., Kim, M., Cooley, M. E., Kiang, P. N., Kim, D., Tang, S., Shi, L., Thiem, L., Kan, P., Peou, S., Touch, C., Chea, P., & Allison, J. (2018). Using narrative intervention for HPV vaccine behavior change among Khmer mothers and daughters: A pilot RCT to examine feasibility, acceptability, and preliminary effectiveness. *Applied Nursing Research*, *40*, 51–60. https://doi.org/10.1016/j.apnr.2017.12.008

Liem, O., Harman, J., Benninga, M. A., Kelleher, K., Mousa, H., & Di Lorenzo, C. (2009). Health utilization and cost impact of childhood constipation in the United States. *The Journal of Pediatrics*, *154*(2), 258–262. https://doi.org/10.1016/j.jpeds.2008.07.060

Loening-Baucke, V. (2005). Prevalence, symptoms and outcome of constipation in infants and toddlers. *The Journal of Pediatrics*, *146*(3), 359–363. https://doi.org/10.1016/j.jpeds.2004.10.046

Malowitz, S., Green, M., Karpinski, A., Rosenberg, A., & Hyman, P. E. (2016). Age of onset of functional constipation. *Journal of Pediatric Gastroenterology and Nutrition*, *62*(4), 600–602. https://doi.org/10.1097/MPG.0000000000001011

Manafo, E., Petermann, L., Mason-Lai, P., & Vandall-Walker, V. (2018). Patient engagement in Canada: A scoping review of the 'how' and 'what' of patient engagement in health research. *Health Research Policy and Systems*, *16*, 5. https://doi.org/10.1186/s12961-018-0282-4

McCready, J. S. (2010). Jamesian pragmatism: A framework for working towards unified diversity in nursing knowledge development. *Nursing Philosophy*, *11*(3), 191–203. https://doi.org/10.1111/j.1466-769X.2010.00444.x

Mousa, H. M., Barsamian, J. M., Benitez, A. J., Webster, J. M., & Fiorino, K. N. (2020). Pediatric constipation: An epidemiologic and physiologic approach to guide a stepwise approach to its management. *Current Treatment Options in Pediatrics*, *6*(3), 155–169. https://doi.org/10.1007/s40746-020-00199-z

Mugie, S. M., Benninga, M. A., & Di Lorenzo, C. (2011). Epidemiology of constipation in children and adults: A systematic review. *Best Practice & Research Clinical Gastroenterology*, *25*(1), 3–18. https://doi.org/10.1016/j.bpg.2010.12.010

Mugie, S. M., Di Lorenzo, C., & Benninga, M. A. (2011). Constipation in childhood. *Nature Reviews Gastroenterology & Hepatology*, *8*(9), 502–511. https://doi.org/10.1038/nrgastro.2011.130

Müller-Lissner, S. A., Kamm, M. A., Scarpignato, C., & Wald, A. (2005). Myths and misconceptions about chronic constipation. *Official Journal of the American College of Gastroenterology | ACG*, *100*(1), 232–242.

National Collaborating Centre for Women's and Children's Health. (2010). *Constipation in children and young people: Diagnosis and management of idiopathic childhood constipation in primary and secondary care*. RCOG Press. http://www.ncbi.nlm.nih.gov/books/NBK65365/

Neill, S., Roland, D., Jones, C. H., Thompson, M., & Lakhanpaul, M. (2015). Information resources to aid parental decision-making on when to seek medical care for their acutely sick child: A narrative systematic review. *BMJ Open*, *5*(12), e008280. https://doi.org/10.1136/bmjopen-2015-008280

Njeru, J. W., Patten, C. A., Hanza, M. M. K., Brockman, T. A., Ridgeway, J. L., Weis, J. A., Clark, M. M., Goodson, M., Osman, A., Porraz-Capetillo, G., Hared, A., Myers, A., Sia, I. G., & Wieland, M. L. (2015). Stories for change: Development of a diabetes digital storytelling intervention for refugees and immigrants to Minnesota using qualitative methods. *BMC Public Health*, *15*, 1311. https://doi.org/10.1186/s12889-015-2628-y

Pérez Jolles, M., Martinez, M., Garcia, S. J., Stein, G. L., Mentor Parent Group Members, & Thomas, K. C. (2017). Involving Latina/o parents in patient-centered outcomes research: Contributions to research study design, implementation and outcomes. *Health Expectations*, *20*(5), 992–1000. https://doi.org/10.1111/hex.12540

Rajindrajith, S., Devanarayana, N. M., & Benninga, M. A. (2013). Review article: Faecal incontinence in children: epidemiology, pathophysiology, clinical evaluation and management. *Alimentary Pharmacology & Therapeutics*, *37*(1), 37–48. https://doi.org/10.1111/apt.12103

Rajindrajith, S., Devanarayana, N. M., Perera, B. J. C., & Benninga, M. A. (2016). Childhood constipation as an emerging public health problem. *World Journal of Gastroenterology*, *22*(30), 6864. https://doi.org/10.3748/wjg.v22.i30.6864

Restrepo, E., & Davis, L. (2003). Storytelling: Both art and therapeutic practice. *International Journal for Human Caring*, *7*(1), 43–48.

Risjord, M. W. (2010). *Nursing knowledge: Science, practice, and philosophy*. Wiley-Blackwell.

Rowan-Legg, A. & Canadian Paediatric Society. (2011). Managing functional constipation in children. *Paediatrics & Child Health*, *16*(10), 661–665.

Scott, S. D., Hartling, L., O'Leary, K. A., Archibald, M., & Klassen, T. P. (2012). Stories – a novel approach to transfer complex health information to parents: A qualitative study. *Arts & Health*, *4*(2), 162–173. https://doi.org/10.1080/17533015.2012.656203

Selby, J. V., Beal, A., & Frank, L. (2012). The patient-centered outcomes research institute (PCORI) national priorities for research and initial research agenda. *Jama*, *307*(15), 1583–1584.

Snyder, H., & Engström, J. (2016). The antecedents, forms and consequences of patient involvement: A narrative review of the literature. *International Journal of Nursing Studies*, *53*, 351–378. https://doi.org/10.1016/j.ijnurstu.2015.09.008

Soobiah, C., Straus, S. E., Manley, G., Marr, S., Paus Jenssen, E., Teare, S., Hamid, J., Tricco, A. C., & Moore, A. (2019). Engaging knowledge users in a systematic review on the comparative effectiveness of geriatrician-led models of care is possible: A cross-sectional survey using the Patient Engagement Evaluation Tool. *Journal of Clinical Epidemiology*, *113*, 58–63. https://doi.org/10.1016/j.jclinepi.2019.05.015

Sood, M., Lichtlen, P., & Perez, M. C. (2018). Unmet needs in pediatric functional constipation. *Clinical Pediatrics*, *57*(13), 1489–1495. https://doi.org/10.1177/0009922818774343

Stacey, D., & Hill, S. (2013). Patient-direct and patient-mediated KT interventions. In *Knowledge translation in health care* (2nd ed., pp. 197–211). Wiley-Blackwell.

Staley, K. (2015). 'Is it worth doing?' Measuring the impact of patient and public involvement in research. *Research Involvement and Engagement*, *1*, 6. https://doi.org/10.1186/s40900-015-0008-5

Staller, K., & Cash, B. D. (2020). Myths and misconceptions about constipation: A new view for the 2020s. *Official Journal of the American College of Gastroenterology | ACG*, *115*(11), 1741–1745. https://doi.org/10.14309/ajg.0000000000000947

Staniszewska, S., Brett, J., Mockford, C., & Barber, R. (2011). The GRIPP checklist: Strengthening the quality of patient and public involvement reporting in research. *International Journal of Technology Assessment in Health Care*, *27*(4), 391–399. https://doi.org/10.1017/S0266462311000481

Staniszewska, S., & Denegri, S. (2013). Patient and public involvement in research: Future challenges. *Evidence-Based Nursing*, *16*(3), 69–69. https://doi.org/10.1136/eb-2013-101406

Straus, S., Tetroe, J., & Graham, I. D. (Eds.). (2013). *Knowledge translation in health care: Moving from evidence to practice* (2 edition). BMJ Books.

Supple, D., Roberts, A., Hudson, V., Masefield, S., Fitch, N., Rahmen, M., Flood, B., de Boer, W., Powell, P., & Wagers, S. (2015). From tokenism to meaningful engagement: Best practices in patient involvement in an EU project. *Research Involvement and Engagement*, *1*, 5. https://doi.org/10.1186/s40900-015-0004-9

Tabbers, M. M., DiLorenzo, C., Berger, M. Y., Faure, C., Langendam, M. W., Nurko, S., Staiano, A., Vandenplas, Y., & Benninga, M. A. (2014). Evaluation and treatment of functional constipation in infants and children: Evidence-based recommendations from ESPGHAN and NASPGHAN. *Journal of Pediatric Gastroenterology and Nutrition*, *58*(2), 258–274.

Tarlier, D. (2005). Mediating the meaning of evidence through epistemological diversity. *Nursing Inquiry*, *12*(2), 126–134. https://doi.org/10.1111/j.1440-1800.2005.00262.x

Thompson, A. P., MacDonald, S. E., Wine, E., & Scott, S. D. (2020). An evaluation of parents' experiences of patient engagement in research to develop a digital knowledge translation tool: Protocol for a multi-method study. *JMIR Research Protocols*, *9*(8), e19108. https://doi.org/10.2196/19108

Thompson, A. P., MacDonald, S. E., Wine, E., & Scott, S. D. (2021). Understanding parents' experiences when caring for a child with functional constipation: Interpretive description study. *JMIR Pediatrics and Parenting*, *4*(1), e24851. https://doi.org/10.2196/24851

Thompson, A. P., Wine, E., MacDonald, S. E., Campbell, A., & Scott, S. D. (2020). Parents' experiences and information needs while caring for a child with functional constipation: A systematic review. *Clinical Pediatrics*, *60*(3), 154–169. https://doi.org/10.1177/0009922820964457

Thorne, S. (2016). *Interpretive description: Qualitative research for applied practice* (2nd ed.). Routledge.

Thorne, S., Reimer Kirkham, S., & MacDonald-Emes, J. (1997). Interpretive description: A noncategorical qualitative alternative for developing nursing knowledge. *Research in Nursing & Health*, *20*(2), 169–177. https://doi.org/10.1002/(SICI)1098-240X(199704)20:2<169::AID-NUR9>3.0.CO;2-I

Timmerman, M. E. W., Trzpis, M., & Broens, P. M. A. (2019). The problem of defecation disorders in children is underestimated and easily goes unrecognized: A cross-sectional study. *European Journal of Pediatrics*, *178*(1), 33–39. https://doi.org/10.1007/s00431-018-3243-6

van den Berg, M. M., Benninga, M. A., & Di Lorenzo, C. (2006). Epidemiology of childhood constipation: A systematic review. *The American Journal of Gastroenterology*, *101*(10), 2401–2409. https://doi.org/10.1111/j.1572-0241.2006.00771.x

van Engelenburg-van Lonkhuyzen, M. L., Bols, E. M. J., Benninga, M. A., Verwijs, W. A., & de Bie, R. A. (2017). Effectiveness of pelvic physiotherapy in children with functional constipation compared with standard medical care. *Gastroenterology*, *152*(1), 82–91. https://doi.org/10.1053/j.gastro.2016.09.015

van Tilburg, M. A. L., Squires, M., Blois-Martin, N., Williams, C., Benninga, M. A., Peeters, B., & Ulshen, M. (2012). Parental knowledge of fecal incontinence in children. *Journal of Pediatric Gastroenterology and Nutrition*, *55*(3), 283–287. https://doi.org/10.1097/MPG.0b013e31824cf555

Varni, J. W., Bendo, C. B., Nurko, S., Shulman, R. J., Self, M. M., Franciosi, J. P., Saps, M., & Pohl, J. F. (2015). Health-related quality of life in pediatric patients with functional and organic gastrointestinal diseases. *The Journal of Pediatrics*, *166*(1), 85-90.e2. https://doi.org/10.1016/j.jpeds.2014.08.022

Varni, J. W., Nurko, S., Shulman, R. J., Self, M. M., Saps, M., Bendo, C. B., Vaughan Dark, C., Pohl, J. F., & Consortium, for the P. Q. of L. I. G. S. M. T. S. (2015). Pediatric functional constipation gastrointestinal symptom profile compared with healthy controls. *Journal of Pediatric Gastroenterology and Nutrition*, *61*(4), 424–430. https://doi.org/10.1097/MPG.0000000000000869

Vriesman, M. H., Rajindrajith, S., Koppen, I. J. N., van Etten- Jamaludin, F. S., van Dijk, M., Devanarayana, N. M., Tabbers, M. M., & Benninga, M. A. (2019). Quality of life in children with functional constipation: A systematic review and meta-analysis. *The Journal of Pediatrics*, *214*, 141–150. https://doi.org/10.1016/j.jpeds.2019.06.059

Wang, C., Shang, L., Zhang, Y., Tian, J., Wang, B., Yang, X., Sun, L., Du, C., Jiang, X., & Xu, Y. (2013). Impact of functional constipation on health-related quality of life in preschool children and their families in Xi'an, China. *PLoS ONE*, *8*(10), e77273. https://doi.org/10.1371/journal.pone.0077273

Ward, P. R., Thompson, J., Barber, R., Armitage, C. J., Boote, J. D., Cooper, C. L., & Jones, G. L. (2010). Critical perspectives on 'consumer involvement' in health research: Epistemological dissonance and the know-do gap. *Journal of Sociology*, *46*(1), 63–82. https://doi.org/10.1177/1440783309351771

Yang, C. H., & Punati, J. (2015). Practice patterns of pediatricians and trainees for the management of functional constipation compared with 2006 NASPGHAN guidelines. *Journal of Pediatric Gastroenterology and Nutrition*, *60*(3), 308–311. https://doi.org/10.1097/MPG.0000000000000591

Youssef, N. N., Langseder, A. L., Verga, B. J., Mones, R. L., & Rosh, J. R. (2005). Chronic childhood constipation is associated with impaired quality of life: A case-controlled study. *Journal of Pediatric Gastroenterology and Nutrition*, *41*(1), 56–60.

Chapter 2. Paper 1: Parents' Experiences and Information Needs While Caring for a Child with Functional Constipation: A Systematic Review

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Abstract

Pediatric Functional Constipation (FC) reportedly affects at least 1 in 10 children worldwide. Parent and family education is a key component for successful treatment, yet there is little research exploring what information families need and how to best support them. The aim of this review is to synthesize current evidence on the experiences and information needs of parents caring for a child with FC. We systematically searched published research and completed screening against *a priori* inclusion criteria. Thirteen studies, (n=10 quantitative, n=3 qualitative) were included. We found two main themes, *precarious footing* and *profound and pervasive effects*. Heavy caregiving burdens fuelled doubts, misinformation, relationship breakdown, and treatment deviation. In light of clinical recommendations, our findings reveal a potential mismatch between parents' needs and care provision for FC. It is likely that both parents and healthcare providers would benefit from resources and interventions to improve care related to pediatric FC.

Systematic Review Registration: PROSPERO # CRD42020148750, Open Science Framework DOI 10.17605/OSF.IO/GRYDM,

Keywords: constipation, pediatric, parents, information needs, experience

Introduction

Pediatric constipation is common and causes significant problems for children and families. Although reported prevalence rates of pediatric Functional Constipation (FC) vary widely and differ across global contexts, a conservative estimate is that 1 in 10 children worldwide are affected by the condition (Koppen, Vriesman, Saps, et al., 2018; Timmerman et al., 2019). FC is by far the most common type of constipation and occurs without underlying medical or physiological causes. Expert consensus established the ROME IV diagnostic criteria, which are widely used in clinical practice (Benninga et al., 2016; Hyams et al., 2016) (see Supplementary File 1). In contrast to the common misconception that pediatric FC is an inconvenient but harmless condition, research findings have demonstrated the heavy physical, emotional, psychosocial, and financial burdens for children, families, and healthcare systems (Liem et al., 2009; van den Berg et al., 2006; Varni, Bendo, et al., 2015). Symptoms such as recurrent abdominal pain, pain with defecation, fecal incontinence, urinary incontinence, and urinary infections are common. Furthermore, children with FC report psychological, emotional, and social distress such as impaired relationships, increased familial stress, and decreased quality of life (Bongers et al., 2009; Varni, Bendo, et al., 2015; Youssef et al., 2005). The significance of these psychosocial effects is underscored by research indicating that children with FC report lower quality of life than children with organic gastrointestinal disease such as inflammatory bowel diseases (e.g. Crohn disease and Ulcerative Colitis) (Varni, Bendo, et al., 2015).

Additionally, children with FC typically use more healthcare resources such as emergency department visits and specialist care. For example, pediatric FC accounts for upwards of 25% of pediatric gastroenterology visits (Jurgens et al., 2011; Liem et al., 2009). Medical costs related to pediatric FC are estimated to be threefold higher per year than for children without constipation (Liem et al., 2009). In addition to the direct financial expenses, there are substantial indirect costs associated with pediatric FC. For example, the high rate of missed school days amongst children with FC (Liem et al., 2009) creates a domino-like effect related to factors such as parental work absenteeism, parental income, and the child's academic success. The overall burden of illness from pediatric FC is significant and affects multiple aspects of health at individual, family, and system levels.

Parents of children with FC are typically tasked with onerous and long-term treatment regimens. Management of pediatric FC typically involves daily oral medication therapy using osmotic laxatives such as polyethylene glycol 3350, combined with behavioural interventions and additional agents according to individual needs (Brazzelli et al., 2011; Gordon et al., 2016; National Collaborating Centre for Women's and Children's Health, 2010; Rowan-Legg & Canadian Paediatric Society, 2011; Tabbers et al., 2014). Unfortunately, despite the therapeutic options available, pediatric FC remains undertreated in primary care (Borowitz et al., 2005). Delayed or suboptimal treatment is particularly problematic since children often experience a progressively worsening trajectory of the condition due to the cyclical nature of symptoms and digestive physiology. For example, painful defecation related to large stool size exacerbates stool-holding behaviours, which in turn causes increased water absorption from the stool, larger stool size, more difficulty passing stool, and worsening painful defecation. FC rarely resolves without intensive intervention and about 40% of children with FC will develop persistent symptoms lasting for more than one year, while ¼ of affected children will become adults with chronic or lifelong FC (Bongers et al., 2010).

Clinical practice guidelines for pediatric FC identify parent and family education as an important step towards effective treatment (National Collaborating Centre for Women's and Children's Health, 2010; Rowan-Legg & Canadian Paediatric Society, 2011; Tabbers et al., 2014). Similarly, parents have expressed the need for relevant, accessible, and reliable information sources to support their child-health decision-making (Jackson et al., 2008). Although reliable, high-quality evidence about childhood constipation is readily available to clinicians, families do not have the same access to information, nor is it clear if they have the same information needs. Relying on healthcare providers' impressions of parents' information needs is likely to be problematic. When parents are caring for an unwell child, the complexities of family life often create unique and dynamic priorities that may transcend the need for basic factual information (Thompson et al., 2019). In addition, FC tends to be a difficult topic for many parents to discuss, even with healthcare providers. Specifically, a previous study found that parents' experiences with childhood FC were often misunderstood by healthcare professionals (Farrell et al., 2003). Therefore, a comprehensive understanding of parents' experiences and self-identified needs when caring for a child with FC is a necessary step to ensure clinicians can provide relevant support.

There is currently a significant knowledge gap at the intersection of parents' experiences and information needs related to caring for a child with FC. Our search of bibliographic databases and registered protocols did not find any current or planned evidence syntheses examining parents' experiences or needs related to caring for a child with functional constipation. This systematic review aims to understand and give voice to parents' information and support needs. Developing an accurate synthesis of parents' needs related to pediatric FC provides a foundation for creating accurate and relevant patient-direct resources. Patient-direct KT connects families to research evidence, creating opportunities to increase knowledge, improve experiences, optimize health resource use, and encourage effective health behaviours (Jackson et al., 2008; Stacey & Hill, 2013). In order to make informed decisions about their children's health, parents caring for a child with FC deserve accessible resources that answer their questions and meet their support needs.

Objectives

The primary objective of this study is to identify, map, and synthesize current evidence on the experiences and information needs of parents caring for a child with FC. Secondary aims are, a) to strengthen the design and planning of subsequent research in the field by identifying existing gaps, and b) to build an evidence base suitable for creating relevant resources to support parents caring for a child with FC. The study used the following research questions to meet these objectives:

1. What are parents' experiences of caring for a child with FC?
2. What information do parents need to understand pediatric FC, make child-health decisions related to pediatric FC, and feel supported when caring for a child with FC?

Methods

We used systematic review methodology with guidance from the Centre for Reviews and Dissemination (Centre for Reviews and Dissemination, 2008) and followed a registered protocol (PROSPERO # CRD42020148750). Our Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA)(Moher et al., 2009) checklist is included as Supplementary File 2.

Search Strategy

The search strategy was developed through collaboration with a health sciences research librarian with expertise creating and conducing systematic searches. The search used a combination of keywords and subject headings for the following concepts: (a) constipation in children, not limited by etiology and including related terms such as functional, inorganic, nonorganic, idiopathic, fecal incontinence, and encopresis; (b) parents and individuals identified as primary caregivers (e.g. foster parents and extended family in a primary caregiving role); and

(c) information needs, experiences, perspectives, views, practices, beliefs, knowledge, and questions. Relating to the concept of constipation, terms for fecal incontinence and encopresis are included because approximately 95% of fecal incontinence cases are caused by underlying constipation (Loening-Baucke, 2007). Therefore, we included these terms with the expectation that parental perspectives and experiences may be tied to the symptom rather than the underlying diagnosis. The same search approach was not extended to other symptoms of childhood FC (ex. abdominal pain) because of diversity in underlying causes and associated diagnoses. Finally, a pediatric gastroenterologist with expertise in childhood FC and a clinician scientist with expertise in systematic reviews assessed the completeness of the search.

The full systematic search was run on November 27th, 2019 in the following electronic databases: Ovid MEDLINE® and Epub Ahead of Print, In-Process & Other Non-Indexed Citations and Daily, Ovid Embase, Ovid PsycINFO, EBSCO CINAHL Plus with Full-text, Wiley Cochrane Library, ProQuest Dissertations & Theses Global, Scopus, Web of Science Core Collection. All databases were searched from inception to present. No date, language or study design limits were applied. The full search strategies are listed by database in Supplementary File 3.

Eligibility Criteria

We considered primary research focused on the information needs, experiences, beliefs, knowledge, or practices of parents related to childhood FC. Detailed inclusion and exclusion criteria for this review are presented in Table 2.1.

Table 2.1 Eligibility Criteria

	Inclusion Criteria	Exclusion Criteria
Study design	Primary research, any design	Reviews, meta-analyses, editorials, letters, opinion pieces, clinical summaries, reports
Study focus	Relates to the information needs, experiences, beliefs, knowledge or practices of parents or primary caregivers currently or previously caring for a child, aged 0-18 years, with FC / encopresis / fecal incontinence. Must be parents' perspective.	1) Constipation or related symptoms with a known non-functional cause (e.g. anatomical, radiological, histological or pharmacological) 2) Studies measuring interventions related to childhood FC (e.g. program satisfaction, treatment efficacy etc) 3) Studies measuring the prevalence/incidence, or factors correlated with prevalence or etiology of childhood FC 4) Studies measuring attributes, demographics, or characteristics of parents of a child with FC
Study outcomes	Information needs, experiences, beliefs, knowledge or practices of parents related to childhood FC	1) Parental data not reported 2) Parental data reported in combination with professional caregivers 3) Parental data used as proxy for child's perspective
Language	No restriction	--
Setting	No restriction	--
Reporting	Published studies & dissertations	Conference proceedings and abstracts

Although studies examining the prevalence, epidemiology, and demographic correlations with pediatric FC are important areas of research; these types of research questions do not provide insight into the experience of a parent caring for a child with this condition. Similarly, healthcare professionals have valuable expertise about the topic and its treatment, but parental perspectives are different from those of health care providers. For example, parents must prioritize the diverse needs of all family members, the effects of treatment on the psychosocial dynamics within the family, the financial costs that may be associated with the condition, and other complexities that are unique to each family and not experienced by healthcare providers. Similarly, in studies where parents answered questions on behalf of or relating to their child's experience with the condition, the unique perspective of the caregiver is not captured and thus these studies were also excluded. Lastly, intervention studies were excluded when parental perspectives were not evident or only included reflections on the particular intervention or treatment.

Screening and Selection Procedures

Screening and selection followed the process outlined by the PRISMA statement (Moher et al., 2009), with decisions and rationale documented throughout the review process. Covidence® review software was used to manage the identified literature. All duplicates were removed. All levels of screening were completed independently by two reviewers (AT, AC). First-level screening assessed titles and abstracts for relevance using *include*, *exclude,* and *unsure* designations. All *unsure* decisions and any records with reviewer disagreement were included for second-level screening. Full-text articles were screened based on the detailed criteria and categorized as include, exclude, or unsure. Disagreements and unsure decisions were resolved through discussion and third party consultation as required (SS). Non-English records moved through the screening in the same manner and were assessed using available translation services. Preliminary screening of non-English records was based upon available English abstracts and maintained the same inclusion and exclusion criteria.

Quality Assessment

Due to the narrow scope of the review question and the small number of anticipated eligible studies, quality assessment was used to inform the discussion about the state of the research evidence but was not used to determine the inclusion or exclusion of articles. Included articles were critically appraised using the Mixed Methods Appraisal Tool (MMAT) version 2018 (Pluye et al., 2009). The MMAT is inclusive of a wide range of research designs within a single tool, thereby increasing the consistency of appraisal across the research. In addition, the tool is supported by comprehensive guidelines and explanations (Hong et al., 2018) to facilitate use. Each study was assessed independently by two reviewers (AT, AC) with discrepancies resolved through discussion and third-party consultation as needed (SS).

Data Extraction

Data from all included studies were extracted into an Excel® spreadsheet and the extraction process was completed by one reviewer (AT) with a second reviewer (AC) for validation. One study (Banaszkiewicz et al., 2005) was translated to English by a colleague with research expertise and familiarity with the Polish language. The data extraction template was created and piloted with 1-2 articles of each type of literature to ensure comprehensiveness and ease of use. Data extraction fields included, study identification number, first author, title, journal, publication year, study design, theoretical framework/philosophical position, research question or objective, sample method, setting or country, participant characteristics, child's age and duration of FC, data collection method, outcome, instruments, time frame, tool reliability, tool validity, analysis, findings or results, limitations, strategies to support rigor, and general author or reviewer comments. All findings, outcomes, and measures relating to parents' information needs and experiences caring for a child with FC were extracted.

Data Synthesis

The variety of our included studies aligns with the principles of a narrative synthesis approach, which is commonly used when evidence is highly heterogeneous by methodology, outcome, or sample (Centre for Reviews and Dissemination, 2008; Popay et al., 2006). In addition, the method is well supported by extensive guidance by Popay and colleagues (2006), which maintains the rigour of the synthesis process and was used as a guide for our narrative synthesis. We proceeded through the main steps of 1) developing a preliminary synthesis, 2) exploring relationships in the data, and 3) assessing the robustness of the synthesis product (Popay et al., 2006). Discussion of the strength of the body of evidence will be based on the cumulative depth of the data because quantitative assessments of the evidence (e.g., GRADE) are not applicable for syntheses including qualitative studies.

Procedures to Minimize Bias

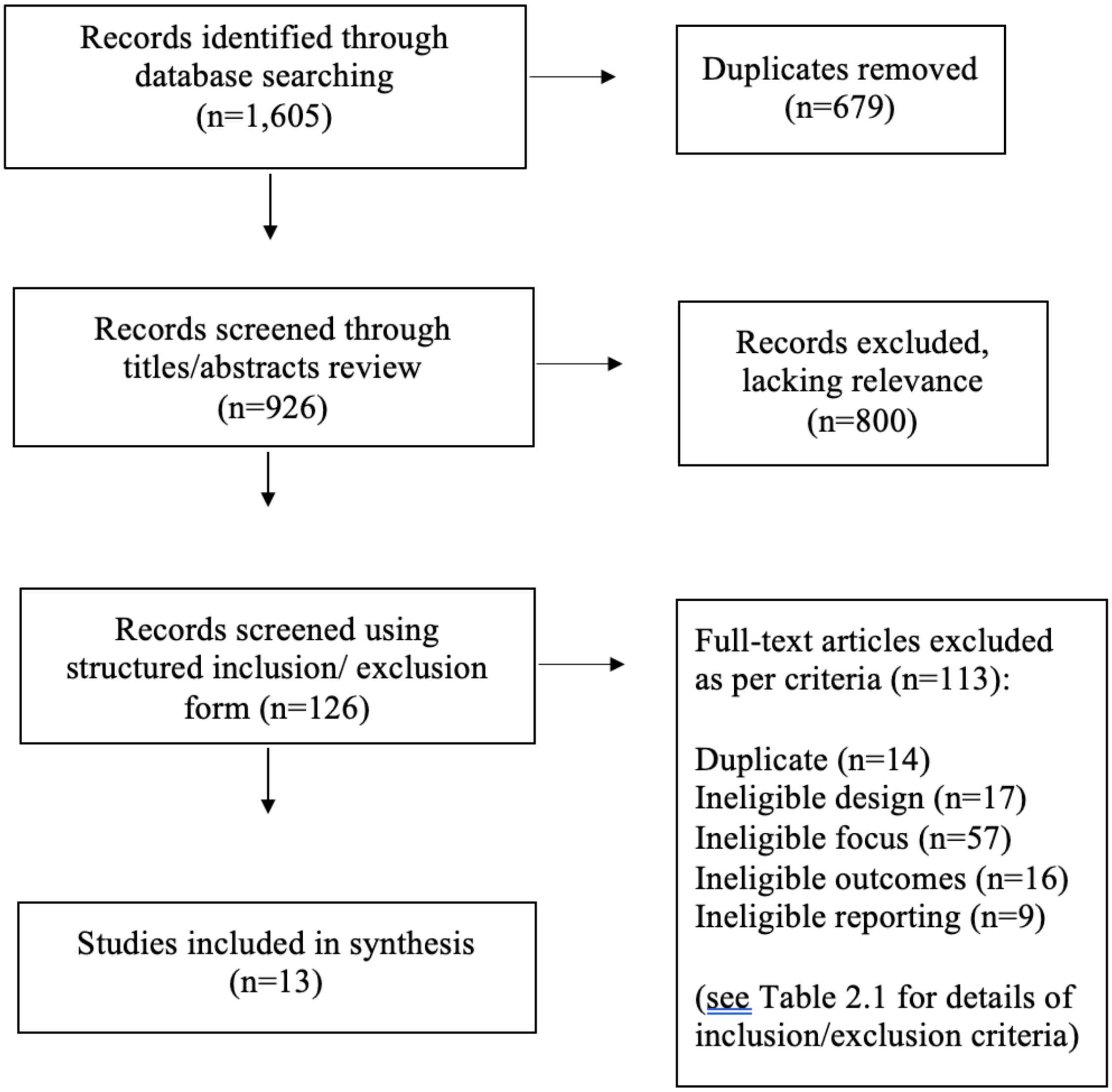
The following processes and decisions were created to minimize the risk of bias throughout the review process with the goal to increase the quality of results. First, the broad approach to including studies of all types, languages, and dates. Second, the use of two independent reviewers for record screening and selection. Third, documentation with rationale and reporting of all exclusion decisions, including a study flow diagram. Fourth, disagreement between reviewers mediated with involvement of a third-party content and process expert. Fifth, data extraction verified by a second reviewer. Finally, the creation and registration of an *a priori* protocol (PROSPERO # CRD42020148750) serves to increase transparency and minimize bias.

Results

Study Characteristics

Thirteen studies met our criteria and were included for quality appraisal and synthesis (Figure 2.1).

Figure 2.1 PRISMA flow diagram of study selection



Publication dates ranged from 1993 – 2018 and median year of publication was 2012. Of the total included studies, ten are quantitative (Banaszkiewicz et al., 2005; Bernardbonnin et al., 1993; Dolgun et al., 2013; Holman, 2012; Klages et al., 2016; Koppen, van Wassenaer, Barendsen, et al., 2018; Kovacic et al., 2015; Samson, 2004; van Tilburg et al., 2012; Wang et al., 2013) and three are qualitative (Brennan-Parsons, 2000; Farrell et al., 2003; Kaugars et al., 2010). Studies reported on a total of 1,146 participants [*studies (Klages et al., 2016; Kovacic et al., 2015) reported data collected from the same sample on different outcomes]. Sample size of quantitative studies ranged from 22 to 410 (mean 114) and qualitative studies ranged 8 to 14 (mean 10). Further study details are provided in Table 2. Due to the narrow scope of the research question and the limited number of relevant studies in the field, our inclusion of qualitative and quantitative designs strengthened our ability to generate meaningful findings across the studies. Qualitative studies provided a comprehensive and rich exploration of parents' experiences while quantitative studies quantified knowledge, quality of life effects, and medication use while caring for a child with FC. Due to the parental focus of our research question, reported data pertaining only to child outcomes were not extracted or synthesized. Quality appraisal information is detailed in Table 3. Since we did not plan to exclude studies based on quality appraisal, one study (Dolgun et al., 2013) with questionable quality assessment remains in our included studies, but the weaknesses of the study did not support substantive inclusion of the authors' findings in our synthesis.

Table 2.2 Overview of Included Studies

Year	First Author	Design	Sample	Country	Child characteristics	Child duration of FC
1993	Bernardbonnin *et al.*	Quantitative - cross-sectional retrospective	n=28 parent/child pairs	Canada	median age 10yrs boys = 20 girls =8	median 2yrs prior to attending clinic and followed by clinic 3.5 yrs (+/-) 0.2 at the time of data collection
2000	Brennan-Parsons	Qualitative - hermeneutic phenomenology	n=8 mothers	Canada	age range 8-13 years 2 females 6 males	minimum 1 year of encopresis, range 2-8 years
2003	Farrell *et al.*	Qualitative - phenomenology	n= parents of 14 children	UK	Sampled according to age and healthcare setting matrix.	Not known

					Further details of final sample not reported	
2005	Banaszkiewicz *et al.*	Quantitative - cross sectional survey	n=60 parents	Poland	mean age 83.2 +/- 48.3 months	29.5 +/- 38.5month duration of constipation
2006	Samson	Quantitative – RCT	n=22	UK	mean age 7.73yrs +/-2.81 82% male 18% female	duration of soiling mean = 2.95 yrs +/-1.73
2010	Kaugars *et al.*	Qualitative - content analysis	n=8 children n=8 parents	USA	mean age 10yrs +/-2.23	Not known

					69% boys	
2012	Holman	Quantitative - cross sectional with matched control	n=68 matched pairs	USA	mean age 4.3 yrs 56% female	Not known
2012	vanTilburg *et al.*	Quantitative - cross sectional	n=232	USA	mean age 7.5years +/- 3.3 70% boys 30% girls 74% white 15% African American 11% Asian ethnicity 10% Hispanic	Not known
2013	Dolgun *et al.*	Quantitative - quasi-experimental	n= 26 children & their mothers	Turkey	mean age 7.21 years +/- 4.12 73.1% boys; 26.9% girls	Not known

2013	Wang *et al.*	Quantitative - cross-sectional with case control	n=152 children and primary caregiver n=176 healthy children and primary caregiver (control group)	China	mean age 4.2 years +/-0.9 boys 53%	5.7 +/-2.3 (months)
2015	Kovacic *et al.*	Quantitative – prospective* (cross-sectional)	n=families of 410 children	USA	mean 7.8yr +/-3.5, 52% male 184 (45%) FC only 226 (55%) FC + FI	age at symptom onset: 3 (3.6) (FC) 3.6 (2.8) (FC+FI) age when help sought: 4 (4) (FC) 4.4 (2.87) (FC+FI) symptom

						duration: 4.2 (3.85) (FC) 4.4 (3.34) (FC+FI)
2016	Klages *et al.*	Quantitative - cross sectional	n=410 caregivers ** same sample as Kovacic - reporting on different tools.	USA	mean age 7.8 years +/-3.5 52% male 184(45%) FC only 226 (55%) FC + FI	age at symptom onset 3-3.6 +/- 2.8-3.6 age when help sought 4-4.4 +/- 2.87-4 symptom duration 4.2-4.4 +/-3.35-3.85

2018	Koppen *et al.*	Quantitative - cross-sectional	n=115	The Netherlands	median age 7.8 yrs 70 female 45 male	median duration of symptoms 42 months

Table 2.3 Quality Assessment (Qualitative)

Mixed Methods Appraisal Tool Questions		First Author of Study		
		Brennan-Parsons	Farrell	Kaugars
Screening Questions	S1. Are there clear research questions?	✓	✓	✓
	S2. Do the collected data allow to address the research questions?	✓	✓	✓
1. Qualitative	1.1. Is the qualitative approach appropriate to answer the research question?	✓	✓	✓
	1.2. Are the qualitative data collection methods adequate to address the research question?	✓	✓	✓
	1.3. Are the findings adequately derived from the data?	✓	✓	✓
	1.4. Is the interpretation of results sufficiently substantiated by data?	✓	✓	✓
	1.5. Is there coherence between qualitative data sources, collection, analysis and interpretation?	✓	✓	✓
	Comments	none	none	none

Table 2.4 Quality Assessment (Quantitative randomized controlled trials)

Mixed Methods Appraisal Tool Questions		First Author of Study Samson
Screening Questions	S1. Are there clear research questions?	✓
	S2. Do the collected data allow to address the research questions?	✓
2. Quantitative randomized controlled trials	2.1. Is randomization appropriately performed?	?
	2.2. Are the groups comparable at baseline?	✓
	2.3. Are there complete outcome data?	✗
	2.4. Are outcome assessors blinded to the intervention provided?	✓
	2.5 Did the participants adhere to the assigned intervention?	✓
	Comments	did not address attrition

Table 2.5 Quality Assessment (Quantitative non-randomized)

Mixed Methods Appraisal Tool Questions		First Author of Study		
		Wang	Holman	Dolgun
Screening Questions	S1. Are there clear research questions?	✓	✓	✓
	S2. Do the collected data allow to address the research questions?	✓	✓	✗
3. Quantitative non-randomized	3.1. Are the participants representative of the target population?	✓	✓	✓
	3.2. Are measurements appropriate regarding both the outcome and intervention (or exposure)?	✓	✓	✓
	3.3. Are there complete outcome data?	✓	?	✓
	3.4. Are the confounders accounted for in the design and analysis?	✓	✗	✗
	3.5. During the study period, is the intervention administered (or exposure occurred) as intended?	✓	?	?
	Comments	worry inversed in both the control and FC groups? error or surprising finding?	concurrent diagnoses not excluded	measurement of FC did not occur at same interval as QOL

Table 2.6 Quality Assessment (Quantitative descriptive)

Mixed Methods Appraisal Tool Questions		First Author of Study					
		Klages	Koppen	Kovacic	Banaszkiewicz	van Tilburg	Bernardbonnin
Screening Questions	S1. Are there clear research questions?	✓	✓	✓	✓	✓	✓
	S2. Do the collected data allow to address the research questions?	✓	✓	✓	✓	✓	✓
4.0 Quantitative descriptive	4.1. Is the sampling strategy relevant to address the research question?	✓	✓	✓	✓	✓	✓
	4.2. Is the sample representative of the target population?	✓	✓	✓	✓	✓	✓
	4.3. Are the measurements appropriate?	✓	✓	✓	?	✓	✓
	4.4. Is the risk of nonresponse bias low?	✓	✓	✓	✓	?	?
	4.5. Is the statistical analysis appropriate to answer the research question?	✓	✓	✓	✓	✓	?
	Comments	none	none	none	none	none	underpowered

Themes Identified

Narrative synthesis of the included studies found two general themes that help explain parents' experiences and information needs when caring for a child with FC. First, *precarious footing* with subthemes related to mis/understanding pediatric FC and treatments. Second, *profound and pervasive effects* describes families' distress while caring for a child with FC. Both themes were underpinned by parents' unmet needs for information, validation, and support.

