Abstract

While much research has been conducted on the experiences of individuals with inflammatory bowel diseases, there remains a dearth of research conducted on those affected by polyposis conditions. As a result, little is known about the lived experiences of those with polyposis conditions, especially in the cases of parents of pediatric patients with these conditions. This study qualitatively explored the lived experiences of parents of children with polyposis conditions, specifically Juvenile Polyposis Syndrome and Peutz-Jeghers Syndrome. Hermeneutic phenomenology was used to explore the lived experiences of seven parents of children diagnosed with polyposis conditions through semi-structured interviews. Collected data was analysed using Lindseth and Norberg (2004)’s Phenomenological Hermeneutical Method for Researching Lived Experience.

In total, four major themes comprising of twelve sub-themes were revealed. Parents discussed feeling grateful for the use of family-centred approaches by their children’s physicians as well as access to medical care for their children, which encouraged them to demonstrate a proactive approach towards their children’s health maintenance. Furthermore, they explained that while seeking information concerning their children’s conditions was anxiety-inducing, discussing their experiences with others with situations similar to theirs was validating and informative. The participants described the importance of advocating for their children within and outside of the medical system, and the responsibility they feel in teaching their children to undertake the advocating process for themselves. Lastly, the parents reflected on the impact their children’s diagnoses have had on their relationships with themselves, their families and their support networks. Overall, the findings from this study are in-line with findings
from prior research, except in the case of the ‘Teaching the children to speak for themselves’ theme which proves to be a novel contribution to the literature.

The shared key aspects of the phenomenon indicate that focus should be placed on the utilization of family-centred care by physicians, the development of support groups for parents, and on educating physicians on how to best facilitate parents as they model advocating behaviours to pediatric patients. This study provides insight into the lived experiences of parents of children with polyposis syndromes, informing the medical community of how the needs of this group can be better met. Furthermore, the qualitative nature of this research will provide the polyposis, chronic illness and rare illness literatures with information it has been lacking, using a valuable methodological perspective.
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Chapter 1: Introduction

Description of the Research Problem

Colorectal cancer is the third most common cancer in women and the fourth most common cancer in men worldwide (Parkin, Bray, Ferlay, & Pisani, 2005). Colorectal cancer can be attributed to individuals’ genetics in approximately 35% of diagnoses (Patel & Ahnen, 2012). Polyposis syndromes confer a significant risk of cancer in those diagnosed with such conditions, as polyps developing in the colon can become malignant (Giardiello, et al., 2000). These conditions are often diagnosed at a young age, prompted by either clinical symptoms or genetic testing based on family history (Ellis, 2004).

Although polyposis syndromes have been studied extensively in the literature from a biological perspective, research on the experiences of individuals and their families living with the conditions is lacking. Research exploring the psychosocial perspectives of individuals living with gastrointestinal conditions, other than polyposis conditions, indicate that affected individuals experience social isolation, and psychological distress (Casati, Toner, de Rooy, Drossman, & Maunder, 2000; Graff, Walker, & Berstein, 2009). Moreover, virtually no research exists on the experiences of parents raising children with polyposis conditions. (The term ‘parent’ will hereafter be used to refer to parents, caregivers, and legal guardians). As children represent an inherently vulnerable portion of the population, the responsibility of caring for polyposis-diagnosed children lays in parents’ hands. With the health psychology literature demonstrating evidence that parents of chronically ill children suffer pronounced responses to their children’s conditions, namely psychological distress, it is
likely that the diagnosis of a child with a polyposis syndrome confers vulnerability upon both pediatric patients and their parents (Kazak et al., 2005).

Unique to polyposis conditions are the conditions’ heightened likelihood of cancer development. Research indicates that individuals who have an increased risk of developing colorectal cancer often do not comply with screening recommendations due to their fear of cancer (Bleiker, et al., 2005). It is of paramount importance that the experiences of parents of children with polyposis syndromes be explored in order to gain insight into the ways in which they cope and adapt to their children’s illnesses and to better understand the effects such experiences have on the medical decisions they make for their children. Exploration would also give us better insight into the needs of these parents and how such needs can be met.

As such, this study seeks to explore the lived experiences of parents of children with polyposis syndromes. This thesis will focus on two polyposis syndromes, Peutz-Jeghers Syndrome and Juvenile Polyposis Syndrome. The qualitative approach guiding this study complements the predominantly quantitative data that currently dominates the research literature. The findings revealed within this study are framed in a way that can be translated to more appropriate support for parents, and subsequently, better care for their children. Furthermore, this research will help to fill the gaps in the literature pertaining to the experiences of parents of children with polyposis conditions, and on a more general level, chronic and rare conditions.

Relevance of this Research

The exploration of this under-investigated topic contributes to both the research literature and practical work of clinicians. The complex nature of this topic indicates that
its findings will be relevant to several different research domains including the rare illness, chronic illness, and parental experiences literature. The qualitative approach of the project will also provide the literature with unique experiential insights, which are currently underrepresented. Practically speaking, it is important to understand the experiences of parents of children with polyposis conditions in order to more effectively serve their psychosocial needs and to better facilitate the medical care of their children.

Outline of the Thesis

This study is divided into five chapters. Following the introductory chapter, in Chapter 2 I summarize the research literature that informs the current study and discuss the objectives and research questions guiding the project. In Chapter 3, I explain the hermeneutical phenomenology methodology and my rationale for its use. I also discuss how the data collection and data analyses were conducted and the means by which methodological rigour was ensured. In Chapter 4 I describe the findings revealed in the data analysis process using participant quotations and explain the relations between the themes. In the final chapter, Chapter 5, I position the findings within the research literature, discuss the implications of the findings, and the final conclusions that can be drawn from the study.
Chapter 2: Literature Review

Peutz-Jeghers Syndrome

Peutz-Jeghers Syndrome is an autosomal-dominant condition caused by a mutation of the STK11 gene (Chow, Meldrum, Crooks, Macrae, Spigelman, & Scott, 2006). Approximately one quarter of Peutz-Jeghers Syndrome cases present as de novo, wherein the mutation presents itself for the first time in the individual (Schreibman, Baker, Amos, & McGarrity, 2005). The condition affects 1 in 50,000 to 1 in 200,000 live births (Burt, 2002; Utsunomiya, Gocho, Miyanaga, Hamaguchi, Kashimuri, Aoki, & Komatsu, 1975). Peutz-Jeghers Syndrome’s most common clinical symptoms include rectal bleeding, pain in the abdomen and anemia as well as potentially fatal obstruction and extrusion of the polyps (Utsunomiya et al., 1975). Individuals diagnosed with Peutz-Jeghers Syndrome generally meet the diagnostic criteria proposed by Giardiello and colleagues (1987). Patients must have pathologic evidence for hamartomatous intestinal polyps and meet two of the following requirements: polyposis of the small intestine, a family history of Peutz-Jeghers Syndrome, and discoloured macules on the oral membrane, lips, fingers, feet and genitals. A diagnosis of Peutz-Jeghers Syndrome predisposes individuals to a higher risk of cancer (Giardiello et al., 1987; Spigelman, Murday, Phillips, 1989; van Lier et al., 2011).

It is recommended that physicians screen at-risk individuals from birth, checking yearly for symptoms of Peutz-Jeghers Syndrome (Giardiello & Trimbath, 2006). Because 30% of Peutz-Jeghers Syndrome patients experience bowel obstruction before 10 years of age, it is also advised that genetic testing be offered to at-risk children at 8 years of age, even if they are asymptomatic (Hinds, Philp, Hyer, & Fell, 2004).


**Juvenile Polyposis**

Juvenile Polyposis is a condition that affects 1 in 160,000 individuals (Chevrel & Gueraud, 1975). In the majority of Juvenile Polyposis Syndrome cases, 75%, present with a family history of the condition, while 25% of cases present as *de novo* (Schreibman, Baker, Amos, & McGarrity, 2005). The term ‘juvenile’ in the condition’s name is used to characterize the type of polyp found in the colon rather than the age of onset of the condition. However, clinical symptoms generally begin to present themselves before 10 years of age (Ellis, 2004). Individuals with Juvenile Polyposis experience chronic bleeding in the gastrointestinal tract, anemia, diarrhea, prolapsed polyps, and abdominal cramps and pain (Merg & Howe, 2004). In order to be diagnosed with Juvenile Polyposis Syndrome, the individual must meet the following diagnostic criteria: Elimination of the possibility of Cowden Syndrome or Bannayan Riley Ruvalcaba Syndrome diagnoses, present with more than 5 juvenile polyps in the gastrointestinal tract or any number of confirmed juvenile polyps with a family history of the condition (Jass, Williams, Bussey, & Morson, 1988). As with Peutz-Jeghers Syndrome, a diagnosis of Juvenile Polyposis also confers a greater risk of developing malignancies (Chow & McCrae, 2005). Experts in the gastroenterology field advise individuals with Juvenile Polyposis diagnoses to participate in regular blood examinations and endoscopies, with removal of found polyps (Dunlop, 2002). It recommended that individuals with Juvenile Polyposis Syndrome undergo routine medical surveillance practices such as endoscopy, every one to two years, although this is dependent on the location and severity of the polyps (Dunlop, 2002). Individuals diagnosed with Juvenile Polyposis Syndrome should also participate in
genetic counseling and testing, with relevant information to be shared with at-risk family members (Chow & McCrae, 2005).

**Family-Centered Care**

Family-centered care is an approach comprising of a partnership between medical practitioners and families in the medical decision-making and medical care process (Kuo, Houtrow, Arango, Kuhlthau, Simmons & Neff, 2012). Although considered to be the pediatric standard of care within the medical community, family-centered care is often inadequately implemented (Kuo et al., 2012). Family centered care comprises of several elements: understanding the importance of the family in the child’s life, respecting the diverse ethnic, religious and socio-economic backgrounds of families, acknowledging strengths within families and different coping methods; encouraging support-seeking behaviours; integrating developmental adaptations to clinical practices, and engaging in medical practices that meet family needs with flexibility (Trivette, 1993; Johnson 1990; Shields, Pratt, Davis, & Hunter, 2007).

Considerable evidence exists demonstrating the advantages of clinicians’ use of a family-centered approach. Research indicates that family centered therapy is related to more effective communication between physicians and families, improved mental and physical health among pediatric patients and increased engagement in medical surveillance practices (Clark et al., 2000; Bonner et al., 2002; Kelly et al., 2000). There is a dearth of evidence, however, exploring the family-centered experiences of pediatric patients suffering from rare illness and their families.
Rare Diseases

Rare diseases are conditions with low prevalence rates, manifesting themselves in fewer than 1 in 2000 individuals by European standards, and fewer than 1 in 20,000 by American standards (Zurynski, Frith, Leonard, & Elliott, 2008). As Peutz-Jeghers Syndrome and Juvenile Polyposis Syndrome have prevalence rates of 1 in 50,000 and 1 in 100,000 respectively, both of these conditions are considered to be rare diseases (Chevrel & Gueraud, 1975; Woo, Sadana, Mauger, Baker, Berk, & McGarrity, 2009). Over 8000 rare diseases have been identified worldwide, creating a greater impact than their categorization would imply (Zurynski, Frith, Leonard, & Elliott, 2008). In total, rare diseases are experienced by 6-10% of the world population, affecting 30 million Europeans and 25 million Americans (Knight & Senior, 2006).

Although rare diseases are experienced by few compared to more common diseases, their rare status belies their psychosocial, physical and economic impact on those affected (Zurynski, Frith, Leonard, & Elliott, 2008). Research into the experiences of individuals with rare diseases is considered to be a newly emerging field, with researchers facing many barriers in recruiting study participants from such small sample pools (Ettore, 2006). Without representation of the experiences of individuals with rare diseases, the illness literature is biased and unrepresentative.

As parents of children with rare diseases play an important role as caretakers, they too experience difficulties during the caretaking and parenting process (Dellve, Samuelsson, Tallborn, Fasth, & Hallberg, 2006). Parents of children with rare illnesses experience heightened psychological stress caused by barriers to appropriate medical care, such as lack of medical expertise, diagnostic delay, uncertainty of future health, and
barriers to necessary medical care (Le Cam, 2007; Zurynski, Frith, Leonard, & Elliott, 2008). Parents of children with rare chronic illnesses also serve different roles within the family than parents of healthy children. They must expend extra energy toward coping with their children’s ailments, accept the reality of the condition, meet their children’s medical and psychosocial needs, advocate for their children, educate individuals in the children’s lives, and create a support team for the children (Dellve, Samuelsson, Tallborn, Fasth, & Hallberg, 2006). In particular, mothers are at an increased risk of experiencing psychosocial distress related to their children’s illnesses compared to the children’s fathers, or mothers with healthy children (Dellve, Samuelsson, Tallborn, Fasth, & Hallberg, 2006). Research indicates that both mothers and fathers of children with rare diseases generally engage in active coping styles, actively seeking out resources to better understand how to meet their children’s needs (Dellve, Samuelsson, Tallborn, Fasth, & Hallberg, 2006). Interestingly, Budych, Helms and Schultz (2002) have found that parents of pediatric patients are more likely to engage in patient-directed interaction, wherein the parents of patients take the lead in informing the physician about their child’s condition, symptoms, than adult patients are likely to advocate for themselves. In sum, evidence demonstrates that parents of children with rare diseases feel that they are under-supported by the medical and research communities and experience significant stresses in caring and advocating for their children with rare conditions (Zurynski, Frith, Leonard, & Elliott, 2008).

Patients affected by a rare disease have significantly different medical experiences than their counterparts with more common conditions. Individuals with rare diseases contradict the general physician-patient interaction model whereby the
physician serves as an expert on the patient’s ailment and the patient actively receives the physician’s expert knowledge (Budych, Helms, Schultz, 2012). In cases of rare disease, physicians often lack the expertise and experience necessary to help the patient, possibly providing patients with inconsistent information (Budych, Helms, & Schultz 2012). As such, patients become experts on their diagnoses and their own biggest advocates (Aymé, Kole, & Groft, 2008; Budych, Helms, & Schultz, 2012). Such a phenomenon is demonstrated by patients’ empowerment as fuelling their search for information and participation in patient support groups (Aymé, Kole, & Groft, 2008).

Positioning patients as medical experts alongside their children’s physicians changes the physician-patient dynamic by altering the roles of the two parties (Budych, Helms, & Schultz, 2012). Research indicates individuals value their professional identities and are resistant to altering their views of themselves in relation to their work (Chreim, Williams, & Hinings, 2007). Because individuals with rare diseases and their families often are experts on the conditions, they are more likely to confront physicians when they feel their or their children’s medical needs are not being met. When they feel their competence is being challenged, physicians sometimes attempt to regain control of the dynamic by unilaterally making decisions for the patients and their families, without taking the patients’ or families’ stances into account (Budych, Helms, & Schultz, 2012).

Preliminary research has also begun to explore the psychosocial experiences of patients with rare diseases versus the experiences of their counterparts with more common diseases. Research by Nispen, van Rijken and Heijmans (2003) indicates that individuals with rare chronic illnesses experience lower physical and psychosocial quality of life than patients with more common chronic conditions. These individuals
also demonstrated a greater need and want for social support, either through other patients of rare diseases, clinicians, or physicians (Nispen, van Rijken, & Heijmans, 2003).

**Increased Risk of Cancer**

Parents of children with polyposis conditions must not only contend with the rarity of the conditions but must also cope with the reality that their children’s conditions confer an increased risk of the development of cancer (Boardman et al., 1998; Jass, Williams, Bussey, & Morson, 1988). Evidence demonstrates Peutz-Jeghers Syndrome to be closely related to the development of both gastrointestinal and non-gastrointestinal cancers, while Juvenile Polyposis Syndrome is most strongly associated with the development of colorectal cancer (Giardiello, et al., 2000; Jass, Williams, Bussey, & Morson, 1988).

Peutz-Jeghers Syndrome confers a risk for cancer that is 18 times greater than that of the healthy population (Giardiello et al., 1987). In a meta-analysis by Giardiello and colleagues (2000), the researchers concluded that Peutz-Jeghers patients have a cumulative risk of developing cancer at 93% between the ages of 15 and 64 years of age. Peutz-Jeghers Syndrome is most strongly linked to the development of gastrointestinal cancer, although cases of breast, ovarian, cervical, uterine, thyroid, prostate and lung cancer have also been documented in the research literature (Boardman, et al., 1998). In particular, women are more vulnerable to developing colorectal cancer as a result of Peutz-Jeghers Syndrome than men with the condition (Boardman, et al., 1998). Furthermore, the risk conferred by Peutz-Jeghers Syndrome on women to develop breast cancer is comparable to the risk conferred by hereditary breast and ovarian cancer-
related $BRCA1$ and $BRCA2$ germline mutations (Giardiello, et al., 2000).