Precarious Footing

The theme and subthemes highlight a typical experience of parents working with incomplete knowledge, creating risks to derail family relationships and treatment plans for pediatric FC. Ambiguity permeated multiple aspects of parents' experiences, including understanding the diagnosis and implementing complex interventions while navigating healthcare and family relationships. Parents caring for a child with FC typically had a combination of accurate knowledge and misconceptions about the condition and treatments. Information gaps and misinformation negatively affected parental confidence and child-health decision making. Consequently, parents often lacked a solid foundation for caregiving, which led to the identification of our first theme: precarious footing.

Mis/understanding pediatric FC

How and when families received a diagnosis of pediatric FC was not explored as a primary research question in any of the studies; however, data suggest that symptoms of pediatric FC were often present for months or years prior to seeking help (Bernardbonnin et al., 1993; Klages et al., 2016; Kovacic et al., 2015). Furthermore, parents recollected frustratingly long timelines to receive a clear, explicitly communicated diagnosis (Brennan-Parsons, 2000; Farrell et al., 2003). It was typical for parents to describe multiple encounters with healthcare providers in order to understand their child's symptoms. For example, "*I had him to the doctor I* *don't know how many times… they did blood work and stuff like that and couldn't find anything*" (Brennan-Parsons, 2000, p. 85). Similarly, another parent reported, "*the least helpful was when doctors would look up and tell you that it's nothing, it's just a little 'he wants attention'*" (Brennan-Parsons, 2000, p. 85). In other cases, FC was recognized but parents felt dismissed or were implied to be overreacting to a benign concern; "*But it was awful because they were just fobbing us off. The way I felt was that they have got more urgent problems, more people to see than a little girl or little boy with constipation: It isn't a problem. They will grow out of it. Don't worry about it*" (Farrell et al., 2003, p. 485). Similarly, another parent reported her experience with a healthcare provider, "*And I said, 'I'm not happy because it feels like you say to me ''He'll grow out of it'', and he's not getting any better''*" (Farrell et al., 2003). Even after receiving a diagnosis of FC, parents' reflections often included persistent ambiguity about the cause of the child's symptoms (Bernardbonnin et al., 1993; van Tilburg et al., 2012).

Furthermore, one study highlighted the importance of providing adequate information to parents (Samson, 2004). In a randomized controlled trial, the group who received written information about this condition in addition to standard care had better outcomes (a measured by fewer soiling episodes) at the end of treatment compared to the group who received only standard care (Samson, 2004). Even though many parents were able to identify common physiological and psychological elements of pediatric FC such as intestinal dysfunction and stool withholding due to pain (Bernardbonnin et al., 1993); misconceptions remained a poignant element of parents' experiences. Parents actively sought information, but often did not find answers to their questions (Bernardbonnin et al., 1993; Brennan-Parsons, 2000; Farrell et al., 2003; Kaugars et al., 2010; van Tilburg et al., 2012). For example, one parent described her efforts to understand the condition, "*I looked it up on the Internet. I looked it up at the library* *and I asked questions. There really isn't a lot out there*" (Brennan-Parsons, 2000, p. 91). Overall, there were two predominant findings related to misinformation that are concerning for the well-being and clinical care of families living with pediatric FC. First, qualitative studies (Brennan-Parsons, 2000; Farrell et al., 2003) highlighted the interplay between etiologic uncertainty, parental guilt, and dismissive attitudes. One mother reflected on her ambiguity and the resulting self-blame, "*I know this is a medical problem and all that, but you can't help but think, in the back of your mind, was it something I did? What's wrong with me and why does my child have it you know… And you feel guilt. It is very upsetting*" (Brennan-Parsons, 2000, p. 68). In addition, encounters with family, friends, teachers, and even healthcare professionals contributed to parents' misunderstanding of the condition and feelings of being judged (Brennan-Parsons, 2000; Kaugars et al., 2010). One parent explained, "*I did go to the doctor, and I never got any satisfaction. I was thinking it can't be anything medical because he would know about it…You think it's your child [only] and a bit of shame creeps in there*" (Brennan-Parsons, 2000, p. 81). Furthermore, some parents' negative healthcare experiences propagated parental guilt, and eroded relationships (Brennan-Parsons, 2000; Farrell et al., 2003). Specifically, some providers implicitly or explicitly suggested parents' bore responsibility; "*I actually went to a doctor here who blamed me. He did! He said [pause], because she was older now, if I had been stricter, if I had been keeping to the regime, if I had been more forceful…she would be taking this lactulose. This after months and months of taking it and he basically blamed me*" (Brennan-Parsons, 2000, p. 70).

Second, there was a subset of parents who expressed a concerning lack of understanding about the etiology of FC and believed that fecal incontinence was caused by negative personality traits or intentional misbehaviour rather than as a sequela of the condition (Bernardbonnin et al., 1993; Brennan-Parsons, 2000; Kaugars et al., 2010; van Tilburg et al., 2012). For example, one mother stated, "*We thought he was just too lazy. You know, the first thing that comes to your mind, he's just too lazy and doesn't want to take the time*." (Brennan-Parsons, 2000, p. 66). One study found that approximately 10% (Bernardbonnin et al., 1993) of parents believed soiling was a deliberate act of defiance or done purposely to gain attention. Similarly, another study found 13% of parents felt soiling occurred because the child was not trying hard enough (Samson, 2004). In addition, parents described shame about their child's condition as a reason to avoid discussing the topic with peers or healthcare providers (Kaugars et al., 2010). For example, one parent described the struggle to anticipate how others would react to the condition, *"I think that they [other people] have the same understanding that I've had all these years—that he is being lazy"* (Kaugars et al., 2010). Furthermore, one study found parents' beliefs about fecal incontinence fell into two broad categories. "Blame and punish" reflected parental attitudes and beliefs that the child's soiling was intentional, and discipline measures were an appropriate intervention (van Tilburg et al., 2012). Conversely, "worry and help" existed when parents attributed the child's symptoms to constipation and sought support and information. Although the attitudes reflected in these categorizations were relatively dichotomous, the authors suggested many parents oscillated between the two positions (van Tilburg et al., 2012). In summary, although many parents expressed a basic understanding of pediatric FC, feelings of guilt or shame, lingering misconceptions about the physiology of FC, and receiving mixed messages from care providers all contributed to parental uncertainty about the condition.

Mis/understanding treatments

Dietary changes, medication use, and behavioural interventions were all commonly identified by parents as important components of treating pediatric FC (Banaszkiewicz et al., 2005; Bernardbonnin et al., 1993; Brennan-Parsons, 2000; Koppen, van Wassenaer, Barendsen, et al., 2018; van Tilburg et al., 2012). To assess parents' knowledge of dietary changes related to FC, one study measured parents' ability to accurately identify high and low fibre foods (Banaszkiewicz et al., 2005). Results showed that only 15% of parents in the study were able to correctly identify at least 75% of the high-fibre foods presented. Furthermore, parents who reported that they had received previous information about the fibre content of common foods were not more successful at identifying high-fibre foods (Banaszkiewicz et al., 2005). In addition to having difficulty applying dietary knowledge, parents described practical challenges to implementing dietary changes. "*We had to cut out fat in his diet and [give him] a high fibre diet. But it is hard you know. Because I find it very hard to put a ten year old on a high fibre diet*" (Brennan-Parsons, 2000, p. 77). Similarly, another parent described her child's refusal, "*he didn't like the bran, didn't want it, wouldn't eat it. Meal time in the hospital was torture, torture. It is also at home actually. I've tried to get him to eat. He says he doesn't want to eat. He is always full*" (Brennan-Parsons, 2000, p. 77).

In addition to the practical challenges and potential lack of knowledge associated with implementing dietary changes for their child (Banaszkiewicz et al., 2005), parents also expressed doubts about medications used to treat pediatric FC (Koppen, van Wassenaer, Barendsen, et al., 2018). Specifically, a recent study found that 60% of children were found to be nonadherence to polyethylene glycol treatment (Koppen, van Wassenaer, Barendsen, et al., 2018). Higher levels of medication adherence were associated with parents who had positive beliefs about the treatment convenience and treatment satisfaction (Koppen, van Wassenaer, Barendsen, et al., 2018). In contrast, parents of children who were nonadherent with medication expressed more concerns, such as fears of medication overuse or dependence on laxatives (Farrell et al., 2003;

Koppen, van Wassenaer, Barendsen, et al., 2018). In addition, parents who identified a heavy emotional burden related to their child's FC were more likely to be nonadherent with medication use (Koppen, van Wassenaer, Barendsen, et al., 2018). Likewise, the emotional burden felt by parents was exacerbated when medication use was a source of parent-child conflict. One parent reported, "*Taking the mineral oil was a big fight. I was giving it to him in a syringe and I had to put it at the back of his throat because he would never take it. All of this was very frustrating and hard on me…she would say you have to hold him down and force it into him. I said well I don't like to do that, I'd like it to be voluntary. But it was the only way we could get it into him*" (Brennan-Parsons, 2000, p. 78). Another parent described low treat satisfaction due to side effects of the medication, "*Then when the mineral oil started to come out of his bum, it would be an orange liquid, bright, bright orange. He was ruining furniture, he was ruining his clothes, he was ruining his bed, he had it over everything*" (Brennan-Parsons, 2000, p. 78). Moreover, authors of the included studies highlighted the importance of identifying and recognizing potential barriers to medication adherence for families living with pediatric FC (Brennan-Parsons, 2000; Farrell et al., 2003; Koppen, van Wassenaer, Barendsen, et al., 2018).

Although making dietary changes and medication use were identified as challenges for parents, implementing behavioural interventions was consistently identified as exceedingly frustrating (Brennan-Parsons, 2000; Kaugars et al., 2010). In contrast to the potential knowledge gaps related to dietary changes (Banaszkiewicz et al., 2005) and medication use, parents understood behavioural interventions, but typically struggled with the familial conflict that arose (Brennan-Parsons, 2000; Kaugars et al., 2010). For example, one parent described attempting to promote a toilet routine with her child, "S*he's not listening. She totally tunes it all out. No matter how hard I try and put it in happy terms, or stay away from the mad, angry, or whatever. Still* *she blocks it out*" (Brennan-Parsons, 2000, p. 75). Behaviour-based treatments were more commonly associated with parent-child conflict and induced emotionally laden reactions from parents. "*It's just one task after another, it's a battle, it's a fight. He gets frustrated, I get frustrated. He gets mad, I get mad and it is just horrible. It's horrible*" (Brennan-Parsons, 2000, p. 93). Conversely, studies identified positive interactions with healthcare providers as a supportive influence, enabling parents to persevere with difficult interventions (Brennan-Parsons, 2000; Farrell et al., 2003). For example, in one encounter the healthcare provider was enthusiastic about care planning, "*Straight away, he responds great: 'That's it, bring (name) along with you next time you come. That's it now, I'll see (name).' And I thought 'Oh...this is brilliant'. And straight away, you know: 'Let's get a plan going for him'*"(Farrell et al., 2003, p. 486). Overall, included studies reveal that treatment regimens for pediatric FC were associated with diverse challenges related to lack of information and as a result of the conflict that was associated with implementing behavioural interventions (Banaszkiewicz et al., 2005; Bernardbonnin et al., 1993; Brennan-Parsons, 2000; Farrell et al., 2003; Kaugars et al., 2010; Koppen, van Wassenaer, Barendsen, et al., 2018).

Profound and Pervasive Effects

Parents experienced diverse and often hidden effects of their child's condition which reverberated throughout the family and beyond the immediate physical effects. A common objective of included studies was examining the effects of pediatric FC on health-related quality of life of caregivers and family functioning (Dolgun et al., 2013; Holman, 2012; Kaugars et al., 2010; Klages et al., 2016; Kovacic et al., 2015; Wang et al., 2013). Parents of a child with FC scored significantly lower on physical, emotional, social, cognitive, and communication scores compared to caregivers of healthy children (Wang et al., 2013). In addition, families affected by pediatric FC reported impaired daily activities and relationships compared to healthy controls (Wang et al., 2013). One study further examined groups of children living with FC based on their predominant symptom pattern, such as withholding/avoiding, pain, or fecal incontinence (Klages et al., 2016). Pain and fecal incontinence were associated with more disease burden, caregiver distress, worry about social impacts, and challenges with the medical team than experienced by parents of youth in the withholding/avoiding group (Klages et al., 2016). Findings suggested that positive symptoms such as pain and fecal incontinence generate stronger reactions in caregivers (Klages et al., 2016). Likewise, another study found the symptom of fecal incontinence was correlated with more impairments to family functioning and higher parental stress when compared to pediatric FC without soiling (Kovacic et al., 2015). Families affected by pediatric FC with fecal incontinence had lower scores (lower functioning) on emotional and social functioning, worry, communication, family relationships, and overall family functioning (Kovacic et al., 2015). In addition, parenting stress was higher among families affected by FC with fecal incontinence. Specifically, communication, emotional distress, and role functioning were more challenging than compared to those with FC alone (Kovacic et al., 2015). Similarly, another study found a negative association between parental stress scores and children’s response to an intervention to reduce soiling (Samson, 2004). The authors suggested that children responded better to the intervention when parents were less stressed (Samson, 2004), which underscores the importance of acknowledging and addressing parents’ needs for support.

In addition to recognizing negative effects on parental and family quality of life, evidence from qualitative studies echoed the findings and detailed the profound and pervasive consequences felt by families (Brennan-Parsons, 2000; Farrell et al., 2003; Kaugars et al., 2010).

Effects on caregivers and the family unit were magnified by symptoms of the condition when combined with the burden of the treatment regimen. Parents faced emotional, financial, social, and physical effects of pediatric FC. In particular, they described enormous pressure to continuously monitor their child's bowel movements, while also balancing the increased costs and time associated with laundry, behavioural interventions, attending appointments, and administering medications (Brennan-Parsons, 2000; Farrell et al., 2003; Kaugars et al., 2010). One parent described, *"It's become a daily conversation item in the house, you know, between my husband [and me], and one thing we talk about every day is, has he pooped? You know? Did he clog the toilet or whatever.. It's seeped into every part of our life—even his little sister checking on him"* (Kaugars et al., 2010). Caregivers' lived experiences mirrored the broad range of effects noted in the quality of life studies (Dolgun et al., 2013; Holman, 2012; Klages et al., 2016; Kovacic et al., 2015; Wang et al., 2013). *"Everything is different. Everything from the way that we feed her to the way that we discipline. Everything"* (Kaugars et al., 2010). Similarly, another parent reported, *"It is consuming. It is all-consuming… it is continual. I mean you are living with it every hour in the day"* (Brennan-Parsons, 2000, p. 92). Furthermore, parents reported alarming levels of frustration and potential risks to child well-being. For example, one parent admitted the toll on her child, "*After 4 years of dealing with this, of constantly getting yelled at and getting grounded, yeah, it's gotta make someone feel like crap"*(Kaugars et al., 2010, p. 750). Another parent recognized a precarious emotional burden, "*But I can see it leading to more than that because it is very bad…I can see if you have a tendency for child abuse to occur because that's how bad it is"* (Brennan-Parsons, 2000, p. 93). Likewise, another parent described overwhelming frustration, "*with me, it works on my nerves. I really get sometimes so I just want to choke her and choke the poop out or something"* (Kaugars et al., 2010, p. 751).

In addition to risks within the family, included studies (Brennan-Parsons, 2000; Kaugars et al., 2010) also reported the fear and concern caregivers commonly experience about a child's social well-being, particularly in school and community settings. Parents were highly sensitive to the implications that altered bowel habits and soiling could have on developing peer relationships. For example, one parent described worries of ostracism due to fecal odours, "*Because I've had experiences where he, like, when he was still at school, when school was in session, and he came out of his classroom, it was like (sharp breath) and he was like totally oblivious to the fact that he smelled"*(Kaugars et al., 2010, p. 750). Another parent shared similar fears, *"Will other children want to play with him… I worried whether [son] would soil himself and turn his friends away. That was one of my biggest worries, that one of the kids at school would make fun of him"* (Brennan-Parsons, 2000, p. 82). In addition, parents worried about long-term mental health, beyond the school and peer environments, "*I worry too, what is it doing to him. Is it going to affect him when he grows up…affect what type of person he is going to be*?" (Brennan-Parsons, 2000, p. 90). Furthermore, parents anticipated lasting physical consequences of pediatric FC, "*We don't know how long it's going to go on. And we're worried. The doctors have told us that if her bowels stay large by the time she's an adult she'll have problems for the rest of her life"* (Brennan-Parsons, 2000, p. 90). Overall, most included studies (Bernardbonnin et al., 1993; Dolgun et al., 2013; Farrell et al., 2003; Holman, 2012; Kaugars et al., 2010; Klages et al., 2016; Kovacic et al., 2015; Wang et al., 2013) highlighted the profound and pervasive influence pf pediatric FC on the well-being of families.

Discussion

Practice Implications

We will discuss our findings in light of current clinical evidence and recommendations to highlight directions to improve care for families affected by pediatric FC. Although the number of included studies is relatively small, results across the studies were consistent. Firstly, our findings confirm that parents typically access the healthcare system after harboring their own concerns and living with the child's symptoms for months or years (Rowan-Legg & Canadian Paediatric Society, 2011). Although a seemingly minor point, clinicians may significantly underestimate the burden and/or magnitude of parents' concerns, particularly at first or early contact. Our findings align with previous studies indicating pediatric FC is often undertreated in primary care (Borowitz et al., 2005), and that delays in treatment may worsen the prognosis for long-term resolution (Bongers et al., 2010; Tabbers et al., 2014; van den Berg et al., 2005). It is important to establish the duration of symptoms and also to recognize the cumulative effects of FC on the child, parents, and family unit that may contribute to parents' reluctance to seek medication attention. For example, clinicians are trained to assess patients for alcohol use disorders with a multi-dimensional approach to decrease the risk of social desirability bias (such as underreporting intake) and to generate a more accurate understanding of harmful life effects (Spithoff & Kahan, 2015). Similarly, shame, embarrassment, and guilt are common barriers faced by parents when accessing healthcare for their child with FC. Healthcare providers may need to adjust their assessment approach to allow parents to openly discuss FC symptoms, including physical, psychological, social, financial, and family functioning effects. Parents' experiences of feeling dismissed created negative impressions of healthcare providers and hindered families' access to appropriate care. Therefore, clinicians may benefit from adopting a more attentive and supportive stance with families presenting with bowel symptoms to more effectively establish a therapeutic relationship (Rowan-Legg & Canadian Paediatric Society, 2011).

Secondly, our findings align with clinical recommendations that promote parental and family education as a mainstay of treatment for pediatric FC (National Collaborating Centre for Women's and Children's Health, 2010; Rowan-Legg & Canadian Paediatric Society, 2011; Tabbers et al., 2014). The intentional provision of support and education for families affected by pediatric FC should parallel the care and concern that is offered to parents receiving other pediatric chronic conditions diagnoses such as type 1 diabetes or asthma. Whether from friends, family, or healthcare providers, parents were often given incorrect reassurances the child would "grow out of it" with time. Our findings suggest that parents' knowledge of pediatric FC and treatment is limited. Although dietary changes such as high-fibre diets and adequate fluid intake are commonly discussed in conjunction with constipation, dietary interventions are known to be insufficient as primary treatment for pediatric FC (National Collaborating Centre for Women's and Children's Health, 2010; Rowan-Legg & Canadian Paediatric Society, 2011; Tabbers et al., 2014). Parents across the included studies typically felt dietary changes were integral and may overestimate the role of diet in the treatment of pediatric FC. This misconception may be supported by exposure to high- fibre messaging through media sources, family, friends, and even from healthcare providers. Specifically, a 2005 study of pediatric FC in primary care found that physicians recommended dietary changes in approximately half of cases (Borowitz et al., 2005). Furthermore, treatment success was positively correlated with more aggressive treatment recommendations (Borowitz et al., 2005). Therefore, healthcare providers may need to clarify dietary changes are more aptly considered a goal of nutritionally balanced eating, than as an active treatment of pediatric FC.

Additionally, it is likely that barriers such as misinformation about the condition and lack of emotional support contribute to low treatment adherence and a high proportion of persistent symptoms. Regardless of the time since diagnosis, parents should be offered detailed teaching about the pathophysiology of FC, with particular attention given to explain episodes of soiling (National Collaborating Centre for Women's and Children's Health, 2010; Rowan-Legg & Canadian Paediatric Society, 2011; Tabbers et al., 2014). Unfortunately, our findings suggest that parents typically did not receive adequate information about the condition, nor was their understanding of pediatric FC consistent over time. For example, the emotional burden of symptoms and caregiving may erode parents' knowledge; meaning reassurance and ongoing support are integral components of parental education. Specifically, behavioural interventions often precipitated parent-child conflict and quickly became exhausting for parents. Parents expressed feelings of being emotionally overwhelmed and felt they were lacking support rather than knowledge when implementing behavioural interventions. Thus, it may be extremely difficult for specialty care providers and busy primary care practice environments to adequately meet dynamic parental needs. Conversely, interdisciplinary care models that can accommodate more frequent visits and as-needed contact with healthcare providers during symptomatic exacerbations or periods of uncertainty have shown promise (Devitt et al., 2007; Ismail et al., 2011; Sullivan et al., 2006; Tappin et al., 2013). Based on our concerning findings about the negative effects of misconceptions and parental frustration on family relationships and child outcomes, finding innovative ways to meets parents' information and support needs is critically important to promote both parent and child well-being.

Similarly, our findings demonstrate that parents' understanding of medication use and safety is often an impediment to treatment adherence. For example, parents frequently had unanswered questions about whether long-term laxative use would cause dependence and were ill-prepared to manage side effects or to titrate doses to achieve optimum results. Clinicians routinely access a myriad of reliable medication information sources, but parents do not have the same access, nor are professional resources likely to be appropriate for parental audiences. Recent research suggests that parents may overestimate their comprehension of discharge instructions, with low health literacy and complex treatment plans further hindering parental understanding (Glick et al., 2020). Additionally, parents' decision support needs are more complex than simple provision of information (Jackson et al., 2008). Parental child health decision-making is supported by ensuring opportunities to talk with others and fostering parents' sense of control (Jackson et al., 2008), which may be improved through the use of shared-decision making approaches in clinical practice (Wyatt et al., 2015). For example, exploring barriers to medication use and explicit consideration of larger contextual influences (e.g. family dynamics, school or work timing, financial resources etc) may offer parents a greater sense of control and an improved ability to implement complex treatment plans.

Research Implications

Given the important disconnect that we have found between clinical care recommendations and parents' experiences caring for a child with FC, there are a number of areas that merit further exploration. It is evident that parents have unmet information needs and it would be helpful to have greater insight into parents' preferences for accessing resources (i.e. digital, in-person, pamphlet etc). For example, when do parents want to learn about pediatric FC and how should information be organized to reflect dynamic needs? In addition, understanding parents' emotional support needs is an important direction for examination. Specifically, what types of support are most and least helpful when parents encounter challenges? Lastly, healthcare providers play a critical role in educating and supporting families when caring for a child with FC. Our findings indicate clinical encounters can be a source of misinformation and exacerbate parental feelings of guilt. Further examination of the clinical environment is likely necessary to understand how to improve care. For example, providing comprehensive education can be time consuming and system constraints such as billing models may be a barrier to providing optimal care to families. Therefore, research is also needed to identify what systems, supports, and resources healthcare providers need to optimize care when working with families affected by pediatric FC.

Limitations

Although our findings highlight key areas where clinical care can shift to better meet parents' information and support needs when caring for a child with FC, the small number of included studies is an important limitation. The small number of studies and the generally small sample sizes within the studies mean the results may not fully reflect the variability of parents' experiences or capture less common occurrences.

Conclusion

Our systematic review provides a comprehensive exploration of parents' experiences caring for a child with FC to highlight current gaps in knowledge and care. Accurate information and misconceptions typically coexisted for parents, while the burden of caregiving fuelled doubts, misinformation, relationship breakdown, and treatment deviation. Parents often lacked a clear understanding of the physiology of the condition. For example, parents may misunderstand overflow fecal incontinence as an intentional behaviour. Inaccurate information and lack of support were detrimental to treatment plans, family dynamics, and parent-provider relationships. In addition, healthcare providers may underestimate and minimize parents' experiences, which further erodes access to reliable resources and supports. Results from our systematic review can be used by clinicians, researchers, and policy-makers to address gaps in clinical encounters, the research evidence, and systems that support families affected by pediatric FC. Our findings lie in contrast to the current clinical recommendations for pediatric FC, meaning both parents' and healthcare providers may benefit from resources or interventions to improve care.

Abbreviations

FC: Functional Constipation

KT: Knowledge Translation

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Authors' contributions

SS and AT conceived and led the design of the study. SS, SM, and EW provided substantial input into the study design. AT developed the search strategy in collaboration with a research librarian. AT and AC screened, appraised and extracted the studies. AT prepared the initial draft of the manuscript, which SS, SM, EW and AC revised. All authors read and approved the final manuscript. SS obtained the research funds through which this research was conducted.

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Competing Interests

The authors declare they have no competing interests.

Ethics approval and consent to participate

Not applicable. Ethical review and approval were not required for this study. All data sources are from published articles and no human participants were included.

Availability of data and materials

The data analyzed during the current review are from published studies and available through the following electronic databases: Ovid MEDLINE® and Epub Ahead of Print, In-Process & Other Non-Indexed Citations and Daily, Ovid Embase, Ovid PsycINFO, EBSCO CINAHL Plus with Full-text, Wiley Cochrane Library, ProQuest Dissertations & Theses Global, Scopus, Web of Science Core Collection. Complete search strategies are included as Supplementary file 3.

References

Banaszkiewicz, A., Topczewska-Cabanek, A., Malewicz, K., & Szajewska, H. (2005). Awareness of high-fiber products by parents of children with constipation: Questionnaire study. *Pediatria Wspolczesna*, *7*(1), 33–35.

Benninga, M. A., Nurko, S., Faure, C., Hyman, P. E., St. James Roberts, I., & Schechter, N. L. (2016). Childhood functional gastrointestinal disorders: Neonate/toddler. *Gastroenterology*, *150*(6), 1443-1455.e2. https://doi.org/10.1053/j.gastro.2016.02.016

Bernardbonnin, A. C., Haley, N., Belanger, S., & Nadeau, D. (1993). Parental and patient perceptions about encopresis and its treatment. *Journal of Developmental and Behavioral Pediatrics*, *14*(6), 397–400.

Bongers, M. E. J., van Dijk, M., Benninga, M. A., & Grootenhuis, M. A. (2009). Health related quality of life in children with constipation-associated fecal incontinence. *The Journal of Pediatrics*, *154*(5), 749-753.e1. https://doi.org/10.1016/j.jpeds.2008.11.029

Bongers, M. E. J., van Wijk, M. P., Reitsma, J. B., & Benninga, M. A. (2010). Long-term prognosis for childhood constipation: Clinical outcomes in adulthood. *Pediatrics*, *126*(1), e156–e162. https://doi.org/10.1542/peds.2009-1009

Borowitz, S. M., Cox, D. J., Kovatchev, B., Ritterband, L. M., Sheen, J., & Sutphen, J. (2005). Treatment of childhood constipation by primary care physicians: Efficacy and predictors of outcome. *Pediatrics*, *115*(4), 873–877. https://doi.org/10.1542/peds.2004-0537

Brazzelli, M., Griffiths, P. V., Cody, J. D., & Tappin, D. (2011). Behavioural and cognitive interventions with or without other treatments for the management of faecal incontinence in children. *Cochrane Database of Systematic Reviews*, *12*. https://doi.org/10.1002/14651858.CD002240.pub4

Brennan-Parsons, S. (2000). *Mothers' experiences in living with a child with encopresis- A phenomenological study.pdf*. Memorial University of Newfoundland.

Centre for Reviews and Dissemination. (2008). *Systematic reviews: CRD's guidance for undertaking reviews in health care*. University of York.

Devitt, P., Thornley, E., & Hinks, M. (2007). An evaluation of an inter-disciplinary constipation clinic for childhood constipation. *Journal of Research in Nursing*, *12*(5), 539–547. https://doi.org/10.1177/1744987107079589

Dolgun, E., Yavuz, M., Çelik, A., & Ergün, M. O. (2013). The effects of constipation on the quality of life of children and mothers. *The Turkish Journal of Pediatrics*, 7.

Farrell, M., Holmes, G., Coldicutt, P., & Peak, M. (2003). Management of childhood constipation: Parents' experiences. *Journal of Advanced Nursing*, *44*(5), 479–489. https://doi.org/10.1046/j.0309-2402.2003.02831.x

Glick, A. F., Farkas, J. S., Rosenberg, R. E., Mendelsohn, A. L., Tomopoulos, S., Fierman, A. H., Dreyer, B. P., Migotsky, M., Melgar, J., & Yin, H. S. (2020). Accuracy of parent perception of comprehension of discharge instructions: Role of plan complexity and health literacy. *Academic Pediatrics*, *20*(4), 516–523. https://doi.org/10.1016/j.acap.2020.01.002

Gordon, M., MacDonald, J. K., Parker, C. E., Akobeng, A. K., & Thomas, A. G. (2016). Osmotic and stimulant laxatives for the management of childhood constipation. *Cochrane Database of Systematic Reviews*, *8*. https://doi.org/10.1002/14651858.CD009118.pub3

Holman, K. S. (2012). *Parent and child functioning in a community sample of children with constipation*. University of Wisconsin - Milwaukee.

Hong, Q. N., Pluye, P., Bartlett, G., Boardman, F., Cargo, M., Dagenais, P., Gagnon, M.-P., Griffiths, F., Nicolau, B., O'Cathain, A., Rousseau, M.-C., & Vedel, I. (2018). *Mixed Methods Appraisal Tool (MMAT), version 2018* (Registration of Copyright #1148552). Canadian Intellectual Property Office, Industry Canada. http://mixedmethodsappraisaltoolpublic.pbworks.com/w/file/fetch/127916259/MMAT_2018_criteria-manual_2018-08-01_ENG.pdf

Hyams, J. S., Di Lorenzo, C., Saps, M., Shulman, R. J., Staiano, A., & van Tilburg, M. (2016). Childhood functional gastrointestinal disorders: Child/adolescent. *Gastroenterology*, *150*(6), 1456-1468.e2. https://doi.org/10.1053/j.gastro.2016.02.015

Ismail, N., Ratchford, I., Proudfoot, C., & Gibbs, J. (2011). Impact of a nurse-led clinic for chronic constipation in children. *Journal of Child Health Care*, *15*(3), 221–229. https://doi.org/10.1177/1367493511406568

Jackson, C., Cheater, F. M., & Reid, I. (2008). A systematic review of decision support needs of parents making child health decisions. *Health Expectations*, *11*(3), 232–251. https://doi.org/10.1111/j.1369-7625.2008.00496.x

Jurgens, H., Oster, C., & Fereday, J. (2011). Management of chronic functional constipation in children: A review of the literature. *Neonatal, Paediatric & Child Health Nursing*, *14*(2), 23–28.

Kaugars, A. S., Silverman, A., Kinservik, M., Heinze, S., Reinemann, L., Sander, M., Schneider, B., & Sood, M. (2010). Families' perspectives on the effect of constipation and fecal incontinence on quality of life. *Journal of Pediatric Gastroenterology and Nutrition*, *51*(6), 747–752. https://doi.org/10.1097/MPG.0b013e3181de0651

Klages, K. L., Berlin, K. S., Silverman, A. H., Mugie, S., Di Lorenzo, C., Nurko, S., Ponnambalam, A., Sanghavi, R., & Sood, M. R. (2016). Empirically derived patterns of pain, stooling, and incontinence and their relations to health-related quality of life among youth with chronic constipation. *Journal of Pediatric Psychology*, jsw068. https://doi.org/10.1093/jpepsy/jsw068

Koppen, I. J. N., van Wassenaer, E. A., Barendsen, R. W., Brand, P. L., & Benninga, M. A. (2018). Adherence to polyethylene glycol treatment in children with functional constipation is associated with parental illness perceptions, satisfaction with treatment, and perceived treatment convenience. *The Journal of Pediatrics*, *199*, 132-139.e1. https://doi.org/10.1016/j.jpeds.2018.03.066

Koppen, I. J. N., Vriesman, M. H., Saps, M., Rajindrajith, S., Shi, X., van Etten-Jamaludin, F. S., Di Lorenzo, C., Benninga, M. A., & Tabbers, M. M. (2018). Prevalence of functional defecation disorders in children: A systematic review and meta-analysis. *The Journal of Pediatrics*, *198*, 121-130.e6. https://doi.org/10.1016/j.jpeds.2018.02.029

Kovacic, K., Sood, M. R., Mugie, S., Di Lorenzo, C., Nurko, S., Heinz, N., Ponnambalam, A., Beesley, C., Sanghavi, R., & Silverman, A. H. (2015). A multicenter study on childhood constipation and fecal incontinence: Effects on quality of life. *The Journal of Pediatrics*, *166*(6), 1482-1487.e1. https://doi.org/10.1016/j.jpeds.2015.03.016

Liem, O., Harman, J., Benninga, M. A., Kelleher, K., Mousa, H., & Di Lorenzo, C. (2009). Health utilization and cost impact of childhood constipation in the United States. *The Journal of Pediatrics*, *154*(2), 258–262. https://doi.org/10.1016/j.jpeds.2008.07.060

Loening-Baucke, V. (2007). Prevalence rates for constipation and faecal and urinary incontinence. *Archives of Disease in Childhood*, *92*(6), 486–489. https://doi.org/10.1136/adc.2006.098335

Moher, D., Liberati, A., Tetzlaff, J., & Altman, D. G. (2009). Preferred reporting items for systematic reviews and meta-analyses: The PRISMA statement. *BMJ*, *339*, b2535. https://doi.org/10.1136/bmj.b2535

National Collaborating Centre for Women's and Children's Health. (2010). *Constipation in children and young people: Diagnosis and management of idiopathic childhood constipation in primary and secondary care*. RCOG Press. http://www.ncbi.nlm.nih.gov/books/NBK65365/

Pluye, P., Gagnon, M.-P., Griffiths, F., & Johnson-Lafleur, J. (2009). A scoring system for appraising mixed methods research, and concomitantly appraising qualitative, quantitative and mixed methods primary studies in mixed studies reviews. *International Journal of Nursing Studies*, *46*(4), 529–546. https://doi.org/10.1016/j.ijnurstu.2009.01.009

Popay, J., Roberts, H., Sowden, A., Petticrew, M., Arai, L., Rodgers, M., & Britten, N. (2006). *Guidance on the conduct of narrative synthesis in systematic reviews: A product from the ESRC methods programme* (Version 1).

Rowan-Legg, A. & Canadian Paediatric Society. (2011). Managing functional constipation in children. *Paediatrics & Child Health*, *16*(10), 661–665.

Samson, D. (2004). *Encopresis and family attitudes* [University of Leicester (United Kingdom)]. https://regroup-production.s3.amazonaws.com/documents/ReviewReference/100380230/Encopresis%20 and%20family%20attitudes.pdf?AWSAccessKeyId=AKIAJBZQODCMKJA4H7DA&Expires=1591192943&Signature=H3jSIlJ9T9KnFsaKatWFYqAVA98%3D

Spithoff, S., & Kahan, M. (2015). Primary care management of alcohol use disorder and at-risk drinking: Part 1: screening and assessment. *Canadian Family Physician*, *61*(6), 509–514.

Stacey, D., & Hill, S. (2013). Patient-direct and patient-mediated KT interventions. In *Knowledge translation in health care* (2nd ed., pp. 197–211). Wiley-Blackwell.

Sullivan, P. B., Burnett, C. A., & Juszczak, E. (2006). Parent satisfaction in a nurse led clinic compared with a paediatric gastroenterology clinic for the management of intractable, functional constipation. *Archives of Disease in Childhood*, *91*(6), 499–501. https://doi.org/10.1136/adc.2005.087486

Tabbers, M. M., DiLorenzo, C., Berger, M. Y., Faure, C., Langendam, M. W., Nurko, S., Staiano, A., Vandenplas, Y., & Benninga, M. A. (2014). Evaluation and treatment of functional constipation in infants and children: Evidence-based recommendations from ESPGHAN and NASPGHAN. *Journal of Pediatric Gastroenterology and Nutrition*, *58*(2), 258–274.

Tappin, D., Nawaz, S., McKay, C., MacLaren, L., Griffiths, P., & Mohammed, T. A. (2013). Development of an early nurse led intervention to treat children referred to secondary paediatric care with constipation with or without soiling. *BMC Pediatrics*, *13*(1), 1–9. https://doi.org/10.1186/1471-2431-13-193

Thompson, A. P., Nesari, M., Hartling, L., & Scott, S. D. (2019). Parents' experiences and information needs related to childhood fever: A systematic review. *Patient Education and Counseling*. https://doi.org/10.1016/j.pec.2019.10.004

Timmerman, M. E. W., Trzpis, M., & Broens, P. M. A. (2019). The problem of defecation disorders in children is underestimated and easily goes unrecognized: A cross-sectional study. *European Journal of Pediatrics*, *178*(1), 33–39. https://doi.org/10.1007/s00431-018-3243-6

van den Berg, M. M., Benninga, M. A., & Di Lorenzo, C. (2006). Epidemiology of childhood constipation: A systematic review. *The American Journal of Gastroenterology*, *101*(10), 2401–2409. https://doi.org/10.1111/j.1572-0241.2006.00771.x

van den Berg, M. M., van Rossum, C. H., de Lorijn, F., Reitsma, J. B., Di Lorenzo, C., & Benninga, M. A. (2005). Functional constipation in infants: A follow-up study. *The Journal of Pediatrics*, *147*(5), 700–704. https://doi.org/10.1016/j.jpeds.2005.05.032

van Tilburg, M. A. L., Squires, M., Blois-Martin, N., Williams, C., Benninga, M. A., Peeters, B., & Ulshen, M. (2012). Parental knowledge of fecal incontinence in children. *Journal of Pediatric Gastroenterology and Nutrition*, *55*(3), 283–287. https://doi.org/10.1097/MPG.0b013e31824cf555

Varni, J. W., Bendo, C. B., Nurko, S., Shulman, R. J., Self, M. M., Franciosi, J. P., Saps, M., & Pohl, J. F. (2015). Health-related quality of life in pediatric patients with functional and organic gastrointestinal diseases. *The Journal of Pediatrics*, *166*(1), 85-90.e2. https://doi.org/10.1016/j.jpeds.2014.08.022

Wang, C., Shang, L., Zhang, Y., Tian, J., Wang, B., Yang, X., Sun, L., Du, C., Jiang, X., & Xu, Y. (2013). Impact of functional constipation on health-related quality of life in preschool children and their families in Xi’an, China. *PLoS ONE*, *8*(10), e77273. https://doi.org/10.1371/journal.pone.0077273

Wyatt, K. D., List, B., Brinkman, W. B., Prutsky Lopez, G., Asi, N., Erwin, P., Wang, Z., Domecq Garces, J. P., Montori, V. M., & LeBlanc, A. (2015). Shared decision making in pediatrics: A systematic review and meta-analysis. *Academic Pediatrics*, *15*(6), 573–583. https://doi.org/10.1016/j.acap.2015.03.011

Youssef, N. N., Langseder, A. L., Verga, B. J., Mones, R. L., & Rosh, J. R. (2005). Chronic childhood constipation is associated with impaired quality of life: A case-controlled study. *Journal of Pediatric Gastroenterology and Nutrition*, *41*(1), 56–60.

Chapter 3. Paper 2: Understanding Parents' Experiences when Caring for a Child with Functional Constipation: Interpretive Description Study

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Abstract

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

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

Methods: This qualitative design used Interpretive Descriptive methodology to generate findings aimed towards improving clinical care. One-on-one, in-depth interviews were completed either in-person or via online teleconferencing to explore parents' perspectives. Data collection and analysis occurred concurrently.

Results: Analysis of sixteen interviews generated four major themes; 1) *living in the shadows*, 2) *not taken seriously* - with a subtheme of 2a) *persevering and advocating*, 3) *missing information and misinformation*, 4) *self-doubt and strained relationships*. One minor theme of *affirmative influences foster resilience and hope* was identified.

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

Keywords pediatric functional constipation; childhood constipation; parents' experiences; parents' information needs; qualitative research

Introduction

Constipation among children is common and often mistaken for a mundane nuisance rather than a serious medical condition. More than 95% of pediatric constipation cases are attributed to Functional Constipation (FC), which occurs without a particular medical, genetic, anatomic, or physiologic cause. Estimates are that at least 1 in 10 children worldwide are affected by pediatric FC (Koppen, Vriesman, Saps, et al., 2018; Timmerman et al., 2019). FC can present with severe symptoms, such as recurrent abdominal pain, painful defecation, fecal incontinence, urinary incontinence, and urinary infections. Pain, toilet avoidance, and stool withholding behaviours worsen the condition by further perpetuating fear of defecation, causing colonic dilation, and dampening neural feedback about the need to defecate. Despite being very common, pediatric FC is often underrecognized and undertreated (Borowitz et al., 2005). Without effective treatment, most children will develop chronic FC, with symptoms continuing through their adult years (Bongers et al., 2010). Additionally, children and families suffer from significant psychological, emotional, and social consequences of FC (Bongers et al., 2009; Varni, Bendo, et al., 2015; Youssef et al., 2005). For example, school attendance and peer relationships are understandably compromised by pain and incontinence. Families also report high levels of stress and decreased quality of life (Bongers et al., 2009; Varni, Bendo, et al., 2015; Youssef et al., 2005). Finally, pediatric FC is a financial burden on families and healthcare systems (Liem et al., 2009). Families face inflated expenses such as medications, laundry, and clothing, in addition to indirect effects such as lost income due to caregiving. Similarly, healthcare systems are burdened with preventable urgent care visits and high usage rates of specialist services (Jurgens et al., 2011; Liem et al., 2009).

Clinical practice guidelines (CPGs) describe a variety of treatment options (Brazzelli et al., 2011; Gordon et al., 2016; National Collaborating Centre for Women’s and Children’s

Health, 2010; Rowan-Legg & Canadian Paediatric Society, 2011; Tabbers et al., 2014); however, the bulk of responsibility for implementing, monitoring, and adjusting therapies falls to parents. Certainly, clinicians can provide parents with accurate information about the condition and treatments, but improving care also requires that healthcare professionals move beyond their own perspective of the condition and acknowledge the unique experiences of families living with a child affected by FC. Specifically, parental experiences critically shape their information and support needs (Thompson, Nesari, et al., 2020). Therefore, an in-depth understanding of parents' experiences and self-identified needs when caring for a child with FC is a necessary step to ensure clinicians can provide *relevant* education and support. Although parental education is an important part of treatment for pediatric FC (National Collaborating Centre for Women's and Children's Health, 2010; Rowan-Legg & Canadian Paediatric Society, 2011; Tabbers et al., 2014), there is a lack of research about parental perspectives of pediatric FC. A recent systematic review on the topic included only thirteen studies examining parents' experiences caring for a child with functional constipation (Thompson, Wine, et al., 2020). The primary cited limitation of the review was the small number of included studies (Thompson, Wine, et al., 2020). Furthermore, there was a predominance of quantitative studies focused on quality of life measures; which are helpful to substantiate the familial effects of childhood FC but are not optimal to understand how healthcare providers can help mitigate negative experiences and outcomes (Thompson, Wine, et al., 2020). Suggestions for future research included a more in-depth exploration of how to best meet parents' information and support needs in light of the dynamic nature of the condition and its profound effects on families (Thompson, Wine, et al., 2020). The initial aim of this study was to understand and give voice to parents' experiences and information needs when caring for a child with FC. The ultimate objective was to build an evidence base suitable for creating a digital Knowledge Translation (KT) tool to better support parents caring for a child with FC.

Methods

The study sought to answer the research question: “What are parents’ experiences and information needs when caring for a child with FC?” Because our ultimate objective was to develop knowledge that could be used to inform and improve clinical practice, we chose Interpretive Description (ID) methodology (Thorne, 2016) to foster applicability of our results. ID methodology was developed specifically for practice-oriented sciences, to generate findings aimed towards improving clinical care (Thorne, 2016), which aligns with our pragmatic philosophical approach for this research project.

Recruitment

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

Ethical Considerations

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

Data Collection Methods

We used one-on-one, in-depth interviews to explore parents' experiences caring for a child with FC. The interviews were completed either in-person or via online teleconferencing, depending on participant preference and geographic location. The interviewer (AT) had experience conducting qualitative interviews and also as a clinician providing care for children with FC. The interviewer did not have any pre-existing personal or professional relationships with the participants. The interviewer spoke with participants at the beginning of the interview to discuss the reasons for conducting this research (to understand parental experiences and subsequently develop resources for parents) and to share the interviewer's relevant clinical background; caring for families affected by pediatric FC and noting the challenges they often encountered managing the condition. The interview style was conversational, and participants were encouraged to discuss aspects of their experiences they deemed most important. The interviewer also used a semi-structured guide (multimedia appendix 1) with open-ended questions. Interview questions were developed based on previous research (Archibald et al., 2015; Meherali et al., 2019; Thompson, Le, et al., 2020) and clinical experience of the team. Prompts and spontaneous questions were used to facilitate participant comfort and collection of high-quality data. Interviews were recorded and transcribed verbatim by a professional transcriptionist. Data were de-identified (i.e. removal of identifying data such as city names, people names, institution names) to ensure confidentiality.

Sample

The sample included sixteen parents or caregivers of children with functional constipation who provided informed consent. Participants were included if their child met diagnostic criteria for pediatric FC (see multimedia appendix 2) and they were willing to discuss their experiences with the interviewer. Screening was conducted by the interviewer as a preamble to the interview to ensure participants' stories reflected experiences of childhood FC rather than other conditions. Since recruitment was most successful through online platforms, participants came from diverse geographical locations across North America.

Based on existing literature examining parental perspectives of pediatric FC and methodological recommendations, we anticipated a sample size between 10 and 20 participants would be adequate to generate clinically significant knowledge(Farrell et al., 2003; Thorne, 2016). The decision to end data collection was an ongoing topic of discussion within the research team and based on the processes of data analysis. Specifically, the occurrence of redundancy within the themes, and rich substantiation suggested that data collection could be stopped.

Data Analysis

We followed guidance from the applied methodology of Interpretive Description (Thorne, 2016) throughout data collection and analysis. We conducted data collection and analysis concurrently to promote data immersion as an important step to move towards a more thorough interpretation of experiences (Thorne, 2016). Interview transcripts were exported into NVIVO12 software to manage the data. Our analytic approach avoided quantification, instead using thematic and inductive traditions (Braun & Clarke, 2006; Vaismoradi et al., 2013). Our analysis followed the processes of engaging with the data, organizing the data, finding patterns within the data, making sense of the patterns, and finally developing patterns and associations into meaningful findings for applied practice (Thorne, 2016). The process was initiated by the interviewer/first author and then verified by the author team. Reflexive journaling and field notes were used during data collection and analysis to examine potential bias, build an audit trail, and support rigour.

Rigor

Developers of ID emphasize that the clinical expertise of researchers (Thorne, 2016) strengthens the design and rigor of the research (Thorne, 2016; Thorne et al., 1997); therefore, the experiences of clinicians on our research team was seen as an benefit. One member of the research team conducted all the interviews to maintain consistency. The interview guide was reviewed by topic experts and a parent advisory group to enhance credibility and ensure the questions could elicit meaningful information from participants. A study log was maintained during the research to document and account for methodological decisions. Data were analyzed and findings were collaboratively critiqued by the research team with the intent to develop epistemological integrity, representative credibility, analytic logic, and interpretive authority (Thorne, 2016) to ensure high-quality research. Following ID guidance, we did not conduct member-checking due to the risks of swaying interpretation and impeding the formation of meaningful clinical implications (Thorne, 2016; Thorne & Darbyshire, 2005). The manuscript follows the Standards for Reporting Qualitative Research (SRQR) (O'Brien et al., 2014) (see multimedia appendix 3).

Results

Our analysis generated four major themes; 1) *living in the shadows*, 2) *not taken seriously* - with a subtheme of 2a) *persevering and advocating*, 3) *missing information and misinformation*, 4) *self-doubt and strained relationships*. We identified one minor theme of *affirmative influences foster resilience and hope*. Demographic details of participants are presented in Table 1. All the particpants in this study self-identified as the caregiver with primary responsibiltiy for managing FC. One of the parents interviewed had more than one child with FC. Participant interviews were randomly assigned a numerical code which is used as a reference marker (e.g. P3) for quotes presented to support the themes in our results.

Table 3.1 Participant Characteristics

Characteristics		N
Preferred Gender Identity		
	Female	16
Number of Children		
	1	4
	2	8
	3	2
	4 or more	2
Affected Child's Age (years)		
	3	1
	4	4
	5	4
	6	5
	7	0
	8	0
	9 or older	2
Education Level		
	High School	1
	Post-Secondary	15
Yearly Family Income		
	< $20,000	1
	$20,000 - $40,000	1
	$40,000 - $60,000	4

	$60,000 - $80,000	2
	> $80,000	8
Duration of Symptoms		
	Less than 1yr	1
	1-2 yrs	3
	More than 2 yrs	12
Number of constipation-related healthcare visits (total)		
	0-5	4
	6-10	6
	More than 10	6

Living in the Shadows

Parents in our study expressed strong feelings of isolation attributed to living with a condition that is considered taboo. Discussing bowel habits and incontinence was thought to be a difficult or inappropriate topic in social circles and within the healthcare context. For example, when parent themselves were open to the conversation, most had experienced or anticipated negative reactions from others. One parent related her sense of isolation, "*Nobody talks about it…. So, you feel alone… And nobody wants to talk about poop"* (P3). Similarly, another parent explained, "*I think, that for myself… because I don't know a lot of other parents that are – I don't know if people just don't talk about it, so I don't know how common it is"* (P4). To combat feelings of isolation, parents had typically searched for resources without success to meet their social support needs. Parents were surprised about the lack of discussion groups because many described how it seems there is an online forum for almost every rare disease or condition. "*Something… so you're not alone, right. Because that's the thing and you don't understand why* *your kid is having so many problems. It's like somebody or something that explains like oh my kids have this issue, so you don't feel like you're the only one. ... Just something you can go to whether it's like a chat group or a parent group or something*" (P5). Another parent described how she would change things to improve other families' experiences with pediatric FC. "*You know, I think it's one of those things that people could really benefit from a support group because it's something that's so like people don't wanna talk about, they're embarrassed about it*" (P9). Another parent simply expressed, "*I just feel like we were very much left on our own*" (P14).

Not Taken Seriously

Parents shared stories of encounters with healthcare professionals who did not take their concerns about constipation seriously. In some cases, parents were explicitly told that the symptoms were nothing to be concerned about and other times parents were implicitly given the impression they were overreacting. One parent shared her care provider's dismissive response to her child's symptoms, "*I was always told it would pass, it would pass. Probably listen to the patient a little bit better because they know their body, right, and I – me living with her, I know what's going on with her. So, listen a little bit closer and maybe have better options than prune juice*." (P13). Similarly, another parent said, "*I wish I had been taken seriously right away. You know, not just like she'll grow out of it, she'll grow out of it. It's normal, she'll grow out of it. It's like this wasn't. I don't know if it ever was*" (P9). One shared the widespread effects of her child's FC and the trivializing response, "*I get that pediatricians are really busy with other things that are, you know, more important than constipation, but like now that he's in school, it's affecting his whole class. It's affecting his teacher. It's affecting him and his friends. Like it affects a lot of things and it affects us daily. It takes up our time as parents and his time away from his activities and the only real thing that we hear is, oh don't worry, it'll end soon. Like* *how*?(P14). One parent reflected on her desire for healthcare providers to change*, "I guess I wish they would learn – they would take it a bit more seriously and understand how it impacts lives and how it impacts – I mean children's lives"* (P7). In parallel to instances of healthcare providers not taking the condition seriously, parents' themselves described periods of questioning the legitimacy or validity of their own concerns. For example one parent shared, "*I think we could have maybe helped him a lot sooner if I wasn't so scared to start the Lax-A-Day but I also didn't want to make an appointment, take someone else's doctor time…I hate wasting doctors time on what I consider a silly thing… I know it's not the right way to think of it but like to my point, it had to be urgent enough*" (P3). Similarly, another parent said, "*you're like, oh is that normal or not normal and you kind of doubt yourself*" (P2).

Persevering and Advocating

As a result of symptoms and concerns not being taken seriously, parents demonstrated perseverance and became stronger advocates for their child's health. One parent described her feelings about healthcare encounters, "*I had talked to my doctor about it. Like our doctor and the doctor said like, oh you know, she's still really young. She'll grow out of it, all that kind of stuff…eventually after lots of kind of like advocating, I ended up – I was like I need another opinion on this"* (P9). Similarly, another parent stated, "*we found that we've gone to the doctor a couple of times now and they haven't been super helpful….and then we wound up back at the doctor because we're still – she's still having accidents*" (P16). Parents returned to healthcare providers repeatedly and asked for referrals to other providers because their child's condition was worsening without adequate treatment. For example, "*I'd asked many times for her to be seen by somebody else just because I need this figured out"* (P13). Parental frustration frequently became the catalyst for advocacy. One parent expressed, "*They don't take it serious enough… it would just be nice if there was a doctor that would take you a little more serious. I* *know lots of kids have it and I get that, but when they get to be older and it's a school issue, I think like we push. I think we asked – my doctor was out of town so we asked the stand in and then we asked the walk-in clinic and then we asked my doctor*" (P5).

Missing Information and Misinformation

Parents caring for a child with FC frequently has unanswered questions about the condition, causes, symptoms, prognosis, and treatment. One parent said, "*maybe I wouldn't have been so upset about it or, you know, it wouldn't have been such an overly concern for me if I'd had a little bit more information*" (P2). Similarly, another parent explained the lack of teaching provided about pediatric FC. "*I'm saying like you go into the doctor and you're like this is an issue and they don't give you…like there's nothing, they give you nothing. My doctor was just very much like, oh it's super common and.. like not giving you any further advice or resources*" (P6). Parents frequently questioned if there was an underlying medical cause for the constipation. For example, one parent stated, "M*aybe something else medically. Like maybe she's lactose intolerant – we thought well maybe there's some issues with milk or dairy which, of course, would not be constipation… but we were convinced it was something she was eating. Maybe it was gluten, maybe it was this, maybe it was that*" (P1). Episodes of incontinence often caused parents to question the underlying reason. One parent wondered, "*I don't know if it's medical or constipation or is it just laziness?*" (P5). Similarly, another parent stated, "*we had no idea whether she actually like did she have control, did she not have control. Could she feel it, could she not feel it? Was she just ignoring it? Did she need to pay more attention? Like all of these huge question marks"* (P9). Questions about treatment for pediatric FC were also common. A parent shared concerns about medication use, "*You read the Lax-A-Day thing it says, "Adults only, blah, blah, blah. So, I'm like 'Are you sure?' Like it feels wrong…But then,* *again we're trying to cut back now on the Lax-A-Day because you can't be on Lax-A-Day forever, can he? Like I don't know*" (P3).

In addition to having questions about pediatric FC, parents shared instances of being provided misinformation that was detrimental to their child's care. As explored above in the theme of not being taken seriously, parents were often incorrectly told the condition would resolve on its own. One parent shared the common false reassurances she received, "*it was very much like, no, no, no, he's fine. And it's just constipation and he'll grow out of it and like I feel like everybody I talked to said, he'll grow out of it. He'll grow out of it. He'll grow out it. And now, two years later, he's not growing out of it"* (P14). Parents were also commonly given misinformation about dietary changes as treatment. "*We were just told to increase fibre, increase water, skip the junk food, but we eat all whole foods anyways*" (P4). Similarly, another parent shared, "The doctor said, it'll get better. You know, just make sure she's eating healthy, which she does, and it'll get better. It'll get better" (P16). Dietary misinformation was problematic because it was ineffective, difficult for families to manage, and delayed further treatment. "*The nurse said don't give her any dairy. And so, we were off dairy for a while and then we were off wheat for a while and it was just like a – none, none of that seemed to make much difference" (P9).* Similarly, another parent reported, "C*ut [cheese] out and try to increase the fruits, the vegetables, take away the bread. It was like a constant diet struggle*." (P3).

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

Self-Doubt and Strained Relationships

Perhaps the most resounding theme from parents' stories was the overarching sense of frustration that developed while caring for a child with FC. One parent shared the emotional fragility that pediatric FC has created for her as a parent. "*It's pretty terrible actually. Like I should know how to deal with this. I'm a nurse. Like I was a pediatric nurse. (crying). I should know and everything that I've tried didn't work and I didn't have any guidance or any help. Like I called the doctor, well it's you know, the pediatrician – it's six months to get into her, so I, you know. I'm just trying things on my own. I'm googling how do you deal with this and you know, information and none of it is working and it makes me feel like – I don't know. Like I should know how to do this, and I don't*" (P14). Self-doubt and conflict were strongly tied to the previous themes of living in the shadows, not being taken seriously, and missing information and misinformation. One parent clearly expressed the situation stating, "*it was just like extremely frustrating because I felt like I wasn't getting – I wasn't getting enough support or information from the medical – like the health professionals we were dealing with… Like it's so frustrating. I'm like if this is so common, why does no one have answers? – it's just so, so frustrating*" (P9). Symptoms and physiology of pediatric FC were further sources of emotional turmoil for parents. *"We are very frustrated and, again, the accidents, I don't know if it's because of this issue or because she's lazy or like because she's so constipated…it's the accidents that are driving us crazy*" (P5). Another parent explained, "*We'll tell him fifteen times to go to the bathroom and he won't and then he'll have an accident and you feel like – you just get to your boiling point sometimes and you don't want to yell and get angry, but sometimes you do"* (P14). Lastly, relationships frequently became strained as a result of pediatric FC. "*It impacts a whole family* *dynamic, you know. Like our world, it seems like I mean this might sound dramatic, but our world has literally revolved around her bathroom habits for the last three years"* (P16). Another parent expressed the strain related to behavioural interventions, "*Like it's always a fight to get her on the toilet"* (P6). Another parent stated, *"There's been lots of fights. Lots of fights. Lots of I hate yous"* (P10). Emotional burden related to pediatric FC also sparked conflict between parents and eroded parental self-efficacy. "*We're both feeling – neither one of us are confident in our parenting. So, we're frustrated, and we can argue about it, for sure… I really felt like a failure as a mom. (pause). I don't know and I still don't know what to do. I don't feel like we're making progress and I don't feel like I have the confidence to fix it. And then I feel like that kind of –permeates, I guess, into our whole situation. Like into everything. Like if I can't figure out constipation, how can I figure out big things?*" (P14).

Affirmative Influences Foster Resilience and Hope

Despite the predominantly despondent themes that were reflected in parents' stories, there were small but significant moments of affirmation that helped to bolster parents' confidence. This is a minor theme of our analysis because the occurrence of positive encounters and resources was unfortunately infrequent. After episodes of misinformation, accurate and understandable explanations of the condition and symptoms were critically important for parents. "*They explained the encopresis is like the fact that like you know, when she did get constipated, the accidents would just be like the new poop coming around the old stuff that's not coming out… it's just like your muscles are just weak because like they've been holding it for so long. Yeah, and I was just like – at first, it just kinda blew my mind and I'm like, why the hell has no one told me about this?"*(P9). Validation came from a variety of sources and was always highlighted as an important event within the caregiving experience. For example, one parent found support through the school system, "*And it was really just brushed off and it's still being* *brushed off until like finally – now that he's taking up so much time from his teacher, the principal has become involved and she has been our only real advocate and our only – like the principal of the school. Like she's not a health care provider. You know, like she's the only person that has really like tried to help at all"* (P14). Parents identified encounters that met their support and informational needs as turning points that rekindled hope and buoyed their confidence. Unfortunately, affirmative influences were meaningful but scarce in parents' experiences. Specifically, many parents did not relate any positive encounters or supports at all throughout their caregiving journey. One parent explained, "*I told them this has been an ongoing issue. This isn't getting any better. This isn't an issue we've had for six months. This is an issue we've had for over three years now*" (P16).

Discussion

Findings from our exploration of parents' experiences with pediatric FC parallel and expand upon results from previous research in the field. In a 2003 study, researchers examined parents' healthcare encounters related to childhood constipation and found similar themes of "dismissed and fobbed off, asserting the need for action, and validation and acknowledgment" (Farrell et al., 2003, p. 483). The continuity of these findings with ours suggests that parents' perceptions of encounters with healthcare providers related to pediatric FC have not improved significantly over the last 17 years. Despite widespread prevalence of the condition (Koppen, Vriesman, Saps, et al., 2018; Timmerman et al., 2019) and advances in understanding childhood FC, (Bongers et al., 2010; Borowitz et al., 2005) parents' concerns continue to be minimized and clinicians' treatment discussions lag behind or are incongruent with symptom severity. In other words, when healthcare providers acknowledge that pediatric FC requires treatment (which in itself may occur belatedly, if at all), the level of intervention is often inadequate for the advanced nature of symptoms described by parents.

Similar to exploring patient and family experiences, measuring quality of life is considered an way important to understand the effects of a health condition or treatment on "patients' lives, rather than just on their bodies" (Addington-Hall & Kalra, 2001, p. 1417). Numerous studies have highlighted the diminished quality of life of parents and families living with pediatric FC (Dolgun et al., 2013; Kaugars et al., 2010; Klages et al., 2016; Kovacic et al., 2015; Wang et al., 2013). For example, three studies found increased family conflict, impaired family functioning, and increased parental worry or stress were related to the presence of fecal incontinence (Kaugars et al., 2010; Klages et al., 2016; Kovacic et al., 2015). Furthermore, Wang and colleagues' found that caregivers of children with FC gave lower ratings of their daily activities and family relationships, in addition to reporting lower physical, emotional, social, cognitive, and communication scores compared to those of caregivers and families with healthy children (Wang et al., 2013). Although quality of life data provide a broad assessment of the effects of a health condition and are a central contribution to the field, qualitative methods are helpful to add important context by exploring why and how families are affected. In our study, parental perspectives provide insight into the significant physical, emotional, and psychological burden on caregivers. Parents' feelings of isolation and frustration were related to incontinence and further compounded by non-supportive interactions and misinformation. Parents' experiences of being told erroneously that pediatric FC would resolve, feeling blamed for the condition or lack of treatment success, and struggling to talk about the condition may help explain the widespread and profound impairments in quality of life for families affected by pediatric FC (Dolgun et al., 2013; Kaugars et al., 2010; Klages et al., 2016; Kovacic et al., 2015; Wang et al., 2013).

A 2019 study examining the prevalence of defecation disorders in children concluded that childhood constipation is likely underestimated by parents who may not consider symptoms sufficient to be labelled a medical condition (Timmerman et al., 2019). The findings seem in contrast to our data which found parents were more frequently dismissed by healthcare providers rather than dismissive of the child's symptoms. One potential explanation for this difference could be the relative disease severity of surveyed parents in the two studies. Specifically, the cross-sectional study included a random selection of parents from the general population and was therefore more likely to include parents with early or mild manifestations compared to parents included in our study whose children all met full diagnostic criteria for pediatric FC. The findings from our study offer a relevant counterpoint; meaning that while parents and families may underestimate early symptoms, once the magnitude of the condition becomes evident, healthcare providers may be more of a barrier to recognition and diagnosis than parents.

Clinical Implications

Our exploration of parents' experiences caring for a child with FC provide important insight towards improving clinical care for this difficult condition. CPGs, which are intended to support clinicians and optimize care, identify family education about pediatric FC as a key component of treatment (National Collaborating Centre for Women's and Children's Health, 2010; Rowan-Legg & Canadian Paediatric Society, 2011; Tabbers et al., 2014). Unfortunately, our results suggest this step is commonly missing in healthcare encounters and that some providers even contribute to misinformation. Since our data was focused on parental perspectives, we cannot report the reasons for CPG deviations. Given the time-consuming nature of consultations to provide emotional support and education it is possible that care providers may be tempted to defer, rush through, or simply struggle to fit these practices into already busy schedules. Based on parents' reluctance to initiate discussions about bowel concerns, it may be prudent for professionals to recognize that effects may be more severe and have persisted for a significant duration by the time these issues are brought to their attention. In contrast to the temptation to offer hasty reassurance, clinicians may need to reframe their thinking towards acknowledgment, education, and active treatment. For example, explaining that the condition is common can be a method of validating parents' concerns and mitigating parental feelings of guilt, but should not be conflated with suggesting the symptoms are normal or do not require treatment. Improving the quality of healthcare encounters may require education or interventions to improve responses and treatment knowledge of healthcare providers. Similar to findings from a previous study about medication adherence (Koppen, van Wassenaer, Barendsen, et al., 2018), parents commonly expressed a lack of information about medication use; therefore, discussions about dosing, duration of use, side-effects, and safety are likely to be well-received by parents. Lastly, clinicians should be attuned to inquiring about parental experiences of isolation and lack of social support during assessment and include these factors as part of treatment plans (National Collaborating Centre for Women's and Children's Health, 2010; Rowan-Legg & Canadian Paediatric Society, 2011; Tabbers et al., 2014). In addition to existing system constraints that disincentivize lengthy consultations, it is unlikely that specialty care providers or primary care clinicians alone can adequately meet complex parental needs. Consideration of alternative care models, such as integration of nursing and allied health members may be helpful to more accurately and consistently meet parents' support needs when caring for a child with FC (Houghton et al., 2016; Ismail et al., 2011; Sullivan et al., 2006).

Future Steps

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

Limitations

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

Conclusion

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

Abbreviations

FC: functional constipation

ID: Interpretive Description

KT: knowledge translation

Authors' contributions

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

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Conflicts of interest/Competing interests

The authors declare they have no conflicts or competing interests.

Ethics approval

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

Consent to participate

All participants provided informed consent.

Availability of data and material

The data that support the findings of this study are available on request from the corresponding author (SS). The data are not publicly available due to ethics restrictions that could compromise the privacy of the research participants.

References

Addington-Hall, J., & Kalra, L. (2001). Who should measure quality of life? *BMJ*, *322*(7299), 1417–1420. https://doi.org/10.1136/bmj.322.7299.1417

Archibald, M. M., Caine, V., Ali, S., Hartling, L., & Scott, S. D. (2015). What is left unsaid: An interpretive description of the information needs of parents of children with asthma. *Research in Nursing & Health*, *38*(1), 19–28. https://doi.org/10.1002/nur.21635

Bongers, M. E. J., van Dijk, M., Benninga, M. A., & Grootenhuis, M. A. (2009). Health related quality of life in children with constipation-associated fecal incontinence. *The Journal of Pediatrics*, *154*(5), 749-753.e1. https://doi.org/10.1016/j.jpeds.2008.11.029

Bongers, M. E. J., van Wijk, M. P., Reitsma, J. B., & Benninga, M. A. (2010). Long-term prognosis for childhood constipation: Clinical outcomes in adulthood. *Pediatrics*, *126*(1), e156–e162. https://doi.org/10.1542/peds.2009-1009

Borowitz, S. M., Cox, D. J., Kovatchev, B., Ritterband, L. M., Sheen, J., & Sutphen, J. (2005). Treatment of childhood constipation by primary care physicians: Efficacy and predictors of outcome. *Pediatrics*, *115*(4), 873–877. https://doi.org/10.1542/peds.2004-0537

Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology*, *3*(2), 77–101. https://doi.org/10.1191/1478088706qp063oa

Brazzelli, M., Griffiths, P. V., Cody, J. D., & Tappin, D. (2011). Behavioural and cognitive interventions with or without other treatments for the management of faecal incontinence in children. *Cochrane Database of Systematic Reviews*, *12*. https://doi.org/10.1002/14651858.CD002240.pub4

Dolgun, E., Yavuz, M., Çelik, A., & Ergün, M. O. (2013). The effects of constipation on the quality of life of children and mothers. *The Turkish Journal of Pediatrics*, 7.

Farrell, M., Holmes, G., Coldicutt, P., & Peak, M. (2003). Management of childhood constipation: Parents' experiences. *Journal of Advanced Nursing*, *44*(5), 479–489. https://doi.org/10.1046/j.0309-2402.2003.02831.x

Gordon, M., MacDonald, J. K., Parker, C. E., Akobeng, A. K., & Thomas, A. G. (2016). Osmotic and stimulant laxatives for the management of childhood constipation. *Cochrane Database of Systematic Reviews*, *8*. https://doi.org/10.1002/14651858.CD009118.pub3

Houghton, D., Horgan, L., & Boldy, D. (2016). An exploratory study of aspects related to current inpatient and nurse-led clinic management of idiopathic childhood constipation. *Collegian*, *23*(2), 151–158. https://doi.org/10.1016/j.colegn.2015.04.002

Ismail, N., Ratchford, I., Proudfoot, C., & Gibbs, J. (2011). Impact of a nurse-led clinic for chronic constipation in children. *Journal of Child Health Care*, *15*(3), 221–229. https://doi.org/10.1177/1367493511406568

Jurgens, H., Oster, C., & Fereday, J. (2011). Management of chronic functional constipation in children: A review of the literature. *Neonatal, Paediatric & Child Health Nursing*, *14*(2), 23–28.