Research into the role of Juvenile Polyposis Syndrome in the development of cancer has been less extensive than that of Peutz-Jeghers Syndrome. Rates in the development of cancers in Juvenile Polyposis Syndrome cases vary greatly, with research demonstrating a range of 17% to 55% of diagnosed patients developing gastrointestinal cancers. While the discrepancy between the two studies’ risk of malignancy evaluations is possibly be due to the genetic nature of the Juvenile Polyposis Syndromes - Coburn and colleagues (1995) included both familial and de novo forms of Juvenile Polyposis while Howe and colleagues (1998) exclusively studied participants with familial Juvenile Polyposis- one cannot discount the role research methodology plays in revealing results. Factors such as sample size, and the populations comprising the samples (clinical vs. population-based) for example, differ in the aforementioned studies and likely play a role in the results achieved. At this time, the literature demonstrates a large range of reported penetrance for gastrointestinal cancers from polyposis conditions.

As malignancy is a very likely development in the Peutz-Jeghers Syndrome and Juvenile Polyposis Syndrome conditions, physicians advise patients to actively engage in disease management programs, although recommendations vary by professional (Syngal, Brand, Church, Giardiello, Hampel, & Burt, 2015). Physicians will generally recommend that individuals at risk for Peutz-Jeghers Syndrome, such as those with diagnosed immediate relatives, be evaluated yearly for common Peutz-Jeghers symptoms from birth (Giardiello & Trimbath, 2006). Children who are asymptomatic by the age of 8 should still be genetically tested for genes associated with Peutz-Jeghers Syndrome (Giardiello & Trimbath, 2006). As discussed, diagnosed individuals will be
advised to undergo colonoscopy, endoscopy yearly. Moreover, gastroenterologists will advise that polyps larger than 1 centimeter be removed by polypectomy or enterectomy (Giardiello & Trimbath, 2006). As a means to managing cancer risk, individuals with Juvenile Polyposis Syndrome are advised to undergo gastrointestinal colonoscopy and endoscopy once symptoms develop, and every one to two years thereafter (Howe, Mitros, & Summers, 1998). Polyps are removed surgically, with large groups of polyps removed through colectomy (Lindor, McMaster, Lindor, & Greene, 2008). Genetic testing for Juvenile Polyposis Syndrome seeks to evaluate pathogenic variations of the BMPR1A and SMAD4 genes (Haidle & Howe, 2015). Consultation with a genetic counsellor is recommended to at-risk and affected individuals in order to better understand the inheritance and nature of the condition. In addition to close surveillance of the colon for malignancies, it is also advised that physicians begin to screen young adult patients for breast, ovarian, testicular, and pancreatic cancers (Syngal, Brand, Church, Giardiello, Hempel, & Burt, 2015).

**Fear of Cancer**

For many of the individuals living the polyposis experience, cancer is “synonymous with death” (Ivanovich & Whelan, 1997, 204). For the parents of children with increased likelihood toward the development of malignancy, a fear of such a development has been demonstrated in the literature (Aronson, 2009). In adult patients, these attitudes impact individuals’ likeliness to engage in illness management behaviours such as surveillance behaviours and genetic testing. The decision to engage in surveillance behaviours can be potentially lifesaving, as colorectal cancer can be effectively managed if treated early enough in its development (Lieberman, 1994).
While such beliefs and attitudes concerning the development of cancer strongly impact diagnosed adult individuals’ experiences of illness while also impacting their familial relationships, the impact of how such attitudes held by parents of pediatric patients impact both parent and child experiences has not been as closely explored.

Physicians will often advise parents of children at risk for a polyposis syndrome to undergo genetic testing (Giardiello & Trimbath, 2006). Genetic testing is an important tool in an at-risk individual’s surveillance, diagnosis and development of treatment protocol (MacDonald & Lessick, 2000). However, genetic testing in children is a much more complicated process as issues of consent, family dynamics, the child’s ability to comprehend and cope with the results present themselves (Tischkowitz & Rosser, 2004). Genetic testing can incite both positive and negative reactions from individuals, depending on their results, coping styles and expectations (Shiloh, Koehly, Jenskins, Martin, & Hadley, 2008).

Parents must be mindful that pre-symptomatic genetic testing of their children may interfere with normal family interaction, changing the way children and siblings are treated (Tischkowitz & Rosser, 2004). If a child is found to be a carrier of cancer-related gene, parents may experience guilt for transmitting the gene. Moreover, non-carrier siblings of the carrier child may experience survivor’s guilt. Parents must also be prepared to aid their children to cope with the feelings of anxiety about future health and evolving identity (Wertz, Fanos, & Reilly, 1994).

Parents often decide whether or not to have their child undergo genetic testing when their children are not yet able to consent to the process. If they choose to participate in the process, parents must weigh the costs and benefits of the testing.
Furthermore, they must decide when they will inform the child of the results and whether family members and friends should be notified on behalf of the child (Tischkowitz & Rosser, 2004).

In a study assessing the experiences of 67 first-degree family members of individuals with colon cancer, Bastani and colleagues (2001) found that only 19% of participants correctly believed themselves to have an increased vulnerability of developing the malignancy. The researchers also determined participants’ ‘fear of finding cancer’ to be the greatest barrier in preventing high-risk individuals of engaging in screening measures, such as colonoscopy or genetic testing (Bastani, Gallardo, Maxwell, 2001). These findings are consistent with Bleiker and colleagues’ (2005) results in which one of the main reported reasons for not complying with screening recommendations was fear of the positive colorectal cancer results. Research assessing the experiences of individuals with a family history of Hereditary Nonpolyposis Colorectal Cancer found that more than half of participants declined the opportunity to undergo genetic screening (Lerman, et al., 1999). Aronson (2009) suggests that high-risk individuals who actively choose not to engage in screening behaviours may refrain from doing so as an avoidance coping mechanism caused by the anxiety and stress of developing cancer. These findings reveal to us that polyposis patients’ fear of cancer is prominent enough to inhibit the use of genetic testing and possibly life-saving treatments. As will be discussed below, parents are the advocates and decision-makers for their children's medical care, and such a fear of cancer may play a role into the medical decisions made.
Parents’ Management of Chronic Illness in Children

There exists an extensive literature on the experience of parenting children with chronic illness. Research indicates that parents and the families of children with chronic illnesses are greatly impacted by their children’s illnesses from the initial diagnosis through ongoing treatment (Tomiak, Samson, Miles, Choquette, Chakraborty, & Jacob, 2007).

Psychological distress. As both Peutz-Jeghers Syndrome and Juvenile Polyposis Syndrome confer a high risk of cancer, the findings related to parental coping with children’s cancer experiences are highly relevant. This line of research will be used to shed light on the experiences of parental coping with children’s chronic illness, as research on the parenting of children with polyposis syndromes is virtually non-existent.

Parents aiding their children through the cancer experience are at increased risk of developing anxiety, depression and even post-traumatic stress symptoms (Dalquist, Czyzewski, & Jones, 1996; Kazak, Boeving, Alderfer, Hwang, & Reilly, 2005; Sawyer, Antoniou, Toogood, Rice, & Baghurst, 1993). Specifically, research demonstrates that mothers and fathers differ in their risk factors for maladaptation. The height of such distress is experienced at the time of the cancer diagnosis of the child, with continued parental distress throughout the illness (Kazak et al., 2005; Magni, Carli, DeLeo, Tshilolo, & Zanesco, 1983). Research findings are unclear as to whether such intense emotional reactions dissipate over time or whether the level of distress present at the time of diagnosis persists throughout the treatment regime (Dalquist et al, 1996; Kazak et al., 2005; Sloper, 2000).
**Risk and protective factors.** Dockerty, Williams, McGee, and Skegg (2000) argue that parents of children with chronic illness do not comprise a homogenous group. Therefore, risk and protective factors should be taken into consideration. According to Sloper (2000), mothers who anticipated the experience of parenting a child with a chronic illness to be psychologically, physically taxing or who were unsure of the abilities to cope with such a situation were more likely to experience distress. Fathers who also made negative appraisals, were unemployed or whose children were admitted to the hospital on a regular basis were also more prone to psychological distress (Sloper, 2000). Finally, both mothers and fathers were likelier to prevent psychological distress if they were part of a cohesive family (Sloper, 2000).

One protective factor that is consistently found to be influential in individuals’ adaptation in the face of adversity is social support. Social support is a resource comprising of the ability of an individual’s social network to aid the individual in coping with life stressors (Grootenhuis & Last, 1997). A lack of social support has been found to be associated with poorer adjustment in parents of children with cancer, specifically in the domains of psychological health and ability to cope (Dockerty, Williams, McGee, & Skegg, 2000). Moreover, as parents experience increasing psychological distress, the less likely they are to be satisfied with their social support resources (Hoekstra-Weebers, Jaspers, Kamps, & Klip, 1999).

**Parental coping styles.** In line with these findings, the use of the social support seeking coping style is also associated with less psychosocial distress in parents of children with cancer (1999). Coping styles play an important role in the ways parents manage, and as a result, experience stressors. Within the coping literature, there exist
two main coping styles that have proven to be effective in coping with chronic illness: Active and avoidant coping. Active coping consists of managing stressors in an engaged, problem-focused manner while avoidance coping focuses primarily on emotional coping and distraction from the stressor (Kliwer & Lewis, 1995). These empirically valid and reliable constructs (Suls & Fletcher, 1985) have both been shown to be effective in contexts of chronic illness despite their seemingly contradictory processes. It has been argued that avoidant coping strategies are particularly effective in contexts where individuals have little control, such as in the case of chronic illness. In a study by Dahlquist, Czyzewski, Copeland, Jones, Taub, and Vaughan (1993), parents of newly diagnosed children with cancer were found to demonstrate more avoidant coping styles than active styles. The authors postulate that parents utilized avoidant coping behaviours because the situation was out of their control (Dahlquist et al., 1993). Moreover, the use avoidant coping behaviours aided the parents to ignore overwhelming stressors and focus on surviving the short-term medical obstacles. It has also been proposed that avoidant coping mechanisms only become maladaptive if they begin to interfere with health behaviours or if they support the presence of intrusive thoughts (Shontz, 1975; Suls & Fletcher, 1985). Conversely, extensive research also supports the effectiveness of active coping mechanisms in parents of children with cancer. Prior research indicates that parents who engaged in active coping behaviours were less likely to develop depression and anxiety (Norberg, Lindblad, & Boman, 2005). An active coping style can also be considered crucial in securing the medical care necessary for a child with a rare disease (Dellve, Samuelsson, Tallborn, Fasth, & Hallberg, 2006).
Samson, Siam, and Lavigne (2007) have developed an integrative model demonstrating the ways in which individuals cope with illness-related stress. The model is a combination of Samson (2006), Cohen and Lazarus (1979), Moos and Tsu (1977), Corr (1992) and Samson and Clark (2007)’s models, as all are premised on the use of adaptive tasks as a means of coping with illness (Samson, Siam, & Lavigne, 2007).

Samson, Siam and Lavigne’s (2007) Integrated Model begins with acknowledging that every individual's context is unique and should therefore be taken into consideration. Samson, Siam and Lavigne (2007) organize contextual information into 3 groups: Personal history and characteristics, illness-related factors and features of the physical and socio-cultural environment. These factors will influence the individual’s primary cognitive appraisal of the diagnosis, a concept initially developed by Cohen and Lazarus (1979). Those diagnosed will appraise their diagnosis to be either benign or warranting stress. Individuals will then assess their resources to cope with the diagnosis (secondary appraisal). Samson, Siam and Lavigne (2007) then theorize that certain adaptive tasks will be effective in coping with the diagnosis. The researchers, building on Corr (1992)’s adaptive task model, prescribe adaptive tasks in the following experiential domains: Physical, psychological, social, spiritual, and vocational. Adaptive tasks include creating personal meaning of the illness experience and cultivating social support groups (Samson, Siam and Lavigne, 2007). Participating or refraining from engaging in these adaptive tasks will play a large role in the coping behaviour utilized by the individual.

Various styles and behaviours have been identified, with varying degrees of effectiveness in illness contexts. Coping skills differ in their focuses, with some based on meeting emotional needs while others focus on actively managing contextual cues (Samson, Siam,
& Lavigne, 2007). The coping skills that are used will ultimately mitigate the illness experience, creating a “new state of psycho-social equilibrium” (p. 24) or psychosocial maladaptation (Samson, Siam, Lavigne, 2007).

**Experience of hope.** Research by Samson, Tomiak, Dimillo, Lavigne, Miles, Choquette, Chakraborty, and Jacob (2009) demonstrates that the ‘fabric of hope’ of parents of children with Duchenne muscular dystrophy is strongly influenced by parents’ cognitive appraisals of the situation, and as a result, evolves. The researchers found that at the time of initial diagnosis, parents appraised the situation to be a crisis and focused their resources toward hoping for advancements in research and treatments. As the illness progressed, the parents become more aware of their resources and were better able to redirect their focus from hoping for a cure to actively coping with their child’s needs (Samson, et al., 2009). As the parents begin to adapt to their new reality, they began to allow the child’s identity to flourish rather than focus solely on the disease. As medical complications began to arise and death becomes imminent, parents’ hope took on a spiritual component (Samson et al., 2009).

**Family dynamics.** Parents’ psychological adjustment to their children’s illness is predictive of their marital satisfaction (Dahlquist, Czyzewski, & Jones, 1996). Although parents of children with cancer do not experience a higher divorce rate, they do report to be less satisfied in their marriages (Kazak, 1989). Discrepancies in partners’ level of distress have also been shown to predict marital maladjustment (Dahlquist, Czyzewski, Copeland, Jones, Taub, & Vaughan, 1993). Dalquist and colleagues (1996) hypothesize that marital partners represent primary resources of social support and therefore play an important role in the parental experience of raising a child with cancer.
Tomiak and colleagues’ (2007) research on the experiences of parents of children with Duchenne Muscular Dystrophy demonstrates that parents of children with the condition must make significant psychosocial adjustments to their unique realities. The authors discovered that mothers and fathers differ in their experiences of their child’s illness, with mothers oftentimes serving as the child’s primary caregiver and fathers acting as supporting parent (Tomiak et al., 2007). This difference in roles can lead to discrepancies in the ways the parenting of a chronically ill child is experienced, such as in the ways parents cope with their child’s diagnosis and everyday obstacles. Mothers generally seek social support as a means of coping with their child’s illness while fathers view the situation as a series of obstacles to be overcome (Tomiak et al., 2007). Such discrepancies in roles and experiences can affect the mental health of each partner differently, the family dynamic and the care of the child.

**Experiences of Polyposis Syndromes**

Although considerable research has been conducted on the clinical and biological aspects of Peutz-Jeghers Syndrome and Juvenile Polyposis Syndrome, virtually no research has been conducted on the psychosocial aspects of parenting a child living with the conditions. This dearth of research in the literature represents an important void in our understanding of polyposis syndromes. As symptoms of both Peutz-Jeghers Syndrome and Juvenile Polyposis Syndrome generally present themselves during youth, parents play a pivotal role in advocating for their children’s medical needs and supporting their children through the experience (Woo, et al, 2009). As previously discussed, parents of children with chronic illness are vulnerable to psychological distress (Kazak et al., 2005; Magni, Carli, DeLeo, Tshilolo, & Zanesco, 1983). Peutz-
Jeghers Syndrome and Juvenile Polyposis Syndrome are rare and heritable conditions that add a layer of complexity to parents’ experiences, warranting further exploration, as it cannot be assumed that the cancer literature adequately represents the polyposis experience. While no research has been conducted on the experiences of parents of children with polyposis syndromes, review of the experiences of individuals with these conditions may give us insight into the issues with which parents must contend.

In a study conducted by Woo and colleagues (2009), individuals with Peutz-Jeghers Syndrome were found to experience significant distress regarding the possibility of developing cancer. Van Lier and colleagues (2010) also reported that individuals’ with Peutz-Jeghers Syndrome felt limited by emotional difficulties, suffered poorer mental health and rated their health to be poorer than that of controls. The researchers found participants who believed Peutz-Jeghers Syndrome to have a significant negative impact of their lives and who felt emotionally impacted by the condition were more likely to experience a diminished quality of life (Van Lier et al., 2010). Participants in the study were also found to suffer from mild depression.

Peutz-Jeghers Syndrome played a significant role in participants’ important life decisions; the condition was cited as being the reason many participants were hesitant to have biological children- for fear of transmitting the condition (Woo et al., 2009). Van Lier and colleagues (2012) report findings similar to Woo and colleagues (2009)'s, in a study where a majority of participants chose not to have biological children for fear of transmitting the disease. These participants also had a higher incidence of cancer and reported greater fear of the development of cancer than participants who did not refrain from having biological children (Van Lier et al., 2012).
Consistent with the rare diseases literature, the researchers also determined that the participants felt a ‘Societal Burden’, indicating dissatisfaction with the medical community’s knowledge and care of Peutz-Jeghers Syndrome (Woo et al., 2009). Sugars (2011) recounts her firsthand experience with Peutz-Jeghers Syndrome and her creation of a Peutz-Jeghers and Juvenile Polyposis Syndrome internet support group for those affected by the conditions, both diagnosed individuals and their friends and families, as a means to supplement the gaps in the knowledge by the medical community and creating an organized support system. Part of the impetus for creating the web forum was the author’s own experience of isolation and fear (Sugars, 2011). Furthermore, Sugars (2011) writes that amongst the research on Peutz-Jeghers Syndrome and Juvenile Polyposis, there is a lack of research depicting the experiences of those diagnosed with these conditions.