Kaugars, A. S., Silverman, A., Kinservik, M., Heinze, S., Reinemann, L., Sander, M., Schneider, B., & Sood, M. (2010). Families' perspectives on the effect of constipation and fecal incontinence on quality of life. *Journal of Pediatric Gastroenterology and Nutrition*, *51*(6), 747–752. https://doi.org/10.1097/MPG.0b013e3181de0651

Klages, K. L., Berlin, K. S., Silverman, A. H., Mugie, S., Di Lorenzo, C., Nurko, S., Ponnambalam, A., Sanghavi, R., & Sood, M. R. (2016). Empirically derived patterns of pain, stooling, and incontinence and their relations to health-related quality of life among youth with chronic constipation. *Journal of Pediatric Psychology*, jsw068. https://doi.org/10.1093/jpepsy/jsw068

Koppen, I. J. N., van Wassenaer, E. A., Barendsen, R. W., Brand, P. L., & Benninga, M. A. (2018). Adherence to polyethylene glycol treatment in children with functional constipation is associated with parental illness perceptions, satisfaction with treatment, and perceived treatment convenience. *The Journal of Pediatrics*, *199*, 132-139.e1. https://doi.org/10.1016/j.jpeds.2018.03.066

Koppen, I. J. N., Vriesman, M. H., Saps, M., Rajindrajith, S., Shi, X., van Etten-Jamaludin, F. S., Di Lorenzo, C., Benninga, M. A., & Tabbers, M. M. (2018). Prevalence of functional defecation disorders in children: A systematic review and meta-analysis. *The Journal of Pediatrics*, *198*, 121-130.e6. https://doi.org/10.1016/j.jpeds.2018.02.029

Kovacic, K., Sood, M. R., Mugie, S., Di Lorenzo, C., Nurko, S., Heinz, N., Ponnambalam, A., Beesley, C., Sanghavi, R., & Silverman, A. H. (2015). A multicenter study on childhood constipation and fecal incontinence: Effects on quality of life. *The Journal of Pediatrics*, *166*(6), 1482-1487.e1. https://doi.org/10.1016/j.jpeds.2015.03.016

Liem, O., Harman, J., Benninga, M. A., Kelleher, K., Mousa, H., & Di Lorenzo, C. (2009). Health utilization and cost impact of childhood constipation in the United States. *The Journal of Pediatrics*, *154*(2), 258–262. https://doi.org/10.1016/j.jpeds.2008.07.060

Meherali, S., Campbell, A., Hartling, L., & Scott, S. (2019). Understanding parents' experiences and information needs on pediatric acute otitis media: A qualitative study. *Journal of Patient Experience*, *6*(1), 53–61. https://doi.org/10.1177/2374373518771362

National Collaborating Centre for Women's and Children's Health. (2010). *Constipation in children and young people: Diagnosis and management of idiopathic childhood constipation in primary and secondary care*. RCOG Press. http://www.ncbi.nlm.nih.gov/books/NBK65365/

O'Brien, B., Harris, I., Beckman, T., Reed, D., & Cook, D. (2014). Standards for reporting qualitative research: A synthesis of recommendations. *Academic Medicine*, *89*(9), 1245–1251. https://doi.org/10.1097/ACM.0000000000000388

Rowan-Legg, A. & Canadian Paediatric Society. (2011). Managing functional constipation in children. *Paediatrics & Child Health*, *16*(10), 661–665.

Sullivan, P. B., Burnett, C. A., & Juszczak, E. (2006). Parent satisfaction in a nurse led clinic compared with a paediatric gastroenterology clinic for the management of intractable, functional constipation. *Archives of Disease in Childhood*, *91*(6), 499–501. https://doi.org/10.1136/adc.2005.087486

Tabbers, M. M., DiLorenzo, C., Berger, M. Y., Faure, C., Langendam, M. W., Nurko, S., Staiano, A., Vandenplas, Y., & Benninga, M. A. (2014). Evaluation and treatment of functional constipation in infants and children: Evidence-based recommendations from ESPGHAN and NASPGHAN. *Journal of Pediatric Gastroenterology and Nutrition*, *58*(2), 258–274.

Thompson, A. P., Le, A., Hartling, L., & Scott, S. D. (2020). Fading confidence: A qualitative exploration of parents' experiences caring for a febrile child. *Journal of Clinical Nursing*, *29*(5–6), 964–973. https://doi.org/10.1111/jocn.15165

Thompson, A. P., Nesari, M., Hartling, L., & Scott, S. D. (2020). Parents' experiences and information needs related to childhood fever: A systematic review. *Patient Education and Counseling*, *103*(4), 750–763. https://doi.org/10.1016/j.pec.2019.10.004

Thompson, A. P., Wine, E., MacDonald, S. E., Campbell, A., & Scott, S. D. (2020). Parents' experiences and information needs while caring for a child with functional constipation: A systematic review. *Clinical Pediatrics*, *60*(3), 154–169. https://doi.org/10.1177/0009922820964457

Thorne, S. (2016). *Interpretive description: Qualitative research for applied practice* (2nd ed.). Routledge.

Thorne, S., & Darbyshire, P. (2005). Land mines in the field: A modest proposal for improving the craft of qualitative health research. *Qualitative Health Research*, *15*(8), 1105–1113. https://doi.org/10.1177/1049732305278502

Thorne, S., Reimer Kirkham, S., & MacDonald-Emes, J. (1997). Interpretive description: A noncategorical qualitative alternative for developing nursing knowledge. *Research in Nursing & Health*, *20*(2), 169–177. https://doi.org/10.1002/(SICI)1098-240X(199704)20:2<169::AID-NUR9>3.0.CO;2-I

Timmerman, M. E. W., Trzpis, M., & Broens, P. M. A. (2019). The problem of defecation disorders in children is underestimated and easily goes unrecognized: A cross-sectional study. *European Journal of Pediatrics*, *178*(1), 33–39. https://doi.org/10.1007/s00431-018-3243-6

Vaismoradi, M., Turunen, H., & Bondas, T. (2013). Content analysis and thematic analysis: Implications for conducting a qualitative descriptive study. *Nursing & Health Sciences*, *15*(3), 398–405. https://doi.org/10.1111/nhs.12048

Varni, J. W., Bendo, C. B., Nurko, S., Shulman, R. J., Self, M. M., Franciosi, J. P., Saps, M., & Pohl, J. F. (2015). Health-related quality of life in pediatric patients with functional and organic gastrointestinal diseases. *The Journal of Pediatrics*, *166*(1), 85-90.e2. https://doi.org/10.1016/j.jpeds.2014.08.022

Wang, C., Shang, L., Zhang, Y., Tian, J., Wang, B., Yang, X., Sun, L., Du, C., Jiang, X., & Xu, Y. (2013). Impact of functional constipation on health-related quality of life in preschool children and their families in Xi'an, China. *PLoS ONE*, *8*(10), e77273. https://doi.org/10.1371/journal.pone.0077273

Youssef, N. N., Langseder, A. L., Verga, B. J., Mones, R. L., & Rosh, J. R. (2005). Chronic childhood constipation is associated with impaired quality of life: A case-controlled study. *Journal of Pediatric Gastroenterology and Nutrition*, *41*(1), 56–60.

Multimedia Appendix 1: Interview Guide

DRAFT INTERVIEW GUIDE*

Project Title - Collaborating with Parents to Understand and Address Information Needs when Caring for a Child with Functional Constipation

1. Tell me about your experiences of having a child with constipation?
2. What did you understand about constipation when your child was first diagnosed?
    - What information were you provided with from health care providers when your child was first diagnosed? What, if anything, was helpful about this information?
    - When did they give you this information?
    - In what format did the health care providers give you this information? Written, verbal, demonstration, combination?
3. How would you have liked to receive this information?
4. How did this information influence your experience with your child's illness?
5. If your friend's child had constipation and she asked you for information, what would you teach her about it?
    - When would you give her this information?
6. Today, do you feel you have enough information about your child's constipation?
    - If no, what would you like more information on?
7. How have your learning needs changed over time (comparison between time of diagnosis and now?)
    - Better access to information?
    - Relationship with health care personnel?
8. How has your confidence to manage your child's illness changed over time? What has influenced your confidence level over time?
9. What was has been the hardest part of having a child with constipation? How has that changed over time?
10. What would you like health providers to know about your experiences of living with a child who has constipation?
11. What is important to you in terms of your child's health outcomes?
12. How do you "define" when your child has a good day? E.g. Number of stools per day? No complaints of pain? No soiling? Child attending school or activities? Parents being able to go to work?

Thank you for your thoughtful feedback to my questions. Do you have any questions or concerns?

Multimedia Appendix 2: ROME IV Diagnostic Criteria for Pediatric Functional Constipation

ROME IV Diagnostic Criteria for Functional Constipation in Infants & Toddlers

Must include 1 month of at least 2 or more of the following in infants up to 4 years of age:
2 or fewer defecations per week
History of excessive stool retention
History of painful or hard bowel movements
History of large diameter stools
Presence of a large fecal mass in the rectum
In toilet-trained children, the following addition criteria may be used:
At least 1 episode/week of incontinence after the acquisition of toilet skills
History of large diameter stools that may obstruct the toilet

Benninga MA, Nurko S, Faure C, Hyman PE, St. James Roberts I, Schechter NL. Childhood Functional Gastrointestinal Disorders: Neonate/Toddler. Gastroenterology. 2016;150:1443-1455.e2.

ROME IV Diagnostic Criteria for Functional Constipation in Children

Must include 2 or more of the following occurring at least once per week for a minimum of 1 month with insufficient criteria for a diagnosis of irritable bowel syndrome:
2 or fewer defecations in the toilet per week in a child of a developmental age of at least 4 years
At least 1 episode of fecal incontinence per week
History of retentive posturing or excessive volitional stool retention
History of painful or hard bowel movements

Presence of a large fecal mass in the rectum
History of large diameter stools that can obstruct the toilet
After appropriate evaluation, the symptoms cannot be fully explained by another medical condition

Hyams JS, Di Lorenzo C, Saps M, Shulman RJ, Staiano A, van Tilburg M. Childhood Functional Gastrointestinal Disorders: Child/Adolescent. Gastroenterology. 2016;150:1456-1468.e2.

Multimedia Appendix 3: Standards for Reporting Qualitative Research (SRQR) checklist

Standards for Reporting Qualitative Research (SRQR)*

http://www.equator-network.org/reporting-guidelines/srqr/

Title and abstract

	Page #*
Title - Concise description of the nature and topic of the study Identifying the study as qualitative or indicating the approach (e.g., ethnography, grounded theory) or data collection methods (e.g., interview, focus group) is recommended	1
Abstract - Summary of key elements of the study using the abstract format of the intended publication; typically includes background, purpose, methods, results, and conclusions	2

Introduction

Problem formulation - Description and significance of the problem/phenomenon studied; review of relevant theory and empirical work; problem statement	4-5
Purpose or research question - Purpose of the study and specific objectives or questions	5-6

Methods

Qualitative approach and research paradigm - Qualitative approach (e.g., ethnography, grounded theory, case study, phenomenology, narrative research) and guiding theory if appropriate; identifying the research paradigm (e.g., postpositivist, constructivist/ interpretivist) is also recommended; rationale**	6

Researcher characteristics and reflexivity - Researchers' characteristics that may influence the research, including personal attributes, qualifications/experience, relationship with participants, assumptions, and/or presuppositions; potential or actual interaction between researchers' characteristics and the research questions, approach, methods, results, and/or transferability	7
Context - Setting/site and salient contextual factors; rationale**	6,8
Sampling strategy - How and why research participants, documents, or events were selected; criteria for deciding when no further sampling was necessary (e.g., sampling saturation); rationale**	8
Ethical issues pertaining to human subjects - Documentation of approval by an appropriate ethics review board and participant consent, or explanation for lack thereof; other confidentiality and data security issues	6-7
Data collection methods - Types of data collected; details of data collection procedures including (as appropriate) start and stop dates of data collection and analysis, iterative process, triangulation of sources/methods, and modification of procedures in response to evolving study findings; rationale**	7-8
Data collection instruments and technologies - Description of instruments (e.g., interview guides, questionnaires) and devices (e.g., audio recorders) used for data collection; if/how the instrument(s) changed over the course of the study	7 & Multmedia Appendix 1
Units of study - Number and relevant characteristics of participants, documents, or events included in the study; level of participation (could be reported in results)	8
Data processing - Methods for processing data prior to and during analysis, including transcription, data entry, data management and security, verification of data integrity, data coding, and anonymization/de-identification of excerpts	8-9
Data analysis - Process by which inferences, themes, etc., were identified and developed, including the researchers involved in data analysis; usually references a specific paradigm or approach; rationale**	8-9
Techniques to enhance trustworthiness - Techniques to enhance trustworthiness and credibility of data analysis (e.g., member checking, audit trail, triangulation); rationale**	9

Results/findings

Synthesis and interpretation - Main findings (e.g., interpretations, inferences, and themes); might include development of a theory or model, or integration with prior research or theory	10-19
Links to empirical data - Evidence (e.g., quotes, field notes, text excerpts, photographs) to substantiate analytic findings	10-19

Discussion

Integration with prior work, implications, transferability, and contribution(s) to the field - Short summary of main findings; explanation of how findings and conclusions connect to, support, elaborate on, or challenge conclusions of earlier scholarship; discussion of scope of application/generalizability; identification of unique contribution(s) to scholarship in a discipline or field	19-23
Limitations - Trustworthiness and limitations of findings	24

Other

Conflicts of interest - Potential sources of influence or perceived influence on study conduct and conclusions; how these were managed	26
Funding - Sources of funding and other support; role of funders in data collection, interpretation, and reporting	25

*The authors created the SRQR by searching the literature to identify guidelines, reporting standards, and critical appraisal criteria for qualitative research; reviewing the reference lists of retrieved sources; and contacting experts to gain feedback. The SRQR aims to improve the transparency of all aspects of qualitative research by providing clear standards for reporting qualitative research.

**The rationale should briefly discuss the justification for choosing that theory, approach, method, or technique rather than other options available, the assumptions and limitations implicit in those choices, and how those choices influence study conclusions and transferability. As appropriate, the rationale for several items might be discussed together.

Reference:

O'Brien BC, Harris IB, Beckman TJ, Reed DA, Cook DA. **Standards for reporting qualitative research: a synthesis of recommendations.** *Academic Medicine*, Vol. 89, No. 9 / Sept 2014
DOI: 10.1097/ACM.0000000000000388

* Page numbers correspond to pagination in published manuscript

Chapter 4. Paper: 3 An Evaluation of Parents' Experiences of Patient Engagement in Research to Develop a Digital Knowledge Translation Tool: Protocol for a Multi-Method Study

Paper three has been published as: Thompson, A. P., MacDonald, S. E., Wine, E., & Scott, S. D. (2020). An evaluation of parents' experiences of patient engagement in research to develop a digital knowledge translation tool: Protocol for a multi-method study. *JMIR Research Protocols*, *9*(8), e19108. https://doi.org/10.2196/19108

Abstract

Background: Over the past ten years, there have been increasing calls for patient and public involvement in health-related research. The impetus for this shift is twofold; an ideological shift towards more equitable and less hierarchical methods of knowledge development, and an effort to increase the usability and relevance of knowledge as evidenced by improved outcomes in clinical practice. Patient engagement includes a spectrum ranging from informing patients to giving complete decision-making autonomy; wherein, patient is an umbrella term that includes individuals, groups, communities, caregivers, friends, and families who have personal experience and knowledge of a health issue. Despite increased use of patient engagement methods in health research, evaluation has lagged, resulting in a meagre evidence base for the processes and outcomes of patient engagement in research. The current knowledge gap makes it difficult to foster capacity and sustainability for patients and researchers alike since little is known about how effective patient collaborations in research are built, maintained, or improved upon. The context of this study centers on pediatric functional constipation; a very common condition worldwide, which causes significant problems for children and families. Since parents play a pivotal role in the effective treatment of pediatric functional constipation, they are an optimal group to engage with to collaboratively improve the resources and support available for them.

Objectives: This study aims to: 1) use patient-engagement methods to establish a research collaboration with parents to co-create a digital Knowledge Translation (KT) tool for parents caring for a child with functional constipation (FC), 2) formally evaluate the patient engagement processes within this project to build the science of patient engagement in research.

Methods: Members of the Parent Collaborator Group (PCG) will be recruited from previous participants who expressed interest in digital Knowledge Translation (KT) tool development.

Target size of the PCG is between four and twelve members. The PCG will collaborate with the research team to co-create a digital KT tool to address patients' support and information needs when caring for a child with functional constipation. Upon tool completion, evaluation of the PCG will use a multi-method design with both quantitative and qualitative components. Data will be digitally and anonymously collected from all members of the PCG, using the validated Public and Patient Engagement Evaluation Tool (PPEET) – patient questionnaire. Descriptive statistics will be used to report group characteristics and question responses. Qualitative analysis will be used to understand open-ended question responses. Specifically, directed content analysis will be used in relation to the themes of the Patient Engagement in Research (PEIR) Framework with a combination of deductive and inductive analyses. Findings from the quantitative and qualitative data will be integrated in the discussion if there are sufficient commonalities and inter-relationships. The final manuscript will include reporting each element described by the Good Reporting of A Mixed Methods Study (GRAMMS) criteria.

Results: Recruitment for the PCG is planned for June 2020. Data collection for the evaluation of patient engagement processes will occur upon completion of the digital KT tool. Results of this study are expected to be published by the end of 2020.

Conclusions: This study will provide valuable information about parents' experiences participating in child-health research and is a fundamental step in building the science of patient engagement in research.

Keywords: patient engagement, patient-oriented research, knowledge translation, caregivers, parents

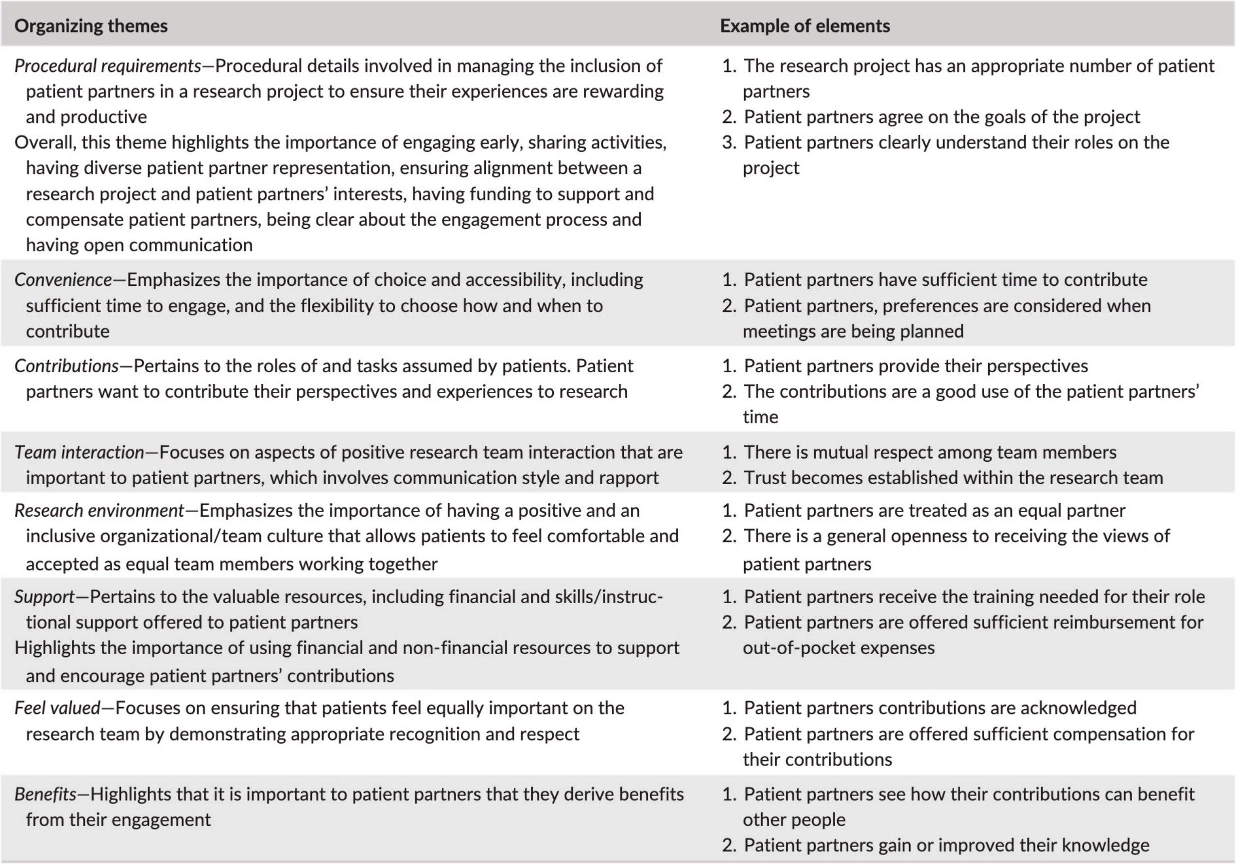
Introduction

Health research programs have historically been considered the exclusive domain of professional scientists. Whereas families' experiential knowledge and input in the clinical environment has been prioritized for many years, the research context has been slower to consider patients as contributors in knowledge development. Despite the intention to create clinically relevant knowledge, research programs have continued to develop knowledge in isolation from patient input. Consequently, patients and families have been at the centre of a paradox between the ideological positions of clinical practice and research (Ward et al., 2010), while questions about the usability and relevance of research findings to improve clinical care have persisted. Over the past ten years, there have been increasing calls for patient and public involvement in health-related research. The impetus for this shift is twofold; an ideological shift towards more equitable and less hierarchical methods of knowledge development (Canadian Institutes of Health Research, 2014; Domecq et al., 2014), and an effort to increase the usability and relevance of knowledge as evidenced by improved outcomes in clinical practice.

Although terminology varies around the world, in Canada, the terms *patient-oriented research* and *patient engagement* are commonly used in healthcare, aligning with guidance from the Canadian Institutes of Health Research. Patient engagement is defined as "meaningful and active collaboration in governance, priority setting, conducting research and knowledge translation" (Canadian Institutes of Health Research, 2014). Furthermore, the word patient is an umbrella term that includes individuals, groups, communities, caregivers, friends, and families who have personal experience and knowledge of a health issue (Canadian Institutes of Health Research, 2014). Although including patients and families as part of the research team is a fairly straightforward ideal, diversity in operationalization has slowed knowledge development related to effectiveness and best-practices of patient engagement (Esmail et al., 2015; Staniszewska et al., 2008; Supple et al., 2015). Similarly, evaluation of the processes and outcomes of patient engagement in research has lagged, resulting in a meagre evidence base for patient-oriented research (Brett et al., 2014b; Domecq et al., 2014; Esmail et al., 2015; Lavallee et al., 2012; Staley, 2015; Staniszewska & Denegri, 2013). The current lack of evidence regarding patient engagement in research makes it difficult to foster capacity and sustainability for patients and researchers alike, since little is known about how effective patient collaborations in research are built, maintained, or improved upon. Furthermore, parents are a unique subgroup of the patient engagement population that merits further exploration because of their dual roles; inherently representing both themselves as caregivers and their children as patients (Amirav et al., 2017; Curran et al., 2018; Pérez Jolles et al., 2017). Specifically, in this study we are engaging with parents caring for a child with functional constipation (FC). FC is a type of constipation that occurs without underlying medical or physiological causes. Prevalence rates amongst North American children are reported in the range of 9% - 18% (Koppen, Vriesman, Saps, et al., 2018) and these patients often have higher rates of emergency department visits and specialist care. Specifically, pediatric FC accounts for upwards of 25% of pediatric gastroenterology visits (Jurgens et al., 2011; Liem et al., 2009). Parents of children with FC are critical stakeholders in the successful management of pediatric FC because the treatment regime is ideally provided and monitored at home. As such, collaborating with parents of a child with FC offers an innovative approach to ensure clinicians can provide relevant support and parents have resources tailored to their needs. We are engaging with patients in pediatric FC research both to improve clinical care for families and to evaluate parents' experiences participating in child-health research, as a fundamental step in building the science of patient engagement in research. That is, the patient engagement process is widely applicable, meaning others can use this protocol to guide patient engagement processes and evaluation in any number of study populations.

There is a significant body of literature that is helpful to conceptualize and operationalize the elements of patient engagement within this study (Canadian Institutes of Health Research, 2014; Government of Canada, 2012; Hamilton et al., 2018; International Association for Public Participation Canada, n.d.; Lavallee et al., 2012). Patient engagement is often considered a spectrum ranging from informing stakeholders to giving stakeholders complete decision-making autonomy. The intention for patient engagement in this project aligns with the term collaboration; wherein a partnership is formed, decision-making is a shared responsibility between the researchers and the patient group, and is inclusive of their knowledge, experience, and preferences. The process goal for our patient engagement approach is based upon identified meta-criterion (Lavallee et al., 2012) of respect, trust, legitimacy, fairness, competence, and accountability in the development of knowledge. To operationalize this intent, we will use the Patient Engagement in Research (PEIR) framework (Hamilton et al., 2018) (see Figure 4.1) to guide the actions and strategies of our patient engagement approach. Whereas the meta-criterion help guide the goals of patient engagement, the PEIR framework highlights key themes that can be used as scaffolding for *how* to conduct meaningful patient engagement in research. Therefore, explicit planning and reporting of the patient engagement approach and activities within the project will be an important foundation of this study.

Figure 4.1 Organizing themes of the PEIR Framework with examples of corresponding elements (reprinted with permission from authors)



Organizing themes	Example of elements
Procedural requirements—Procedural details involved in managing the inclusion of patient partners in a research project to ensure their experiences are rewarding and productive Overall, this theme highlights the importance of engaging early, sharing activities, having diverse patient partner representation, ensuring alignment between a research project and patient partners' interests, having funding to support and compensate patient partners, being clear about the engagement process and having open communication	1. The research project has an appropriate number of patient partners 2. Patient partners agree on the goals of the project 3. Patient partners clearly understand their roles on the project
Convenience—Emphasizes the importance of choice and accessibility, including sufficient time to engage, and the flexibility to choose how and when to contribute	1. Patient partners have sufficient time to contribute 2. Patient partners, preferences are considered when meetings are being planned
Contributions—Pertains to the roles of and tasks assumed by patients. Patient partners want to contribute their perspectives and experiences to research	1. Patient partners provide their perspectives 2. The contributions are a good use of the patient partners' time
Team interaction—Focuses on aspects of positive research team interaction that are important to patient partners, which involves communication style and rapport	1. There is mutual respect among team members 2. Trust becomes established within the research team
Research environment—Emphasizes the importance of having a positive and an inclusive organizational/team culture that allows patients to feel comfortable and accepted as equal team members working together	1. Patient partners are treated as an equal partner 2. There is a general openness to receiving the views of patient partners
Support—Pertains to the valuable resources, including financial and skills/instructional support offered to patient partners Highlights the importance of using financial and non-financial resources to support and encourage patient partners' contributions	1. Patient partners receive the training needed for their role 2. Patient partners are offered sufficient reimbursement for out-of-pocket expenses
Feel valued—Focuses on ensuring that patients feel equally important on the research team by demonstrating appropriate recognition and respect	1. Patient partners contributions are acknowledged 2. Patient partners are offered sufficient compensation for their contributions
Benefits—Highlights that it is important to patient partners that they derive benefits from their engagement	1. Patient partners see how their contributions can benefit other people 2. Patient partners gain or improved their knowledge

Hamilton, C. B., Hoens, A. M., Backman, C. L., McKinnon, A. M., McQuitty, S., English, K., & Li, L. C. (2018). An empirically based conceptual framework for fostering meaningful patient engagement in research. *Health Expectations*, *21*(1), 396–406. https://doi.org/10.1111/hex.12635

The purpose of this study is to: 1) use patient-engagement methods to establish a research collaboration with parents to co-create a digital Knowledge Translation (KT) tool for parents caring for a child with FC, 2) formally evaluate the patient engagement processes within this project to build the science of patient engagement in research.

Methods

Patient Engagement

This study forms part of a multi-stage research project to improve care and resources for families living with pediatric functional constipation (FC) (see diagnostic criteria Appendix 1) (Benninga et al., 2016; Hyams et al., 2016). The preceding stage of qualitative, Interpretive Descriptive (Thorne, 2016) research frames this proposed patient-engagement phase and will be the primary recruitment source of our collaborators. The purpose of the qualitative research stage was to develop an in-depth understanding of parents' experiences and information needs when caring for a child with FC. Recruitment was through community and social media information posts, shared in the summer and fall of 2019. Interested parents contacted the research team for further details. We recognize that parents who volunteer for such research are unlikely to reflect the general population and we will explicitly cite this limitation in our findings. After sharing the information letter and discussing any questions, 18 parents consented and participated in semi-structured interviews. At the conclusion of the interview, parents were asked if they would like to allow the research team to keep their contact information and be notified about the subsequent stage; patient-engagement to co-create a digital KT tool.

Operationalization of patient engagement in this project is through the creation of a parent collaborator group (PCG) and is detailed as follows. A PCG will be formed by inviting all participants from the qualitative portion of the research to move forward in a new role as a member of the PCG. Through collaboration we will work together to establish priorities and co-create a digital KT tool for parents caring for a child with FC. This stage of the research fits within the *tailoring knowledge* portion of the Knowledge-to-Action (KTA) framework (Straus et al., 2013). The patient engagement process and activities described in this stage are meant to provide a framework rather than a rigid protocol because the PCG has not been formed and their contributions to shaping the research process are critical to uphold the legitimacy of parents' collaborative role in this stage.

We did not locate any evidence to support best practice about the optimal group size for patient engagement in research. Instead, we will build the PCG group based on practical considerations and recommendations of co-authors with extensive experience working with parent groups. Specifically, the size of the group should foster meaningful engagement. That is – we strive to develop a group that is large enough in size to be able to have discussion and everyone has the opportunity to share ideas. Conversely, we do not want a group so large that it is unmanageable. Lastly, we remain cognisant that these are parents with children, and they may not be able to attend every session, so we aim to have enough flexibility in our meetings to accommodate for all of these factors (ex. online synchronous and asynchronous access). We anticipate a reasonable target size of between four and twelve members will be sufficient to build meaningful engagement. Although the primary source of collaborators in this stage will be from the preceding qualitative stage of the project, additional parents who have experience with childhood FC will be welcomed to join the PCG as they become known to other members of the group (friends or community members known to be have a child with FC).

At our first meeting, detailed information about the study commitment will be provided verbally and in writing. Informed consent will be sought from interested participants. Members may revoke their consent to participate at any time. The first meeting will be facilitated by a registered professional (i.e. psychologist, social worker) with extensive group facilitation experience to establish group norms and support effective group process. Subsequent meetings will be co-facilitated by the researcher and parents. The aim of the project will be discussed including the following six key points. First parent participation is explicitly being sought to ensure this project will accurately address the challenges and improve the experiences of families living with pediatric FC. Second, parents will be supported to develop new skills if desired, but their experiential knowledge already qualifies them as valuable partners in this project. Third, parents will share decision-making responsibility with the researchers for the content, form, and style of the KT tool. Decision-making processes within the group will be documented and determined by the group. For example, the group may choose to use a modified Delphi technique (Hasson et al., 2000) or focus on robust discussion to generate consensus. Fourth, although individual input is desired, participation will also involve interacting with other parents affected by pediatric FC. Fifth, differing perspectives amongst group members are expected and considered beneficial because the aim is to advocate for the needs of the larger parent community as a whole. That is, participants need not aim for unanimous agreement on topics of discussion. Finally, the concepts of respect, trust, legitimacy, fairness, competence, and accountability will be our guideposts for the work of the PCG.

Expected activities and commitment of the PCG will also be discussed. The time commitment is based on previous experience of co-authors and is anticipated to be approximately one-hour meetings held every 3-4 weeks for a period of 1-3 months. This timeline is flexible and will be adapted based on the progress and needs of the PCG. Meeting locations will be central to parents, accessible by public transportation, and include childcare and light refreshments. The content of the digital KT tool will stem from two primary sources, 1) best practice guidelines and clinical recommendations for the management of pediatric FC, and 2) the themes and experiences generated from the qualitative inquiry of the preceding stage. The methods and process for developing the KT tool is based on existing literature(Archibald et al., 2018; Hartling et al., 2010; Reid et al., 2017; Scott et al., 2012) and previous experience with creating KT tools for parents. This research is situated within a larger program of research in a nationally funded knowledge mobilization network, Translating Emergency Knowledge for Kids (TREKK) (*Translating Emergency Knowledge for Kids*, n.d.), where a clinical team develops bottom line recommendations, developed by exploring practice guidelines and the best available synthesized research evidence. All bottom line recommendations are vetted through a large, clinical focused national committee in TREKK. The format of the KT tool will be determined by the PCG while building on the strengths of a narrative-based medium. For example, previously successful KT tools have been whiteboard videos and digital storybooks. A graphic designer and creative writer will be available to support the development of a high-quality digital KT tool. The design team of the writer and graphic designer will be provided with a story outline that reflects the combined experiences and most salient themes from the qualitative inquiry. The PCG will work with the design team to revise and build the KT tool through iterations to address questions of clarity, potential bias or marginalizing factors, ease of use, relevance and other factors as determined by the PCG. Upon completion of the KT tool, the final component of the project will be to evaluate the process of patient engagement in the project. Although not directly part of this stage of the research project, the KT tool (after completion) will be formally evaluated and tested for usability. The KT tool will also be made widely available on digital and social media platforms.

Evaluation

Design: The evaluation of the PCG will use a multi-method design with both quantitative and qualitative components. Multi-method design was chosen to answer two related but distinct research questions. First, the quantitative component will use the Public and Patient Engagement Evaluation Tool (PPEET) – participant questionnaire (Abelson et al., 2016; *Public & Patient Engagement | PPE Evaluation Tool*, n.d.) survey questions with Likert response options to examine the question, “To what degree did the patient engagement processes of the research meet the intended meta-criterion of respect, trust, legitimacy, fairness, competence, and accountability (Lavallee et al., 2012)?” The qualitative component will use open-ended questions to explore in more detail “Why or how did/didn’t the patient engagement processes of this research project meet the meta-criterion?” The rationale for use of both quantitative and qualitative methods in this stage of the research aligns with the purpose of *expansion or enhancement* by using an additional method to augment and further detail the findings (Bryman, 2006; Greene et al., 1989). Due to the focused nature of the evaluation and the small size of the PCG, both the quantitative and qualitative aspects of the study will be limited to descriptive methodologies.

Sample: All caregivers who participate in the PCG will be invited to participate in the evaluation phase. Parents who did not continue for the full duration of the project will also be included in the sample if they are willing. In addition, parents who were invited to participate in the group but declined will be asked if they are willing to share any feedback about what may have influenced their decision not to join the group.

Data Collection: Data collection will occur after the completion of the KT tool development. The PPEET – patient questionnaire (Abelson et al., 2016) will be copied into a digital format by entering the questions and response fields into the secure surveying platform SimpleSurvey. Parents will receive digital access to the questionnaire, which can be completed anonymously. Demographic questions which are considered indirect identifiers will be optional data fields. The survey instructions will include an explanation that if the demographic questions are answered, respondent’s data will remain confidential but may no longer be anonymous (to the researchers). The aim of the tool is to generate data in relation to the key features of the engagement approach and the participants perceptions of impact (Abelson et al., 2016). The PPEET includes 14 survey questions with five Likert-scale response options ranging from strongly agree to strongly disagree. In addition, the tool includes open-ended questions querying how the results may be used, the best aspect of the engagement and areas for improvement. Qualitative analysis will be used to understand the open-ended question portion of the PPEET to generate more in-depth data. In addition, documents from the PCG meetings such as agendas, minutes, and decision processes will be used as additional data sources to more comprehensively answer the research questions.

Analysis: The two types of data collected will be analyzed and reported separately. The findings from the quantitative and qualitative data will be integrated in the discussion if there are sufficient commonalities and inter-relationships.

Data from the Likert-scale questions will be entered into Statistical Package for Social Sciences (SPSS) version 25. Descriptive statistics will be used to report group characteristics and question responses, including mean, median, and/or the mode (as appropriate), and range (or interquartile-range, as appropriate) of scores. Frequency and percentages will be reported for categorical demographic information. No further analysis is planned because there is no comparative element of the design.

We will use directed content analysis (Hsieh & Shannon, 2005) to explore participant responses in relation to the themes of the PEIR Framework (Hamilton et al., 2018) using a combination of deductive and inductive analyses. Documents from PCG meetings (agendas, minutes, decision processes etc) will also be used as data sources for qualitative analysis. Data will be cleaned and transferred into NVIVO® version 11. Using the themes of the PEIR

Framework, all responses will be explored in relation to the codes: *procedural requirements, convenience, contributions, support, team interaction, research environment, feeling valued and benefits* (Hamilton et al., 2018). All text will be coded using the predetermined categories wherever possible. Text that cannot be coded into one of these categories will coded with another label that captures the meaning of the response. Finally, we will compare the extent to which the data are supportive of the PEIR framework versus how much represents different themes. To maintain a collaborative relationship of patient engagement, interested members of the PCG will also be invited to contribute to the analysis and dissemination of the evaluation findings. The manuscript produced from this stage of the research will include reporting each element described by the Good Reporting of A Mixed Methods Study (GRAMMS) criteria (O'Cathain et al., 2008).

Ethics: Approval from the appropriate University Health Research Ethics Board is complete for this project (#Pro00087548). Each participant will receive an information sheet that will provide details on the purpose of the study, identify the potential risks/benefits, and explain the voluntary nature of their participation. Participants may choose not to answer particular questions and can revoke consent at any time during the PCG. Evaluation data will be collected anonymously; therefore, individual participant data cannot be removed after it is collected. Data will be kept confidential with the exception of the duty to report any information relating to child welfare. Any information disclosed that falls under mandatory reporting laws (e.g. safety and well-being of a child) would be shared first with the disclosing participant. Eligible participants will receive a written consent form to be read and signed before partaking in the study. All data will be stored using secured software on a password protected server (LAN).