Coping, an important mitigating factor in the illness experience has been shown to be socialized from parent to child in families where parents and children suffer from the same illness. In a study by Miller, Bauman, Friedman, and DeCosse, (1986), individuals with Familial Adenomatous Polyposis (FAP) whose parents with the same condition took an active role in guiding their children through the illness experience, were less likely to experience anxiety about their future health. Kliewer, Fearnnow and Miller (1996) have developed a model in which coping strategies are transmitted from parent to child, based on research in a non-clinical population. The researchers include parental coping suggestions and parental modeling as means of transmission, with influences from family dynamics (Kliewer, Fearnnow, & Miller, 1996). In their study using questionnaires, the authors found that mothers have a greater impact on their children’s
coping skills than fathers (Kliewer, Fearnlow, & Miller, 1996). The quality of the parent-child relationship was also found to be associated with stronger coping socialization effects (Kliewer, Fearnlow & Miller, 1996). The transmission of coping methods in hereditary diseases such as polyposis syndromes poses a unique complexity in illness experiences, as parents’ own experience with illness will likely influence their parenting behaviors of their children with the same illness.

In sum, the literature available on rare diseases, experiences of parents of children with chronic illness and experiences of polyposis creates a strong rationale for further research into the experiences of parents of children with polyposis syndromes. While the lines of research on parents’ experiences of raising children with cancer is currently practitioners’ most relevant resource, it cannot be assumed that these contexts are generalizable to the polyposis experience. Because of the unique properties of polyposis syndromes, it also cannot be assumed that prior research or theoretical frameworks represent the experiences of individuals with children suffering with polyposis syndromes. Cancer research has revealed common themes within the cancer experience, such as experiencing stigma (DiMillo et al., 2013), and fear of dying (Murray, Grant, Grant, & Kendall, 2003) that cannot necessarily be attributed to polyposis syndromes without further research. As such, this exploratory, qualitative study presents an opportunity for researchers to strengthen the polyposis literature and more effectively engage clinical practitioners.

**Objectives and Research Questions**

The main objective of this study is to explore the experiences of parents of children and teenagers under the age of 20 (hereinafter referred to as ‘children’) with
polyposis syndromes using a hermeneutic phenomenological approach. The following research question will direct this exploratory study: What are the lived experiences of parents of children with polyposis syndromes?

**Contributions to Knowledge and Practice**

The research literature demonstrates a clear void in knowledge about the experiential impact of parenting a child with a polyposis syndrome. The results from this study and the means by which it is collected provide the research literature with invaluable information. Furthermore, the findings generated by this study aid physicians and psychosocial practitioners working with parents of children with polyposis to understand the complexities of their lived experiences and ultimately offer better care to both pediatric patients and their parents.
CHAPTER 3: Methodology

The literature review provides context for this study. In this chapter I discuss the philosophical foundations guiding this study as well as the hermeneutical phenomenological approach I used. Furthermore, I delineate my position as a researcher. I also discuss the strategies I used to develop my data collection instruments as well as the ways I recruited participants, collected the data, and analysed it. Lastly, I highlight the steps I took to establish methodological rigour.

This study was conducted using an inductive, qualitative hermeneutic phenomenological approach. This approach was informed by Lindseth and Norberg (2004)’s Phenomenological Hermeneutical Method for Researching Lived Experience. Thematic analysis of the participants’ transcribed interviews was undertaken to develop to reveal the common essences of the participants’ shared experiences. As identifying essences is the “methodological goal” of phenomenological research, the essences revealed in this research serve as the study’s findings (Dahlberg, 2009, p. 11).

Philosophical Foundations: Hermeneutical Phenomenology

Phenomenological research seeks to reveal the “essence or structure of an experience” (Merriam, 2002, p. 7). One of the central tenets of hermeneutic phenomenology is that “complex meanings are built out of simple units of direct experience” (Merriam, 2002, p. 7). Hermeneutic phenomenology, developed by Martin Heidegger, builds on the work of Edmund Husserl, the father of transcendental phenomenology. Husserl (1931) posited that phenomenologists should engage in bracketing when exploring a phenomenon, whereby the researchers attempt to suspend their own biased views of the world in order to observe and analyse the experience in question in its truest form. Heidegger, a student of Husserl, deviated from the
transcendental phenomenological viewpoints to develop hermeneutic phenomenology, wherein phenomenologists are believed to be unable to suspend their biases and therefore are encouraged to focus their attention to the awareness of their influence on the exploration (Connelly, 2010). Rather, Heidegger (1962) argued that a researcher’s prior knowledge can be beneficial to the research process and can actually enrich the research process. Heidegger (1962) also highlighted the notion of *co-constitutionality*, wherein both participants and researchers are viewed as partners in developing and interpreting meaning from participants’ narratives (Koch, 1995). However, hermeneutic phenomenologists must ensure that the derived meanings are reflective of participants’ experiences, well-reasoned, and reflect the implications of the meanings in various practical fields (Annells, 1996).

In exploring the various manifestations of the phenomenological approach, I reflected on my own epistemological and ontological viewpoints, as well as my own role as researcher. Upon reflection, I realized that my biases, views, and experiences might influence my interpretation of the data. As such, I continued to engage in self-reflection and document the ways in which I felt I influenced the progression of this study. This reflection served to enrich the research process.

**Rationale for the use of the Hermeneutical Phenomenological Approach**

As mentioned, this study was conducted using a hermeneutical phenomenological approach. This approach allows the researcher to play an active role in collecting, organizing and interpreting data in a way that is reflective of the lived experiences of the parents of children with polyposis syndromes. I have chosen the phenomenological approach to guide the development and actualization of this research because of the
focus of the approach on the interpretation of shared lived experiences. I appreciate the recognition that this approach has for context in subjective experiences. As I am choosing to describe and interpret the lived experiences of the parents of children with polyposis diagnoses, I believe that this approach is best suited.

A limited amount of qualitative research has been conducted in the domain of parental coping of children with chronic illness, with the majority of studies employing quantitative analysis using structured questionnaires. The sole use of empirical methodologies as a means to representing complex experiences risks losing important experiential details in data represented by numbers. The use of qualitative data in understanding the experiences of parents of children with polyposis syndromes is integral. Polyposis syndromes demonstrate various unique properties dissimilar to many illnesses studied in the literature. Peutz-Jeghers Syndrome and Juvenile Polyposis Syndrome are rare conditions that strongly predispose those diagnosed to cancer. Moreover, these conditions are hereditary and chronic. It is not yet known how the combination of these factors affects the experiences of parents in the context of polyposis syndromes. Many interrelated factors play a role in the experiences of parents of children with polyposis syndromes and the hermeneutical phenomenological approach allows for the most integrative and meaningful collection and analysis of these factors. While research on the experiences of parents of children with cancer is currently our greatest source of information into the experiences of parents of children with polyposis, it cannot be assumed that these phenomena can be generalized to polyposis experiences. Because of the unique properties of polyposis syndromes, it also cannot be assumed that prior research represents the experiences of individuals with children
suffering with polyposis syndromes. As such, hermeneutical phenomenology serves as a useful tool in beginning the exploration of these experiences.

Hermeneutical phenomenology positions participants as the experts of their experiences, an important feature of qualitative research (Marques & McCall, 2005). Participants will be able to focus on themes most salient to them, thereby allowing them to create their own narratives. As I am seeking to describe and understand the meaning behind the parents’ experiences of their children’s illnesses, hermeneutical phenomenology serves as an appropriate approach as it will allow me to delve and explore the lived experiences of these individuals (Creswell, 2013).

**Researcher Position**

Prior to the start of my graduate studies, I worked with a health psychology research group that studied the experiences and health behaviours of individuals with cutaneous melanoma. Such experience ignited my passion for health psychology and interest in the ways in which individuals cope with adversity, specifically health challenges. As an intern counsellor during my graduate studies, I learned the depths to which context, environment and psychological factors play in creating experience and mitigating coping strategies. Furthermore, my training in counselling has allowed me to be able to effectively convey empathy and encourage clients to speak openly about their experiences; a skill I feel was effectively utilized in the data collection process. I, myself, do not suffer from a polyposis condition, nor am I a parent to a child with such a diagnosis.
Participants

Eligibility criteria. In order to be eligible to participate in this study, participants had to be the parent, caretaker, guardian, or individual serving within a parental role of a pediatric patient under the age of 20 years diagnosed with either Peutz-Jeghers Syndrome or Juvenile Polyposis Syndrome at the time of the study. Both clinical and molecular diagnoses of these conditions were accepted. Participants must not have had a psychiatric or medical illness that would prevent them from participating in the interview or giving informed consent. Participants needed to be able to communicate in either French or English, as interviews were only conducted in either of these languages. Participants meeting these criteria were recruited for the study.

Recruitment Process. Physicians at a pediatric health sciences centre in Ontario, identified participants from their patient lists fitting the eligibility criteria. Once the list of eligible individuals was completed, participants were contacted and informed about the study. Both parents of each pediatric patient were invited to participate, allowing for both a mother and father of the shared child to participate. Participants were recruited using a criterion sampling method, wherein eligibility for the study was dependent on having lived the aforementioned phenomenon. As discussed, small population sizes are an inherent challenge of studies studying rare illnesses (Ettore, 2006). Once the eligible patients and their parents were identified, the physicians addressed and mailed envelopes containing the Study Description and Permission to Contact Participants forms (Appendices B and C, respectively), as well as institution-addressed stamped envelopes so that the recipients could easily mail back their responses. The physicians notified myself, the primary researcher, once she received the returned envelopes, at which point
I contacted parents consenting to be contacted by their preferred means, either by telephone or by email. In the follow-up contact, I informed parents about the objectives of the study and what their participation would entail. Once their questions were answered, parents were invited to participate in the study. I then scheduled a date and time to meet at the pediatric health centre to conduct the interview, at which point the Participant Information and Consent Forms (Appendix D) were completed.

Parents who did not respond to the recruitment letters within three weeks were sent a reminder recruitment package containing the same documents. In total, thirteen individual parents (of eleven diagnosed children) were contacted to participate in this study, with seven parents indicating their interest and consent to participate in the study. All parents who were contacted regarding the study were sent both the initial recruitment forms as well as the final, identical reminder recruitment forms. The parents who were contacted but who ultimately did not participate in the study indicated their disinterest in participating in the study by not responding to either of the two contact attempts for recruitment.

Instrument Development

Development of the Interview Protocol. The interview protocol was created by consulting relevant research and medical professionals working in the gastroenterology and genetics fields. Although very little research has yet to be conducted on the experiences of parents of children with polyposis syndromes, findings of relevant research indicate that parents of children with chronic illnesses suffer an increased risk of mental health and emotional problems (Dalquist, Czyzewski, & Jones, 1996; Kazak, Boeving, Alderfer, Hwang, & Reilly, 2005; Sawyer, Antoniou, Toogood, Rice, & Baghurst,
As a means of managing the stress conferred by raising a child with chronic illness, parents generally turn to their partners, with an increased likelihood of influencing family dynamics (Budych, Helms, & Schultz, 2012; Dellve, Samuelsson, Tallborn, Fasth, & Hallberg, 2006). The newly emerging literature on rare diseases also demonstrates individuals with rare diseases undergo significantly different experiences as patients than individuals with more common conditions, as patients become experts on their diagnoses and their own biggest advocates (Aymé, Kole, & Groft, 2008; Budych, Helms, & Schultz, 2012). As such, questions were compiled specifically to explore these domains (i.e. Psychological impact, social impact, medical care seeking experience, adaptation to illness, view of child’s health, family planning decisions).

Although there exists research on the various influential factors of being a parent of child with chronic illness, it cannot be assumed that such findings are generalizable to parents of polyposis syndromes. The interview protocol ensured participants with the flexibility to focus on particularly salient issues in their experiences while probing evidence-based topics. Interview questions were organized and ordered to facilitate the development of trust between the researcher and participant. Open-ended questions, humanistic in nature, were also used in order to promote the participants’ introspection and exploration of their experiences.

**Development of Demographic Questionnaire.** Participants were asked to complete a demographic questionnaire in order to enable to the researcher to more appropriately contextualize and understand the collected data. Variables assessed within the demographic questionnaire were included based on their demonstrated relevance in prior research. Participants were asked to list the resources within their support
networks in order to better understand their social support experiences. Furthermore, the parents were asked to describe their marital statuses and to list the number of children they parent in order to gain further insight into their family situation. Lastly, participants were asked to specify details of their own and their children’s medical histories in order to build a rich description of the sample group.

**Demographics.** As mentioned, a total of seven participants (representing six patients, as two of the participants had formerly been married) were recruited to take part in the interview process. All participants at the time of the interview were married, except for one participant who identified himself as a single-parent widower. Combined, the participants had an average of two children, with a range of one to four children. All of the participants had either one or two children diagnosed with Juvenile Polyposis Syndrome, except for one participant whose child was diagnosed with Peutz-Jeghers Syndrome. None of the participants suffered from Juvenile Polyposis Syndrome or Peutz-Jeghers Syndrome themselves. Only one participant indicated that his Juvenile Polyposis diagnosed children had been identified as having a family history of the condition; his wife had passed away from complications relating to the condition.
Table 1

*Participants’ Demographics*

<table>
<thead>
<tr>
<th>Item</th>
<th>n (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Sex</strong></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>2 (29)</td>
</tr>
<tr>
<td>Female</td>
<td>5 (71)</td>
</tr>
<tr>
<td><strong>Primary language of communication</strong></td>
<td></td>
</tr>
<tr>
<td>English</td>
<td>4 (57)</td>
</tr>
<tr>
<td>French</td>
<td>3 (43)</td>
</tr>
<tr>
<td><strong>Marital Status</strong></td>
<td></td>
</tr>
<tr>
<td>Married</td>
<td>6 (86)</td>
</tr>
<tr>
<td>Single</td>
<td>1 (14)</td>
</tr>
<tr>
<td><strong>Highest level of education achieved</strong></td>
<td></td>
</tr>
<tr>
<td>High School</td>
<td>1 (14)</td>
</tr>
<tr>
<td>CEGEP/Technical College</td>
<td>2 (29)</td>
</tr>
<tr>
<td>University (Undergraduate degree)</td>
<td>2 (29)</td>
</tr>
<tr>
<td>University (Graduate degree)</td>
<td>2 (29)</td>
</tr>
<tr>
<td><strong>Participant diagnosis of polyposis condition</strong></td>
<td>0 (0)</td>
</tr>
<tr>
<td><strong>Number of children</strong></td>
<td></td>
</tr>
<tr>
<td>1</td>
<td>1 (14)</td>
</tr>
<tr>
<td>2</td>
<td>3 (43)</td>
</tr>
<tr>
<td>3</td>
<td>1 (14)</td>
</tr>
<tr>
<td>4</td>
<td>2 (29)</td>
</tr>
<tr>
<td><strong>Number of children with polyposis diagnosis</strong></td>
<td></td>
</tr>
<tr>
<td>1</td>
<td>5 (71)</td>
</tr>
<tr>
<td>2</td>
<td>2 (29)</td>
</tr>
<tr>
<td><strong>Polyposis diagnosis of children</strong></td>
<td></td>
</tr>
<tr>
<td>Juvenile Polyposis Syndrome</td>
<td>6 (86)</td>
</tr>
<tr>
<td>Peutz-Jeghers Syndrome</td>
<td>1 (14)</td>
</tr>
<tr>
<td><strong>Use of genetic testing</strong></td>
<td>7 (100)</td>
</tr>
<tr>
<td><strong>Individuals part of support system</strong></td>
<td></td>
</tr>
<tr>
<td>Partner/spouse</td>
<td>5 (71)</td>
</tr>
<tr>
<td>Family</td>
<td>5 (71)</td>
</tr>
<tr>
<td>Friends</td>
<td>5 (71)</td>
</tr>
<tr>
<td>Physicians</td>
<td>5 (71)</td>
</tr>
<tr>
<td>Co-workers</td>
<td>0 (0)</td>
</tr>
<tr>
<td>Psychological services</td>
<td>1 (14)</td>
</tr>
<tr>
<td>Internet forums</td>
<td>1 (14)</td>
</tr>
</tbody>
</table>

**Data Collection**

Participants met the primary researcher at a health sciences institution, where they were brought to a private room located in the Gastroenterology unit. This location
ensured that participants were afforded privacy and confidentiality as they discussed their personal experiences. The site also served as a familiar site to participants, minimizing undue stress concerning attending the interview.

Prior to commencing each interview, the researcher reviewed the Participant Information and Confidentiality Form (Appendix D) and ensured the participant understood the interview process. She then witnessed the participant sign two copies of the Participant Information and Confidentiality Form (Appendix D), one of which was kept by the researcher, and one kept by the participant for their files. Participants were reimbursed $20.00 for their parking fees by the researcher. Participants were then asked to complete the Demographic Information Form (Appendix A). The information collected from this document was used to create the context with which participants’ data was interpreted.

Data was collected through the use of the semi-structured Interview Protocol (Appendix E). The semi-structured interview process was chosen to afford the interviewer the flexibility to probe particularly salient themes with participants. The effectiveness of semi-structured interviews was also enhanced as it was used with concurrent data analysis and collection, which allows for initial interviews to inform the organization of subsequent interviews. Interviews were digitally recorded and transcribed verbatim as a means of ensuring accurate records and analyses. All files were kept on the primary researcher’s password-protected computer. Participants’ identities were protected by pseudonym, with identity-revealing information omitted from final transcripts.
Interview recordings were immediately transcribed after each interview session in order for the researcher to best recall the interview session as a whole. After each transcribing process, the researcher highlighted and took note of possible themes that should be added to subsequent interview protocols in the form of questions or probes.

**Data Analysis**

As previously discussed, the hermeneutical phenomenological approach does not prescribe a specific methodology (Colaizzi, 1978; Hein & Austin, 2001). The data collected for this study was analyzed using Lindseth and Norberg (2004)’s outlined approach to hermeneutical phenomenological analysis. I chose this type of analysis as it was specifically designed to explore lived experiences within health care contexts (Lindseth & Norberg, 2004). The authors’ structure for analysis aims to “elucidate essential meaning as it is lived in human experience” (Lindseth & Norberg, 2004, p.146). Such elucidation of meaning is revealed through story-telling and expression of one’s experiences (Lindseth & Norberg, 2004). Lindseth and Norberg (2004) also argue that interpretation of the lived experiences should be done using textual material (transcribed copies of participants’ recorded interviews) through the use of thematic analysis.

Following Lindseth and Norberg (2004)’s guidelines, I began the data analysis process by taking meticulous notes during the interview process. Within hermeneutical phenomenology, it is understood that to a certain extent interviewers help to shape the final narrative product (Lindseth & Norberg, 2004). Once I began the data collection process, recorded interviews were transcribed verbatim as quickly as possible in order to most effectively capture each participant’s narrative through text. Data was collected
and analysed concurrently, with data from earlier interviews informing the formation and organization of subsequent interview questions. Once all the interviews were transcribed, I engaged in what Lindseth and Norberg (2004) call a naïve reading, wherein I read the transcripts multiple times in order to understand the meaning of the narratives as a whole.

Once the naïve reading was completed and I felt that I had gained insight into the meaning of the participants’ experiences (i.e., their lifeworlds), I began the structural analysis of the data analysis process. While phenomenological structure analysis can be achieved through various methods, I chose to engage in thematic structural analysis. Lindseth and Norberg (2004) describe the identification and formulation of themes (units of meaning) as “methodical instances of interpretation” (p. 149). Whole sentences, fragments of sentences, and paragraphs that revealed commons meanings were grouped together and revised, refined, until they were reflective and consistent with the understanding built from the naïve reading. The themes were situated within the phenomenon –that is the lived experiences of the participants. The themes were then once again refined using experiential and colloquial language. My thesis supervisor served as an auditor of the data analysis process and engaged in the same data analysis process as me. When differences were found between our findings, we then reconciled our discrepancies through discussion.

**Methodological Rigour**

This study maintained and examined methodological rigour using the concepts and standards outlined by Lincoln and Guba to achieve trustworthiness (1985): credibility, transferability, dependability and confirmability.
Lincoln and Guba (1985)’s concept of transferability is defined as the ability to apply the results of one context to another context. In order to do so, researchers would need the detailed information of the original context in which the study was developed (Lincoln & Guba, 1985). As the sample group comprises of individuals experiencing a very unique and rare phenomenon, transferability of results may be difficult to acquire. However, background contexts of the participants were assessed in the demographic questionnaire, the data of which has been included in this manuscript. Furthermore, the many interview questions were formulated in a way to encourage participants to discuss histories, contexts. The literature review included in this manuscript was also written with the goal of being able to easily situate this study within the current available research.

In order to ensure the credibility of the data obtained, a standard that aims to ensure the congruence of results with reality, established qualitative research methods were followed, medical specialists working with polyposis families as well as relevant literatures were consulted in order for me to familiarize myself with some of the realities and experienced faced by patients and their families prior to data collection. The interview protocol was also formulated to encourage participants to speak honestly, with the interview summarized and recounted back to the participant to ensure that the interviewer understood and interpreted their responses correctly. My ability to follow the direction of emerging themes presented by the participant enriched the inductive process and delved into the components of the experience most pertinent to the participants. Furthermore, a gastroenterologist was asked to review the themes revealed in this study to assess the credibility of the interpretations.
The dependability of the results is strong, as the steps taken to develop the research design and procedure have been outlined in great detail. Lastly, in order to increase the level of confirmability of this study, the use of a second researcher auditing the coding process served to limit the potential for bias. The primary researcher has also provided a statement indicating her positioning as a researcher and possible biases that may have affected the progression of the research process.
Chapter 4: Findings

This chapter discusses the revealed essences of participants’ shared experiences. As essences were revealed using thematic analysis, the terms ‘essences’ and ‘themes’ will be used interchangeably. The data analysis process revealed that while the participating parents may have come from varying backgrounds, their lived experiences shared many commonalities. The analyses reveal that these shared commonalities—essences—are essential to the phenomenon of parenting a child with a polyposis syndrome. In this chapter, I identify and describe the essences revealed in the data analysis process.

Use of Identifiers

In order to retain the anonymity and protect the privacy of the study’s participants, participant names were replaced with numbered identifiers, such as ‘Participant 1’ and all identifiable information has been removed.

Description of Essences

In total, four themes and twelve underlying sub-themes were revealed. The following themes comprise the shared, meaningful experiences of the participants from the time of their children’s diagnoses onward: (1) Building healing relationships within the medical community (a. Coming together for the wellbeing of the child; b. Always on the lookout: Doing everything possible to maintain health; c. Feeling grateful: It could be worse); (2) Peeking through covered eyes at illness literature (a. Seeking validation in shared experiences; b. Tuning out anxiety-inducing information); (3) Perservering until needs are met (a. Teaching the children to speak for themselves; b. Trusting maternal intuition; c. Taking it one day at a time because the future is uncertain); (4) Creating a new ‘normal’ as everything becomes unsettled (a. Becoming a different type of parent; b.
Trusting they will speak so they can be heard; c. Putting families to the test: coping with the illness as a family; d. Feeling as though no one understands. Diagrams are included within the chapter to visually represent the relations between the themes and sub-themes. Figure 1, shown below, depicts the sub-themes comprising the 'Building healing relationships within the medical community' theme.

**Figure 1.** Visual representation of the Building healing relationships with the medical community theme and sub-themes

**Building healing relationships within the medical community**

The following three subthemes were found to contribute to the understanding of the theme “Building healing relationships within the medical community”: (a) Coming together for the wellbeing of the child; (b) Always on the lookout: Doing everything possible to maintain the child’s health; (c) Feeling grateful: It could be worse. In this section, I discuss how the physicians’ way of interacting and relating with the patients’ parents ultimately shape the parents’ cognitions, emotions and behaviours. As their children’s conditions progressed, parents reported feeling heard and included in the
medical process by their children’s physicians, they described feeling less anxiety and uncertainty about their children’s health, more gratitude for the health of their children as well as the available medical resources, and were more committed to ensuring their children were appropriately followed by the medical team.

**Coming Together for the Wellbeing of the Child.** When describing their lived experiences, parents described their relationship with their child’s physician to be particularly influential in affecting the way in which they coped with their child’s diagnosis. While some participants discussed difficult initial experiences with their family physicians, the remainder of the participant group discussed positive experiences with their children’s physicians. They described relationships in which physicians engaged them in the collaborative care of their child. By collaborating with parents to ensure their patients received the most appropriate medical care, physicians were able to build foundations of trust with parents. For example, participant 5 explained that in her meetings with her child’s gastroenterologist, that she felt comfortable engaging the doctor in a discussion about her concerns for her child. She stated:

Well, um, I felt very safe. I felt very comforted. I felt free to voice my stress, my angst, my anxiety, my concerns. I felt listened to and I felt heard. I felt this was a very safe place to have all these discussions. Um, I mean to the point where we, my ex-husband and I, had an open and candid conversation with [the doctor] about our marital breakdown and whether the stress of that may have impacted our child’s health. And she did not shy away from that conversation, we didn’t shy away from that conversation. It was just a very open and honest environment to
consider everything that might impact this child’s health. It was really a collective effort.

Participant 1 also reflected having a similar experience, in that he also felt comfortable asking questions to the doctor, which helped to reduce his anxiety. He explained:

Interviewer: Et pour vous, être suivit par [le médecin], c’est la meilleure façon de diminuer l’anxiété?

Participant 1: Oui, c’est plus rassurant quand tu sais qu’ils sont suivit de prof. Elle fait toute temps à l’estomac, les intestine, être sur qu’il y a rien nul part. Oh oui, c’est pas mal...Non, c’est tranquille.

Interviewer: Et est ce que vous vous sentiez confortable de poser des questions [au médecin]?

Participant 1: Oui, oui.

Working within a collaborative relationship with physicians also promoted honesty and an opportunity for dialogue on the part of parents who were unhappy with physicians’ recommendations, as Participant 7 revealed learning while serving as an advocate for her son. She encouraged parents in similar situations to ensure the physicians are made aware of their concerns and to create dialogues even when doctors are hesitant. She explained:

And don’t always, I’m learning now you don’t have to do what [the doctors] want you to do. You don’t have to take all this garbage [medication] if you don’t want to take it. Find me another solution. Find me a better solution. ‘I don’t have to do
that. ‘Well, the health system says that you have to.’ ‘Okay, well, find me another solution because this one isn’t working for me’.

In three of the seven parent-physician relationships described, participants described experiencing difficult challenges in trying to convey to their family physicians the seriousness of their children’s symptoms. In these cases, participants were placed in positions where advocating for their children’s needs proved to be necessary, as the children’s conditions were revealed to be life-threatening. The findings demonstrate the lack of a patient and family-centered approach negatively impacted the experiences of the parents and their affected children.

Participant 2 described her initial experience in trying to learn more about her daughter’s health as such:

Well, for some time, since she was in about grade six, so about twelve, she hadn’t been feeling that great. We had gone to the family doctor a couple of times and you know, sometimes they just sort of, you know, poo poo you. You know, it’s like, “Oh well, you know, she’s becoming a teenager, and they’re just irritable or they tend to get tired a lot”, because she was tired a lot. And, um, she was eating tonnes and tonnes of ice. Just like cup after cup after cup of ice. And I kept saying to the doctor, “This is not normal for someone to eat this amount of ice and to be this tired all the time”. And I was ignored.

The participant further described meeting with the doctor multiple other times, eventually convincing the doctor to allow her daughter to undergo testing. Ultimately, the participant’s daughter was found to be severely anemic and necessitating emergency
surgery due to a polyp blocking her colon. When asked how she felt about the experience in seeking medical care for her child, she explained:

    Well, it was mostly at the time a sense of frustration that no one was listening to us... She switched family doctors after that because she felt that our, the family doctor that I still have, had dropped the ball on her.

Participant 3 experienced a similar situation, wherein her family physician also discounted her concerns about her child’s health. She stated:

    And then so I took her to the doctor. I remember I was really, really sick. I had a double ear infection and I had a little baby, like a 3-month old. And I got into a big fight with the doctor because I wanted her tested, I wanted her blood work tested. I said “I think she’s anemic”. And he told me I would be laughed out of [the hospital], were his exact words. He said, “I’ve seen anemic children and this child is not anemic” because she was wired. She was running around and this was a walk-in clinic and she said, he said, “You’re gonna be laughed out of [the hospital]”. So I said, “Can you just give...” and I literally fought with him. I said, “I’m so sick, can you please?” Thank God he agreed, because he almost wasn’t going to give it to me and then he gave... I got my husband to take her in, I think the next day, he got a blood work. Then that clinic called us and told us we had to bring her immediately to [the hospital] because she was so anemic, she might need a blood transfusion.

    The participant then stated that she “called that doctor and [she] gave him a piece of [her] mind. And he told [her] it was the biggest lesson of his medical career.”
The same participant, when meeting with her other child’s physician, once again had to advocate for her child’s needs to an unreceptive audience. She explained:

As a third roadblock, when I went to my family doctor, I said “[My daughter’s] got polyposis”... [My younger son] is anemic, [he] has got blood in his stool, [he] has got all the same stuff, you know, can you please refer him to [the gastroenterologist]? And she gave me a hard time. “Well, you know, if it wasn’t for [your daughter], I probably would not. I’d probably just automatically...” This should be a no-brainer... He should have been here before. And so I had to like “Please, can you, you know, against your better judgment, please send him to Dr...?” Like, yeah, it really... I don’t know. I don’t know.

In both experiences, the physicians, when first discussing the patients’ symptoms, did not take the mothers’ concerns seriously. In the case of Participant 2, it is particularly evident that the physician did not view the participant’s experiences of her children’s symptoms as valid, positioning herself as the expert and devaluing the mother’s role.

If the dynamic between physician and patient/advocate is to be truly collaborative, the conscious decision by doctors to positions themselves and patients/advocates as equal partners in the medical process is paramount. Such a perspective is in opposition to the positioning of the physician as “expert”, with parents passively receiving information (Elwyn et al., 2012). My findings reveal that in cases where parents’ concerns were invalidated by physicians during the initial phases of their children’s medical care, physicians often positioned themselves as experts unwilling to consider parents’ perspectives. Such is demonstrated in the following excerpt from the interview with Participant 3.
Participant 3: Because every time as a mom I’ve come in and said, I’ve told them what I think and I don’t think they want a mom going on the internet or making their own judgment calls and uh, I don’t know.

Interviewer: So maybe the culture of medicine of they’re the experts...

Participant 3: Yeah, I think so. Like that one doctor that we got into a big argument with. I was so mad at him. I said like, “You see her for 2 minutes. I see her every day. And how dare you think you know her better than me?” Like, even if like you know, she was the happiest, healthiest-looking kid, like for a 2-minute blip, it’s not a reflection of the entire day, you know? And it’s just also mother’s intuition, I think as well. Big time. They don’t take that seriously. And that is valid.

For some of the participants, consultations with family physicians comprised of invalidating the parents’ experiences of their children’s’ health as well as behaving in a manner unreceptive to the parents’ concerns.

**Always on the lookout: Doing everything possible to maintain the child’s health.** When discussing their experiences in evaluating their child’s diagnoses and processing their related emotions, the majority of parents interviewed indicated medical follow-ups as playing an integral role in their attitude toward their child’s condition. Simply having their child assessed by a physician yearly afforded parents the opportunity to reframe their stress, create space for hope, and even perceive their experience within the medical system as “positive”. Overall, yearly medical examinations by their child’s physicians allowed the parents to perceive the nature of their child’s conditions to be less threatening and uncertain.
Participant 1, whose wife had passed away as a result of complications of Juvenile Polyposis Syndrome, expressed the relief he felt in knowing his children were being medically followed. He shared:

Ça a bien été avec le médecin. Il a confirmé qu’ils avaient le polyposé. Ma fille s’est suivie. Ils l’ont poigné jeune, ils ont dit qu’ils avaient beaucoup de risque que s’aggrave puisque qu’ils sont pas suivis... Le moment qu’on sait, on porte plus attention. [Le médecin] et son équipe, elle m’avait dit que ça n’arrivera pas comme ça par que c’est moins surveiller. Elle a dit que c’est à date et c’est clair qu’est ce qui en ait. J’espère que ça .... contre ça. On sait qu’ils sont suivi et qu’ils s’occupent les affaires et c’est beaucoup moins stressant.

Participant 5 explained that for her, her child’s medical appointments also provide reassurance. She stated:

Um, and [the doctor] herself has, I think, made this experience very positive for us, in the sense that uh, you know, she always reassured us and said “He’s on our radar screen and better we monitor him and if something turns up, we’ll catch it early and if not, you know, no harm done kind of thing”. He will have been followed into his adult years.

Furthermore, the consistency with which appointments are scheduled allows parents to challenge their anxiety and uncertainty concerning their child’s condition by meeting with their child’s physician and discussing the progression of their child’s condition. Such regularity of consultation affords parents the opportunity to build their confidence and hope in their child’s future health as well as plan accordingly with the physician. Participant 3 stated:
Yeah, so the mystery has been taken out of it a bit. We know what we’re dealing with. We’re seeing a slight pattern. And we’re seeing a digression, which is great. So things are positive. So I’m being positive with that, you know?

The medical follow-ups were seen as integral in maintaining the children’s health, both current and future. Furthermore, the medical visits aid parents to ease their fears of the development of cancer in their children, a theme that will be discussed further in the remainder of this section.