Data Management: Survey data will be collected on participants' computer or tablet devices through the surveying platform called SimpleSurvey®. SimpleSurvey® is a secure online platform with secure servers in Canada. It is protected by several firewalls and three physical layers of security. Data collected through this online platform is completely anonymous and cannot be traced back to any one individual. The data is stored on SimpleSurvey® servers until data collection for the specific survey/project is complete. Once data is downloaded onto University of Alberta servers, it will be deleted from SimpleSurvey® storage. Data will be stored on a secure drive, which is hosted by the University of Alberta, Faculty of Nursing secure server system. The server is backed-up twice a day. Files can be recovered if accidentally deleted/lost/corrupt. In case of a system-wide corruption, an external hard-drive is used to back up the data once a month. This hard-drive is kept in a locked area in a locked office.

Results

Recruitment for the PCG will begin in April of 2020. Once the PCG is formed, development of the digital KT tool for parents caring for a child with FC is expected to take 3-4 months. Data collection for the evaluation of patient engagement processes will occur upon completion of the digital KT tool and is expected to take 2-4 weeks to optimize the number of responses. Results of this study are expected to be published by the end of 2020.

Discussion

This study will include the development of a relevant and accessible digital KT tool created *with* and *for* parents caring for a child with FC. In addition, the findings will make an important contribution to fill the current evidence gaps about the processes of patient engagement in research. Our reported patient engagement processes are widely applicable, meaning others can use this protocol to guide patient engagement and evaluation in a variety of contexts. Specifically, the results can inform future research collaborations to ensure contributions by patient stakeholders are optimized, and challenges recognized and planned for accordingly. For example, avoiding tokenism, fostering inclusivity, and building capacity are knowledge gaps within patient engagement methods in research that may be better understood through widespread evaluations and dissemination. Results of this study can help build the science of patient engagement in research. Limitations of the study and findings will be discussed. Despite our planning and intentions, it is possible this study may face challenges such as a small sample size or significant attrition. We commit to full disclosure of the barriers encountered and the potential implications for the results. Given the emergent nature of PE evaluation, we suggest that studies with negative or limited findings are equally important to understand the barriers to further development of this field.

This study fits within the KTA framework (Straus et al., 2013) as a component of *tailoring knowledge* by creating a KT tool. Future projects related to this research will plan and examine integration of the KT Tool into the *action cycle* of the KTA framework (Straus et al., 2013). For example, assessing usability of the tool by a broader audience contributes to adapting the knowledge to the local context and can also help identify potential barriers to use. In addition to the creation of a digital, patient-direct KT tool, knowledge translation activities will be woven throughout this research. Specifically, the topic of FC aligns with priority areas of research identified by a national needs assessment of care providers; therefore, the foundation for this research stems from an existing relationship with clinical knowledge stakeholders. Use of a patient engagement approach in this research allows for explicit and ongoing inclusion of stakeholders; thus, integrating end-users of the knowledge into the development processes. Lastly, dissemination of the findings from this study will include tailored presentations to stakeholder groups and manuscript publication to target healthcare researchers.

Abbreviations

FC: Functional Constipation

KT: Knowledge Translation

KTA: Knowledge-to-Action

PCG: Parent Collaborator Group

PEIR: Patient Engagement in Research

PPEET: Public and Patient Engagement Evaluation Tool

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Ethics approval and consent to participate

University of Alberta Health Research Ethics Board approval #Pro00087548.

Competing Interests

The authors declare they have no competing interests.

References

Abelson, J., Li, K., Wilson, G., Shields, K., Schneider, C., & Boesveld, S. (2016). Supporting quality public and patient engagement in health system organizations: Development and usability testing of the Public and Patient Engagement Evaluation Tool. *Health Expectations*, *19*(4), 817–827. https://doi.org/10.1111/hex.12378

Amirav, I., Vandall-Walker, V., Rasiah, J., & Saunders, L. (2017). Patient and researcher engagement in health research: A parent's perspective. *Pediatrics*, *140*(3), e20164127. https://doi.org/10.1542/peds.2016-4127

Archibald, M. M., Hartling, L., Ali, S., Caine, V., & Scott, S. D. (2018). Developing "My Asthma Diary": A process exemplar of a patient-driven arts-based knowledge translation tool. *BMC Pediatrics*, *18*(1), 186. https://doi.org/10.1186/s12887-018-1155-2

Benninga, M. A., Nurko, S., Faure, C., Hyman, P. E., St. James Roberts, I., & Schechter, N. L. (2016). Childhood functional gastrointestinal disorders: Neonate/toddler. *Gastroenterology*, *150*(6), 1443-1455.e2. https://doi.org/10.1053/j.gastro.2016.02.016

Brett, J., Staniszewska, S., Mockford, C., Herron-Marx, S., Hughes, J., Tysall, C., & Suleman, R. (2014). A systematic review of the impact of patient and public involvement on service users, researchers and communities. *The Patient - Patient-Centered Outcomes Research*, *7*(4), 387–395. https://doi.org/10.1007/s40271-014-0065-0

Bryman, A. (2006). Integrating quantitative and qualitative research: How is it done? *Qualitative Research*, *6*(1), 97–113. https://doi.org/10.1177/1468794106058877

Canadian Institutes of Health Research. (2014). *Strategy for patient-oriented research—Patient engagement framework*. http://www.cihr-irsc.gc.ca/e/48413.html

Curran, J. A., Bishop, A., Chorney, J., MacEachern, L., & Mackay, R. (2018). Partnering with parents to advance child health research. *Healthcare Management Forum*, *31*(2), 45–50. https://doi.org/10.1177/0840470417744568

Domecq, J. P., Prutsky, G., Elraiyah, T., Wang, Z., Nabhan, M., Shippee, N., Brito, J. P., Boehmer, K., Hasan, R., Firwana, B., Erwin, P., Eton, D., Sloan, J., Montori, V., Asi, N., Abu Dabrh, A. M., & Murad, M. H. (2014). Patient engagement in research: A systematic review. *BMC Health Services Research*, *14*, 89. https://doi.org/10.1186/1472-6963-14-89

Esmail, L., Moore, E., & Rein, A. (2015). Evaluating patient and stakeholder engagement in research: Moving from theory to practice. *Journal of Comparative Effectiveness Research*, *4*(2), 133–145. https://doi.org/10.2217/cer.14.79

Government of Canada, C. I. of H. R. (2012, January 10). *Patient engagement—CIHR*. http://www.cihr-irsc.gc.ca/e/45851.html

Greene, J. C., Caracelli, V. J., & Graham, W. F. (1989). Toward a conceptual framework for mixed-method evaluation designs. *Educational Evaluation and Policy Analysis*, *11*(3), 255–274. https://doi.org/10.3102/01623737011003255

Hamilton, C. B., Hoens, A. M., Backman, C. L., McKinnon, A. M., McQuitty, S., English, K., & Li, L. C. (2018). An empirically based conceptual framework for fostering meaningful patient engagement in research. *Health Expectations*, *21*(1), 396–406. https://doi.org/10.1111/hex.12635

Hartling, L., Scott, S., Pandya, R., Johnson, D., Bishop, T., & Klassen, T. P. (2010). Storytelling as a communication tool for health consumers: Development of an intervention for parents of children with croup. *BMC Pediatrics*, *10*, 64. https://doi.org/10.1186/1471-2431-10-64

Hasson, F., Keeney, S., & McKenna, H. (2000). Research guidelines for the Delphi survey technique. *Journal of Advanced Nursing*, *32*(4), 1008–1015. https://doi.org/10.1046/j.1365-2648.2000.t01-1-01567.x

Hsieh, H.-F., & Shannon, S. E. (2005). Three approaches to qualitative content analysis. *Qualitative Health Research*, *15*(9), 1277–1288. https://doi.org/10.1177/1049732305276687

Hyams, J. S., Di Lorenzo, C., Saps, M., Shulman, R. J., Staiano, A., & van Tilburg, M. (2016). Childhood functional gastrointestinal disorders: Child/adolescent. *Gastroenterology*, *150*(6), 1456-1468.e2. https://doi.org/10.1053/j.gastro.2016.02.015

International Association for Public Participation Canada. (n.d.). *IAP2 Canada—Public participation spectrum*. Retrieved 12 April 2018, from http://iap2canada.ca/page-1020549

Jurgens, H., Oster, C., & Fereday, J. (2011). Management of chronic functional constipation in children: A review of the literature. *Neonatal, Paediatric & Child Health Nursing*, *14*(2), 23–28.

Koppen, I. J. N., Vriesman, M. H., Saps, M., Rajindrajith, S., Shi, X., van Etten-Jamaludin, F. S., Di Lorenzo, C., Benninga, M. A., & Tabbers, M. M. (2018). Prevalence of functional defecation disorders in children: A systematic review and meta-analysis. *The Journal of Pediatrics*, *198*, 121-130.e6. https://doi.org/10.1016/j.jpeds.2018.02.029

Lavallee, D. C., Williams, C. J., Tambor, E. S., & Deverka, P. A. (2012). Stakeholder engagement in comparative effectiveness research: How will we measure success? *Journal of Comparative Effectiveness Research*, *1*(5), 397–407.

Liem, O., Harman, J., Benninga, M. A., Kelleher, K., Mousa, H., & Di Lorenzo, C. (2009). Health utilization and cost impact of childhood constipation in the United States. *The Journal of Pediatrics*, *154*(2), 258–262. https://doi.org/10.1016/j.jpeds.2008.07.060

O'Cathain, A., Murphy, E., & Nicholl, J. (2008). The quality of mixed methods studies in health services research. *Journal of Health Services Research & Policy*, *13*(2), 92–98. https://doi.org/10.1258/jhsrp.2007.007074

Pérez Jolles, M., Martinez, M., Garcia, S. J., Stein, G. L., Mentor Parent Group Members, & Thomas, K. C. (2017). Involving Latina/o parents in patient-centered outcomes research: Contributions to research study design, implementation and outcomes. *Health Expectations*, *20*(5), 992–1000. https://doi.org/10.1111/hex.12540

Public & Patient Engagement | PPE Evaluation Tool. (n.d.). Retrieved 17 June 2020, from https://healthsci.mcmaster.ca/ppe/our-products/public-patient-engagement-evaluation-tool

Reid, K., Hartling, L., Ali, S., Le, A., Norris, A., & Scott, S. D. (2017). Development and usability evaluation of an art and narrative-based knowledge translation tool for parents with a child with pediatric chronic pain: Multi-method study. *Journal of Medical Internet Research*, *19*(12), e412. https://doi.org/10.2196/jmir.8877

Scott, S. D., Hartling, L., O'Leary, K. A., Archibald, M., & Klassen, T. P. (2012). Stories – a novel approach to transfer complex health information to parents: A qualitative study. *Arts & Health*, *4*(2), 162–173. https://doi.org/10.1080/17533015.2012.656203

Staley, K. (2015). 'Is it worth doing?' Measuring the impact of patient and public involvement in research. *Research Involvement and Engagement*, *1*, 6. https://doi.org/10.1186/s40900-015-0008-5

Staniszewska, S., & Denegri, S. (2013). Patient and public involvement in research: Future challenges. *Evidence-Based Nursing*, *16*(3), 69–69. https://doi.org/10.1136/eb-2013-101406

Staniszewska, S., Herron-Marx, S., & Mockford, C. (2008). Measuring the impact of patient and public involvement: The need for an evidence base. *International Journal for Quality in Health Care*, *20*(6), 373–374. https://doi.org/10.1093/intqhc/mzn044

Straus, S., Tetroe, J., & Graham, I. D. (Eds.). (2013). *Knowledge translation in health care: Moving from evidence to practice* (2 edition). BMJ Books.

Supple, D., Roberts, A., Hudson, V., Masefield, S., Fitch, N., Rahmen, M., Flood, B., de Boer, W., Powell, P., & Wagers, S. (2015). From tokenism to meaningful engagement: Best practices in patient involvement in an EU project. *Research Involvement and Engagement*, *1*, 5. https://doi.org/10.1186/s40900-015-0004-9

Thorne, S. (2016). *Interpretive description: Qualitative research for applied practice* (2nd ed.). Routledge.

Translating Emergency Knowledge for Kids. (n.d.). TREKK.Ca. Retrieved 17 June 2020, from https://trekk.ca/

Ward, P. R., Thompson, J., Barber, R., Armitage, C. J., Boote, J. D., Cooper, C. L., & Jones, G. L. (2010). Critical perspectives on ‘consumer involvement’ in health research: Epistemological dissonance and the know-do gap. *Journal of Sociology*, *46*(1), 63–82. https://doi.org/10.1177/1440783309351771

Chapter 5. Paper 4: Development and Usability of a Knowledge Translation Tool for Parents Managing Pediatric Functional Constipation

Paper four is being prepared for submission as: Thompson, A. P., Hartling, L., & Scott, S. D. Development and Usability of a Knowledge Translation Tool for Parents Managing Pediatric Functional Constipation. (Target journal: *Journal for Specialists in Pediatric Nursing,* April 2021).

Abstract

Pediatric functional constipation (FC) is a common condition with poor health outcomes for children and unmet needs for their parents. Providing parents with relevant, accessible, and clear resources to understand the condition and treatment is an important contribution to child health. This descriptive, multi-method research used patient engagement strategies to develop a knowledge translation tool for parents caring for a child with functional constipation. The resource was well-received and found to be highly usable amongst our sample of parents. Usability testing of the knowledge translation tool aligns with previous research findings and indicates that parents appreciate an easy-to-use resource and information that is easy to understand. Parents' perceptions of relevance may be enhanced by using a narrative-based format. Large scale evaluation of impact, including knowledge and user-experience outcomes, is planned to assess the effectiveness in clinical practice and to guide any necessary revisions of the KT tool.

Keywords: pediatric functional constipation, parents, knowledge translation, patient-engagement

Background

Pediatric Functional constipation (FC) is a widespread and debilitating medical condition (Koppen, Vriesman, Saps, et al., 2018; Varni, Nurko, et al., 2015). Children affected by FC and their families struggle with difficult symptoms and report decreased quality of life (Vriesman et al., 2019). Abdominal pain, distention and bloating, decreased appetite, decreased stool frequency, hard stools, painful or difficult defecation, fecal impaction, fecal incontinence, urinary incontinence and increased frequency of urinary tract infections are all common symptoms associated with the condition (van den Berg et al., 2006; Varni, Nurko, et al., 2015). In addition, families affected by pediatric FC frequently experience high levels of stress and conflict (Kaugars et al., 2010; Klages et al., 2016; Kovacic et al., 2015; Varni, Bendo, et al., 2015). Beyond the individual and family-level consequences of FC, financial implications include inflated costs related to increased emergency department visits, diagnostic testing, inpatient admissions, outpatient clinic visits, and medication use (Choung et al., 2011; Liem et al., 2009). For instance, FC accounts for an estimated 25% of pediatric gastroenterology visits (Jurgens et al., 2011) while mean costs of inpatient and outpatient care for children with FC are four times higher than for children without constipation (Choung et al., 2011). Unfortunately, long-term costs related to pediatric FC may be compounded by a low rate of treatment and resolution of symptoms. Specifically, evidence indicates pediatric FC is often undertreated and that up to 50% of children will still have symptoms after 5 years of treatment (Borowitz et al., 2005; Mugie, Di Lorenzo, et al., 2011; Sood et al., 2018; Yang & Punati, 2015). Although a variety of therapeutic options are available for the condition, too many children and families are continuing to suffer (Brazzelli et al., 2011; Gordon et al., 2016; Mousa et al., 2020; Southwell, 2020; van Engelenburg-van Lonkhuyzen et al., 2017). Therefore, interventions to improve health outcomes and to support families are imperative to decrease the burden of pediatric FC for children and the healthcare system.

Although there are a multitude of avenues to target for improvement, clinical practice guidelines for childhood FC highlight the importance of family education as a fundamental component of treatment (National Collaborating Centre for Women's and Children's Health, 2010; Rowan-Legg & Canadian Paediatric Society, 2011; Tabbers et al., 2014). Unfortunately, research indicates that parents struggle to understand and manage the condition (Thompson et al., 2021; Thompson, Wine, et al., 2020). Therefore, our team decided that focusing on supports to empower families with relevant and accessible information could make a meaningful contribution to the field. Knowledge translation (KT) initiatives are meant to decrease gaps between research and practice. Although many KT projects are focused on healthcare providers, connecting parents and families to research evidence has significant potential to improve outcomes (Stacey & Hill, 2013). For instance, patient-direct KT has the potential to increase knowledge, improve experiences, optimize health resource use, and encourage effective health behaviours (Stacey & Hill, 2013). Furthermore, research about parents' information and decision-making support needs suggests resources that integrate parental perspectives, can be accessed easily, are relevant, and foster a sense of self-efficacy are most likely to be effective (Jackson et al., 2008; Neill et al., 2015). Patient-direct and creative KT approaches are well-suited to pediatric practice and may be a promising strategy to improve care for children with FC (Hartling et al., 2013; Scott et al., 2012). Specifically, narrative-based methods such as stories have been highlighted for their ability to make information relatable in healthcare education initiatives (Archibald et al., 2018; Cunningham & Boom, 2013; Lee et al., 2018; Njeru et al., 2015). Therefore, the aim of this research was to first develop and second test the usability of a narrative-based, patient-direct KT tool for parents of a child with FC.

Theoretical Framework

This research used the Knowledge-to Action framework (Straus et al., 2013) to guide the methods and design. Our work mirrors the central elements of using knowledge synthesis to tailor knowledge towards the creation of products and tools in response to an identified problem. Specifically, we address the problem of parent's information and support needs when caring for a child with FC and follow the path of adapting knowledge to the local context by integrating patient engagement methods and finally assess strengths and weaknesses of the tool through usability testing.

Methods

We designed a multi-phase, multi-method research study to develop and assess usability of a patient-direct KT tool for parents of a child with FC. This study is also situated within a larger program of research using patient engagement approaches to develop resources for parents about a variety of common childhood conditions. The subsequent methods section is divided for clarity into subsections of 1) KT tool development and 2) Usability testing.

Ethical Considerations

The University of Alberta research ethics board granted approval (#Pro 00062904) prior to initiation of both the development and usability testing portions of the study. Each potential participant received an information sheet, which provided details on the purpose of the study, identified the potential risks/benefits, and explained the voluntary nature of their participation. Participants were given an opportunity to ask questions about the research and were free to withhold consent for any reason.

KT Tool Development

Development of the patient-direct KT tool for parents built on the findings of the completed research synthesis (Thompson, Wine, et al., 2020) and qualitative inquiry (Thompson et al., 2021), and followed the phases of 1) draft prototype creation, 2) clinician expert input, and 3) patient engagement with Pediatric Parent Advisory Group (P-PAG).

Sample and Setting

The P-PAG group was formed in 2016 with the purpose of sharing parental advice, guidance and knowledge to inform various health research activities (Hartling et al., 2021). Eligibility criteria of the group were: a parent, grandparent or legal guardian of a child (less than 18 years); wanting to contribute to child health research; willingness to work collaboratively with a group; and able to attend regular meetings in-person, by telephone, or online (Hartling et al., 2021). Participants were not required to have any condition specific knowledge or particular experiences with the healthcare system.

Recruitment

Parents and caregivers were invited to join the group with the purpose of contributing to child health research through activities such providing parental perspectives about KT tools, understanding the best ways to work with families in research, and identifying research priorities. The opportunity to participate was advertised in physical spaces related to child health (ie. clinical sites), on the University of Alberta campus, and via digital means (email, social media etc).

Usability Testing

Sample and Setting

We sought to include parents presenting with an ill child to the emergency department of the Stollery Children's Hospital in Edmonton, Canada. The Stollery Children's Hospital is a full-service pediatric hospital and is the only specialized healthcare facility for infants, children and youth in central and northern Alberta, serving a geographical area of over 500,000 (Alberta Health Services, 2021). We chose to assess usability of the resource amongst parents in the emergency department for two main reasons. Firstly, to mirror the heightened stress level and presence of numerous distractions such as those expected in the home environment when parents are seeking information about their child's health which typically occurs during a period of exacerbated symptoms or acuity. Second, the environment had well-established research support which fostered convenience sampling and fiscal responsibility by having a large pool of potential participants from which to recruit. Based on previous usability research, our target sample size was 25-30 parents (Archibald & Scott, 2019; Reid et al., 2017). Power calculation and effect size estimate were not required because the study purpose was descriptive rather than inferential.

Recruitment

Recruitment occurred in November and December, 2020. A research nurse from the Stollery Hospital approached parents in the waiting room and explained the study process. Interested parents were asked to complete a 5-10 minute survey about their perceptions of a digital educational tool. Parents were free to refuse or skip any questions, stop the survey at any time, or withdraw from the study. Digital submission of the survey was accepted as consent to participate.

Data Collection

Once a parent agreed to participate, they were given a study iPad and single-use disposable earbuds to view and listen to the KT tool prototype. After the video finished, the iPad presented a survey to assesses perceptions of the prototype on a 5-point Likert scale. Survey questions were informed by existing research including a systematic search of over 180 usability evaluations and reflected the main elements of 1) usability, 2) aesthetics, 3) language, 4) level of engagement, 5) quality of information, 6) length, 7) preference of form over traditional dissemination venues, and 8) value-added (Hornbæk, 2006; Zapata et al., 2015). The survey also included free-text boxes for parents to provide written feedback about the tool in general including areas that required revisions or more information.

Data Analysis

The data from the usability survey were cleaned and managed according to industry standards. Statistical Package for Social Sciences (SPSS, version 24) was used to generate descriptive statistics. Data in the free text boxes was explored using content analysis (Vaismoradi et al., 2013) to identify barriers and facilitators, and further inform our prototype revisions.

Results

Development Processes

The foundational phases of the project included a systematic review and an in-depth, qualitative exploration of parents experiences and information needs related to pediatric functional constipation, the findings of which are detailed in separate manuscripts (Thompson et al., 2021; Thompson, Wine, et al., 2020). Development of the KT tool began with the findings from the previous phases of the project. In addition, the use of evidence-based, bottom line recommendations (Eltorki et al., 2019) added important clinical context for developing the KT tool. A story-based format was used for the resource to foster audience engagement in the narrative and to facilitate learning (Archibald et al., 2018; Dahlstrom, 2014; Green & Brock, 2000; Jones, 2014; Lee et al., 2018). A draft composite narrative was created, based on the most common and poignant elements from the systematic review findings and of parents' stories from the sixteen qualitative interviews. The goals for the preliminary versions of the tool were to foster recognition of shared experience amongst the audience and integrate the best research evidence for management of FC. The concomitant and yet challenging aim was to keep the length of the narrative accessible (less than 5 minutes duration). The medium selected for the KT tool, narrated video, was determined based on experiences and findings from similar creative KT approaches to support parental education (Archibald & Scott, 2019; Reid et al., 2017). Our team worked with a creative design team, including an illustrator, narrator, graphic designer, and digital technologist to construct the digital video. The process was highly iterative with numerous rounds of revision and refinement enabled through collaboration between the research team and the creative design team.

Clinician Experts

Once a prototype of the digital video was created, we shared the product with a group of clinician experts including, pediatric gastroenterologists, nurses, and advanced practice nurses. We received valuable feedback from clinicians about language and portrayals that could have created misunderstandings. For example, a common diagnostic feature of pediatric FC includes "hard stools", however; clinicians expressed that parents and children frequently do not consider the consistency of stool as much as the difficulty of passing firm stools, so the language was changed to "hard to pass stools" which is intended to convey a similar but more understandable concept. Subsequent to the revisions suggested by clinician experts, the prototype was reviewed and adapted by a parental advisory group.

Pediatric Parent Advisory Group (P-PAG)

P-PAG members provided comprehensive feedback initially at the storyboard phase of tool development and subsequently at the prototype stage when the tool included illustrations and narration. Regularly scheduled meetings were initially held in-person with options for online and telephone participation for those unable to attend in-person. During the Covid-19 pandemic meetings were held online. Meetings were typically on a weekday evening and lasted about 1.5 hours. Reimbursement for parking and childcare was offered and snacks were provided at each meeting. Participants were offered a gift card (approximately Cdn$25) twice a year in recognition of their contributions. Membership in the P-PAG was dynamic and at the time of the group review of the pediatric FC KT tool, there were 16 – 19 who viewed the tool and were invited to share their perspectives. The group was asked to advise on all aspects of the tool and its use. Discussion amongst the P-PAG members generated more profound consideration of the development process by exploring the depth of agreement or dissent of particular aspects of the feedback. For example, an idea brought forward by one individual would typically be discussed in-depth by the group, including expressions of support or disagreement by others, which ultimately refined the feedback. Consensus within the group was not an aim of the process; however, only once (related to a preference for treatment with diet alone) was feedback expressed by an individual or small fraction of the group that was not eventually widely supported. The themes that arose from P-PAG meeting discussions were, 1) comprehensiveness of the content, 2) clarity of information and ease of use, and 3) aesthetics and audience response.

Comprehensiveness of the content

The P-PAG initially felt that more information about management of the condition including side effects and doses of medication and alternate therapies for FC were important additions for the content of the KT tool. Again, the primary challenge was to balance the desire to add more information and detail while maintaining a short duration for the video. Ultimately, the group agreed that attempting to address comprehensive management instructions would be overwhelming for parents and could inadvertently sway parents away from accessing appropriate healthcare resources and supports. There was a suggestion that in the future, creating a subsequent resource with a narrow, detailed focus on a particular aspect of childhood FC and its management may be helpful based on parent demand or if further information gaps are identified

Clarity of information and ease of use

Similar to the insights gained from expert clinicians, parents provided valuable advice about wording and images that could cause misunderstanding amongst parents. For example, in one iteration of the KT tool, there was text that parents felt implied FC did not require medical assessment. The intended message had been that FC typically does not require exhaustive diagnostic testing or assessment by specialists. The wording of the text was promptly revised in the subsequent version to clarify the information for parents. The P-PAG did not have concerns about ease or use or accessibility. They discussed that the format of combining static illustrations with text and complementary narration was exceptionally easy to use because no clicking or navigating is required by the user. Group members described the resource as being easily accessible for parents on a mobile device or computer. Being freely available online was also discussed as a benefit, meaning parents can find and use the resource independently which may help to bypass systemic barriers to accessing healthcare.

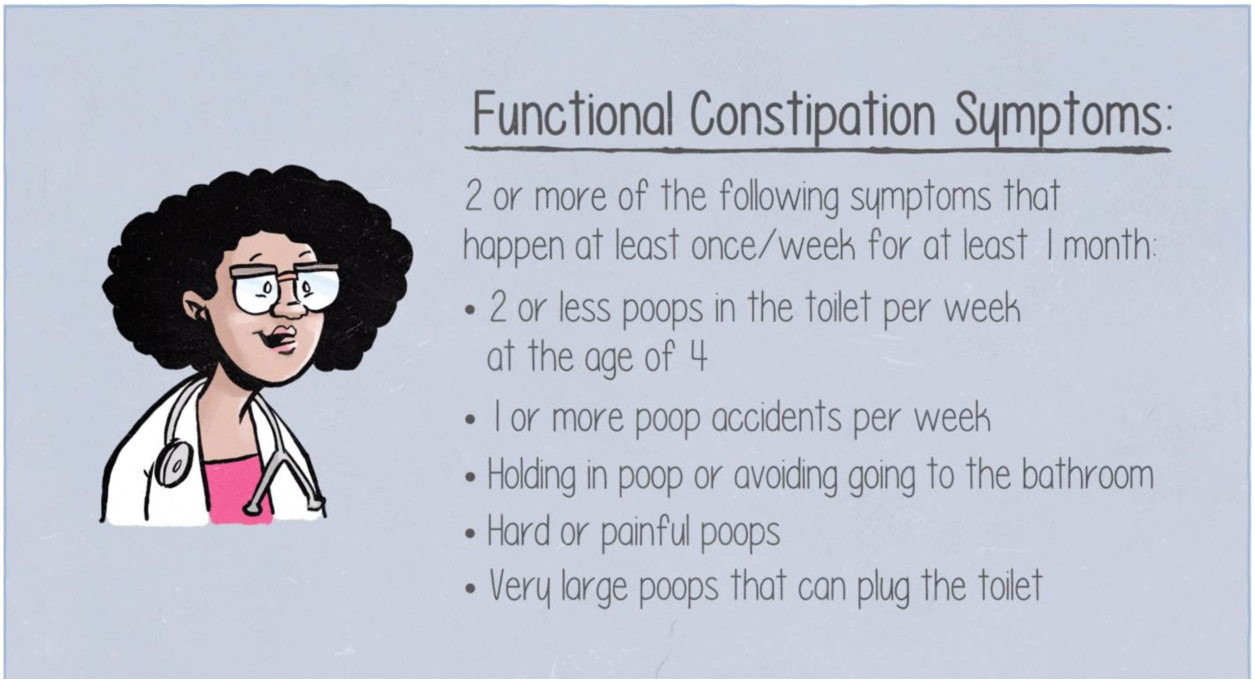
Aesthetics and audience response

P-PAG members agreed that inclusion of racially diverse characters in the illustrations was a positive feature of the KT tool and supported inclusivity amongst the audience. The group also highlighted the importance of beginning to acknowledge parents' emotional responses to pediatric FC. Specifically, the P-PAG wanted to ensure parents understand how common pediatric FC is, as a foundation for addressing information needs and to potentially reduce feelings of isolation or stigma. In addition, the group appreciated explicit statements recognizing parental frustration and providing reassurance that parents are not to the blame for the condition was helpful to foster a supportive theme of the KT tool.

KT Tool

The development processes resulted in the creation of an illustrated, narrated video prototype presenting the story of 5-year-old Ari and his parents, who struggle with pediatric FC. The video is approximately 5 minutes in duration and includes information about prevalence, typical age of onset, common symptoms, what to expect during assessment, physiological cycle of worsening symptoms, how soiling accidents occur, treatment, and further resources. The information is presented in the form of a story, in order to promote engagement with the information and emotional validation through recognition and empathy of the audience with the challenges faced by the characters.   Images from the video are included as Figures 5.1 - 5.6 The KT tool will be freely and widely accessible for both healthcare professionals and the public on www.trekk.ca which is a nationally established platform that hosts other parent tools for common childhood conditions.

Figure 5.1 KT tool symptoms



Functional Constipation Symptoms:

2 or more of the following symptoms that happen at least once/week for at least 1 month:

- 2 or less poops in the toilet per week at the age of 4
- 1 or more poop accidents per week
- Holding in poop or avoiding going to the bathroom
- Hard or painful poops
- Very large poops that can plug the toilet

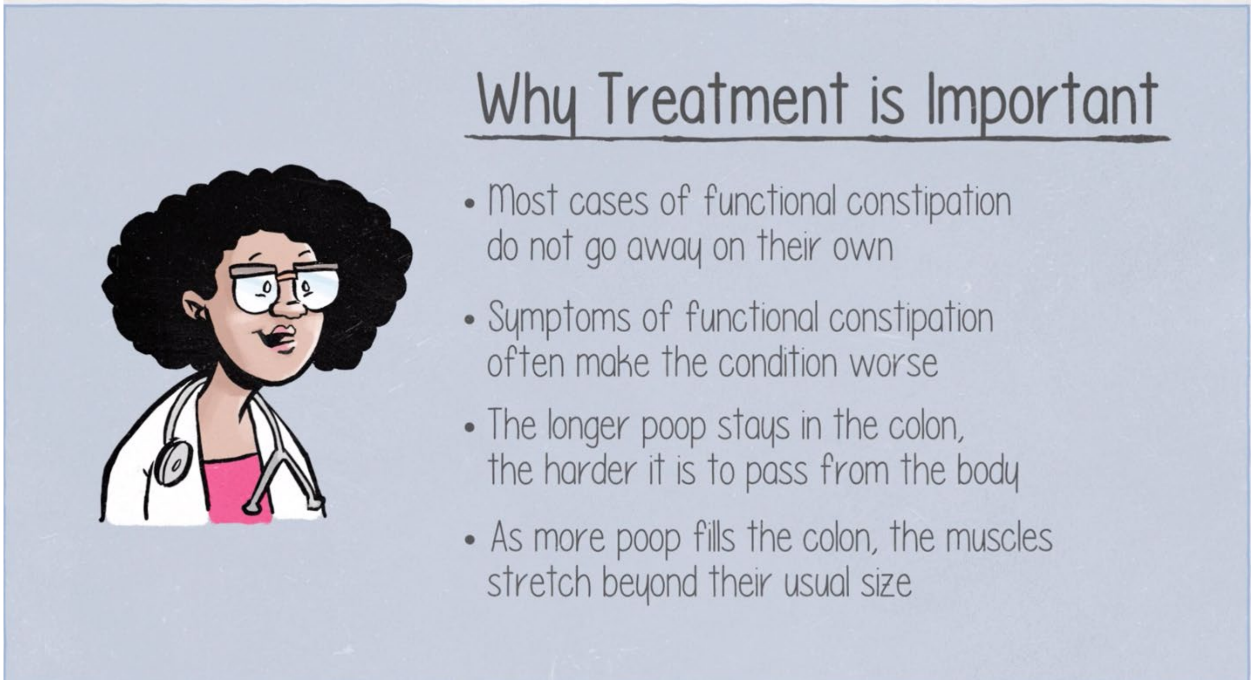
Figure 5.2 KT tool diagnosis

Most children do not need special testing or to see specialists to be diagnosed.

Figure 5.3 KT tool prevalence

Function constipation affects about 1 in 10 children.

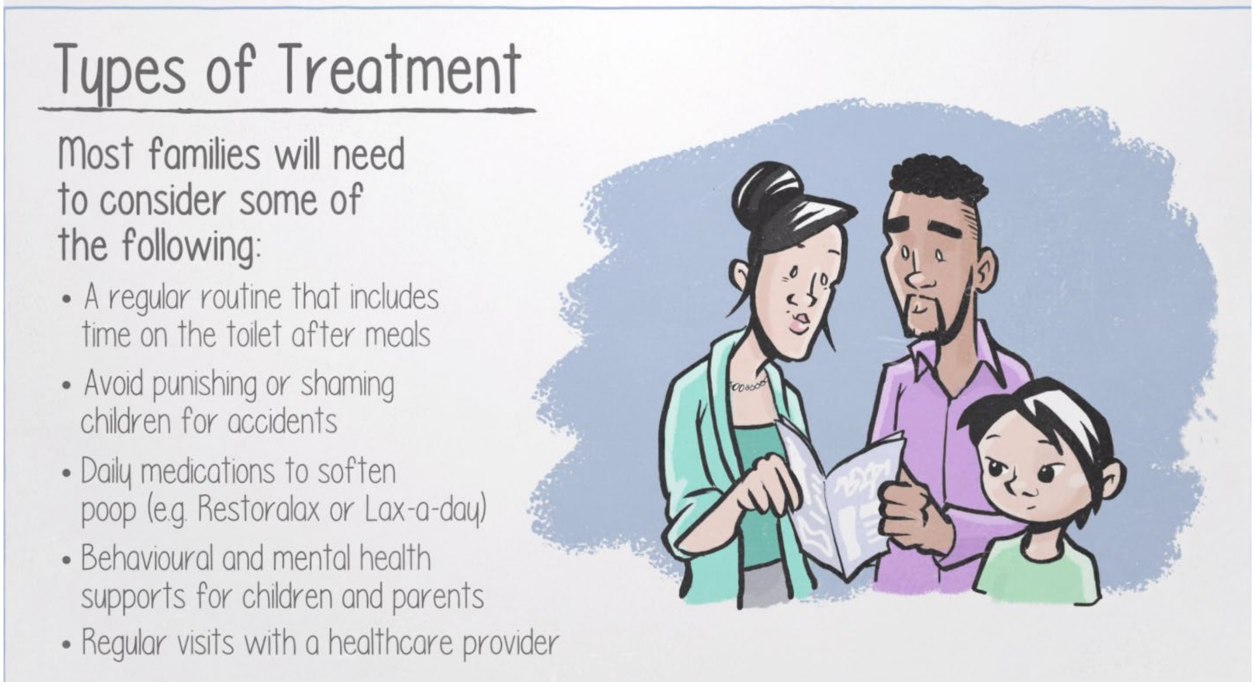
Figure 5.4 KT tool treatment



Why Treatment is Important

- Most cases of functional constipation do not go away on their own
- Symptoms of functional constipation often make the condition worse
- The longer poop stays in the colon, the harder it is to pass from the body
- As more poop fills the colon, the muscles stretch beyond their usual size

Figure 5.5 KT tool types of treatment



Types of Treatment

Most families will need to consider some of the following:

- A regular routine that includes time on the toilet after meals
- Avoid punishing or shaming children for accidents
- Daily medications to soften poop (e.g. Restoralax or Lax-a-day)
- Behavioural and mental health supports for children and parents
- Regular visits with a healthcare provider

Figure 5.6 KT tool reassurance

Try to be patient with yourself and your child.

Usability Testing

30 parents watched the KT video about pediatric FC and answered usability questions.