When discussing their child’s physicians’ recommendations that their child be medically followed with regular follow-ups, all participants expressed the responsibility they feel for ensuring their children attend all follow-ups, scheduled endoscopies. Participant 3 explained that for her, ensuring her child attends all medical follow is her best way of ensuring that her child’s condition does not progress. She explained:

Yeah! And just because I feel like if we weren’t on it, she could, right? But we’re on it. So you know and they assure us all the time, ‘These are never cancerous, these are never cancerous’. So, I believe them. And that is one thing, you know, that it’s not even an issue, it’s not even discussed, it’s sort of like, oh no no no. But obviously it could be at some point but I am under the understanding and I am firmly under the belief, and this is what I’ve read and I’ve heard from them, that it is only when they are left untreated...so I am going to make sure that they are never left untreated. And I guess when they are adults and I can’t drag them to the hospital, well, it’s going to be a different story.
In communicating the importance of medical vigilance, follow-ups for their children, physicians offer parents the opportunity to actively cope with the uncertainty and anxiety resulting from the polyposis diagnosis. Participant 7 stated:

Oui, pour nous le plus... le quoi qui est le plus lourd là dans, c'est de faire passer à [mon fils] les colonoscopies à chaque année ou à, enfin plusieurs fois. Donc, ça évidemment c'est un processus qui est un peu inquiétant mais qui est nécessaire. Alors, pour répondre à la question, on s'est adapté dans le sens que oui, on savait de quoi il s'agissait, on savait également après chaque examen qu'elle était la situation. Donc, on pouvait gérer le portrait avec ça. Alors, je dirai que le facteur le plus important comme dans toutes ces questions, c'est la communication d'information pertinents, la part des médecins, des infirmières aux parents. Et sans informations adéquates, c'est certain que les parents vont s'inquiéter ou ils vont paniquer, qu'ils vont supposer toutes sortes de choses, parce que c'est la nature humaine. Mais si on les offre des renseignements précis et à jour, et qu'on fait de façon régulière, je suis convaincu que ça va calmer une grande partie des inquiétudes des parents.

In the case of Participant 4, medical supervision and follow-ups for her child’s condition served as an adequate course of action to the situation and decreased the level of uncertainty and anxiety related to the diagnosis. She explained:

Yes, that's true and so the other thing is that I have a friend who is a doctor. He's retired, quite elderly now but he had said too at that time when I told him about it, he said 'Oh, if you stay on top of it, it shouldn't be a problem'.
While many parents discussed the anxiety associated with preparing their children for their routine endoscopies and awaiting their results, all participants voiced the importance they place on ensuring their children are medically followed. When asked what advice they would share with other parents of children diagnosed polyposis conditions, many advised that they ensure that their child receives the proper medical care. Participant 1 cautioned:

D'y faire suivres. De ne pas manquez [les suivis], des fois ça change vite.

**Feeling grateful: It could be worse.** When describing their experiences, the majority of participants expressed gratitude as a way of framing their experiences, regardless of the level of severity of their children’s conditions. The participants described two factors for which they are grateful: the fact that their child’s condition is manageable and not “worse”, and the opportunity to be aware of their child’s condition and to be able to remain vigilant of their child’s health.

Parents often described feeling grateful for their child’s condition, as it “could be worse”. Participant 7 explained that for her, focusing on her and her child’s ability to manage his condition was something to be appreciated. She explained:

That’s how you have to make it through; you know you can deal with it.

Participant 5 explained that her child’s diagnosis encouraged her to reflect on the importance of health, explaining:

Well, it certainly forces you to pause and to reflect on the true value of health and being healthy. And um, so I certainly don’t take any of that for granted anymore... I mean, I don't know, gratitude is the foundation of everything, you know? I mean,
um, I feel so blessed. You know, I suppose the diagnosis for our son could have been far worse and uh we could have been in a country where all of this healthcare is not accessible to us. We could have been in the US where it could have cost us thousands of dollars; it hasn’t. [This city] has this amazing physician in [doctor]. Um, yeah, I mean it’s fantastic.

In line with feeling grateful for their children’s health, the parents also indicated gratitude for the opportunities afforded to them in order to be able to maintain their children’s health. Such gratitude helps the parents to be able to reframe their children’s difficult medical procedures relating to their polyposis conditions, and mitigate their experiences of worry. When describing the role of gratitude in obtaining medical care for her child, Participant 4 explained:

I mean, you’re always kind of, not worried, worried is such a strong word, but you kind of think umm ‘hey, my kid is gonna go under general anaesthetic again, that’s not normal. That’s not a normal thing for people to do.’ But, again thankful that we can do it, right? This is a way that we can maintain my daughter’s health, so it’s okay, it doesn’t bother us too much.

Additionally, participants explained that every procedure that reveals positive results provides them with another opportunity to feel grateful.
Peeking through covered eyes at the illness literature

- Seeking validation in shared experiences
- Tuning out anxiety-inducing information

*Figure 2.* Visual representation of the *Peeking through covered eyes at illness* theme and sub-themes.

**Peeking through covered eyes at the illness literature**

During the interview process, the participants revealed their experiences of seeking information regarding their children’s conditions. Consistent with the *Coming Together for the Wellbeing of the Child* theme, the results indicate that parents sought human connection, support in building their knowledge, coping with uncertainty and fear concerning their children’s future health to be important in their adaptation to their new reality. The sub-themes comprising the ‘Peeking through covered eyes at the illness literature’ theme include: a. Seeking validation in shared experiences; b. Tuning out anxiety-inducing information, which is depicted in Figure 2.

**Seeking Validation in Shared Experiences.** Participants described validation of their experiences as an important coping mechanism and learning tool in their lived experiences of parenting a child with a polyposis condition. In addition to seeking information about their children’s conditions online, a few participants discussed
building their own bank of knowledge by meeting with other parents in similar situations. The parents discussed seeking out other parents whose children were diagnosed with polyposis conditions in order to learn about the lives of others’ children, both medically and experientially. Participant 3 described connecting with a parent living in a different country, and comparing her child’s symptoms and medical experience with those of the other parent. The mother explained that through this newly formed friendship, she was able to learn about various procedures and resources available to children with polyps, and explore patterns between the two children’s development of the condition in order to best care for her child. The mother described her conversations with the other mother as more “technical” in nature, explaining:

I’m more looking to learn. I’m very like, I’m pretty technical. I want to know exactly, like you know, do they have the same vitamin deficiencies, do they have the same tendencies? And what do you see when they hit puberty? What do you see moving, like all these sorts of things. In fact, I should probably just start a blog. [Laughs] I really should.

The mother also discussed being very aware of the lack of information available on the topic of polyposis conditions, and feeling that connecting families with similar conditions might be very helpful in creating a forum to share experiences of the conditions. With regards to herself, she explained:

I know. Because I have, I think I have done more than the average person. I really have. I have done this to death. And I have explored it and researched it and everything. So I do think I have something to offer. I really do think I could teach
someone something or I could learn something or whatever. I’m all about piecing this together.

In addition to sharing technical, medical information, Participant 7 found that meeting with older individuals with the same condition as her son enabled her to build hope for her son’s future, as the individuals that she met led happy and successful lives. The participant described her experience of meeting an individual living a gastrointestinal condition as such:

She has no bowel left but she survives, she looks great, I don’t know what meds, I didn’t want to get into her details but she had special surgeries and she’s an advocate... But I guess what I learned is, you know, do any of them feel sorry for themselves? Nope, they’re citizens and they work hard and they... So there’s a positive that comes out of that and I try, I wish my son would have come with me to the event at least because he would have seen that in the world, he’s not alone. And that’s what I want for him, I want him to be able to grow up, have a normal life and not have to stop living because of this disease.

Learning about the experiences of others living with polyposis conditions allowed the participant to observe other individuals’ adaptive coping experiences with polyposis conditions, mitigating the uncertainty and anxiety of what the future holds for her son. The rarity of the conditions hinders both medical information available online as well as the opportunity for discussion about parents’ experiences. Two participants in particular discussed wanting to set up a discussion group for parents of children with polyposis conditions. Participant 2 explained:
So maybe it would be helpful just to compare notes with another parent and see what they're going through, see what they've experienced, you know, what's helped their kids, because there's not a lot of teenagers with this, or young kids.

Participant 3 described her experience in asking her child's gastroenterologist to give her contact information to other parents of patients so that they could share their experiences with one another. However, the participant stated nothing came of her requests, likely due to privacy and confidentiality issues, and that she is therefore on her own in terms of connecting with other parents with similar experiences.

**Tuning out anxiety-inducing information.** Virtually all of the participants indicated that they conducted Internet research on their child's condition following the diagnosis. They described lived experiences of anxiety and uncertainty of their child's future health when seeking information about polyposis conditions. Participant 5 described doing so as a result of “human nature”, as “we all have this tendency, the minute we have something, we Google it, right? We want to know”. Participant 2 explained this tendency as natural, as “everyone’s primary tool for everything nowadays is the internet”.

When conducting their Internet research concerning treatments, lifestyle habits and prognoses, the participant sample as a whole described disappointing results. Upon engaging in the research, two participants described experiencing difficulty in finding information regarding polyposis conditions, especially for younger children. Participant 3 stated:

It’s brutal. There’s nothing. I mean everything is aimed at 50-year-olds, for starters. I mean I can't even get any advice.
Participant 2 also noted the “limited” nature of the information available on the Internet. The same participant also discussed the importance of examining the available literature through a critical lens, lest the available information be detrimental. She explained:

So you have to be careful about what, you know... Luckily, for my daughter, because I was going to be a psychologist at one time and medical training and the statistical training that I received let's me sort of look at things and analyze things from a clinical point of view and sort of “Yes, that’s great but where’s the statistical backup for what you’re saying?”, “Where is the studies, where is this, where is that?” So I’m not likely to fall for whatever gobbledygook they put online. Overwhelmingly, parents discussed their experiences of researching their children’s conditions as anxiety inducing. Reading about other individuals’ “horrible stories” was particularly distressing to the parents. Participant 4 described her experience in researching her child’s condition as such:

I'm not an internet-reader, I don't like to freak myself out so I tend to stay away from the internet. So I don’t need to, I don’t need to go out and look for all the horrible stories because it’s not gonna help us.

Participant 2 echoed this sentiment, stating:

See, I wouldn't be able to tune out the horrible things that they say they're going through.

As a result of the emotions elicited by their online research, parents tended to consciously avoid seeking information from the Internet but rather, placed further trust
in their children’s physicians for informative dialogue about their child’s condition.

Participant 5 explained:

Uh no, I think it’s as my trust in [my child’s physician] grew, that um I realized we were in really good hands. And also, I mean I... You know, I’m just as guilty as the next person, you know? I Googled as well and I found myself getting worked up by you know, you click here, you click here and then you do get to colon cancer, you do get to worst diagnosis possible. And then, you know, soon after I started realizing this, I just stopped and said, you know, how am I helping myself? How is this helping my son? How is this helping us? It’s just causing more angst in the house and we don’t need it. So I had to adopt a very disciplined approach and making a decision that I wouldn’t Google it anymore. It has to be a very cognizant decision that one takes for the benefit of everyone around you or else it can drive someone crazy.

For those who initially read about others’ distressing experiences, they consciously reminded themselves that such an experience would not necessarily be their child’s future. For example, Participant 2 stated:

But the majority of the people that go on these boards are people that are chronically affected by it. So, they’re like, “Oh, you know, I just had another surgery” or “I lost a part of my bowel”, or this or that or the other thing. And why would you bombard yourself with such horrors on a daily basis and read about this stuff? [laughs] I mean, I feel for the people and everything but um their experience is not going to be [my child’s] experience. And maybe she won’t live past 30, maybe she’ll be 90. We don’t know. We can only take it one day at a time.
As a result of the parents’ distressing or ineffective experiences seeking information online, they placed even greater trust in their children’s physicians, and the medical system in its entirety, as competent information-providers. Participant 6 explained his trust in the medical system in providing him with information concerning his son’s health as the following:

"C'est certain lorsqu'on lit dans un article médical, dans des informations médicales, les informations sont souvent générique ou générales ou rédigés pour refléter le situation dans son ensemble. Mais ça peut pas nous informer sur le corps précis de notre enfant parce que la condition est un peu différente, les symptômes peuvent être un peut différentes. Enfin, alors c'est utile pour comprendre l'ensemble du portrait mais on a absolument besoin évidemment des traitements médicaux, des consultations médicales avec les médecins pour bien saisir qu'est ce qui arrive avec les patients qui nous concernent."

Participant 5, when asked how she would advise other parents of children with polyposis conditions, responded with the following:

"I would say 'Have confidence in [your child's physician]. I would say ‘Stay off the internet’. Don’t be, uh, don’t be yourself diagnosis doctor for your child. Stay grounded. Don’t let your imagination run wild because it will. So no need to pre-diagnose anything."

When describing their experiences of learning of their children’s polyposis conditions, the majority of parents highlighted their fear of cancer resulting from the polyps. Furthermore, many explained that this fear continues to be psychologically and emotionally impactful. When discussing the specific moment he was informed of his eldest child’s polyposis diagnoses, Participant 1 stated:
Il y avaient des grosses de craintes. Ils vont devenir cancéreux aux moments donnés. L’esprit disparaître un peu.

For participant 5, cancer was at the forefront of her mind when she was first told of her child’s condition. She explained:

Well, I think it’s instant. Cancer comes into your mind instantaneously. Whether my father-in-law had passed away [of colon cancer] or not, I think, was immaterial. The moment that your son or your child goes through an endoscopy and colonoscopy and the doctor says, “I’ve removed polyps”, regardless of the size, the first instant, you wonder if they’re cancerous. Instant. I mean, that was my reaction.

She further explained her emotional reaction to the possibility of her child having cancer, her reference to the condition as “the C-word” conveying the fear she experienced:

Um, there was initial concern because my father-in-law died of colon cancer, so you know, we always dreaded that C-word. I think there was a lot of concern about that… And then they were sent off to the lab to get tested. And we were fraught with anxiety and stress. I mean, he was a baby. At least, in our minds, he was a baby. Defenseless. So, that was a, you know, tough time but you know, we kept, we stayed positive and focused on the good and in the end, you know, we were very fortunate that the results came back negative and our child is 14 now and he’s thriving and so we’re lucky, we’re very lucky.

With those who continue to fear the development of cancer in their children, uncertainty and the chronic nature of polyposis conditions were found to play
contributing roles to their anxiety. Participant 1 explained the fact that he is acutely aware that although his children have not yet developed cancerous polyps, the possibility of such a development exists. He explained:

Participant: Surtout les émotions, que c’est dur à contrôler...Tu sais qu’ils sont poigné à vie avec ça. Même si c’est pas cancéreux, c’est trop stressant. Les émotions, et ils remontent vites.

Interviewer: Et pour vous, le cancer est encore...

Participant 1: ...plus présent, hein? Ca ne reste pas trop dans le passé, hein? Même s’ils n’ont pas eu jusqu’à date, ca ne veut pas dire qu’ils n’auront pas. Même s’ils ressortent là comme [le médecin] a dit, c’est très rare qu’ils viennent cancéreuses.

The uncertainty of how the conditions of their children will progress, for some, lasts the duration of the condition. Participant 7 explained, in the following excerpt, that for as long as her son continues to live with Juvenile Polyposis Syndrome, she will live with the uncertainty and fear of cancer development in her son.

Interviewer: Was, again this might be a sensitive topic, but was cancer ever a concern for you, or a fear? Can you tell me about that?

Participant: Oh, yes, absolutely. Always. I mean that is from, you know, they tell you ‘We’re not sure, we’re not sure, we’re still checking. We’re not sure’. Well that’s the first thing that you’re going to think. Or will it turn into that? Is it going to be colon cancer? I don’t know. I’ll be thinking about that until I’m dead.

In the case of Participant 3, such fear of cancer and the anxiety surrounding the possibility of its development fostered further anxiety concerning her daughter’s overall health and mistrust of the doctors’ viewpoint.
Like, I keep saying, you know, “Is there any chance she could have a polyp anywhere else in her body that if left untreated could turn to cancer?” Like, I want to know this, even though I know you don’t know for sure, is that still a possibility, you know? “No, no, no, no, no” But, I don’t know if I believe it.

When discussing their experiences with coping with the possibility of their children developing cancer, participants acknowledged both the difficulty of emotions present as well as the conscious decision to persist through their fear. Participant 5 explained:

I would be surprised if there were parents out there who tell you that they don’t think about it. It’s like everywhere, it’s prevalent but then it’s how you manage those emotions and thoughts afterwards.

When asked how she copes with her fear of cancer, Participant 7 stated:

I don’t know. I just do. You just do, yeah. You do as much as you can, I mean I’m not saying that I just have my times when I don’t, I wanna hide under my bed and never come out. But you just have to, you have to for the sake of the kids… No, but quitting is not an option.

Figure 3. Visual representation of the Persevering until needs are met theme and sub-themes.
Persevering until needs are met

Parents discussed advocating for their children's needs, both within and outside of the medical sphere, as an important part of their lived experiences. They discussed the importance of advocating for their children's needs because of the inherent vulnerability associated with pediatric patients, the responsibility they've taken on in modelling advocating behaviours, the importance of their children adopting advocating behaviours due to the chronic nature of their conditions, and the role of maternal intuition in their advocating behaviours. Figure 3 depicts the relationships between the ‘Persevering until needs are met’ theme and its comprising themes (a. Teaching children to speak for themselves; b. Trusting maternal intuition; c. Feeling grateful: I could be worse).

Teaching the children to speak for themselves. Overwhelmingly, parents discussed advocating for their children's health and preparing their children to advocate for themselves to be an important process within their parenting experience. Integral in the parents' advocating for their children's health was the conscious training of their children to become their own advocates. Participants in this study discussed that while they continue to advocate for their children both inside and outside of the medical system, they are beginning to take a step back in order for their children to be able to slowly transition into becoming their own biggest advocates. It is worth noting that the children in question are 17 and 19 years of age, and as such, fall within developmental stages in which individuals play an active role in formulating identity and are tasked with taking on greater responsibility (Munley 1977). The parents noted the difficulty in acting as an “observer...not saying anything”, rather than acting within their general advocate roles. One parent stated that her child’s physician told the mother that her child
had to “speak for himself”, increasing the importance of developing her son’s advocating skills in her own mind. The transition of pediatric patients’ roles as simply patients to that of patient-advocates impels parents to transition from protector-advocate to simply secondary advocate.

In addition to the necessity of parents advocating for their children’s medical needs, participants within the study described taking on the responsibility of advocating for their children’s needs outside of the medical sphere. In particular, participants noted educational settings as sites in which their children’s health posed problems. Some of the mothers explained that because their children’s symptoms were not visible and not easily understood by teachers, it became a necessity to reach out to their children’s teachers to explain their children’s conditions, request flexibility in their children’s study plans and evaluation methods and explain how to proceed if their children became ill in school. Participant 2 explained that she believes that the response of the teachers to her child’s condition and its affect on her school performance have affected her child on a psychological level, stating:

Anyways, so um a lot of them they don’t, they’re not very sympathetic and she’s a very sensitive person so when she senses that they’re not believing her or they start giving her crap for not showing up or falling behind, then she gets anxious about it and then she just doesn’t want to go.

In order to provide her child with an educational setting that would provide her daughter with a more flexible plan to accommodate the child’s symptoms, the participant researched alternative schools for her daughter. However, the participant found that very few education-related resources were available to her daughter, explaining:
They don’t provide a lot of online courses. If someone is chronically sick like she is, then there is no accommodation for them.

As such, the mother has found it to be “a struggle to get her educated”. Participant 7 found that her child’s ability to work a part-time job was also impacted by his condition, as his employer did not allow the individual to use the bathroom as needed. The mother described the ability to go to the bathroom as one pleases as one of the “really ridiculous things that you take for granted”. She took on the responsibility of coaching her son through the process of putting his needs first, stating:

And so I said, “You don’t need the stress, [name of son]. The stress is just gonna kill you, so you know what? Tell them you’re done. Think about what you want to do with yourself. Get yourself healthy. And so that’s where he’s at now.”

Lastly, Participant 3 explained the need to advocate for her child’s needs to the individuals she encounters on a daily basis. The mother explained that her child presents as very hyperactive. The participant explained that such behaviours prompt individuals who did not know her daughter well to judge her and her parenting decisions. She explained,

And no one would give that kid a break. I swear to God, people would just roll their eyes and I’d say to them, like, “She’s anemic. It affects her... It’s beyond her control. She’s been through a lot... They don’t understand that she’s been through hell. And I’m not letting her get away with murder. I’m understanding what she’s been through, you know?

The participant explained that she has had to explain her daughter’s condition to others in order for her daughter to be treated in a way that the mother feels is fair. She
stated that she feels that others sometimes have no compassion because of their lack of understanding of what her daughter has gone through and that she has “always defended her”.

**Trusting maternal intuition.** Mothers in the study described maternal intuition as playing an important part in shaping their lived experiences. When delving into the importance of advocating for their children’s needs, female participants cited their “mother’s intuition” or “gut instinct” as being one of the driving forces behind taking a stand against their doctors’ invalidation of their concerns. Participant 2 stated:

> I just tell anyone, you know any of my friends, that they say something to me about their kid, I say, “Well, go with your gut instinct and if you feel like, you know, doctors are not paying attention to you and you feel there’s something more there, dig in your heels and just go and throw a mummy tantrum on them until someone pays attention.”

The participants explained that they viewed their maternal instinct or gut instinct as being a “valid” tool in their aim to protect their children. From their perspective, their doctors’ invalidation of their concerns and “mother’s intuition” served as a personal affront on the validity of their relationship with their children and knowledge of their children’s experiences, with Participant 3 stating:

> Like that one doctor that we got into a big argument with. I was so mad at him. I said like, “You see her for 2 minutes. I see her every day. And how dare you think you know her better than me?” Like, even if like you know, she was the happiest, healthiest-looking kid, like for a 2-minute blip, it’s not a reflection of the entire
day, you know? And it’s just also mother’s intuition, I think as well. Big time. They
don’t take that seriously. And that is valid.

In the cases of these two participants, their gut instincts or mother’s intuition
allowed them to refuse to accept doctors’ initial incorrect medical advice, as it did not
feel right to them. Such experiences support the call for more dynamic, collaborative
relationships between physicians, patients and their advocates wherein symptoms as
well as experiences are discussed and viewed as valid.

**Taking it one day at a time because the future is uncertain.** When discussing
the chronic nature of their children’s conditions, many parents discussed the uncertainty
of the progression of their children’s conditions as difficult to cope with. Such feelings of
uncertainty were deeply embedded within their lived experiences, with the uncertainty
motivating many of their adaptive behaviours and medical decisions. They also
explained that with their children’s chronic illness comes chronic stress for them as
parents, as they often feel helpless in alleviating their children’s pain. Furthermore, the
inability to imagine what their children’s future health might look like or predict their
child’s next intestinal flare up seems to comprise an important element in their
experience of anxiety. Participant 3 stated:

I mean we just don’t know how these things are going to play out. But that’s the
thing. I mean I get that they don’t know either but you kind of wish you could look
forward where you can go “Oh okay, this is going to be an non-issue in her life
because maybe it’ll just go away”. But the problem is, is that even if they go away,
like who’s to say one can’t just all of a sudden form? A big one, and maybe it’s
really fast. And maybe if she gets pregnant even, it’ll grow, you know? It’s always
going to be this mystery of ‘How fast will they grow?’ And I don’t know if they’ll ever... God, by that time they have any real answers, who knows, right?

Participant 2 explained that coping with such uncertainty is difficult, seemingly impossible and has created an experience of helplessness and hopelessness for her. She explained:

I mean, it’s like a nightmare that you just don’t wake up from. It’s just like... because she’s not well, because it’s almost a daily thing with us, it’s just like constant stress, constant uncertainty, constant like you see your kid struggling and you just want them to be better and they’re not. And there’s no way to make them better. And you don’t have a magic wand and your kid sort of expects you do to make things better. And you can’t. How do you deal with that? How do you adapt to that? You don’t adapt to that. [Laughs]. There’s no adapting to that.

Participant 7 described the uncertainty that transpires when parenting an older child with a polyposis condition, whose mental health has been affected by his condition. She explained:

Oh, honestly, yeah. I mean I’m worried about it getting worse. I’m worried about him losing a bowel. I’m worried about him committing suicide. I guess because in my mind when I see him really in the black zone when he’s really down, I don’t, you don’t know what they’re thinking, right? So you are constantly wondering. Like, there are times where, you know, he’s been in his room too long and I’ll go in there and I’m afraid what I’ll see. So, he certainly doesn’t want to seek any professional help because he sees nothing wrong with what he’s doing so I really
try to talk him through stuff and try to get him to, you know, there are other things...

Moreover, as she explained in her experience in the following interview excerpt, the chronicity of the condition and of the resulting anxiety she feels limits her ability to process her emotions.

Participant 7: ...So you’re more dealing with trying to keep him out of the anger zone that you don’t really have time to think of emotion, you know what I mean? Because I still have to go to work everyday, I still have to function, I still have to you know... It really, I don’t know, I can’t even answer that question. Emotionally, it’s hard. Of course, it’s hard but I don’t have time to be emotional. I don’t...

Interviewer: It seems like you still haven’t even processed it.

Participant 7: No.

Interestingly, two of the participants compared their experiences of their children’s polyposis conditions to their perceptions of other parents’ experiences of their children’s cancer diagnoses. The mothers stated that they felt that the possibility of remission of certain forms of cancer and the non-chronic nature of certain cancers might be easier to cope with than their children’s polyposis conditions. Participant 2 stated:

It was just like, ‘Okay, there’s polyps and that’s what’s causing the internal bleeding, so okay, we know it’s not cancer, phew.’ It’s not that but you know, in hindsight, cancer might have been a better thing because you know, if it’s not that advanced, you can treat it and be done with it whereas this is a lifetime sort of thing, right? It’s not going away. It’s not going anywhere.
Participant 3 described a conversation she had with fellow mother she had met whose son was in remission from cancer and who had initially invalidated the participant’s daughter’s diagnosis of Juvenile Polyposis Syndrome. She explained:

But it’s funny because I ran into her afterwards and I was like, you know, “How’s [your son]?” “Oh, he’s fine, totally clean bill of health” and then she was like, “Oh, how’s [your daughter]?” And that’s when I was like “Oh, yeah”, so now you might get it. Like, your son doesn’t have cancer anymore but my daughter still has this condition. So suddenly what was unimportant – and I just said “Oh, she’s fine”, whatever – but it was a bit of a light bulb moment when I was like, “Yeah, now this seems slightly more important to you.”

The majority of parents stated that remaining vigilant regarding their children’s health and ensuring their children participated in the medical follow-ups was helpful to them in coping with the uncertainty implicated in their children’s conditions. Furthermore, adapting their expectations from hoping their children’s conditions would eventually be cured to simply managing their children’s symptoms was revealed to be helpful to some participants. Participant 7 discussed setting small, achievable goals for her son’s health. She explained:

Well, I think I’m just... I don’t think... Like, I think I just constantly worry. Like, I’m always looking for... You know, you’re in the bathroom a little bit too long, are you okay? Just over the top now, right? He’s pretty good though to tell me when things are really not going well. But they don’t go well a lot, so our relationship now is all about “how do you feel?” You know, not “Oh, how was your day? What did you do today?” It’s just, “Did you have a good day today or was it a regular day?”, you
know? So yes, I don’t know if that will ever change because his condition seems to be something he’s going to have to live with. Our goal is not to have surgery again. Our goal is to keep the inflammation at bay, right? So how do you do that? Because I’m not going to tell him what to eat at night or what to do or what not to do. I’ll have to deal with it when it happens though. I wonder if they’ll let me stay with him in the hospital. [laughs]

Participant 6 explained that while he understands that his son’s condition is chronic, he takes solace in the fact that the illness is under control. He stated:

Donc, c’est pas pour dire que c’est finit pour jamais mais semble être sous contrôле.

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**Figure 4.** Visual representation of the *Creating a new ‘normal’ as everything becomes unsettled* theme and sub-themes
Creating a new ‘normal’ as everything becomes unsettled

The final theme, Creating a new ‘normal’ as everything becomes unsettled, comprises the following sub-themes: a. Becoming a different type of parent, b. Trusting they will speak so they can be heard, c. Putting families to the test: coping with the illness as a family, and d. Feeling as though no one understands. The sub-themes, depicted in Figure 4, expand on how the polyposis diagnosis affected the lived experiences of parents on a relational level. Furthermore, they will discuss the parents’ lived experiences of their evolving identities and how such changes have influenced their relationships with themselves and others. The findings reveal that such changes affect the ways in which the parents seek and receive support.

**Becoming a different type of parent.** When discussing their lived experiences, parents described an evolution in their self-identities as parents, which facilitated and motivated their advocating and medical decision-making. Overwhelmingly, participants stated that once their children were diagnosed with polyposis conditions, they began to view themselves as “protectors” of their children. The participants described ‘protecting’ their children from their conditions, but also noted protecting their children’s innocence, mental health, sense of normalcy and family relationships. Participant 1 discussed trying to create a bubble of protection around his children without smothering them in his role as their protector:

Mais c’est sur que tu te vois comme protecteur un peu... J’essaye de les protégé un peu plus là. Essaye de faire une boule plus autour sans les étouffer. Tu provoques qu’est ce qu’ils font et comment ça va à l’école et t’es plus proche à eux, tu les tiens proches, tu veux te rapprocher un peu plus.
Participant 7 echoed Participant 1’s sentiments in taking on the role of “protector” for her son, although she used to term “The Caregiver” to describe her experience in parenting her son. She explained her evolved role as such:

I mean I sang in a band for 15 years and I was always out and doing that. And then I quit because it was just too much too worry about him and do you know what I mean? You just can’t do it all and be yourself. You can’t be who you are and be mom and be, you know, and then be the caregiver. I call it The Caregiver, I mean you are a parent, you should be but it’s just that extra level of worry that you take for granted.

The mother explained that an important distinction between mother and caregiver is the experience of worry and anxiety she feels as her son’s caregiver. As she explained, in the role of a caretaker, it is necessary to place her child’s needs before her own and prioritize the adopted role of caretaker above other identities she may hold.

**Trusting they will speak so they can be heard.** When discussing their relationships with their children, the majority of parents described a development of trust through communication. Building such a relationship was essential to their parenting experience and plays a meaningful role in their lived experiences. They explained that their children’s level of communication and trust in them as parents served as a necessity, as the parents needed to be aware of their children’s polyposis-related symptoms. Participant 5 described the importance of communication with her children as such:

And I communicate with the kids regularly about everything because if something does show up in their stool, it cannot be an embarrassment for them to tell us. If
whatever is happening is happening to their bodies, they have to have that level of confidence and comfort to come to us. So if anything, I think I’ve probably become any even greater communicator with my kids.

The trust and communication built between parent and child also creates opportunities for parents to teach their children how to manage their conditions thereby removing the stigma of discussing uncomfortable topics such as blood in stool and stool consistency. Participant 3 described her communication with her young children about their conditions as the following:

And the children are trained to look at their stool... Not the older ones. [My children] are always, “Ah mom, there’s blood”, you know. They’re so nonchalant. “I saw blood” “Okay, show me”, you know? So it’s like, ah [sigh]. Anyone else would be like... [Laughs]. I’m just so blasé about it now. You know, and I’ll often have to check their stool. Is it solid? Is it, you know, if they have chronic diarrhoea, that’s a tip off that there’s something going on, meaning that it’s blocking the stool, right?

Children’s communication of their medical symptoms to their parents, especially in the case of younger children, facilitates parents’ roles as advocates within the medical system as well as outside of the medical system as they are better informed of what their child is experiencing. In one case, Participant 7 explained how the trust she places in her child to notify her when he is feeling unwell, helps her to mitigate her anxiety about his condition. She stated:

I think I just constantly worry. Like, I’m always looking for... You know, you’re in the bathroom a little bit too long, are you okay? Just over the top now, right? He’s
pretty good though to tell me when things are really not going well. But they don’t go well a lot, so our relationship now is all about “how do you feel?” You know, not “Oh, how was your day?”

Such lines of communication only become more important as the child grows older and other factors such as puberty, peer pressure and need for independence play a role in the parent-child relationship. Participant 2 explained that as her daughter progresses through her early teenage years, she has found that the emotional impact of Peutz-Jeghers Syndrome has begun to become a topic that is more often discussed in their conversations. She explained,

She needs to have one person that she can always go to no matter what, especially with the depression and anxiety… I think I’m probably closer to my daughter than I would normally be.

While the parent-child relationships of families dealing with polyposis conditions parallel the parent-child relationships of families not affected by the condition, these findings reveal that the development of trust through communication is of paramount importance when raising a child with a chronic illness.

**Putting families to the test: Coping with the illness as a family.** Although they brought forth varying challenges, all the participants discussed their children’s conditions as having an impact on their family dynamics. As previously discussed, the participants served as advocates for their children, built upon a foundation of trust between child and parent. They explained that as their children’s illnesses progressed, their ways of ‘being’ within the family evolved. The participants reported that the impact
of such a close, unique bond between themselves and their affected child affected their children’s relationships with their other parents and siblings.

Participant 2 explained that while her daughter will ask her for advice and while they share a closeness between the two of them, her daughter does not feel the same about her father. She explained that because her husband holds “this sort of dude mentality of ‘Suck it up and get on with it... and just dealing with’, her daughter no longer seeks the support of her father. The mother explained that this can be difficult for her husband, as he “resents” the closeness of their relationship.

Participant 7 also explained that because of her husband’s way of responding to their son’s illness, her son does not feel comfortable seeking support from his father. She explained that she does not feel her son receives “a lot of support from his dad because his dad doesn’t know how to support him without it become a fight. And his father would love to tell him what to do.”