Our sample primarily self-identified as female (80%) and lived in an urban environment (70%).

Full participant characteristics of our sample are presented in Table 5.1 below.

Table 5.1 Participant Characteristics

Question with responses		Frequency n (%)
What is your gender?		
	Female	24 (80.0%)
	Male	6 (20.0%)
Which ethnicities best describes you? (select all that apply)		
	African American or African Canadian	3 (10.0%)
	Asian	2 (6.7%)
	First Nations	2 (6.7%)
	Métis	1 (3.3%)
	Middle Eastern or North African	2 (6.7%)
	South Asian	3 (10.0%)
	White or Caucasian	15 (50.0%)
	Not listed	1 (3.3%)
	Prefer not to answer	1 (3.3%)
What is your age?		
	20-30 years	4 (13.3%)
	31-40 years	20 (66.7%)
	41-50 years	6 (20.0%)
What is your gross annual household income?		
	Less than $25,000	4 (13.3%)
	$25,000-$49,999	5 (16.7%)
	$50,000-$74,999	6 (20.0%)
	$75,000-$99,999	2 (6.7%)
	$100,000-$149,999	6 (20.0%)
	$150,000 and over	5 16.7%)
	Prefer not to answer	2 (6.7%)
What is your highest level of education?		
	Some high school	3 (10.0%)
	High school diploma	2 (6.7%)
	Some post-secondary	3 (10.0%)

Post-secondary certificate/diploma	7 (23.3%)
Post-secondary degree	11 (36.7%)
Graduate degree	4 (13.3%)
Where does your household live?	
City	21 (70.0%)
Town	1 (3.3%)
Suburb	6 (20.0%)
Farm	1 (3.3%)
Other	1 (3.3%)
How many children do you have?	
1	9 (30.0%)
2	10 (33.3%)
3	8 (26.7%)
4	3 (10.0%)
How many times have you visited the emergency department with your children?	
1-5 times	23 (76.7%)
6+ times	7 (23.35%)
Have any of your children ever been admitted to the hospital?	
No	15 (50.0%)
Yes	15 (50.0%)
What is your relationship to the child that you have brought to the emergency department?	
Parent	29 (96.7%)
Guardian	1 (3.3%)

The digital KT tool was very well received by parents, with none of the usability questions receiving negative (disagree or strongly disagree) responses (see details in Figure 5.7). The mean score from the question related to the simplicity of the KT tool (4.50) ranked the highest. Questions about use in the future and whether the tool will help the parent make decisions about the child's health had the lowest means (4.20) (see details in Figure 5.8). Open text response explained that "*if you don't find it* [the condition] *relative, it* [KT tool] *may not hold attention.*" Meaning parents without personal relevance of pediatric FC were less interested.

Figure 5.7 Usability Questions by Response Frequency

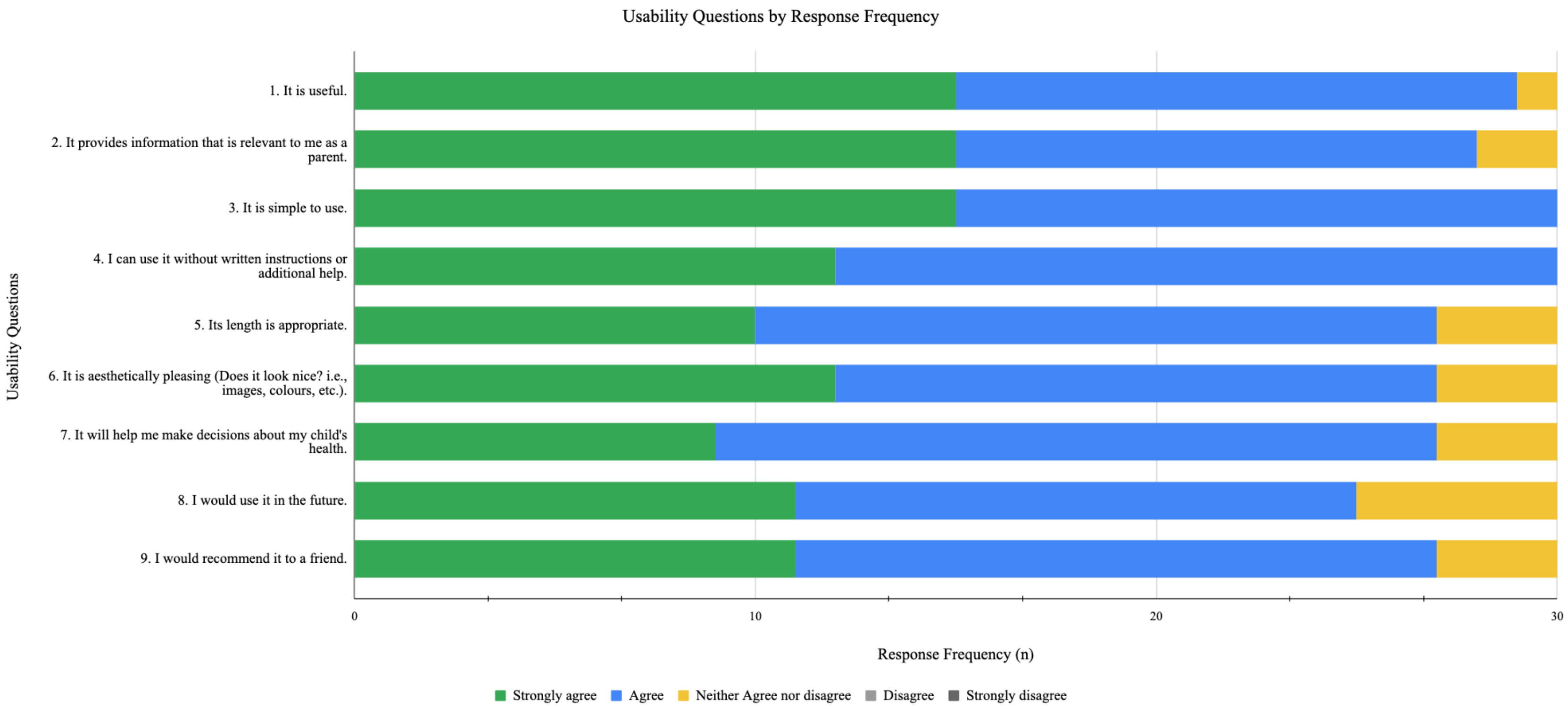
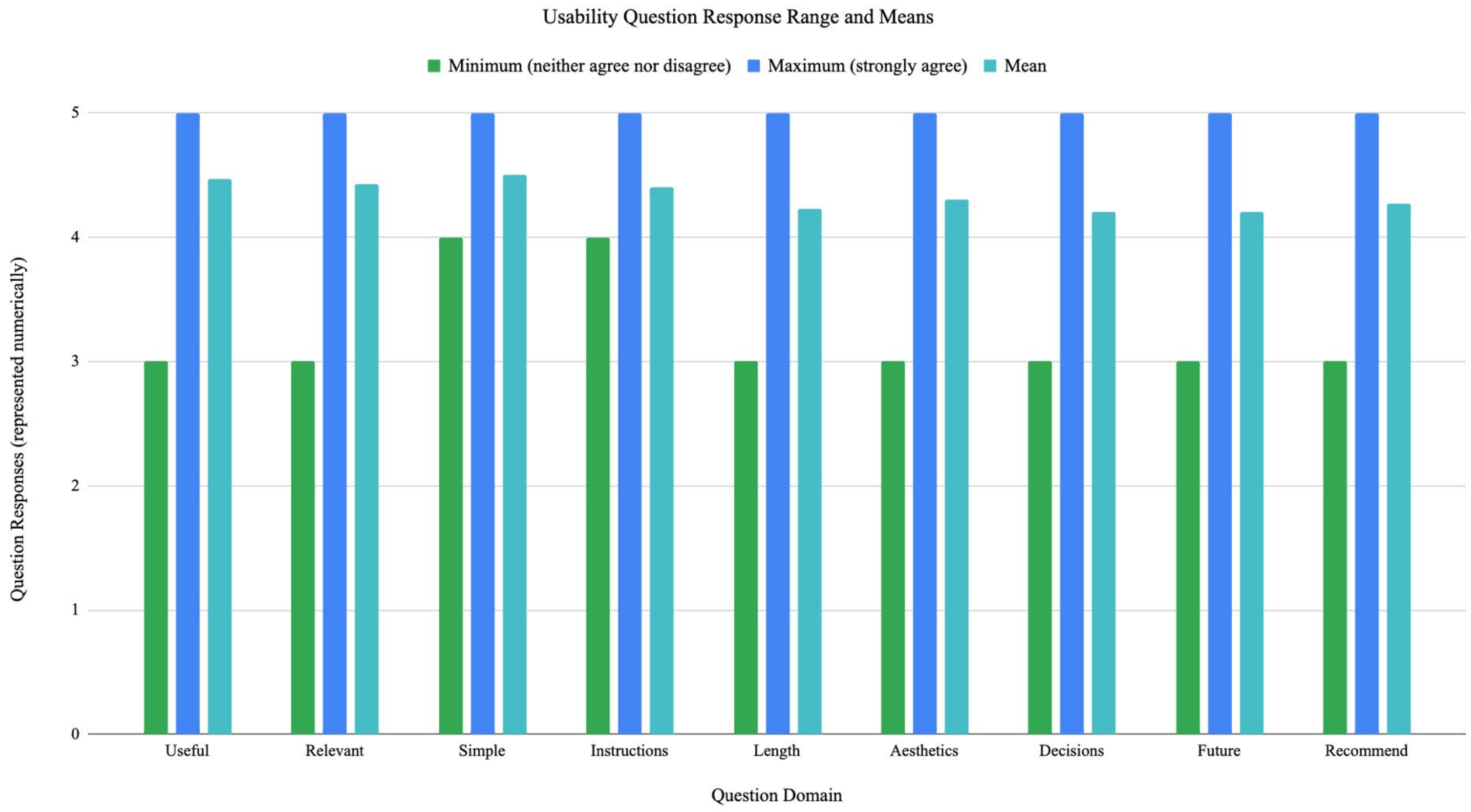


Figure 5.8 Usability Question Response Range and Means



The predominant finding from the open text responses related to strong clarity and ease of use. Multiple responses highlighted the clarity of the information presented. One parent described, "*It was simple and easy to understand. The information was clear and not repetitive*." Another parent described the KT tool as, "*very informative and easy to follow*" with "*clear, concise messages, overall very usable.*" Similarly, another respondent described the video as "*To the point, easy to understand.*" Another theme from the text responses was helpfulness. For example, responses included "*it is very helpful*", "*helping us as parents notice symptoms*", and "*I wish I would have seen it 10 years ago*." Lastly, parents emphasized the effectiveness of the aesthetic aspects of the KT tool. Specifically, responses such as, "*visually pleasing*", "*the use of graphics with text is effective*", "*the voice is very clear, and I like the multicultural family",* and *"good visual aids"* highlight the strengths of the narrated-text, video medium. Open text response suggested the length of the video could potentially be shortened. Two responses included, "*it's a little long*" and "*a bit too long*" as feedback.

Discussion

This study focused on the development and usability evaluation of a novel KT tool for parents caring for a child with FC. The project resulted in the creation of a free and widely accessible digital KT tool about pediatric FC to provide reliable and accurate information to caregivers. In addition, use of patient-engagement methods to develop the KT tool and usability testing are important contributions to the field of patient-direct KT to improve child health outcomes.

Patient Engagement

Our use of a qualitative interviews and the P-PAG meetings to shape the development of the KT tool reflects the growing movement to involve patient voices into research projects and integrated KT. The motivation for this shift includes broad socio-political change towards more equitable methods of knowledge development and a specific intention to optimize KT initiatives with improved outcomes in healthcare (Canadian Institutes of Health Research, 2014; Domecq et al., 2014). Although terminology differs according to country, in Canada, the terms *patient-oriented research* and *patient engagement* are commonly used in healthcare and reflect guidance from the Canadian Institutes of Health Research. Patient engagement is defined as “meaningful and active collaboration in governance, priority setting, conducting research and knowledge translation”(Canadian Institutes of Health Research, 2014). Moreover, the word patient is not restrictive, rather it is used to represent individuals, groups, communities, caregivers, friends, and families who have personal experience and knowledge of a health issue (Canadian Institutes of Health Research, 2014). In this study, parents of children with FC were identified as essential stakeholders because treatment for the condition is primarily provided and monitored by parents and ideally occurs at home. Development of our KT tool using patient engagement methods aligns with recommendations from a systematic review of information resources for parents and offers a meaningful approach to ensure parents have resources tailored to their needs (Neill et al., 2015). Patient engagement reflects a spectrum of activities and responsibilities ranging from informing stakeholders to giving stakeholders complete decision-making autonomy. Our approach in this project aligned with the term collaboration; wherein decision-making is a shared responsibility between the researchers and the patient group, and is inclusive of their knowledge, experience, and preferences (International Association for Public Participation Canada, n.d.). Finally, findings from an evaluation of the P-PAG, as a patient engagement method in pediatric healthcare research, are detailed in a separate manuscript (Hartling, Scott 2021). Results from the evaluation are an important contribution to better understand how to foster meaningful and effective relationships with patients to improve health outcomes (Domecq et al., 2014; Esmail et al., 2015; Lavallee et al., 2012; Staniszewska & Denegri, 2013).

Usability of KT Tool

The purpose of usability testing was to assess how well the developed KT tool meets parents' needs in real life use (Zapata et al., 2015), thereby identifying the barriers to and facilitators of the KT tool's use (Straus et al., 2013). Survey results and open text responses from the parent sample were highly favorable for the usability of the video KT tool. Parents reported the information presented was clear and easy to understand. These findings fit well with a recent systematic review of information resources to support parents caring for a sick child (Neill et al., 2015). For instance, the review explained that resources were most effective when the information provided was relevant and comprehensive, and the resource could be accessed in a relaxed atmosphere (ie. home) (Neill et al., 2015). Although our KT tool was evaluated by parents in the emergency department waiting room, the resource is housed on a digital platform so that parents are able to access at a time and location of their choosing.

In addition, the integration of audio, text, and illustrations into a story-based medium was described by parents as visually pleasing and effective to communicate the information. Our results align with the existing research about the effectiveness of story-telling and video for communicating science information. Specifically, Finkler and León (2019) proposed a framework that included the critical elements of simplification, concreteness, credibility, and emotions to effectively communicate information in story-based videos. Usability results for our KT tool parallel similar domains that made the video relevant and appealing to parents. For example, responses highlighted the accessibility (simple, easy to use) and clarity (informative, effective, easy to understand). Because the parent sample in this study may not have had personal experience with pediatric functional constipation, it is difficult to determine the degree of emotional engagement or validation the narrative format of the tool elicits.

Open text responses were helpful to understand what aspects of the tool were the strongest and what could be improved. Although the question about the length of the KT tool did not garner any negative responses in the survey (disagree or strongly disagree), in the comments portion some respondents suggested the length was slightly too long. As discussed in the development process, video duration is an ongoing and difficult balance of trying to include as much relevant information as possible without creating a tool that becomes inaccessible due to length. Research has demonstrated declining interest and attention span of audiences in relation to increasing video duration (Kim et al., 2014). Optimizing the length of videos for a given audience is a the subject of debate and definitive evidence about duration is scare (Bradbury, 2016). Much of the current research about viewership stems from education settings, where students comprise the audience and the content is more extensive (Brame, 2016; Lau et al., 2018). Therefore, caution should be used when considering applying findings from a student population to a group of parents. In the absence of clear recommendations about an ideal duration for a digital, narrative-based KT tool, we recognize the importance of getting feedback from the users. Consequently, future revisions of the KT tool may need to focus on how to shorten the video by ensuring there is an appropriate limit to the scope or comprehensiveness while also maintaining the clarity of the information presented. Certainly, these can be difficult elements to balance in the development of resources for parents and there is unlikely to be a product that meets all needs for all parents, rather the goal may more realistically be to meet most needs for most parents. Because the comments about the duration of the video were only received from two respondents and there was no indication of redundant or excessive information, it is unclear at this time if content should be removed to shorten the video.

Future Steps

This description of our development and usability evaluation builds a foundation to assess whether the KT tool effectively supports parents' and improves their understanding of FC. Although previous research found no difference between knowledge acquired from an infographic compared to a plain language summary, all the user groups (university students, consumers, and doctors) ranked the reader experience and user-friendliness of the infographic higher (Buljan et al., 2018). Similarly, it is unclear if our use of patient engagement and creative KT methods make a difference to parents' knowledge acquisition; however, providing more user-friendly resources may offer important benefits such as improving patient experiences and fostering positive emotional reactions (i.e. feelings of validation, normalization, or reassurance) (Hartling et al., 2010; Reid et al., 2017; Scott et al., 2012). A randomized-control trial of our KT tool is currently planned and includes knowledge change and user experience outcomes. More rigorous evaluation of the tool will be particularly helpful to understand if the length of the video should be decreased.

Existing research has demonstrated that recognition and treatment of pediatric FC by healthcare providers can be improved (Koppen, Vriesman, Tabbers, et al., 2018; Sood et al., 2018). Although our research is focused on parental outcomes rather than clinicians, educational initiatives for parents may also be helpful to shift healthcare provider behaviours. Specifically, patient-mediated KT interventions focus on the patient audience while also aiming for changes in healthcare provider behaviours (Stacey & Hill, 2013). For example, a systematic review found that strategies where patients are given healthcare information or take part in patient education are amongst the most promising patient-mediated approaches to change healthcare professionals' practice (Fønhus et al., 2018). Therefore, further research to examine the effects of our KT tool on professional practice for childhood FC may also be worthwhile.

Limitations

Since our sample did not require any personal experience with the condition of pediatric FC, there may be elements of the KT tool that will benefit from follow-up assessment. In particular, responses to questions about use in the future and the ability of the KT to support child health decision-making may have been influenced by whether the topic of FC had any personal relevance to the participant(s).

Conclusion

Pediatric FC is a common condition with poor health outcomes for children and unmet needs for their parents. Providing parents with relevant, accessible, and clear resources to understand the condition and treatment is an important contribution to child health. Our use of patient engagement methods to develop a narrative-based KT tool has resulted in a well-received and highly usable resource for parents. Large scale evaluation of impact, including knowledge and user-experience outcomes, is planned to assess the effectiveness in clinical practice and to guide any necessary revisions of the KT tool.

How might this information affect nursing practice?

This research details our work to develop and evaluate usability of a narrative-based, patient-direct KT tool for parents about pediatric FC. Understanding the development process of this KT tool may help healthcare providers to feel confident in the resource and content, which in turn can position them to share the KT tool with families affected by pediatric FC. Promoting awareness of this parental resource is a concrete outcome that can positively affect both clinicians and families in nursing practice. In addition, knowledge of this type of research, using patient engagement strategies to develop resources for families, highlights both a meaningful practice (patient engagement) and a definitive outcome (KT tool) that are widely applicable, meaning others can use this approach to inform their work in any number of populations. In clinical practice, this work has the potential to improve health outcomes for children with FC and their parents. For example, parents may feel supported by having access to relevant information and may be better equipped to advocate for their needs and their child's needs.

References

Alberta Health Services. (2021). *About Us*. Alberta Health Services. https://www.albertahealthservices.ca/stollery/Page14264.aspx

Archibald, M. M., Hartling, L., Ali, S., Caine, V., & Scott, S. D. (2018). Developing “My Asthma Diary”: A process exemplar of a patient-driven arts-based knowledge translation tool. *BMC Pediatrics*, *18*(1), 186. https://doi.org/10.1186/s12887-018-1155-2

Archibald, M. M., & Scott, S. D. (2019). Learning from usability testing of an arts-based knowledge translation tool for parents of a child with asthma. *Nursing Open*, *6*(4), 1615–1625. https://doi.org/10.1002/nop2.369

Borowitz, S. M., Cox, D. J., Kovatchev, B., Ritterband, L. M., Sheen, J., & Sutphen, J. (2005). Treatment of childhood constipation by primary care physicians: Efficacy and predictors of outcome. *Pediatrics*, *115*(4), 873–877. https://doi.org/10.1542/peds.2004-0537

Bradbury, N. A. (2016). Attention span during lectures: 8 seconds, 10 minutes, or more? *Advances in Physiology Education*, *40*(4), 509–513. https://doi.org/10.1152/advan.00109.2016

Brame, C. J. (2016). Effective educational videos: Principles and guidelines for maximizing student learning from video content. *CBE—Life Sciences Education*, *15*(4), es6. https://doi.org/10.1187/cbe.16-03-0125

Brazzelli, M., Griffiths, P. V., Cody, J. D., & Tappin, D. (2011). Behavioural and cognitive interventions with or without other treatments for the management of faecal incontinence in children. *Cochrane Database of Systematic Reviews*, *12*. https://doi.org/10.1002/14651858.CD002240.pub4

Buljan, I., Malički, M., Wager, E., Puljak, L., Hren, D., Kellie, F., West, H., Alfirević, Ž., & Marušić, A. (2018). No difference in knowledge obtained from infographic or plain language summary of a Cochrane systematic review: Three randomized controlled trials. *Journal of Clinical Epidemiology*, *97*, 86–94. https://doi.org/10.1016/j.jclinepi.2017.12.003

Canadian Institutes of Health Research. (2014). *Strategy for patient-oriented research—Patient engagement framework*. http://www.cihr-irsc.gc.ca/e/48413.html

Choung, R. S., Shah, N. D., Chitkara, D., Branda, M. E., Van Tilburg, M. A., Whitehead, W. E., Katusic, S. K., Locke, G. R. I., & Talley, N. J. (2011). Direct medical costs of constipation from childhood to early adulthood: A population-based birth cohort study. *Journal of Pediatric Gastroenterology and Nutrition*, *52*(1), 47–54. https://doi.org/10.1097/MPG.0b013e3181e67058

Cunningham, R. M., & Boom, J. A. (2013). Telling stories of vaccine-preventable diseases: Why it works. *South Dakota Medicine*, 21–26.

Dahlstrom, M. F. (2014). Using narratives and storytelling to communicate science with nonexpert audiences. *Proceedings of the National Academy of Sciences of the United States of America*, *111 Suppl 4*, 13614–13620. https://doi.org/10.1073/pnas.1320645111

Domecq, J. P., Prutsky, G., Elraiyah, T., Wang, Z., Nabhan, M., Shippee, N., Brito, J. P., Boehmer, K., Hasan, R., Firwana, B., Erwin, P., Eton, D., Sloan, J., Montori, V., Asi, N., Abu Dabrh, A. M., & Murad, M. H. (2014). Patient engagement in research: A systematic review. *BMC Health Services Research*, *14*, 89. https://doi.org/10.1186/1472-6963-14-89

Eltorki, M., Landry, P., & TREKK Network. (2019). *Bottom line recommendations for the treatment and management of pediatric constipation*. https://trekk.ca/resources?external_resource_type=Quick_glance&tag_id=D003248

Esmail, L., Moore, E., & Rein, A. (2015). Evaluating patient and stakeholder engagement in research: Moving from theory to practice. *Journal of Comparative Effectiveness Research*, *4*(2), 133–145. https://doi.org/10.2217/cer.14.79

Finkler, W., & Leon, B. (2019). The power of storytelling and video: A visual rhetoric for science communication. *Journal of Science Communication*, *18*(5), A02. https://doi.org/10.22323/2.18050202

Fønhus, M. S., Dalsbø, T. K., Johansen, M., Fretheim, A., Skirbekk, H., & Flottorp, S. A. (2018). Patient-mediated interventions to improve professional practice. *Cochrane Database of Systematic Reviews*, *9*. https://doi.org/10.1002/14651858.CD012472.pub2

Gordon, M., MacDonald, J. K., Parker, C. E., Akobeng, A. K., & Thomas, A. G. (2016). Osmotic and stimulant laxatives for the management of childhood constipation. *Cochrane Database of Systematic Reviews*, *8*. https://doi.org/10.1002/14651858.CD009118.pub3

Green, M. C., & Brock, T. C. (2000). The role of transportation in the persuasiveness of public narratives. *Journal of Personality and Social Psychology*, *79*(5), 701–721. https://doi.org/10.1037/0022-3514.79.5.701

Hartling, L., Elliott, S. A., Buckreus, K., Leung, J., & Scott, S. D. (2021). Development of a parent advisory group to inform a research program for knowledge translation in child health: A patient-oriented evaluation [manuscript submitted for publication]. *Research Involvement and Engagement*.

Hartling, L., Scott, S. D., Johnson, D. W., Bishop, T., & Klassen, T. P. (2013). A randomized controlled trial of storytelling as a communication tool. *PLOS ONE*, *8*(10), e77800. https://doi.org/10.1371/journal.pone.0077800

Hartling, L., Scott, S., Pandya, R., Johnson, D., Bishop, T., & Klassen, T. P. (2010). Storytelling as a communication tool for health consumers: Development of an intervention for parents of children with croup. *BMC Pediatrics*, *10*, 64. https://doi.org/10.1186/1471-2431-10-64

Hornbæk, K. (2006). Current practice in measuring usability: Challenges to usability studies and research. *International Journal of Human-Computer Studies*, *64*(2), 79–102. https://doi.org/10.1016/j.ijhcs.2005.06.002

International Association for Public Participation Canada. (n.d.). *IAP2 Canada—Public participation spectrum*. Retrieved 12 April 2018, from http://iap2canada.ca/page-1020549

Jackson, C., Cheater, F. M., & Reid, I. (2008). A systematic review of decision support needs of parents making child health decisions. *Health Expectations*, *11*(3), 232–251. https://doi.org/10.1111/j.1369-7625.2008.00496.x

Jones, M. D. (2014). Communicating climate change: Are stories better than 'just the facts'? *Policy Studies Journal*, *42*(4), 644–673. https://doi.org/10.1111/psj.12072

Jurgens, H., Oster, C., & Fereday, J. (2011). Management of chronic functional constipation in children: A review of the literature. *Neonatal, Paediatric & Child Health Nursing*, *14*(2), 23–28.

Kaugars, A. S., Silverman, A., Kinservik, M., Heinze, S., Reinemann, L., Sander, M., Schneider, B., & Sood, M. (2010). Families' perspectives on the effect of constipation and fecal incontinence on quality of life. *Journal of Pediatric Gastroenterology and Nutrition*, *51*(6), 747–752. https://doi.org/10.1097/MPG.0b013e3181de0651

Kim, J., Guo, P. J., Seaton, D. T., Mitros, P., Gajos, K. Z., & Miller, R. C. (2014). Understanding in-video dropouts and interaction peaks in online lecture videos. *Proceedings of the First ACM Conference on Learning @ Scale Conference*, 31–40. https://doi.org/10.1145/2556325.2566237

Klages, K. L., Berlin, K. S., Silverman, A. H., Mugie, S., Di Lorenzo, C., Nurko, S., Ponnambalam, A., Sanghavi, R., & Sood, M. R. (2016). Empirically derived patterns of pain, stooling, and incontinence and their relations to health-related quality of life among youth with chronic constipation. *Journal of Pediatric Psychology*, jsw068. https://doi.org/10.1093/jpepsy/jsw068

Koppen, I. J. N., Vriesman, M. H., Saps, M., Rajindrajith, S., Shi, X., van Etten-Jamaludin, F. S., Di Lorenzo, C., Benninga, M. A., & Tabbers, M. M. (2018). Prevalence of functional defecation disorders in children: A systematic review and meta-analysis. *The Journal of Pediatrics*, *198*, 121-130.e6. https://doi.org/10.1016/j.jpeds.2018.02.029

Koppen, I. J. N., Vriesman, M. H., Tabbers, M. M., Di Lorenzo, C., & Benninga, M. A. (2018). Awareness and implementation of the 2014 ESPGHAN/NASPGHAN guideline for childhood functional constipation. *Journal of Pediatric Gastroenterology and Nutrition*, *66*(5), 732–737. https://doi.org/10.1097/MPG.0000000000001786

Kovacic, K., Sood, M. R., Mugie, S., Di Lorenzo, C., Nurko, S., Heinz, N., Ponnambalam, A., Beesley, C., Sanghavi, R., & Silverman, A. H. (2015). A multicenter study on childhood constipation and fecal incontinence: Effects on quality of life. *The Journal of Pediatrics*, *166*(6), 1482-1487.e1. https://doi.org/10.1016/j.jpeds.2015.03.016

Lau, K. H. V., Farooque, P., Leydon, G., Schwartz, M. L., Sadler, R. M., & Moeller, J. J. (2018). Using learning analytics to evaluate a video-based lecture series. *Medical Teacher*, *40*(1), 91–98. https://doi.org/10.1080/0142159X.2017.1395001

Lavallee, D. C., Williams, C. J., Tambor, E. S., & Deverka, P. A. (2012). Stakeholder engagement in comparative effectiveness research: How will we measure success? *Journal of Comparative Effectiveness Research*, *1*(5), 397–407.

Lee, H., Kim, M., Cooley, M. E., Kiang, P. N., Kim, D., Tang, S., Shi, L., Thiem, L., Kan, P., Peou, S., Touch, C., Chea, P., & Allison, J. (2018). Using narrative intervention for HPV vaccine behavior change among Khmer mothers and daughters: A pilot RCT to examine feasibility, acceptability, and preliminary effectiveness. *Applied Nursing Research*, *40*, 51–60. https://doi.org/10.1016/j.apnr.2017.12.008

Liem, O., Harman, J., Benninga, M. A., Kelleher, K., Mousa, H., & Di Lorenzo, C. (2009). Health utilization and cost impact of childhood constipation in the United States. *The Journal of Pediatrics*, *154*(2), 258–262. https://doi.org/10.1016/j.jpeds.2008.07.060

Mousa, H. M., Barsamian, J. M., Benitez, A. J., Webster, J. M., & Fiorino, K. N. (2020). Pediatric constipation: An epidemiologic and physiologic approach to guide a stepwise approach to its management. *Current Treatment Options in Pediatrics*, *6*(3), 155–169. https://doi.org/10.1007/s40746-020-00199-z

Mugie, S. M., Di Lorenzo, C., & Benninga, M. A. (2011). Constipation in childhood. *Nature Reviews Gastroenterology & Hepatology*, *8*(9), 502–511. https://doi.org/10.1038/nrgastro.2011.130

National Collaborating Centre for Women’s and Children’s Health. (2010). *Constipation in children and young people: Diagnosis and management of idiopathic childhood* *constipation in primary and secondary care*. RCOG Press. http://www.ncbi.nlm.nih.gov/books/NBK65365/

Neill, S., Roland, D., Jones, C. H., Thompson, M., & Lakhanpaul, M. (2015). Information resources to aid parental decision-making on when to seek medical care for their acutely sick child: A narrative systematic review. *BMJ Open*, *5*(12), e008280. https://doi.org/10.1136/bmjopen-2015-008280

Njeru, J. W., Patten, C. A., Hanza, M. M. K., Brockman, T. A., Ridgeway, J. L., Weis, J. A., Clark, M. M., Goodson, M., Osman, A., Porraz-Capetillo, G., Hared, A., Myers, A., Sia, I. G., & Wieland, M. L. (2015). Stories for change: Development of a diabetes digital storytelling intervention for refugees and immigrants to Minnesota using qualitative methods. *BMC Public Health*, *15*, 1311. https://doi.org/10.1186/s12889-015-2628-y

Reid, K., Hartling, L., Ali, S., Le, A., Norris, A., & Scott, S. D. (2017). Development and usability evaluation of an art and narrative-based knowledge translation tool for parents with a child with pediatric chronic pain: Multi-method study. *Journal of Medical Internet Research*, *19*(12), e412. https://doi.org/10.2196/jmir.8877

Rowan-Legg, A. & Canadian Paediatric Society. (2011). Managing functional constipation in children. *Paediatrics & Child Health*, *16*(10), 661–665.

Scott, S. D., Hartling, L., O’Leary, K. A., Archibald, M., & Klassen, T. P. (2012). Stories – a novel approach to transfer complex health information to parents: A qualitative study. *Arts & Health*, *4*(2), 162–173. https://doi.org/10.1080/17533015.2012.656203

Sood, M., Lichtlen, P., & Perez, M. C. (2018). Unmet needs in pediatric functional constipation. *Clinical Pediatrics*, *57*(13), 1489–1495. https://doi.org/10.1177/0009922818774343

Southwell, B. R. (2020). Treatment of childhood constipation: A synthesis of systematic reviews and meta-analyses. *Expert Review of Gastroenterology & Hepatology*, *14*(3), 163–174. https://doi.org/10.1080/17474124.2020.1733974

Stacey, D., & Hill, S. (2013). Patient-direct and patient-mediated KT interventions. In *Knowledge translation in health care* (2nd ed., pp. 197–211). Wiley-Blackwell.

Staniszewska, S., & Denegri, S. (2013). Patient and public involvement in research: Future challenges. *Evidence-Based Nursing*, *16*(3), 69–69. https://doi.org/10.1136/eb-2013-101406

Straus, S., Tetroe, J., & Graham, I. D. (Eds.). (2013). *Knowledge translation in health care: Moving from evidence to practice* (2 edition). BMJ Books.

Tabbers, M. M., DiLorenzo, C., Berger, M. Y., Faure, C., Langendam, M. W., Nurko, S., Staiano, A., Vandenplas, Y., & Benninga, M. A. (2014). Evaluation and treatment of functional constipation in infants and children: Evidence-based recommendations from ESPGHAN and NASPGHAN. *Journal of Pediatric Gastroenterology and Nutrition*, *58*(2), 258–274.

Thompson, A. P., MacDonald, S. E., Wine, E., & Scott, S. D. (2021). Understanding parents' experiences when caring for a child with functional constipation: Interpretive description study. *JMIR Pediatrics and Parenting*, *4*(1), e24851. https://doi.org/10.2196/24851

Thompson, A. P., Wine, E., MacDonald, S. E., Campbell, A., & Scott, S. D. (2020). Parents' experiences and information needs while caring for a child with functional constipation: A systematic review. *Clinical Pediatrics*, *60*(3), 154–169. https://doi.org/10.1177/0009922820964457

Vaismoradi, M., Turunen, H., & Bondas, T. (2013). Content analysis and thematic analysis: Implications for conducting a qualitative descriptive study. *Nursing & Health Sciences*, *15*(3), 398–405. https://doi.org/10.1111/nhs.12048

van den Berg, M. M., Benninga, M. A., & Di Lorenzo, C. (2006). Epidemiology of childhood constipation: A systematic review. *The American Journal of Gastroenterology*, *101*(10), 2401–2409. https://doi.org/10.1111/j.1572-0241.2006.00771.x

van Engelenburg-van Lonkhuyzen, M. L., Bols, E. M. J., Benninga, M. A., Verwijs, W. A., & de Bie, R. A. (2017). Effectiveness of pelvic physiotherapy in children with functional constipation compared with standard medical care. *Gastroenterology*, *152*(1), 82–91. https://doi.org/10.1053/j.gastro.2016.09.015

Varni, J. W., Bendo, C. B., Nurko, S., Shulman, R. J., Self, M. M., Franciosi, J. P., Saps, M., & Pohl, J. F. (2015). Health-related quality of life in pediatric patients with functional and organic gastrointestinal diseases. *The Journal of Pediatrics*, *166*(1), 85-90.e2. https://doi.org/10.1016/j.jpeds.2014.08.022

Varni, J. W., Nurko, S., Shulman, R. J., Self, M. M., Saps, M., Bendo, C. B., Vaughan Dark, C., Pohl, J. F., & Consortium, for the P. Q. of L. I. G. S. M. T. S. (2015). Pediatric functional constipation gastrointestinal symptom profile compared with healthy controls. *Journal of Pediatric Gastroenterology and Nutrition*, *61*(4), 424–430. https://doi.org/10.1097/MPG.0000000000000869

Vriesman, M. H., Rajindrajith, S., Koppen, I. J. N., van Etten- Jamaludin, F. S., van Dijk, M., Devanarayana, N. M., Tabbers, M. M., & Benninga, M. A. (2019). Quality of life in children with functional constipation: A systematic review and meta-analysis. *The Journal of Pediatrics*, *214*, 141–150. https://doi.org/10.1016/j.jpeds.2019.06.059

Yang, C. H., & Punati, J. (2015). Practice patterns of pediatricians and trainees for the management of functional constipation compared with 2006 NASPGHAN guidelines. *Journal of Pediatric Gastroenterology and Nutrition*, *60*(3), 308–311. https://doi.org/10.1097/MPG.0000000000000591

Zapata, B. C., Fernández-Alemán, J. L., Idri, A., & Toval, A. (2015). Empirical studies on usability of mhealth apps: A systematic literature review. *Journal of Medical Systems*, *39*(2), 1. https://doi.org/10.1007/s10916-014-0182-2

Chapter 6. Discussion and Conclusion

Overview of Findings

My dissertation focused on improving care for families affected by pediatric FC. Through this research, I found two primary knowledge gaps in the research literature. Firstly, despite the important role parents play in treatment for pediatric FC, little was known about how parents experienced and understood the condition. Secondly, PE is a growing method used in research to optimize processes and outcomes; however, the lack of evaluation has delayed evidence-based development in the field. Unfortunately, my attempt to form a PCG was not successful, so the planned evaluation of this PE strategy did not generate results. The published study protocol remains a contribution to the field by delineating the PE process, the use of theory to guide both the PE approach and the planned evaluation, and lastly by the use of validated tools. To further my research program, my plans shifted to pursue usability testing of the developed KT tool. The cumulative results from this dissertation make a substantive contribution to improving care for families affected by pediatric FC and advancing knowledge of pediatric FC, patient-direct KT, and PE in research. In this concluding chapter, I provide an overview of the knowledge that developed from the four dissertation papers in relation to the previous gaps in the fields. I also discuss the relevance of this dissertation to clinical care, KT, and PE. I present the strengths and limitations of the work and conclude by outlining directions for future research.