The participants also described the impact of their children’s disease in the way their children relate to their siblings. Participant 3 explained that the changes she has made in her parenting style and her role in the family since her child’s diagnosis has not only impacted her child with Juvenile Polyposis Syndrome but her other children as well. She stated:

Oh yeah, well it’s definitely put us on a different path for sure. Just everything, like the concept of our family in some ways and yeah, I sense her... Like my eldest, I can tell that he treats [child with Juvenile Polyposis diagnosis] differently. I can tell that, you know, my second, my daughter, my second eldest, I don’t think she really gets it, like she does but I can see with my oldest, he’s concerned but not
saying it, you know. And so I can see how it plays on them a bit. And then um I mean it’s going to be something that is always there, you know? And yeah. Participant 7 explained that her son’s diagnosis and experience of his condition have had longstanding effects on her son’s relationship with his sister. She stated:

And he has an older sister who, I think she’s almost resentful at this point because she feels that everybody worries about [her brother], right? And it’s ‘cause it was, that’s the way it was. I mean we had a family holiday booked and we had to cancel because he wasn’t allowed to go anywhere. And she was in her, I can’t even think what she would have been in, he was 13, she would have been 16 or 17, I guess. So, it was probably the end of school, I forget. It was an important year for her. But we couldn’t, I didn’t have the energy to do it all, right? And now they don’t even have a relationship because he resents her for being resentful, right? It almost sounds like... I think he interprets it as she’s being very jealous of him, which sounds like she is. When now that she’s 24, she can get over it, and she wants a relationship, but he is not there yet. So she lives in [another city], she goes to [school]. And it’s good that they’re not together right now, but it’s sad. It’s sad because they were very close.

Finally, participants described their children’s diagnoses as having negatively impacted their relationships with their spouses initially, but ultimately creating an opportunity and need to reflect on their relationship for the sake of their children. Participant 5 described the impact of her child’s diagnosis on her marriage as the following:
Yeah. I mean, I suppose it did. How can it not, right? But I think there was so many other facts that impacted our relationship that that was just one of many. So it was one of many stressors, which eventually led to the breakdown of the marriage. I wouldn’t say it was it was the sole stressor, far from it. I wouldn’t even say it was the biggest stressor, far from it. If anything though, I think the diagnosis forced his father and I just to stop bickering and stop the dynamics of our relationship and have a different relationship for this kid, which is hugely beneficial today because his father has re-married, I have remarried, this kid has lots of step-siblings, and everybody gets along, you know? We get a long very well with his father and his new wife. So, you know, we’re very lucky. There’s no um, there’s no acrimony between the families, the children all seem to get along, so we’re quite lucky.

Participant 3 described a similar experience, in that she and her husband were able to adapt their relationship over time to create healthier dynamics in order to better support their child. She explained:

Well I kind of know what to expect now. Now I know exactly what to expect. And I still get stressed but like you know the last time we went in, [my son] went in the last time, I felt, like I didn’t feel like I was having a full-on anxiety attack, like I was going to throw up. So it’s like like, ‘okay, I’ve come a long way’. My husband is pretty good, he just draws and zones out. And he’s just like “You can’t stress, it’s beyond your control”, you know? But I will. And then I tend to sort of spend my energy or get frustrated at the lack of support. And that seems to be a focus but whatever...I guess my husband and I are probably closer now, kind of coming
through on the other end. There was a time when our marriage was very strained.

And uh, I'm sure this played a big role. For sure.

In sum, the majority of participants discussed tension arising when individuals’ emotional and social support needs were not met within the family. The needs, specifically in the cases of the children with Juvenile Polyposis Syndrome and their siblings, paralleled developmental needs of their healthy peers. However, the stressors inflicted by the conditions increase the severity and importance of these needs.

**Feeling as though no one understands.** Parents expressed not having their emotional and social support needs met by their support networks, as impactful and concerning part of their lived experiences. Participants brought forth a wide variety of experiences of sharing their children’s diagnosis and overall health with family members and friends. My findings reveal several factors affecting the quality of social support the participants received. The majority of participants described not receiving social support in line with their needs. Participant 3 explained that her family did not meet her emotional needs during the early years of her daughter’s diagnosis. She stated:

Oh, I’m going to get emotional about it. [Crying] Um, because we had really poor support from our family, that was the hardest thing, I think. Because we had to do a ... Our families for some reason treat it as though it is completely nothing. They act like, “Oh, whatever”, like “Get over it”. And when we have to do um, when we have to do uh the fasting and stuff, it’s very stressful because we have other children, especially when [our child] was younger ... Our parents were just not there for us at all. Like, we were like, oh my God, we were so stressed out. And I got strep throat. I had [my daughter] and [my son] go in back to back for a
procedure and it was the first time [my son] had gone in and I was terrified of what they were going to find. Um, luckily it was only one, but you don’t know until you go in, right? And he had quite a bit of blood. Um, he kept saying that he was bringing up stuff and that he could taste blood in it. And, so I mean, and he was little. And uh, my mom, every time would see our kids go in, because [my daughter] has been in 9 times I think, [my son]’s been in twice, anyway, my mom would conveniently leave town and my mom and my sister didn’t even… They didn’t call me the day before, when they knew that I was a wreck, they called me, we had the procedure in the morning I think, they called me like 6:00 the next night. And I just lost it on them. I just said like, like whatever, it’s over, like... You know, like sure they weren’t diagnosed with cancer but in our eyes, it was terrifying. So, that wasn’t good for sure. Um, it was very stressful on my husband and I, without a doubt. I think we’ve kind of come over it. The worst was when they went back to back, that was hard [Crying].

When asked how she would have liked to her family to have responded to her needs, the mother explained:

I just would have loved someone to have called and said “It’s going to be okay, don’t worry about it”, you know? … Even just that, or good luck or anything. You know, because we’re always very aware, like it’s scary when anyone is put out. There is the chance that you don’t wake up, you know? There is. Um, so I just couldn’t believe that uh... I just don’t know. I just don’t. I really don’t know. It was to the point of almost eye-rolling.
Such responses from her family members dissuaded the mother from seeking future support, she explained:

I don’t even; I don’t think I would even bring it up again really with them.

The mother further elaborated that she felt her family did not fully appreciate the seriousness of the condition, with very little understanding of the condition. She stated:

They don’t think it’s a big deal because they look at it as like... They all say the same thing... Like my sister-in-law says, ‘Well, my mom has a polyp’ and so and so has a polyp, but it’s a condition in 50-year-olds, it’s common. So to them it’s not a big deal, so it’s ‘I know tons of people with polyps and tons of people who have colonoscopies and get over it’. Well, hello. This is a child. And have they had 50 polyps removed? ...They don’t seem to think it’s a big deal. They’re like, ‘just take them out’. And when I talk about it, I sort of say like... Like, sometimes they’ll say things to me and it’s like they don’t understand at all and I’ll be... Like, my sister was like, “Oh, I talked to my friend and she said you should take this because it makes polyps go away” and I’m like, “No, no, you don’t understand. Like, this isn’t like polyps like grandpa had or whatever, you know? Like, this is different.”

Participant 2 echoed similar sentiments, reporting that she felt others in her life did not understand her child’s condition. She explained:

They just, they don’t get it. They have never heard of it, they just don’t know what this means and like I said, you say ‘polyps’ and it sounds so cute, you know? What could a polyp do to you, you know? People have heard of polyps and they’re like... “Well that doesn’t sound so bad, does it? You know, you remove it and you’re
good.” But when they’re constantly growing and you don’t know at what rate, you can’t see them on the outside...

Participant 5’s experience of sharing the news of her child’s diagnosis with her family demonstrated the value of conveying information and fostering understanding about the condition. She stated:

Uh, no. I think everybody was just kind of just “Oh, what is it? What does that mean? What are the risks? What are the possible diagnoses?” And so it just led to very factual conversations and keeping people apprised of what was happening. And um, I mean our families were very supportive and very understanding, and so were our friends and employers. So, yeah, I don’t um, I don’t recall anything negative about that either, just very factual.

Part of the reason why others are likely to underestimate the seriousness of polyposis conditions and ill-appraise the affected parties’ need for support, as explained by Participant 3, is the fact that the condition oftentimes does not prevent children from presenting differently from a healthily developing child. The following excerpt demonstrates how the participant’s daughter’s outward appearance does not reflect the child’s overall experience and health challenges:

Interviewer: And especially because you can’t tell outwardly.

Participant 3: Exactly.

Interviewer: And your daughter is full of energy. She’s probably the last person you’d assume.

Participant 3: Exactly. If she was sickly, then she would have... But they don’t understand that by being like that, it’s still sickly.
The difficulty of individuals to reconcile illness with an outwardly ‘healthy’ appearance extends not only to those unfamiliar with polyposis conditions but to physicians as well. My findings reveal that physicians often discounted parents’ initial requests for further examination of their children’s symptoms or overall health, with physicians sometimes invalidating parents concerns by stating the children’s symptoms were simply part of normal development. Experiences such as these demonstrate a lack of support from physicians, ushering in experiences where, for many parents, support is not available.

Another reason why individuals, including physicians, are unlikely to initially understand the complexities and seriousness of the condition is the rarity of disease. Participant 3 explained:

The rarity of it doesn’t give it any exposure, it doesn’t give it anything.

While the rarity of the condition hinders education about the condition, it also makes it very difficult of individuals affected by the condition to find others who “get it”. When discussing whether being in contact with other families affected by polyposis conditions would be helpful, Participant 2 responded:

And maybe, like, maybe if... Like, how would you find another parent with, you know, a kid with Peutz-Jeghers? How would you even go about that? Like, are there other parents in the [city] area? How would you even locate them? Like, there’s doctor-patient confidentiality and you know, it’s not like [the doctor] is going to say, well, ‘Oh there’s like one or two other people’... Yeah, I think it actually would be helpful, yeah. And then even for my daughter, maybe, to meet someone else her own age with the same thing and sort of say “Well, you know”...
No one... She can explain to her friends but no one really understands what she’s going through.

In addition to others being ill informed about polyposis conditions, Participant 3 also noted the stigma attached to polyposis conditions because they affect the rectum. The mother discussed the discomfort she believes others feel as a possible reason why others are hesitant to discuss her child’s condition. She stated:

Participant 3: It’s a weird, it is a weird thing, right? Because it’s to do with the bum, right? So people don’t want to go there, they don’t want to talk about that area.

With regards to parents’ experiences of social support, it is important to discuss parents’ psychological readiness to receive support. Participant 2 discussed not wanting others to engage her in conversation about her daughter’s health, possibly due to her expectation that the support will not meet her needs. She stated:

You can smile and “Oh, everything’s fine” but it’s not and you just don’t want to be asked. Um and because it doesn’t run in our family, no one really understands.

Lastly, Participant 2 discussed hesitating whether to tell her family members of their children’s conditions for fear of burdening them. She explained:

At first, because I told my husband my mother has high blood pressure, both sets of grandparents are still alive, I said ‘I am not telling my parents until we know exactly what this means and then when we do know exactly what this means, then I’ll decide how much or what they need to know.’ Because, um, you know, it’s just upsetting to my mother and she’ll just worry a lot and then it just puts her blood pressure up, you know.
Descriptive Identification of the Phenomenon Structure

In dealing with the anxiety and uncertainty brought on by their children’s diagnoses, our participants acted proactively in working to ensure their children’s current and future health, thereby allaying their own distress. Their means of adaptation reflect the use of relationship-building behaviours to meet these ends, including forming relationships with their children’s physicians to ensure proper medical care, strengthening relationships with their affected children in order to facilitate advocating on their behalf, evaluation of their relationships with family members in order to better adapt family dynamics and reflection upon their relationships with themselves in order to better understand their identities as parents.

Physicians who were able to effectively communicate information while also making parents feel heard, served as valuable sources of support, which my findings indicate parents of children with rare, chronic illnesses desperately lack. Furthermore, physicians who were able to earn the trust of parents by engaging them in family-centered care were able to effectively convey to the parents the importance of continued medical surveillance, an important component for the continued medical care of individuals with chronic illness. Parents also discussed the importance of creating relationships with other individuals in situations similar to theirs, as the rarity and stigma of their children’s conditions can be isolating. Positive experiences in which parents are able to share their concerns and stories with others, whether professionals or non-professionals, are meaningful to parents, evidenced by the gratitude they expressed in the interviews.
In parenting their children, the parents acted upon their felt responsibility to teach their children the importance of advocating for their needs, both within the medical system and within other contexts. They saw this as an integral part of their parenting role due to the chronic nature of their children's conditions, and the importance of continued medical surveillance to the health of their children. They also discussed how their roles as parents and individuals have changed, having taken on the roles of 'Protector' and 'Caregiver', and with it, and the evolution of their family dynamics. Specifically, parents expressed consciously fostering effective means of communication and trust with their diagnosed children in order to better understand their day-to-day health and more effectively advocate for their children's needs to the physicians. The usual social support systems of the parents oftentimes did not present the forms of support helpful to the parents in their times of needs, and as such, they reported feeling isolated in their experiences.
Chapter 5: Discussion

This chapter positions my findings within the research literature and discusses their relevance to the medical and counselling domains. All of my revealed findings align well with current chronic illness, rare diseases and polyposis literatures. The ‘Teaching children to speak for themselves’ theme proved to be the most novel of the themes, in that the current research literature has not yet explored this matter in depth.

Furthermore, in this chapter I bring forth limitations that should be considered when reviewing this study’s findings. Lastly, my final conclusions regarding this study and possible avenues for future research based on my findings are discussed.

Summary of Findings

My findings reveal that a diagnosis of a polyposis condition brings with it a great amount of fear and uncertainty. In order to adapt and combat these feelings, parents have developed various tools for allaying their fears and ensuring their children remain healthy. The parents discussed how they experienced gratitude and reassurance when physicians worked collaboratively with them to maintain their children's health. When parents felt their children’s needs were not being met or their instincts about their children’s health not recognized, they did not hesitate to advocate for their children. In order to combat the uncertainty the parents felt regarding their children’s future and to best prepare their children to maintain their own health, the parents discussed their experiences in teaching their children to advocate for themselves. The participants also discussed their experience in building trusting, communicative relationships with their children, as it is integral for them to be aware of their children’s health. Lastly, parents discussed their parent identities and family dynamics evolving with the progression of
their children’s illnesses, with most individuals not understanding their experiences. They discussed seeking others who share their experiences in order to receive the validation denied to them by the individuals who simply do not understand their experiences, and to build their knowledge about their children’s conditions.

Discussion of Findings

The aim of this research study was to gain qualitative insight into the lived experiences of parents of children with polyposis diagnoses. The findings compiled in this study add to the limited research literature on the experiences of parents of children with chronic, rare illnesses as well as the polyposis literature. Overall, the findings of this study parallel those of prior research and contribute novel insight on the lived experience of this phenomenon.

When reviewing the collected data, one of the major themes that emerged was parents’ need to have their experiences and concerns heard and validated by those within their support networks, which includes physicians. When discussing their experiences, all participants discussed in-depth their physicians’ abilities to support their families by listening to their concerns, informing them of how to best care for their children, and supporting them through their process of adapting to their children’s diagnosis. While parents witnessed their children receiving medical care, their focus of discussion during the interviews prioritized their experiences of the support they received (or didn’t receive) and its impact on their experience. Parents also discussed their need to be able to share their experiences with other parents of children with polyposis conditions, to once again feel heard, have their experiences validated, and possibly benefit from other individuals’ insight. Our study demonstrates the importance
of parents of children with rare and chronic conditions having an opportunity to express their experiences and needs as well as the importance of hearing others’ stories to know that they are not alone. In this way, the relationships that are built are healing.

The findings reveal that participants’ relationships with their children’s physician plays a key role in determining the quality of their experiences. Parents’ whose children’s physicians took the time to explore their concerns and assess their children’s symptoms accordingly demonstrated trust in their medical overall system and expressed having many of their concerns assuaged. In a study by McCormick and colleagues (2012), family physicians were found to invalidate patients’ experiences of gastrointestinal pain, resulting in patients’ emotional distress. As with my findings, Drossman, Chang, Schneck, Blackman, Norton, and Norton (2009) found having their experiences and concerns understood and acknowledged by their physicians aided Irritable Bowel Syndrome patients’ ability to function in their day-to-day lives. Drossman, Creed, Olden, Svedlund, Toner, and Whitehead (1999) in their review of psychosocial experiences of individuals living with gastrointestinal disorders suggest physicians use a patient-centred approach when discussing patients’ experiences, exploring both psychosocial and physical components. My findings suggest that physicians’ use of patient-centred practice should extend to the parents of pediatric patients, as well, utilizing a family-centered approach.

As quality of care has been demonstrated to play a significant role in health-related quality of life, patient-centered medical practice continues to prove its value in patient-physician relationships (van der Eijk et al., 2004). Even seemingly unimportant gestures that patients interpret as ‘courteous’ have been found to significantly benefit health-related quality of life (van der Eijk et al., 2004). The participants expressed the
the importance they place in trusting in their physicians, with such trust allowing the participants to better cope with their uncertainty and anxiety. Specifically, my findings reveal that a collaborative relationship with their child’s physician enables parents to play a more active role in their children’s medical care, thereby affording parents an opportunity to actively cope with their feelings of helplessness. Trust demonstrated by patients in their physician has been shown to be a predictor of adherence to medical practices, an important factor in cancer-related conditions such as polyposis (Nguyen, LaVeist, Harris, Datta, Bayless, & Brant, 2009). Taken together, my findings indicate that while consulting their children’s physicians and reflecting on their children’s conditions can be a very anxious time for certain parents, follow-ups with their children’s physicians, especially when the doctors facilitate a collaborative relationship, ultimately play an important factor in reducing parents’ anxiety. Specifically, scheduled follow-ups and physicians’ surveillance allows parents to somewhat allay their fears of the development of cancer and unforeseen medical issues.