Implications for Clinical Care

This research makes a unique contribution to understanding the experiences and information needs of parents caring for a child with FC. Bringing parents' perspectives to the forefront of clinical practice details an avenue to improve care for families and creates opportunities to better support parents.

Knowledge Synthesis

Firstly, from the systematic review, I established that research specifically exploring parents' experiences with pediatric FC was scarce. Given that pediatric FC is a very common condition and the important role parents play in treatment, the research exploring parents' viewpoints was insufficient to meaningfully inform improvements in practice. For instance, parents were more often included in research as a proxy for a young child unable to participate first-hand (Inan et al., 2007; Oostenbrink et al., 2010). Similarly, research with parents as participants was frequently tied to satisfaction with a particular intervention rather than related to care for pediatric FC generally (Kuizenga-Wessel et al., 2016; Lu et al., 2018; Sullivan et al., 2006). Several of the included studies measured parents' quality of life and while this captured the broad burden weighing on caregivers, it was difficult to understand how to translate this information into improved care.

Despite the small number of included studies in our systematic review, findings across the studies were consistent. Concerningly, our results revealed that parents are typically reluctant to access the healthcare system—often living with the child's symptoms for months or years. Although parents were typically deeply concerned about their child's symptoms, clinicians mistakenly underestimated the significance of parents' distress and the severity of children's symptoms. These findings parallel recent evaluations of clinical practice that suggest treatment and symptom severity are often mismatched (Sood et al., 2018). Unfortunately, this seems to be an enduring trend, with evidence from more than 15 years ago similarly describing pediatric FC as undertreated by clinicians (Borowitz et al., 2005). Although reluctance to access healthcare services may stem from feelings of shame, embarrassment, and guilt, parents risked being further marginalized through dismissive encounters with healthcare providers.

Another significant finding from our review was that parents' knowledge of pediatric FC was limited, with a need for increased family education about the condition and its treatment. Unfortunately, parents were often given incorrect reassurances their child would grow out of the symptoms with time. Whether provided from friends, family, or healthcare providers, the effects of false information are likely to further delay or impede parents' child health decision-making. In addition, our results suggested that parents may overestimate the role of dietary changes such as increased fluid and fibre intake in the treatment of pediatric FC, whereas dietary interventions are known to be insufficient as a primary treatment for pediatric FC (Mugie, Di Lorenzo, et al., 2011; Tabbers et al., 2014). Therefore, healthcare providers should take care to present dietary changes as a goal of nutritionally balanced eating, rather than as an active treatment of pediatric FC.

Similarly, our results demonstrated that parents' understanding of medication use and safety was a barrier to treatment adherence. For example, questions about causing laxative dependence and safety of long-term use were common. Furthermore, parents expressed hesitancy about managing side effects and titrating doses. It is a substantial burden to task parents with giving high-dose medications while also trying to differentiate between symptoms such as abdominal pain or loose stools which can equally stem from medication side-effects and progressive worsening of FC. Recent evidence noted that parents may overestimate their understanding of discharge instructions, particularly when combined with low health literacy or complex treatment plans (Glick et al., 2020). Therefore, acknowledging potential barriers to medication use and honest discussion of larger contextual influences (e.g., family dynamics, school or work timing, financial resources etc) may be needed to support parents' information needs related to medication use for pediatric FC.

Lastly, our findings highlighted the negative effects of ongoing symptoms combined with arduous caregiving on parents' knowledge and confidence. In other words, parents require reassurance and ongoing support as critical components of parental education. For example, studies revealed that behavioural interventions may become the source of parent-child conflicts and are emotionally taxing for parents. Given the negative effects of misinformation and parental frustration on family relationships and treatment outcomes, finding innovative ways to meet parents' information and support needs is critically important to improve care for families living with pediatric FC.

Qualitative Inquiry

Building from the results of the systematic review, I conducted a qualitative inquiry using interpretive description methodology to further explore parents' experiences caring for a child with FC. Our findings from this study of parents' experiences with pediatric FC echoed results from previous research in the field. In particular, a 2003 study of parents' healthcare encounters for pediatric FC found themes of "dismissed and fobbed off, asserting the need for action, and validation and acknowledgment" (Farrell et al., 2003). The similarity of our findings revealed that problematic encounters with healthcare professionals for pediatric FC are a longstanding issue. Prevalence of pediatric FC is high, so healthcare providers should seemingly anticipate cases in practice, yet findings suggested clinicians frequently discounted parental concerns and underestimated symptom severity.

One major finding from our study was that despite explicit recommendations to build treatment plans around family education, parents generally received little to no information about the condition. Specifically, parents had longstanding unanswered questions and held misconceptions about the condition and treatment. Even worse, some healthcare providers perpetuated misinformation. In addition, our results revealed clinicians may need to be cautious about how messages are interpreted by parents. For example, parents described that emphasizing how common the condition could be perceived as an (admittedly unintended) message of normality rather than validation. It is critical to recognize most parent's context for pediatric FC. Parents in our study were typically living with significant physical, emotional, and psychological burdens which magnified their feelings of vulnerability when discussing pediatric FC. In particular, parents identified strong feelings of isolation and frustration which were often triggered by symptoms such as soiling and further exacerbated by negative interactions or misinformation.

Given the time-consuming nature of consultations to provide emotional support and education it is understandable that healthcare professionals are challenged to fit these sessions into already busy schedules and within systems that disincentivize lengthy appointments. More comprehensive service models may be necessary to adequately meet families' complex needs related to pediatric FC. For example, integration of nursing and allied health members may be helpful to more accurately and consistently meet parents' support needs when caring for a child with FC (Houghton et al., 2016; Ismail et al., 2011; Sullivan et al., 2006) .

Integration of Results

Integrating the major findings from the systematic review and the qualitative study with central perspectives from the research literature exposed key areas that may improve care for pediatric FC. The following list presents elements that were recurring in our findings and supported by related research in the field of pediatric FC.

1. The need for improved healthcare models that facilitate comprehensive care and include more accessible opportunities for regular follow-up (Houghton et al., 2016; Ismail et al., 2011; Sullivan et al., 2006)
2. An opportunity to develop interventions or resources for clinicians (Koppen, Vriesman, Tabbers, et al., 2018; Sood et al., 2018; Yang & Punati, 2015) to address the gaps between parents' experiences and clinical practice recommendations.
3. The need for more relevant and accessible resources to address parents' questions about pediatric FC including:
    - Recognition and validation of caregiver burden for pediatric FC
    - Physiological changes related to FC
    - Explanation of soiling pathophysiology
    - Medication use and safety
    - Treatment goals and duration

The intersection of the existing research and our findings produced compelling evidence about the need to better support parents when caring for a child with FC. Consequently, the next phase of this research involved translating the findings into a resource for parents that accurately reflected their needs. The development of practice relevant knowledge and a novel resource are substantive contributions towards the goal of improving care for families affected by pediatric FC.

Implications for Knowledge Translation Science

This program of research embodies the integration of PE in research and KT with the goal of creating a relevant, accessible, and clear resource for parents. In addition to developing knowledge to address gaps in the literature about pediatric FC, this research built upon the research findings to create a patient-direct KT tool for parents. To begin the iterative process, I created a draft composite narrative based on the most poignant of elements of parents' stories from the qualitative interviews and the themes from the systematic review findings. The broad goals for the process were to foster recognition of shared experience amongst the audience and integrate the best research evidence for management of FC. The use of a story-based format aligned with research about effective mediums to foster engagement and facilitate learning (Archibald et al., 2018; Dahlstrom, 2014; Green & Brock, 2000; Jones, 2014; Lee et al., 2018). Narrative methods were purposely chosen with the intention to heighten audience engagement, build relevance, and mirror parents' experiences to promote identification with the information. I chose to use a narrated-video to convey the story based on personal experiences with digital learning and findings from similar creative KT approaches to support parental education (Archibald & Scott, 2019; Reid et al., 2017). The resulting KT tool presents the story of 5-year-old Ari and his parents, who struggle with pediatric FC. The video is approximately 5 minutes in duration and includes information about prevalence, typical age of onset, common symptoms, what to expect during assessment, physiological cycle of worsening symptoms, how soiling accidents occur, treatment, and further resources.

Usability of the KT tool was assessed by thirty parents in the waiting room of a pediatric emergency department with highly favorable results. Parents felt the information was clear and easy to understand. Our findings align with a recent systematic review of information resources to support parents caring for a sick child (Neill et al., 2015). The authors reported that resources were most effective when the information provided was relevant and comprehensive (Neill et al., 2015). In addition, our usability results fit with existing research about the effectiveness of storytelling and video for communicating science information. More specifically, the developers of a framework proposed the elements of simplification, concreteness, credibility, and emotions were central to effectively communicate information in story-based videos (Finkler & Leon, 2019). Usability results for our KT tool parallel similar domains that made the video relevant, easy to understand, simple, and appealing for parents.

As described previously, video duration was an ongoing and difficult balance of trying to provide comprehensive information without creating a tool that was unappealing due to its length. Not surprisingly, some usability comments suggested the length was slightly too long. Ideal KT tool duration is an important concern because there is conflicting research about interest and attention span in relation to increasing video duration (Bradbury, 2016; Kim et al., 2014). For instance, much of the research about viewership comes from education settings with students and therefore, the content is more extensive (Brame, 2016; Lau et al., 2018). Undoubtedly, length and content are difficult elements to balance in the development of KT tools for parents. It is implausible to try to create a resource that meets all needs for all parents; instead, it may be more realistic to aim to meet most needs for most parents.

Although the focus of my dissertation was on parents rather than clinicians, it is interesting to note that KT interventions that integrate patient perspectives may have dual benefits for patients and healthcare professionals. In particular, patient-mediated KT is a broader term that includes patient-direct KT and refers to initiatives that are aimed at changing clinicians' behaviour through interactions with patients, or through information provided by or to patients (Fønhus et al., 2018). A systematic review of patient-mediated approaches found that strategies where patients are given healthcare information and strategies where patients take part in patient education are amongst the most promising to change healthcare professionals' practice (Fønhus et al., 2018). In addition to providing parents with relevant and accessible information, improving care for families affected by pediatric FC also requires changes in practice. Therefore, this research may form an integral connection to future fields of inquiry about the effects of patient-direct KT to shift healthcare providers practice.

Implications for Patient Engagement Science

Involving parents in the development of our KT tool for pediatric FC was an essential design element of this research and also served to ensure the resource would ultimately meet their needs. As discussed previously, the planned PCG for this research did not occur, so the method of PE changed. Instead of a group of parents collaborating for a condition-specific cause, I was able to shift towards working with an existing P-PAG to actively incorporate parental input for the KT tool. Our use of a qualitative interviews and the P-PAG meetings to shape the development of the KT tool reflects the growing movement to involve patient voices into research projects and integrated KT (Banner et al., 2019; Rolfe et al., 2018). The group was invited to advise on all aspects of the KT tool and discussion with P-PAG members was integral to refine and adapt the images and content of the KT tool. For instance, parents identified a statement that was intended to reassure parents that specialized testing or diagnostic imaging is rarely required for pediatric FC but instead it was mistaken to mean pediatric FC did not need medical care—certainly not the desired message! The primary tension of the process was to balance parents' eagerness to add more information and detail with the desire to keep the length accessible. Ultimately, the group determined that detailed management instructions could be overwhelming for parents and might be better addressed in a subsequent resource.

Although I experienced first-hand the meaningful contribution of PE in this research, the planned evaluation did not occur, meaning I cannot offer guidance for future projects or PE approaches. Despite this setback, the published PE evaluation protocol may still be helpful to develop PE science. In particular, recommendations to strengthen the field of PE science identify priorities areas for research (Esmail et al., 2015; Manafo et al., 2018; Staniszewska & Denegri, 2013). I purposely addressed these areas in the study protocol through: 1) the use of an *a priori* evaluation protocol, 2) explicit meta-criterion as the foundation for patient engagement (Lavallee et al., 2012), 3) an empirically based conceptual framework to guide operationalization (Hamilton et al., 2018), and 4) a validated tool for data collection (Abelson et al., 2016). These elements of design and planning underscore the rigor of the proposed study but remain widely applicable to other contexts. That is, others can use the protocol to guide patient engagement processes and evaluation across diverse conditions and populations.

Strengths and Limitations

A strength of my dissertation is the clinically driven relevance of the contribution to understanding parents' information needs related to pediatric FC. Specific strengths and limitations related to each phase of the research are also discussed.

Representation

In the systematic review (paper 1), although the results across the studies were consistent, the small number of included studies is an important limitation. The small number of studies and the generally small sample sizes within the studies mean the results may not fully reflect the variability of parents' experiences or have captured less common occurrences.

In the qualitative study (paper 2) I had significant success with online recruitment through various social media platforms. I was pleased to have a strong interest from parents and felt the interviews generated rich data; however, self-selection of participants may have created a potential sampling bias. In particular, parents with dramatic experiences or worse symptom severity may have been disproportionally motivated to participate. Conversely, sampling and recruitment methods that are mediated by a documented diagnosis or access to healthcare (i.e., research liaison, use of electronic medical records, healthcare referral etc.) can inadvertently exclude participants who are less connected to healthcare systems. In addition, fathers' perspectives were underrepresented in the qualitative study. Despite additional efforts to recruit fathers, the sample was exclusively mothers. Data from the interviews revealed that all the mothers identified themselves as the caregiver with primary responsibility for managing pediatric FC; therefore, the disproportionate lack of fathers may reflect a common occurrence in the real-life care of pediatric FC. Lastly, parents who shared their story for the study were typically from higher education and income levels; meaning, experiences of parents with lower levels of education or income may not be adequately captured in the findings.

In the usability testing (paper 4), our sampling criteria did not require or assess for any personal experience with the condition of pediatric FC. Consequently, there may be elements of the KT tool that will benefit from a more targeted inquiry. For example, details of the information presented may need to be refined or revised to more accurately address user needs. Similarly, responses to questions about use in the future and the ability of the KT tool to support child health decision-making may differ for parents with personal experience with pediatric FC.

Methodology and Design

One key strength of this research is the alignment between the philosophical, methodological, and practical elements of the dissertation. Specifically, approaching qualitative inquiry from a pragmatic stance parallels the clinical orientation emphasized in ID. Additionally, the use of PE strategies to develop a KT tool for parents mirrors a vital relational aspect of knowledge generation that aligns with my nursing practice, research aims, and philosophical position. Lastly, the creation of a lasting resource for parents parallels my pragmatic stance towards research and working to improve clinical care for families affected by pediatric FC.

Future Directions

Throughout the course of this dissertation, I was pleased to consider how the knowledge I was contributing to could be used in the future to inform subsequent research. Based on the important disconnect revealed between parents' experiences caring for a child with FC and clinical care, there are several areas that merit further exploration. Firstly, although it is clear that parents have unmet information needs, further studies would be helpful to generate insight into parents' preferences for accessing resources. Additionally, parents' need for emotional support was a key finding from this research and this work only begins to address this need. Although there are many condition specific elements, it may be possible to gain valuable insight through a scoping review of emotional supports offered to parents caring for an ill child.

The findings from this research identified that clinical encounters can be a source of misinformation and exacerbate parental feelings of guilt. Further examination of the clinical environment is likely necessary to understand how to improve care. For example, observation of interactions between healthcare providers and parents may help clarify challenges and barriers to improving care provision for this condition. Furthermore, research is also needed to identify what systems, supports, and resources healthcare providers need to optimize care when working with families affected by pediatric FC.

Usability testing of the KT tool builds a foundation to assess whether the resource effectively supports parents' and/or improves their understanding of FC. Towards this end, more rigorous evaluation of the impact of the KT tool is needed. A randomized-control trial of our KT tool is currently planned and includes parental knowledge change and user experience outcomes.

As discussed previously, exploring the role of patient-direct KT (as a specific subset of patient-mediated KT) in changing healthcare providers practice is a fascinating direction of inquiry that can be explored from this dissertation. Therefore, further research to examine the effects of our KT tool on professional practice for childhood FC may also be worthwhile.

Conclusion

My dissertation has generated a comprehensive understanding of parents' experiences and information needs when caring for a child with FC. In addition to developing knowledge, my research resulted in the creation of an enduring, patient-direct KT tool for parents. The development of practice relevant knowledge and a novel resource are substantive contributions to the field of pediatric FC, with implications for PE and KT science. I conducted a multi-phase, progressive research project that aligned with theoretical guidance and research evidence to fill knowledge gaps that may hold promise to improve care for families affect by pediatric FC. I started by identifying and synthesizing relevant research evidence using systematic review methods. This step highlighted the incomplete picture of how parents were living with and managing pediatric FC. The findings substantiated the worrisome caregiving toll of the condition for parents. In addition to clarifying a variety of information gaps, the results revealed that healthcare encounters were potential sources of misinformation for parents. To address the scant research on the topic, I conducted a qualitative inquiry on the same topic to generate more in-depth findings. Using ID methodology positioned the research towards producing clinically relevant results, which aligned with my overarching goal for this dissertation to make a positive contribution to the care of families living with pediatric FC. I used the findings from the first two phases to inform the development of a KT tool for parents. Specifically, I used PE methods by collaborating with an existing parent group to refine and revise the KT tool. By publishing an evaluation protocol that targets the priority areas needed to strengthen the field of PE, I made a needed methodological contribution to this emerging science. Lastly, I tested the usability of the KT tool to understand if the resource adequately met parents' needs in a real-life environment. This research has resulted in a tangible output to improve clinical care for pediatric FC including a relevant, accessible, and patient-driven KT tool to support parents. The research findings, methodological, and practical contributions of this dissertation have the potential to improve health outcomes for families while also creating a foundation for future research.

References

Abelson, J., Li, K., Wilson, G., Shields, K., Schneider, C., & Boesveld, S. (2016). Supporting quality public and patient engagement in health system organizations: Development and usability testing of the Public and Patient Engagement Evaluation Tool. *Health Expectations*, *19*(4), 817–827. https://doi.org/10.1111/hex.12378

Archibald, M. M., Hartling, L., Ali, S., Caine, V., & Scott, S. D. (2018). Developing "My Asthma Diary": A process exemplar of a patient-driven arts-based knowledge translation tool. *BMC Pediatrics*, *18*(1), 186. https://doi.org/10.1186/s12887-018-1155-2

Archibald, M. M., & Scott, S. D. (2019). Learning from usability testing of an arts-based knowledge translation tool for parents of a child with asthma. *Nursing Open*, *6*(4), 1615–1625. https://doi.org/10.1002/nop2.369

Banner, D., Bains, M., Carroll, S., Kandola, D. K., Rolfe, D. E., Wong, C., & Graham, I. D. (2019). Patient and public engagement in integrated knowledge translation research: Are we there yet? *Research Involvement and Engagement*, *5*(1), 8. https://doi.org/10.1186/s40900-019-0139-1

Borowitz, S. M., Cox, D. J., Kovatchev, B., Ritterband, L. M., Sheen, J., & Sutphen, J. (2005). Treatment of childhood constipation by primary care physicians: Efficacy and predictors of outcome. *Pediatrics*, *115*(4), 873–877. https://doi.org/10.1542/peds.2004-0537

Bradbury, N. A. (2016). Attention span during lectures: 8 seconds, 10 minutes, or more? *Advances in Physiology Education*, *40*(4), 509–513. https://doi.org/10.1152/advan.00109.2016

Brame, C. J. (2016). Effective educational videos: Principles and guidelines for maximizing student learning from video content. *CBE—Life Sciences Education*, *15*(4), es6. https://doi.org/10.1187/cbe.16-03-0125

Dahlstrom, M. F. (2014). Using narratives and storytelling to communicate science with nonexpert audiences. *Proceedings of the National Academy of Sciences of the United States of America*, *111 Suppl 4*, 13614–13620. https://doi.org/10.1073/pnas.1320645111

Esmail, L., Moore, E., & Rein, A. (2015). Evaluating patient and stakeholder engagement in research: Moving from theory to practice. *Journal of Comparative Effectiveness Research*, *4*(2), 133–145. https://doi.org/10.2217/cer.14.79

Farrell, M., Holmes, G., Coldicutt, P., & Peak, M. (2003). Management of childhood constipation: Parents' experiences. *Journal of Advanced Nursing*, *44*(5), 479–489. https://doi.org/10.1046/j.0309-2402.2003.02831.x

Finkler, W., & Leon, B. (2019). The power of storytelling and video: A visual rhetoric for science communication. *Journal of Science Communication*, *18*(5), A02. https://doi.org/10.22323/2.18050202

Fønhus, M. S., Dalsbø, T. K., Johansen, M., Fretheim, A., Skirbekk, H., & Flottorp, S. A. (2018). Patient-mediated interventions to improve professional practice. *Cochrane Database of Systematic Reviews*, *9*. https://doi.org/10.1002/14651858.CD012472.pub2

Glick, A. F., Farkas, J. S., Rosenberg, R. E., Mendelsohn, A. L., Tomopoulos, S., Fierman, A. H., Dreyer, B. P., Migotsky, M., Melgar, J., & Yin, H. S. (2020). Accuracy of parent perception of comprehension of discharge instructions: Role of plan complexity and health literacy. *Academic Pediatrics*, *20*(4), 516–523. https://doi.org/10.1016/j.acap.2020.01.002

Green, M. C., & Brock, T. C. (2000). The role of transportation in the persuasiveness of public narratives. *Journal of Personality and Social Psychology*, *79*(5), 701–721. https://doi.org/10.1037/0022-3514.79.5.701

Hamilton, C. B., Hoens, A. M., Backman, C. L., McKinnon, A. M., McQuitty, S., English, K., & Li, L. C. (2018). An empirically based conceptual framework for fostering meaningful patient engagement in research. *Health Expectations*, *21*(1), 396–406. https://doi.org/10.1111/hex.12635

Houghton, D., Horgan, L., & Boldy, D. (2016). An exploratory study of aspects related to current inpatient and nurse-led clinic management of idiopathic childhood constipation. *Collegian*, *23*(2), 151–158. https://doi.org/10.1016/j.colegn.2015.04.002

Inan, M., Aydiner, C. Y., Tokuc, B., Aksu, B., Ayvaz, S., Ayhan, S., Ceylan, T., & Basaran, U. N. (2007). Factors associated with childhood constipation. *Journal of Paediatrics and Child Health*, *43*(10), 700–706. https://doi.org/10.1111/j.1440-1754.2007.01165.x

Ismail, N., Ratchford, I., Proudfoot, C., & Gibbs, J. (2011). Impact of a nurse-led clinic for chronic constipation in children. *Journal of Child Health Care*, *15*(3), 221–229. https://doi.org/10.1177/1367493511406568

Jones, M. D. (2014). Communicating climate change: Are stories better than 'just the facts'? *Policy Studies Journal*, *42*(4), 644–673. https://doi.org/10.1111/psj.12072

Kim, J., Guo, P. J., Seaton, D. T., Mitros, P., Gajos, K. Z., & Miller, R. C. (2014). Understanding in-video dropouts and interaction peaks in online lecture videos. *Proceedings of the First ACM Conference on Learning @ Scale Conference*, 31–40. https://doi.org/10.1145/2556325.2566237

Koppen, I. J. N., Vriesman, M. H., Tabbers, M. M., Di Lorenzo, C., & Benninga, M. A. (2018). Awareness and implementation of the 2014 ESPGHAN/NASPGHAN guideline for childhood functional constipation. *Journal of Pediatric Gastroenterology and Nutrition*, *66*(5), 732–737. https://doi.org/10.1097/MPG.0000000000001786

Kuizenga-Wessel, S., Heckert, S. L., Tros, W., van Etten-Jamaludin, F. S., Benninga, M. A., & Tabbers, M. M. (2016). Reporting on outcome measures of functional constipation in children-a systematic review. *Journal of Pediatric Gastroenterology & Nutrition*, *62*(6), 840–846. https://doi.org/10.1097/MPG.0000000000001110

Lau, K. H. V., Farooque, P., Leydon, G., Schwartz, M. L., Sadler, R. M., & Moeller, J. J. (2018). Using learning analytics to evaluate a video-based lecture series. *Medical Teacher*, *40*(1), 91–98. https://doi.org/10.1080/0142159X.2017.1395001

Lavallee, D. C., Williams, C. J., Tambor, E. S., & Deverka, P. A. (2012). Stakeholder engagement in comparative effectiveness research: How will we measure success? *Journal of Comparative Effectiveness Research*, *1*(5), 397–407.

Lee, H., Kim, M., Cooley, M. E., Kiang, P. N., Kim, D., Tang, S., Shi, L., Thiem, L., Kan, P., Peou, S., Touch, C., Chea, P., & Allison, J. (2018). Using narrative intervention for HPV vaccine behavior change among Khmer mothers and daughters: A pilot RCT to examine feasibility, acceptability, and preliminary effectiveness. *Applied Nursing Research*, *40*, 51–60. https://doi.org/10.1016/j.apnr.2017.12.008

Lu, P. L., Koppen, I. J. N., Orsagh-Yentis, D. K., Leonhart, K., Ambeba, E. J., Deans, K. J., Minneci, P. C., Teich, S., Diefenbach, K. A., Alpert, S. A., Benninga, M. A., Yacob, D., & Lorenzo, C. D. (2018). Sacral nerve stimulation for constipation and fecal incontinence in children: Long-term outcomes, patient benefit, and parent satisfaction. *Neurogastroenterology & Motility*, *30*(2), e13184. https://doi.org/10.1111/nmo.13184

Manafo, E., Petermann, L., Mason-Lai, P., & Vandall-Walker, V. (2018). Patient engagement in Canada: A scoping review of the 'how' and 'what' of patient engagement in health research. *Health Research Policy and Systems*, *16*, 5. https://doi.org/10.1186/s12961-018-0282-4

Mugie, S. M., Di Lorenzo, C., & Benninga, M. A. (2011). Constipation in childhood. *Nature Reviews Gastroenterology & Hepatology*, *8*(9), 502–511. https://doi.org/10.1038/nrgastro.2011.130

Neill, S., Roland, D., Jones, C. H., Thompson, M., & Lakhanpaul, M. (2015). Information resources to aid parental decision-making on when to seek medical care for their acutely sick child: A narrative systematic review. *BMJ Open*, *5*(12), e008280. https://doi.org/10.1136/bmjopen-2015-008280

Oostenbrink, R., Jongman, H., Landgraf, J. M., Raat, H., & Moll, H. A. (2010). Functional abdominal complaints in pre-school children: Parental reports of health-related quality of life. *Quality of Life Research: An International Journal of Quality of Life Aspects of Treatment, Care and Rehabilitation*, *19*(3), 363–369. https://doi.org/10.1007/s11136-009-9583-y

Reid, K., Hartling, L., Ali, S., Le, A., Norris, A., & Scott, S. D. (2017). Development and usability evaluation of an art and narrative-based knowledge translation tool for parents with a child with pediatric chronic pain: Multi-method study. *Journal of Medical Internet Research*, *19*(12), e412. https://doi.org/10.2196/jmir.8877

Rolfe, D. E., Ramsden, V. R., Banner, D., & Graham, I. D. (2018). Using qualitative Health Research methods to improve patient and public involvement and engagement in research. *Research Involvement and Engagement*, *4*(1), 49. https://doi.org/10.1186/s40900-018-0129-8

Sood, M., Lichtlen, P., & Perez, M. C. (2018). Unmet needs in pediatric functional constipation. *Clinical Pediatrics*, *57*(13), 1489–1495. https://doi.org/10.1177/0009922818774343

Staniszewska, S., & Denegri, S. (2013). Patient and public involvement in research: Future challenges. *Evidence-Based Nursing*, *16*(3), 69–69. https://doi.org/10.1136/eb-2013-101406

Sullivan, P. B., Burnett, C. A., & Juszczak, E. (2006). Parent satisfaction in a nurse led clinic compared with a paediatric gastroenterology clinic for the management of intractable, functional constipation. *Archives of Disease in Childhood*, *91*(6), 499–501. https://doi.org/10.1136/adc.2005.087486

Tabbers, M. M., DiLorenzo, C., Berger, M. Y., Faure, C., Langendam, M. W., Nurko, S., Staiano, A., Vandenplas, Y., & Benninga, M. A. (2014). Evaluation and treatment of functional constipation in infants and children: Evidence-based recommendations from ESPGHAN and NASPGHAN. *Journal of Pediatric Gastroenterology and Nutrition*, *58*(2), 258–274.

Yang, C. H., & Punati, J. (2015). Practice patterns of pediatricians and trainees for the management of functional constipation compared with 2006 NASPGHAN guidelines. *Journal of Pediatric Gastroenterology and Nutrition*, *60*(3), 308–311. https://doi.org/10.1097/MPG.0000000000000591

Bibliography

Abelson, J., Li, K., Wilson, G., Shields, K., Schneider, C., & Boesveld, S. (2016). Supporting quality public and patient engagement in health system organizations: Development and usability testing of the Public and Patient Engagement Evaluation Tool. *Health Expectations*, *19*(4), 817–827. https://doi.org/10.1111/hex.12378

Addington-Hall, J., & Kalra, L. (2001). Who should measure quality of life? *BMJ*, *322*(7299), 1417–1420. https://doi.org/10.1136/bmj.322.7299.1417

Alberta Health Services. (2021). *About Us*. Alberta Health Services. https://www.albertahealthservices.ca/stollery/Page14264.aspx

Amirav, I., Vandall-Walker, V., Rasiah, J., & Saunders, L. (2017). Patient and researcher engagement in health research: A parent's perspective. *Pediatrics*, *140*(3), e20164127. https://doi.org/10.1542/peds.2016-4127

Archibald, M. M., Caine, V., Ali, S., Hartling, L., & Scott, S. D. (2015). What is left unsaid: An interpretive description of the information needs of parents of children with asthma. *Research in Nursing & Health*, *38*(1), 19–28. https://doi.org/10.1002/nur.21635

Archibald, M. M., Hartling, L., Ali, S., Caine, V., & Scott, S. D. (2018). Developing "My Asthma Diary": A process exemplar of a patient-driven arts-based knowledge translation tool. *BMC Pediatrics*, *18*(1), 186. https://doi.org/10.1186/s12887-018-1155-2

Archibald, M. M., & Scott, S. D. (2019). Learning from usability testing of an arts-based knowledge translation tool for parents of a child with asthma. *Nursing Open*, *6*(4), 1615–1625. https://doi.org/10.1002/nop2.369

Banaszkiewicz, A., Topczewska-Cabanek, A., Malewicz, K., & Szajewska, H. (2005). Awareness of high-fiber products by parents of children with constipation: Questionnaire study. *Pediatria Wspolczesna*, *7*(1), 33–35.

Banner, D., Bains, M., Carroll, S., Kandola, D. K., Rolfe, D. E., Wong, C., & Graham, I. D. (2019). Patient and public engagement in integrated knowledge translation research: Are we there yet? *Research Involvement and Engagement*, *5*(1), 8. https://doi.org/10.1186/s40900-019-0139-1

Benninga, M. A., Nurko, S., Faure, C., Hyman, P. E., St. James Roberts, I., & Schechter, N. L. (2016). Childhood functional gastrointestinal disorders: Neonate/toddler. *Gastroenterology*, *150*(6), 1443-1455.e2. https://doi.org/10.1053/j.gastro.2016.02.016

Bernardbonnin, A. C., Haley, N., Belanger, S., & Nadeau, D. (1993). Parental and patient perceptions about encopresis and its treatment. *Journal of Developmental and Behavioral Pediatrics*, *14*(6), 397–400.

Boivin, A., L'Espérance, A., Gauvin, F.-P., Dumez, V., Macaulay, A. C., Lehoux, P., & Abelson, J. (2018). Patient and public engagement in research and health system decision making: A systematic review of evaluation tools. *Health Expectations*, *21*(6), 1075–1084. https://doi.org/10.1111/hex.12804

Bongers, M. E. J., van Dijk, M., Benninga, M. A., & Grootenhuis, M. A. (2009). Health related quality of life in children with constipation-associated fecal incontinence. *The Journal of Pediatrics*, *154*(5), 749-753.e1. https://doi.org/10.1016/j.jpeds.2008.11.029

Bongers, M. E. J., van Wijk, M. P., Reitsma, J. B., & Benninga, M. A. (2010). Long-term prognosis for childhood constipation: Clinical outcomes in adulthood. *Pediatrics*, *126*(1), e156–e162. https://doi.org/10.1542/peds.2009-1009

Borowitz, S. M., Cox, D. J., Kovatchev, B., Ritterband, L. M., Sheen, J., & Sutphen, J. (2005). Treatment of childhood constipation by primary care physicians: Efficacy and predictors of outcome. *Pediatrics*, *115*(4), 873–877. https://doi.org/10.1542/peds.2004-0537

Bradbury, N. A. (2016). Attention span during lectures: 8 seconds, 10 minutes, or more? *Advances in Physiology Education*, *40*(4), 509–513. https://doi.org/10.1152/advan.00109.2016

Brame, C. J. (2016). Effective educational videos: Principles and guidelines for maximizing student learning from video content. *CBE—Life Sciences Education*, *15*(4), es6. https://doi.org/10.1187/cbe.16-03-0125

Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology*, *3*(2), 77–101. https://doi.org/10.1191/1478088706qp063oa

Brazzelli, M., Griffiths, P. V., Cody, J. D., & Tappin, D. (2011). Behavioural and cognitive interventions with or without other treatments for the management of faecal incontinence in children. *Cochrane Database of Systematic Reviews*, *12*. https://doi.org/10.1002/14651858.CD002240.pub4

Brennan-Parsons, S. (2000). *Mothers' experiences in living with a child with encopresis- A phenomenological study.pdf*. Memorial University of Newfoundland.

Brett, J., Staniszewska, S., Mockford, C., Herron-Marx, S., Hughes, J., Tysall, C., & Suleman, R. (2014a). Mapping the impact of patient and public involvement on health and social care research: A systematic review. *Health Expectations*, *17*(5), 637–650. https://doi.org/10.1111/j.1369-7625.2012.00795.x

Brett, J., Staniszewska, S., Mockford, C., Herron-Marx, S., Hughes, J., Tysall, C., & Suleman, R. (2014b). A systematic review of the impact of patient and public involvement on service users, researchers and communities. *The Patient - Patient-Centered Outcomes Research*, *7*(4), 387–395. https://doi.org/10.1007/s40271-014-0065-0

Bryman, A. (2006). Integrating quantitative and qualitative research: How is it done? *Qualitative Research*, *6*(1), 97–113. https://doi.org/10.1177/1468794106058877

Buljan, I., Malički, M., Wager, E., Puljak, L., Hren, D., Kellie, F., West, H., Alfirević, Ž., & Marušić, A. (2018). No difference in knowledge obtained from infographic or plain language summary of a Cochrane systematic review: Three randomized controlled trials. *Journal of Clinical Epidemiology*, *97*, 86–94. https://doi.org/10.1016/j.jclinepi.2017.12.003

Canadian Institutes of Health Research. (2014). *Strategy for patient-oriented research—Patient engagement framework*. http://www.cihr-irsc.gc.ca/e/48413.html

Centre for Reviews and Dissemination. (2008). *Systematic reviews: CRD's guidance for undertaking reviews in health care*. University of York.

Chinn, P. L., & Kramer, M. K. (2015). *Knowledge development in nursing: Theory and process* (9th ed.). Elsevier Mosby.

Choung, R. S., Shah, N. D., Chitkara, D., Branda, M. E., Van Tilburg, M. A., Whitehead, W. E., Katusic, S. K., Locke, G. R. I., & Talley, N. J. (2011). Direct medical costs of constipation from childhood to early adulthood: A population-based birth cohort study. *Journal of Pediatric Gastroenterology and Nutrition*, *52*(1), 47–54. https://doi.org/10.1097/MPG.0b013e3181e67058

Christian, B. J. (2017). Translational research—The intersection between sociocultural and environmental factors and the health of children and families. *Journal of Pediatric*

Nursing: Nursing Care of Children and Families, *37*, 127–131. https://doi.org/10.1016/j.pedn.2017.09.013

Cunningham, R. M., & Boom, J. A. (2013). Telling stories of vaccine-preventable diseases: Why it works. *South Dakota Medicine*, 21–26.

Curran, J. A., Bishop, A., Chorney, J., MacEachern, L., & Mackay, R. (2018). Partnering with parents to advance child health research. *Healthcare Management Forum*, *31*(2), 45–50. https://doi.org/10.1177/0840470417744568

Dahlstrom, M. F. (2014). Using narratives and storytelling to communicate science with nonexpert audiences. *Proceedings of the National Academy of Sciences of the United States of America*, *111 Suppl 4*, 13614–13620. https://doi.org/10.1073/pnas.1320645111

Devitt, P., Thornley, E., & Hinks, M. (2007). An evaluation of an inter-disciplinary constipation clinic for childhood constipation. *Journal of Research in Nursing*, *12*(5), 539–547. https://doi.org/10.1177/1744987107079589

Doane, G. H., & Varcoe, C. (2007). Relational practice and nursing obligations. *Advances in Nursing Science*, *30*(3), 192–205. https://doi.org/10.1097/01.ANS.0000286619.31398.fc

Doane, G., & Varcoe, C. (2005). Toward compassionate action: Pragmatism and the inseparability of theory/practice. *Advances in Nursing Science*, *28*(1), 81–90.

Dolgun, E., Yavuz, M., Çelik, A., & Ergün, M. O. (2013). The effects of constipation on the quality of life of children and mothers. *The Turkish Journal of Pediatrics*, 7.

Domecq, J. P., Prutsky, G., Elraiyah, T., Wang, Z., Nabhan, M., Shippee, N., Brito, J. P., Boehmer, K., Hasan, R., Firwana, B., Erwin, P., Eton, D., Sloan, J., Montori, V., Asi, N., Abu Dabrh, A. M., & Murad, M. H. (2014). Patient engagement in research: A systematic review. *BMC Health Services Research*, *14*, 89. https://doi.org/10.1186/1472-6963-14-89

Eltorki, M., Landry, P., & TREKK Network. (2019). *Bottom line recommendations for the treatment and management of pediatric constipation*. https://trekk.ca/resources?external_resource_type=Quick_glance&tag_id=D003248

Esmail, L., Moore, E., & Rein, A. (2015). Evaluating patient and stakeholder engagement in research: Moving from theory to practice. *Journal of Comparative Effectiveness Research*, *4*(2), 133–145. https://doi.org/10.2217/cer.14.79

Evans, D., Coad, J., Cottrell, K., Dalrymple, J., Davies, R., Donald, C., Laterza, V., Long, A., Longley, A., Moule, P., Pollard, K., Powell, J., Puddicombe, A., Rice, C., & Sayers, R. (2014). *Public involvement in research: Assessing impact through a realist evaluation*. http://www.ncbi.nlm.nih.gov/books/NBK260168/

Farrell, M., Holmes, G., Coldicutt, P., & Peak, M. (2003). Management of childhood constipation: Parents' experiences. *Journal of Advanced Nursing*, *44*(5), 479–489. https://doi.org/10.1046/j.0309-2402.2003.02831.x

Finkler, W., & Leon, B. (2019). The power of storytelling and video: A visual rhetoric for science communication. *Journal of Science Communication*, *18*(5), A02. https://doi.org/10.22323/2.18050202

Fix, G. M., Houston, T. K., Barker, A. M., Wexler, L., Cook, N., Volkman, J. E., & Bokhour, B. G. (2012). A novel process for integrating patient stories into patient education interventions: Incorporating lessons from theater arts. *Patient Education and Counseling*, *88*(3), 455–459. https://doi.org/10.1016/j.pec.2012.06.012

Fønhus, M. S., Dalsbø, T. K., Johansen, M., Fretheim, A., Skirbekk, H., & Flottorp, S. A. (2018). Patient-mediated interventions to improve professional practice. *Cochrane Database of Systematic Reviews*, *9*. https://doi.org/10.1002/14651858.CD012472.pub2

Frank, L., Forsythe, L., Ellis, L., Schrandt, S., Sheridan, S., Gerson, J., Konopka, K., & Daugherty, S. (2015). Conceptual and practical foundations of patient engagement in research at the patient-centered outcomes research institute. *Quality of Life Research*, *24*(5), 1033–1041. https://doi.org/10.1007/s11136-014-0893-3

Glick, A. F., Farkas, J. S., Rosenberg, R. E., Mendelsohn, A. L., Tomopoulos, S., Fierman, A. H., Dreyer, B. P., Migotsky, M., Melgar, J., & Yin, H. S. (2020). Accuracy of parent perception of comprehension of discharge instructions: Role of plan complexity and health literacy. *Academic Pediatrics*, *20*(4), 516–523. https://doi.org/10.1016/j.acap.2020.01.002

Goding, A. (2013). *Storytelling: Reflecting on oral narratives and cultures*. Cognella.

Goodman, M. S., Ackermann, N., Bowen, D. J., & Thompson, V. (2019). Content validation of a quantitative stakeholder engagement measure. *Journal of Community Psychology*, *47*(8), 1937–1951. https://doi.org/10.1002/jcop.22239

Gordon, M., MacDonald, J. K., Parker, C. E., Akobeng, A. K., & Thomas, A. G. (2016). Osmotic and stimulant laxatives for the management of childhood constipation. *Cochrane Database of Systematic Reviews*, *8*. https://doi.org/10.1002/14651858.CD009118.pub3

Government of Canada, C. I. of H. R. (2012, January 10). *Patient engagement—CIHR*. http://www.cihr-irsc.gc.ca/e/45851.html

Green, M. C., & Brock, T. C. (2000). The role of transportation in the persuasiveness of public narratives. *Journal of Personality and Social Psychology*, *79*(5), 701–721. https://doi.org/10.1037/0022-3514.79.5.701

Greene, J. C., Caracelli, V. J., & Graham, W. F. (1989). Toward a conceptual framework for mixed-method evaluation designs. *Educational Evaluation and Policy Analysis*, *11*(3), 255–274. https://doi.org/10.3102/01623737011003255

Hamilton, C. B., Hoens, A. M., Backman, C. L., McKinnon, A. M., McQuitty, S., English, K., & Li, L. C. (2018). An empirically based conceptual framework for fostering meaningful patient engagement in research. *Health Expectations*, *21*(1), 396–406. https://doi.org/10.1111/hex.12635

Hamilton, C. B., Leese, J. C., Hoens, A. M., & Li, L. C. (2017). Framework for advancing the reporting of patient engagement in rheumatology research projects. *Current Rheumatology Reports*, *19*(7), 38. https://doi.org/10.1007/s11926-017-0666-4

Hartling, L., Elliott, S. A., Buckreus, K., Leung, J., & Scott, S. D. (2021). Development of a parent advisory group to inform a research program for knowledge translation in child health: A patient-oriented evaluation [manuscript submitted for publication]. *Research Involvement and Engagement*.

Hartling, L., Scott, S. D., Johnson, D. W., Bishop, T., & Klassen, T. P. (2013). A randomized controlled trial of storytelling as a communication tool. *PLOS ONE*, *8*(10), e77800. https://doi.org/10.1371/journal.pone.0077800

Hartling, L., Scott, S., Pandya, R., Johnson, D., Bishop, T., & Klassen, T. P. (2010). Storytelling as a communication tool for health consumers: Development of an intervention for parents of children with croup. *BMC Pediatrics*, *10*, 64. https://doi.org/10.1186/1471-2431-10-64

Hasson, F., Keeney, S., & McKenna, H. (2000). Research guidelines for the Delphi survey technique. *Journal of Advanced Nursing*, *32*(4), 1008–1015. https://doi.org/10.1046/j.1365-2648.2000.t01-1-01567.x

Holman, K. S. (2012). *Parent and child functioning in a community sample of children with constipation*. University of Wisconsin - Milwaukee.

Hong, Q. N., Pluye, P., Bartlett, G., Boardman, F., Cargo, M., Dagenais, P., Gagnon, M.-P., Griffiths, F., Nicolau, B., O'Cathain, A., Rousseau, M.-C., & Vedel, I. (2018). *Mixed Methods Appraisal Tool (MMAT), version 2018* (Registration of Copyright #1148552). Canadian Intellectual Property Office, Industry Canada. http://mixedmethodsappraisaltoolpublic.pbworks.com/w/file/fetch/127916259/MMAT_2018_criteria-manual_2018-08-01_ENG.pdf

Hornbæk, K. (2006). Current practice in measuring usability: Challenges to usability studies and research. *International Journal of Human-Computer Studies*, *64*(2), 79–102. https://doi.org/10.1016/j.ijhcs.2005.06.002

Houghton, D., Horgan, L., & Boldy, D. (2016). An exploratory study of aspects related to current inpatient and nurse-led clinic management of idiopathic childhood constipation. *Collegian*, *23*(2), 151–158. https://doi.org/10.1016/j.colegn.2015.04.002

Houston, T. K., Cherrington, A., Coley, H. L., Robinson, K. M., Trobaugh, J. A., Williams, J. H., Foster, P. H., Ford, D. E., Gerber, B. S., Shewchuk, R. M., & Allison, J. J. (2011). The art and science of patient storytelling-harnessing narrative communication for behavioral interventions: The ACCE project. *Journal of Health Communication*, *16*(7), 686–697. https://doi.org/10.1080/10810730.2011.551997

Hsieh, H.-F., & Shannon, S. E. (2005). Three approaches to qualitative content analysis. *Qualitative Health Research*, *15*(9), 1277–1288. https://doi.org/10.1177/1049732305276687

Hyams, J. S., Di Lorenzo, C., Saps, M., Shulman, R. J., Staiano, A., & van Tilburg, M. (2016). Childhood functional gastrointestinal disorders: Child/adolescent. *Gastroenterology*, *150*(6), 1456-1468.e2. https://doi.org/10.1053/j.gastro.2016.02.015

Inan, M., Aydiner, C. Y., Tokuc, B., Aksu, B., Ayvaz, S., Ayhan, S., Ceylan, T., & Basaran, U. N. (2007). Factors associated with childhood constipation. *Journal of Paediatrics and Child Health*, *43*(10), 700–706. https://doi.org/10.1111/j.1440-1754.2007.01165.x

International Association for Public Participation Canada. (n.d.). *IAP2 Canada—Public participation spectrum*. Retrieved 12 April 2018, from http://iap2canada.ca/page-1020549

Ismail, N., Ratchford, I., Proudfoot, C., & Gibbs, J. (2011). Impact of a nurse-led clinic for chronic constipation in children. *Journal of Child Health Care*, *15*(3), 221–229. https://doi.org/10.1177/1367493511406568

Jackson, C., Cheater, F. M., & Reid, I. (2008). A systematic review of decision support needs of parents making child health decisions. *Health Expectations*, *11*(3), 232–251. https://doi.org/10.1111/j.1369-7625.2008.00496.x

Joinson, C., Heron, J., Buter, U., von Gontard, A., & Avon Longitudinal Study of Parents and Children Study Team. (n.d.). Psychological differences between children with and without soiling problems. *Pediatrics*, *117*(5), 1575–1584.

Jones, M. D. (2014). Communicating climate change: Are stories better than ‘just the facts’? *Policy Studies Journal*, *42*(4), 644–673. https://doi.org/10.1111/psj.12072

Jurgens, H., Oster, C., & Fereday, J. (2011). Management of chronic functional constipation in children: A review of the literature. *Neonatal, Paediatric & Child Health Nursing*, *14*(2), 23–28.

Kaugars, A. S., Silverman, A., Kinservik, M., Heinze, S., Reinemann, L., Sander, M., Schneider, B., & Sood, M. (2010). Families' perspectives on the effect of constipation and fecal incontinence on quality of life. *Journal of Pediatric Gastroenterology and Nutrition*, *51*(6), 747–752. https://doi.org/10.1097/MPG.0b013e3181de0651

Kim, J., Guo, P. J., Seaton, D. T., Mitros, P., Gajos, K. Z., & Miller, R. C. (2014). Understanding in-video dropouts and interaction peaks in online lecture videos. *Proceedings of the First ACM Conference on Learning @ Scale Conference*, 31–40. https://doi.org/10.1145/2556325.2566237

Klages, K. L., Berlin, K. S., Silverman, A. H., Mugie, S., Di Lorenzo, C., Nurko, S., Ponnambalam, A., Sanghavi, R., & Sood, M. R. (2016). Empirically derived patterns of pain, stooling, and incontinence and their relations to health-related quality of life among youth with chronic constipation. *Journal of Pediatric Psychology*, jsw068. https://doi.org/10.1093/jpepsy/jsw068

Koppen, I. J. N., van Wassenaer, E. A., Barendsen, R. W., Brand, P. L., & Benninga, M. A. (2018). Adherence to polyethylene glycol treatment in children with functional constipation is associated with parental illness perceptions, satisfaction with treatment, and perceived treatment convenience. *The Journal of Pediatrics*, *199*, 132-139.e1. https://doi.org/10.1016/j.jpeds.2018.03.066

Koppen, I. J. N., Vriesman, M. H., Saps, M., Rajindrajith, S., Shi, X., van Etten-Jamaludin, F. S., Di Lorenzo, C., Benninga, M. A., & Tabbers, M. M. (2018). Prevalence of functional defecation disorders in children: A systematic review and meta-analysis. *The Journal of Pediatrics*, *198*, 121-130.e6. https://doi.org/10.1016/j.jpeds.2018.02.029

Koppen, I. J. N., Vriesman, M. H., Tabbers, M. M., Di Lorenzo, C., & Benninga, M. A. (2018). Awareness and implementation of the 2014 ESPGHAN/NASPGHAN guideline for childhood functional constipation. *Journal of Pediatric Gastroenterology and Nutrition*, *66*(5), 732–737. https://doi.org/10.1097/MPG.0000000000001786

Kovacic, K., Sood, M. R., Mugie, S., Di Lorenzo, C., Nurko, S., Heinz, N., Ponnambalam, A., Beesley, C., Sanghavi, R., & Silverman, A. H. (2015). A multicenter study on childhood constipation and fecal incontinence: Effects on quality of life. *The Journal of Pediatrics*, *166*(6), 1482-1487.e1. https://doi.org/10.1016/j.jpeds.2015.03.016

Kuizenga-Wessel, S., Heckert, S. L., Tros, W., van Etten-Jamaludin, F. S., Benninga, M. A., & Tabbers, M. M. (2016). Reporting on outcome measures of functional constipation in children-a systematic review. *Journal of Pediatric Gastroenterology & Nutrition*, *62*(6), 840–846. https://doi.org/10.1097/MPG.0000000000001110

Lau, K. H. V., Farooque, P., Leydon, G., Schwartz, M. L., Sadler, R. M., & Moeller, J. J. (2018). Using learning analytics to evaluate a video-based lecture series. *Medical Teacher*, *40*(1), 91–98. https://doi.org/10.1080/0142159X.2017.1395001

Lavallee, D. C., Williams, C. J., Tambor, E. S., & Deverka, P. A. (2012). Stakeholder engagement in comparative effectiveness research: How will we measure success? *Journal of Comparative Effectiveness Research*, *1*(5), 397–407.

Lee, H., Fawcett, J., & DeMarco, R. (2016). Storytelling/narrative theory to address health communication with minority populations. *Applied Nursing Research*, *30*, 58–60. https://doi.org/10.1016/j.apnr.2015.09.004

Lee, H., Kim, M., Cooley, M. E., Kiang, P. N., Kim, D., Tang, S., Shi, L., Thiem, L., Kan, P., Peou, S., Touch, C., Chea, P., & Allison, J. (2018). Using narrative intervention for HPV vaccine behavior change among Khmer mothers and daughters: A pilot RCT to examine feasibility, acceptability, and preliminary effectiveness. *Applied Nursing Research*, *40*, 51–60. https://doi.org/10.1016/j.apnr.2017.12.008

Liem, O., Harman, J., Benninga, M. A., Kelleher, K., Mousa, H., & Di Lorenzo, C. (2009). Health utilization and cost impact of childhood constipation in the United States. *The Journal of Pediatrics*, *154*(2), 258–262. https://doi.org/10.1016/j.jpeds.2008.07.060

Loening-Baucke, V. (2005). Prevalence, symptoms and outcome of constipation in infants and toddlers. *The Journal of Pediatrics*, *146*(3), 359–363. https://doi.org/10.1016/j.jpeds.2004.10.046

Loening-Baucke, V. (2007). Prevalence rates for constipation and faecal and urinary incontinence. *Archives of Disease in Childhood*, *92*(6), 486–489. https://doi.org/10.1136/adc.2006.098335

Lu, P. L., Koppen, I. J. N., Orsagh-Yentis, D. K., Leonhart, K., Ambeba, E. J., Deans, K. J., Minneci, P. C., Teich, S., Diefenbach, K. A., Alpert, S. A., Benninga, M. A., Yacob, D., & Lorenzo, C. D. (2018). Sacral nerve stimulation for constipation and fecal incontinence in children: Long-term outcomes, patient benefit, and parent satisfaction. *Neurogastroenterology & Motility*, *30*(2), e13184. https://doi.org/10.1111/nmo.13184

Malowitz, S., Green, M., Karpinski, A., Rosenberg, A., & Hyman, P. E. (2016). Age of onset of functional constipation. *Journal of Pediatric Gastroenterology and Nutrition*, *62*(4), 600–602. https://doi.org/10.1097/MPG.0000000000001011

Manafo, E., Petermann, L., Mason-Lai, P., & Vandall-Walker, V. (2018). Patient engagement in Canada: A scoping review of the 'how' and 'what' of patient engagement in health research. *Health Research Policy and Systems*, *16*, 5. https://doi.org/10.1186/s12961-018-0282-4

McCready, J. S. (2010). Jamesian pragmatism: A framework for working towards unified diversity in nursing knowledge development. *Nursing Philosophy*, *11*(3), 191–203. https://doi.org/10.1111/j.1466-769X.2010.00444.x

Meherali, S., Campbell, A., Hartling, L., & Scott, S. (2019). Understanding parents' experiences and information needs on pediatric acute otitis media: A qualitative study. *Journal of Patient Experience*, *6*(1), 53–61. https://doi.org/10.1177/2374373518771362

Moher, D., Liberati, A., Tetzlaff, J., & Altman, D. G. (2009). Preferred reporting items for systematic reviews and meta-analyses: The PRISMA statement. *BMJ*, *339*, b2535. https://doi.org/10.1136/bmj.b2535

Mousa, H. M., Barsamian, J. M., Benitez, A. J., Webster, J. M., & Fiorino, K. N. (2020). Pediatric constipation: An epidemiologic and physiologic approach to guide a stepwise approach to its management. *Current Treatment Options in Pediatrics*, *6*(3), 155–169. https://doi.org/10.1007/s40746-020-00199-z

Mugie, S. M., Benninga, M. A., & Di Lorenzo, C. (2011). Epidemiology of constipation in children and adults: A systematic review. *Best Practice & Research Clinical Gastroenterology*, *25*(1), 3–18. https://doi.org/10.1016/j.bpg.2010.12.010

Mugie, S. M., Di Lorenzo, C., & Benninga, M. A. (2011). Constipation in childhood. *Nature Reviews Gastroenterology & Hepatology*, *8*(9), 502–511. https://doi.org/10.1038/nrgastro.2011.130

Müller-Lissner, S. A., Kamm, M. A., Scarpignato, C., & Wald, A. (2005). Myths and misconceptions about chronic constipation. *Official Journal of the American College of Gastroenterology | ACG*, *100*(1), 232–242.

National Collaborating Centre for Women's and Children's Health. (2010). *Constipation in children and young people: Diagnosis and management of idiopathic childhood constipation in primary and secondary care*. RCOG Press. http://www.ncbi.nlm.nih.gov/books/NBK65365/

Neill, S., Roland, D., Jones, C. H., Thompson, M., & Lakhanpaul, M. (2015). Information resources to aid parental decision-making on when to seek medical care for their acutely sick child: A narrative systematic review. *BMJ Open*, *5*(12), e008280. https://doi.org/10.1136/bmjopen-2015-008280

Njeru, J. W., Patten, C. A., Hanza, M. M. K., Brockman, T. A., Ridgeway, J. L., Weis, J. A., Clark, M. M., Goodson, M., Osman, A., Porraz-Capetillo, G., Hared, A., Myers, A., Sia, I. G., & Wieland, M. L. (2015). Stories for change: Development of a diabetes digital storytelling intervention for refugees and immigrants to Minnesota using qualitative methods. *BMC Public Health*, *15*, 1311. https://doi.org/10.1186/s12889-015-2628-y

O'Brien, B., Harris, I., Beckman, T., Reed, D., & Cook, D. (2014). Standards for reporting qualitative research: A synthesis of recommendations. *Academic Medicine*, *89*(9), 1245–1251. https://doi.org/10.1097/ACM.0000000000000388

O'Cathain, A., Murphy, E., & Nicholl, J. (2008). The quality of mixed methods studies in health services research. *Journal of Health Services Research & Policy*, *13*(2), 92–98. https://doi.org/10.1258/jhsrp.2007.007074

Oostenbrink, R., Jongman, H., Landgraf, J. M., Raat, H., & Moll, H. A. (2010). Functional abdominal complaints in pre-school children: Parental reports of health-related quality of life. *Quality of Life Research: An International Journal of Quality of Life Aspects of Treatment, Care and Rehabilitation*, *19*(3), 363–369. https://doi.org/10.1007/s11136-009-9583-y

Pérez Jolles, M., Martinez, M., Garcia, S. J., Stein, G. L., Mentor Parent Group Members, & Thomas, K. C. (2017). Involving Latina/o parents in patient-centered outcomes research: Contributions to research study design, implementation and outcomes. *Health Expectations*, *20*(5), 992–1000. https://doi.org/10.1111/hex.12540

Pluye, P., Gagnon, M.-P., Griffiths, F., & Johnson-Lafleur, J. (2009). A scoring system for appraising mixed methods research, and concomitantly appraising qualitative, quantitative and mixed methods primary studies in mixed studies reviews. *International Journal of Nursing Studies*, *46*(4), 529–546. https://doi.org/10.1016/j.ijnurstu.2009.01.009

Popay, J., Roberts, H., Sowden, A., Petticrew, M., Arai, L., Rodgers, M., & Britten, N. (2006). *Guidance on the conduct of narrative synthesis in systematic reviews: A product from the ESRC methods programme* (Version 1).

Public & Patient Engagement | PPE Evaluation Tool. (n.d.). Retrieved 17 June 2020, from https://healthsci.mcmaster.ca/ppe/our-products/public-patient-engagement-evaluation-tool

Rajindrajith, S., Devanarayana, N. M., & Benninga, M. A. (2013). Review article: Faecal incontinence in children: epidemiology, pathophysiology, clinical evaluation and management. *Alimentary Pharmacology & Therapeutics*, *37*(1), 37–48. https://doi.org/10.1111/apt.12103

Rajindrajith, S., Devanarayana, N. M., Perera, B. J. C., & Benninga, M. A. (2016). Childhood constipation as an emerging public health problem. *World Journal of Gastroenterology*, *22*(30), 6864. https://doi.org/10.3748/wjg.v22.i30.6864

Reid, K., Hartling, L., Ali, S., Le, A., Norris, A., & Scott, S. D. (2017). Development and usability evaluation of an art and narrative-based knowledge translation tool for parents with a child with pediatric chronic pain: Multi-method study. *Journal of Medical Internet Research*, *19*(12), e412. https://doi.org/10.2196/jmir.8877

Restrepo, E., & Davis, L. (2003). Storytelling: Both art and therapeutic practice. *International Journal for Human Caring*, *7*(1), 43–48.

Risjord, M. W. (2010). *Nursing knowledge: Science, practice, and philosophy*. Wiley-Blackwell.

Rolfe, D. E., Ramsden, V. R., Banner, D., & Graham, I. D. (2018). Using qualitative Health Research methods to improve patient and public involvement and engagement in research. *Research Involvement and Engagement*, *4*(1), 49. https://doi.org/10.1186/s40900-018-0129-8

Rowan-Legg, A. & Canadian Paediatric Society. (2011). Managing functional constipation in children. *Paediatrics & Child Health*, *16*(10), 661–665.

Samson, D. (2004). *Encopresis and family attitudes* [University of Leicester (United Kingdom)]. https://regroup-production.s3.amazonaws.com/documents/ReviewReference/100380230/Encopresis%20and%20family%20attitudes.pdf?AWSAccessKeyId=AKIAJBZQODCMKJA4H7DA&Expires=1591192943&Signature=H3jSIlJ9T9KnFsaKatWFYqAVA98%3D

Scott, S. D., Hartling, L., O'Leary, K. A., Archibald, M., & Klassen, T. P. (2012). Stories – a novel approach to transfer complex health information to parents: A qualitative study. *Arts & Health*, *4*(2), 162–173. https://doi.org/10.1080/17533015.2012.656203

Selby, J. V., Beal, A., & Frank, L. (2012). The patient-centered outcomes research institute (PCORI) national priorities for research and initial research agenda. *Jama*, *307*(15), 1583–1584.

Snyder, H., & Engström, J. (2016). The antecedents, forms and consequences of patient involvement: A narrative review of the literature. *International Journal of Nursing Studies*, *53*, 351–378. https://doi.org/10.1016/j.ijnurstu.2015.09.008

Soobiah, C., Straus, S. E., Manley, G., Marr, S., Paus Jenssen, E., Teare, S., Hamid, J., Tricco, A. C., & Moore, A. (2019). Engaging knowledge users in a systematic review on the comparative effectiveness of geriatrician-led models of care is possible: A cross-sectional survey using the Patient Engagement Evaluation Tool. *Journal of Clinical Epidemiology*, *113*, 58–63. https://doi.org/10.1016/j.jclinepi.2019.05.015

Sood, M., Lichtlen, P., & Perez, M. C. (2018). Unmet needs in pediatric functional constipation. *Clinical Pediatrics*, *57*(13), 1489–1495. https://doi.org/10.1177/0009922818774343

Southwell, B. R. (2020). Treatment of childhood constipation: A synthesis of systematic reviews and meta-analyses. *Expert Review of Gastroenterology & Hepatology*, *14*(3), 163–174. https://doi.org/10.1080/17474124.2020.1733974

Spithoff, S., & Kahan, M. (2015). Primary care management of alcohol use disorder and at-risk drinking: Part 1: screening and assessment. *Canadian Family Physician*, *61*(6), 509–514.

Stacey, D., & Hill, S. (2013). Patient-direct and patient-mediated KT interventions. In *Knowledge translation in health care* (2nd ed., pp. 197–211). Wiley-Blackwell.

Staley, K. (2015). ‘Is it worth doing?’ Measuring the impact of patient and public involvement in research. *Research Involvement and Engagement*, *1*, 6. https://doi.org/10.1186/s40900-015-0008-5

Staller, K., & Cash, B. D. (2020). Myths and misconceptions about constipation: A new view for the 2020s. *Official Journal of the American College of Gastroenterology | ACG*, *115*(11), 1741–1745. https://doi.org/10.14309/ajg.0000000000000947

Staniszewska, S., Brett, J., Mockford, C., & Barber, R. (2011). The GRIPP checklist: Strengthening the quality of patient and public involvement reporting in research. *International Journal of Technology Assessment in Health Care*, *27*(4), 391–399. https://doi.org/10.1017/S0266462311000481

Staniszewska, S., & Denegri, S. (2013). Patient and public involvement in research: Future challenges. *Evidence-Based Nursing*, *16*(3), 69–69. https://doi.org/10.1136/eb-2013-101406

Staniszewska, S., Herron-Marx, S., & Mockford, C. (2008). Measuring the impact of patient and public involvement: The need for an evidence base. *International Journal for Quality in Health Care*, *20*(6), 373–374. https://doi.org/10.1093/intqhc/mzn044

Straus, S., Tetroe, J., & Graham, I. D. (Eds.). (2013). *Knowledge translation in health care: Moving from evidence to practice* (2 edition). BMJ Books.

Sullivan, P. B., Burnett, C. A., & Juszczak, E. (2006). Parent satisfaction in a nurse led clinic compared with a paediatric gastroenterology clinic for the management of intractable, functional constipation. *Archives of Disease in Childhood*, *91*(6), 499–501. https://doi.org/10.1136/adc.2005.087486

Supple, D., Roberts, A., Hudson, V., Masefield, S., Fitch, N., Rahmen, M., Flood, B., de Boer, W., Powell, P., & Wagers, S. (2015). From tokenism to meaningful engagement: Best practices in patient involvement in an EU project. *Research Involvement and Engagement*, *1*, 5. https://doi.org/10.1186/s40900-015-0004-9

Tabbers, M. M., DiLorenzo, C., Berger, M. Y., Faure, C., Langendam, M. W., Nurko, S., Staiano, A., Vandenplas, Y., & Benninga, M. A. (2014). Evaluation and treatment of functional constipation in infants and children: Evidence-based recommendations from ESPGHAN and NASPGHAN. *Journal of Pediatric Gastroenterology and Nutrition*, *58*(2), 258–274.

Tappin, D., Nawaz, S., McKay, C., MacLaren, L., Griffiths, P., & Mohammed, T. A. (2013). Development of an early nurse led intervention to treat children referred to secondary paediatric care with constipation with or without soiling. *BMC Pediatrics*, *13*(1), 1–9. https://doi.org/10.1186/1471-2431-13-193

Tarlier, D. (2005). Mediating the meaning of evidence through epistemological diversity. *Nursing Inquiry*, *12*(2), 126–134. https://doi.org/10.1111/j.1440-1800.2005.00262.x

Thompson, A. P., Le, A., Hartling, L., & Scott, S. D. (2020). Fading confidence: A qualitative exploration of parents' experiences caring for a febrile child. *Journal of Clinical Nursing*, *29*(5–6), 964–973. https://doi.org/10.1111/jocn.15165

Thompson, A. P., MacDonald, S. E., Wine, E., & Scott, S. D. (2020). An evaluation of parents' experiences of patient engagement in research to develop a digital knowledge translation tool: Protocol for a multi-method study. *JMIR Research Protocols*, *9*(8), e19108. https://doi.org/10.2196/19108

Thompson, A. P., MacDonald, S. E., Wine, E., & Scott, S. D. (2021). Understanding parents' experiences when caring for a child with functional constipation: Interpretive description study. *JMIR Pediatrics and Parenting*, *4*(1), e24851. https://doi.org/10.2196/24851

Thompson, A. P., Nesari, M., Hartling, L., & Scott, S. D. (2019). Parents' experiences and information needs related to childhood fever: A systematic review. *Patient Education and Counseling*. https://doi.org/10.1016/j.pec.2019.10.004

Thompson, A. P., Nesari, M., Hartling, L., & Scott, S. D. (2020). Parents' experiences and information needs related to childhood fever: A systematic review. *Patient Education and Counseling*, *103*(4), 750–763. https://doi.org/10.1016/j.pec.2019.10.004

Thompson, A. P., Wine, E., MacDonald, S. E., Campbell, A., & Scott, S. D. (2020). Parents' experiences and information needs while caring for a child with functional constipation: A systematic review. *Clinical Pediatrics*, *60*(3), 154–169. https://doi.org/10.1177/0009922820964457

Thorne, S. (2016). *Interpretive description: Qualitative research for applied practice* (2nd ed.). Routledge.

Thorne, S., & Darbyshire, P. (2005). Land mines in the field: A modest proposal for improving the craft of qualitative health research. *Qualitative Health Research*, *15*(8), 1105–1113. https://doi.org/10.1177/1049732305278502

Thorne, S., Reimer Kirkham, S., & MacDonald-Emes, J. (1997). Interpretive description: A noncategorical qualitative alternative for developing nursing knowledge. *Research in Nursing & Health*, *20*(2), 169–177. https://doi.org/10.1002/(SICI)1098-240X(199704)20:2<169::AID-NUR9>3.0.CO;2-I

Timmerman, M. E. W., Trzpis, M., & Broens, P. M. A. (2019). The problem of defecation disorders in children is underestimated and easily goes unrecognized: A cross-sectional study. *European Journal of Pediatrics*, *178*(1), 33–39. https://doi.org/10.1007/s00431-018-3243-6

Translating Emergency Knowledge for Kids. (n.d.). TREKK.Ca. Retrieved 17 June 2020, from https://trekk.ca/

Vaismoradi, M., Turunen, H., & Bondas, T. (2013). Content analysis and thematic analysis: Implications for conducting a qualitative descriptive study. *Nursing & Health Sciences*, *15*(3), 398–405. https://doi.org/10.1111/nhs.12048

van den Berg, M. M., Benninga, M. A., & Di Lorenzo, C. (2006). Epidemiology of childhood constipation: A systematic review. *The American Journal of Gastroenterology*, *101*(10), 2401–2409. https://doi.org/10.1111/j.1572-0241.2006.00771.x

van den Berg, M. M., van Rossum, C. H., de Lorijn, F., Reitsma, J. B., Di Lorenzo, C., & Benninga, M. A. (2005). Functional constipation in infants: A follow-up study. *The Journal of Pediatrics*, *147*(5), 700–704. https://doi.org/10.1016/j.jpeds.2005.05.032

van Engelenburg-van Lonkhuyzen, M. L., Bols, E. M. J., Benninga, M. A., Verwijs, W. A., & de Bie, R. A. (2017). Effectiveness of pelvic physiotherapy in children with functional constipation compared with standard medical care. *Gastroenterology*, *152*(1), 82–91. https://doi.org/10.1053/j.gastro.2016.09.015

van Tilburg, M. A. L., Squires, M., Blois-Martin, N., Williams, C., Benninga, M. A., Peeters, B., & Ulshen, M. (2012). Parental knowledge of fecal incontinence in children. *Journal of Pediatric Gastroenterology and Nutrition*, *55*(3), 283–287. https://doi.org/10.1097/MPG.0b013e31824cf555

Varni, J. W., Bendo, C. B., Nurko, S., Shulman, R. J., Self, M. M., Franciosi, J. P., Saps, M., & Pohl, J. F. (2015). Health-related quality of life in pediatric patients with functional and organic gastrointestinal diseases. *The Journal of Pediatrics*, *166*(1), 85-90.e2. https://doi.org/10.1016/j.jpeds.2014.08.022

Varni, J. W., Nurko, S., Shulman, R. J., Self, M. M., Saps, M., Bendo, C. B., Vaughan Dark, C., Pohl, J. F., & Consortium, for the P. Q. of L. I. G. S. M. T. S. (2015). Pediatric functional constipation gastrointestinal symptom profile compared with healthy controls. *Journal of Pediatric Gastroenterology and Nutrition*, *61*(4), 424–430. https://doi.org/10.1097/MPG.0000000000000869

Vriesman, M. H., Rajindrajith, S., Koppen, I. J. N., van Etten- Jamaludin, F. S., van Dijk, M., Devanarayana, N. M., Tabbers, M. M., & Benninga, M. A. (2019). Quality of life in children with functional constipation: A systematic review and meta-analysis. *The Journal of Pediatrics*, *214*, 141–150. https://doi.org/10.1016/j.jpeds.2019.06.059

Wang, C., Shang, L., Zhang, Y., Tian, J., Wang, B., Yang, X., Sun, L., Du, C., Jiang, X., & Xu, Y. (2013). Impact of functional constipation on health-related quality of life in preschool children and their families in Xi'an, China. *PLoS ONE*, *8*(10), e77273. https://doi.org/10.1371/journal.pone.0077273

Ward, P. R., Thompson, J., Barber, R., Armitage, C. J., Boote, J. D., Cooper, C. L., & Jones, G. L. (2010). Critical perspectives on 'consumer involvement' in health research: Epistemological dissonance and the know-do gap. *Journal of Sociology*, *46*(1), 63–82. https://doi.org/10.1177/1440783309351771

Wyatt, K. D., List, B., Brinkman, W. B., Prutsky Lopez, G., Asi, N., Erwin, P., Wang, Z., Domecq Garces, J. P., Montori, V. M., & LeBlanc, A. (2015). Shared decision making in pediatrics: A systematic review and meta-analysis. *Academic Pediatrics*, *15*(6), 573–583. https://doi.org/10.1016/j.acap.2015.03.011

Yang, C. H., & Punati, J. (2015). Practice patterns of pediatricians and trainees for the management of functional constipation compared with 2006 NASPGHAN guidelines. *Journal of Pediatric Gastroenterology and Nutrition*, *60*(3), 308–311. https://doi.org/10.1097/MPG.0000000000000591

Youssef, N. N., Langseder, A. L., Verga, B. J., Mones, R. L., & Rosh, J. R. (2005). Chronic childhood constipation is associated with impaired quality of life: A case-controlled study. *Journal of Pediatric Gastroenterology and Nutrition*, *41*(1), 56–60.

Zapata, B. C., Fernández-Alemán, J. L., Idri, A., & Toval, A. (2015). Empirical studies on usability of mhealth apps: A systematic literature review. *Journal of Medical Systems*, *39*(2), 1. https://doi.org/10.1007/s10916-014-0182-2