Some of the participants reported wanting to be able to interact with other individuals in situations similar to theirs, with online support forums not meeting their current needs of gaining social support and collecting information. Although all of the participants reported using the Internet to build their understanding of their children’s conditions, my findings do not entirely parallel those of prior research. Many of the participants cited information found online to be too general (and therefore not necessarily relevant to their children’s specific situation) or too anxiety inducing for them to continue to use the Internet as a source of information. While research demonstrates links between inflammatory bowel diseases and anxiety, the findings
provide insight into specific triggers for such anxiety (Graff, Walker, & Berstein, 2009). In line with prior research in studies focused on inflammatory bowel disease patients, the participants reported viewing their children’s physicians as their preferred source of information (Bernstein, Promislow, Carr, Rawsthorne, Walker, & Bernstein, 2011; Cima et al., 2007). Such findings further highlight the importance of the trust built between parties and the need for parents to be able to ask physicians questions without fear of judgement or disregard.

Interestingly, a few of the participants voiced wanting to be able to both share the information they’ve gathered concerning their children’s polyposis experiences and gain insight into others’ experiences. Such a need seems to be in line with the fact that many of the participants’ experiences felt invalidated by those with whom they initially shared their stories, including family members, friends and even physicians, ultimately creating a sense of isolation. As such, it is likely that the participants are seeking information within an environment in which they feel they are likely to receive support and a shared understanding of their experiences. Shared experiences as a form of information seeking may resonate more with parents and may be more easily understood than scholarly online journal articles or medically-focused Internet pages. A study by van der Marel, and colleagues (2009) found that 57% of online websites posting information concerning inflammatory bowel disease scored either fair or poor using a readability-rating instrument, indicating that the general population may experience difficulty understanding much of the information on the Internet concerning their condition. Certainly, the participants discussing their need for interaction with others in similar situations did not suggest that anecdotal information gleaned from such encounters
could replace consultation with medical experts, however, the findings indicate that
information/experience sharing with other individuals affected by polyposis conditions
offers a form of support, care, and refuge from isolation that physicians alone cannot
offer. Sharing experiences in contexts such as in-person support groups would help
parents reduce their feelings of social isolation, and self-stigmatization. It is worth noting
that a resource for children and their families seeking to meet other families affected by
polyposis conditions is currently available through the Mount Sinai Hospital’s Zane
Cohen Centre for Digestive Diseases located in Toronto, Ontario (Mount Sinai Hospital,
2014). Although no current research exists on the effectiveness of the program, my
findings would suggest that the resource serves an important need within the polyposis
community. Although it is unfortunate that none of the participants included in the study
were aware of the program, I have discussed the possibility of better informing patients
and their families about the program with the physicians in the Genetics and Pediatric
Gastroenterology units.

Participants discussed the important role gratitude plays in framing their
experiences regarding both their children’s health and their experiences with the
medical system. Within the literature, gratitude has been conceptualized as the emotion
resulting from recognizing and appreciating the advantages or rewards one has been
given or experienced (Wood, Froh, & Geraghty, 2010). Within the context of this study,
parents noted feeling grateful for the manageable nature of their children’s condition
due to available medical resources and the comparatively mild nature of the condition.

Within the positive psychology research, gratitude has been positively linked to
well-being. Specifically, McMillen (1999) found that individuals demonstrating high
levels of gratitude while facing stressful life events were likelier to utilize coping skills, adapt their behaviours and reflect on their strengths than those who did not experience gratitude when faced with comparable situations. Within healthy adults, gratitude has also been negatively correlated with anxiety and depression (Petrocchi & Couyoumdjian, 2016).

Within the illness literature, research indicates individuals living with heart failure, a chronic condition, were found to use gratitude as a coping resource, with participants specifically citing medical resources and social support as cause for gratitude (Sacco, Park, Suresh, & Bliss, 2014). While gratitude has also been found to be positively related to emotion-focused coping and utilization of psychological resources in familial caregivers of individuals living with dementia, virtually no research is available on the role gratitude plays in parenting children with chronic illness.

My findings reveal gratitude to be an important coping resource by which parents were able to reframe their anxieties and distress. Although no previous research explores the experiences of gratitude of parents of children with chronic illness, taken together, the findings of this study seem to be in line with the available previous research.

One of the major themes revealed by the data, Becoming a different type of parent, discusses the idea of the ‘Protector Parent’. The term ‘Protector Parent’ is used to portray the essence of the ways in which the parents relate to their children as a function of their conditions. The participants reported that their children’s diagnoses represented a concerning threat to their health, and adopting the ‘Protector Parent’ identity and role were borne of necessity. The role serves to ‘protect’ the children from social isolation.
from society and invalidation of symptoms on the part of physicians. My findings reveal, however, that the role of 'Protector Parent' can influence the parents’ other identities and change social dynamics within the family and support systems.

As discussed in the literature review, participants reported placing high importance on the value of advocating their children's needs within the medical system (Dellve, Samuelsson, Tallborn, Fasth, & Hallberg, 2006; Zurynski, Frith, Leonard, & Elliott, 2008). Interestingly, the findings reveal that in advocating for their children, some of our participants were consciously modelling behaviours they hoped their children would eventually adopt for themselves. Research indicates that a pediatric patient’s developmental maturity, which comprises competencies such as psychological coping, understanding of illness self-management, and communication skills, is the most predictive factor in successful transitions for inflammatory bowel disease illness from pediatric to adult medical care, which was measured in quality-of-life, health, and medical adherence outcomes (Paine et al., 2014). Furthermore, the level of involvement by parents in supporting their children through the transition was also found to have predictive value for successful transition (Paine et al., 2014). As demonstrated through the reports of our participants, transition between pediatric medical care to adult medical care can be a difficult process for both parents and children. Research indicates that adolescents often lack knowledge of their medical history, skills in advocating for their needs and organization of their medical care (Hait, Barendse, Arnold, Valim, Sands, Korzenik,, & Fishman, 2009; Sebastian, Jenkins, McCartney, Ahmad, Arnott, Croft, Russel, & Lindsay, 2012). Within the Social-Ecological Model of Adolescent and Young Adult Readiness to Transition developed by Paine and colleagues (2014), parents serve
as part of the child’s microsystem, the most immediate system in affecting the child’s experience and therefore hold opportune positions to influence their children’s development and behaviour. Further research supports the role of parents in teaching their children to self-advocate (Daly-Cano, Vaccaro, & Newman, 2015), with research also indicating that over-protective parents who voice their concerns about their children’s abilities to self-advocate ultimately hinder their children’s likelihood of success in developing self-advocating skills (Dowrick, Anderson, Heyer, & Acosta, 2005).

My findings support the notion that parents play a pivotal role in preparing their children for such a transition and that their advocacy in their children’s early years benefits their children’s medical care long after they have graduated from pediatric medical care systems.

The findings also indicate that parents’ advocacy for their children extended beyond the medical system. The participants discussed the necessity of advocacy in their children’s schools and places of employment as well. These findings are in-line with current inflammatory bowel disease literature, which has demonstrated that teachers sometimes lack empathy towards the symptoms and experiences of their students with inflammatory bowel disease, generally as a result of lack of knowledge and the lack of visibility of the condition (Gordon, 2015). Research also indicates that parents of children with inflammatory bowel disease also needed to consistently meet with their children’s teachers to discuss re-evaluating their children’s needs and accommodations, responsibilities also taken on by some of the parents in our sample group (Gordon, 2015).
The results also support previous research indicating that families of children with chronic illnesses often experience a change in family dynamics. Specifically, some of our participants indicated their children’s conditions exacerbated difficulties in their marriages, paralleling results demonstrated by Dahlquist and colleagues (1993). In a study conducted by Engstrom (1999), families with children with irritable bowel syndrome were found to experience reduced family functioning, compared to families with children with diabetes, families with children with chronic headaches, and families with healthy children. However, within the group of children affected by inflammatory bowel disease, the author found a subgroup of children who demonstrated healthier psychological states than others with the same illness. This subgroup reported healthier family climates, wherein they had access to an individual in their family, generally their mother, who encouraged them to discuss their feelings about their conditions (Enstrom, 1999). Findings by Nicholas and colleagues (2007, p. 6) reconcile these results, demonstrating through their research that support given to children with inflammatory bowel disease from their parents is most effective when it “includes information and emotional support presented in an environment of honest, age-appropriate and sensitive communication”. The findings support this research, as some of our participants indicated needing to support their children in certain ways in order for their children to accept such support. Specifically, two mother participants of older children indicated that their children preferred to seek help from them because their methods of support were more validating and less directive than that of their husbands.

Prior research and the findings indicate that parents of children with chronic illnesses often take on additional roles for their affected child, such as Advocate and
Caregiver (Kratz, Uding, Trahms, Villareale, & Klekhefer, 2009). The findings reveal the importance of strong parent-child ties, not only in building healthy family but also for the effectiveness of the parent’s role as advocate. Children affected by polyposis conditions need to feel comfortable in confiding in their parents when symptoms, both physical and psychological, arise. Emotional support given to parents of children with chronic conditions has been considered valuable by its recipients (Linblad, Rasmussen, Sandman, 2005; Liptak, Orlando, Yingling, Theurer-Kaufman, Malay, Tompkins, & Flynn, 2006).

Parents highlighted the uncertainty of their children’s health as a major factor in coping with the nature of their children’s conditions. While previous research has demonstrated fear of cancer to be a significant concern of individuals with Peutz-Jeghers Syndrome (Woo et al., 2009), my findings reveal that parents of children with polyposis conditions must cope with both the uncertainty of the development of cancer, as well as what their children’s short and long-term future overall health will look like. As Participant 3 discussed, it is the chronic nature of the condition that has led her to experience chronic stress, chronic uncertainty, and chronic worry.

As previously mentioned, research indicates that individuals with rare diseases require and want greater social support than those affected by more common medical conditions (Nispen, van Rijken, & Heijmans, 2003). The results reveal that the rare illness community, while necessitating greater needs for support, also experience greater challenges and barriers to receiving the social support that meets their needs. The main reasons cited by participants for the lack of support they’ve received included difficulty connecting with others affected by the condition, lack of awareness and understanding of
the condition in the community (general and medical) as well as participants’ own psychological states and fear of burdening others.

Relevant to the chronic illness literature is the differentiation between ‘invisible’ and ‘visible’ illnesses, terms coined by Joachim and Acorn (2000). Invisible conditions are chronic conditions that are not generally externally observable and are therefore more difficult for others to detect. Polyposis conditions fall within the invisible illness category, as most of the symptoms caused by the condition are not visibly apparent. As such, individuals with invisible conditions are at a greater risk of having their symptoms, and experiences invalidated, as was seen in the participants’ experiences in advocating for their children within the medical context and in seeking support from others. However, because of the ‘invisible’ nature of the invisible illnesses, those affected also have the ability to choose for themselves to whom they choose to disclose and discuss the condition. In the case of polyposis conditions, choosing to discuss the illness often includes discussing the ‘disgusting’ symptoms, deemed by both sufferers and society, of bowel diseases (i.e. blood in stool, diarrhea, constipation) (Hall, Rubin, Dougall, Hungin, & Neely, 2005). Such unwillingness to discuss bodily functions can make social support seeking uncomfortable for both those seeking support and those providing it.

Also in line with previous research, my findings reveal that the rare nature of polyposis conditions serves as a barrier for receiving social support. This research illustrates the factors implicated in the social support seeking process of parents of children with polyposis conditions, supporting the previous studies indicating that individuals affected by invisible illnesses experience difficulty in discussing the conditions (Diener, 2001; Stone, 2005).
The quality of social support and its ability to meet recipients’ needs plays a pivotal role in moderating its capacity to affect experiences of distress (Sewitch et al., 2001). My findings reveal that social support can serve as a protective factor, as found by Grootenhuis and Last (1997), with the ability of social support to meet an individual’s needs as paramount. The participants of this study voiced a need to be able to share their experiences with individuals who have gone through similar experiences, as the likelihood of those unfamiliar with the conditions receiving their stories with empathy and understanding is low. It is also important to note that prior findings indicate that as an individual’s psychological distress increases, the likelihood of satisfaction with the social support they receive diminishes (Hoekstra-Weebers, Jaspers, Kamps, & Klip, 1999). Within this study, the participants most vocal about their dissatisfaction with the social support they received were also those whose children were most symptomatic.

The findings also support previous findings in which individuals hesitate in seeking support for fear of burdening others. In study on the experiences of children coping with inflammatory bowel disease, Nicholas and colleagues (2007) found that participants often chose not to share their concerns with their family members in order not to transmit their worry onto others.

It is uncertain whether given social support that met their needs, the participants indicating dissatisfaction with their current social support would more proactively seek to share their experiences with others. My findings further highlight the psychological state of individuals needing social support, and the barriers they face in seeking such resources.
My findings are in line with previous research indicating that fear of cancer is a significant concern of individuals affected by polyposis conditions (Woo et al., 2009). Although the participants in this study discussed experiencing distressing, chronic concerns regarding the development of cancer in their children, such distress did not pose as a hindrance to parents’ engagement in medical management/surveillance procedures. Such findings are notable, as the discovery of cancer has been found to be the most reported reason for adults at risk for colorectal cancer not participating in screening procedures. My findings provide insight into how the parents are able to cope with their fear of cancer while still engaging in active coping techniques such as ensuring their children are properly followed and consulting with their children’s physicians. The participants cited reassurance by their physicians that with appropriate medical surveillance, the chances of cancer development lessen. It would seem that the physicians’ support provided the parents with ways of cognitively reframing their fear, thereby providing them with hope.

**Implications for the Medical Field**

Taken together, the findings from this study can be used by the medical community to better support and serve pediatric patients with polyposis conditions and their parents. While not all parents of children with polyposis conditions may find their experiences reflected in the stories captured within this study, the findings that were obtained can used to both better inform the medical profession and create concrete objectives for medical professionals.

Firstly, my findings suggest that in situations with pediatric patients, physicians’ relationships with their patients’ parents are just as important to the care of the child as
their actual relationship with the child. In building a strong, collaborative rapport with the child’s parents, the physician is directly benefiting the child’s medical experience, and in many cases, the child’s health. As polyposis conditions affect those diagnosed and their families chronically, it may be helpful for physicians to check in with parents regarding their own experiences and mental states while working with their pediatric patients. As discussed, parents of children with rare and chronic illnesses have been shown to demonstrate an elevated risk for developing anxiety and depression. As such, it may be helpful for physicians to be aware of resources available to parents of children with medical conditions who are experiencing difficulty coping.

Continued attention should be placed on the relationship with the patient’s parents, ensuring that their concerns are validated and addressed, with special consideration given to the possibly difficult experience that parents face in having their children’s rare condition diagnosed.

Lastly, as my findings reveal that parents make strong efforts to train their children to self-advocate, we would recommend that physicians remain mindful of this and collaborate with both parent and child when discussing medical treatments, as appropriate, especially during the patient’s transition between child and adult medical care.

**Implications for the Counselling Domain**

My findings suggest that counsellors could play an important role in facilitating parents’ adjustment to their children’s diagnoses, and subsequent every day life. Overall, this study highlights important themes that may arise in the counselling process, such as fear of the development of cancer, change of identity, evolving family dynamics, and
coping strategies. Importantly, many of the participants noted gratitude as an important practice in their every day lives, which has been demonstrated to be effective in fostering well-being (Emmons & Stern, 2013). As coping styles have been shown to be transmitted from parent to child, counselling parents toward more effective coping styles may indirectly also benefit pediatric patients (Kliwer, Fearnow, & Miller, 1996).

Furthermore, as isolation and lack of support were found to be relevant themes to the experiences of the parents interviewed, our findings would suggest that it would be best if these support groups are led by parents. Little research has been conducted on the use and efficacy of support groups for those affected by gastrointestinal disorders. As such, those seeking to develop support groups for polyposis patients and their families are limited by the available literature.

**Limitations**

Although this study has revealed interesting and important findings, the limitations of the research should also be taken into consideration. Firstly, saturation of themes from the data could not be achieved due to the limited sample size of the study. It is worth mentioning once again, however, that small sample size is an inherent challenge when studying the experiences of individuals with rare disease (Ettore, 2006).

Regarding the transferability of these findings to other parents of children with polyposis conditions, the demographics of this particular sample group must also be taken into account. While the majority of the children considered in this study were diagnosed with clinical manifestations of either Juvenile Polyposis or Peutz-Jeghers Syndrome, all but one participant’s children’s conditions were de novo in nature. Furthermore, none of the parents interviewed suffered from a polyposis condition
themselves, which would possibly affect their experiences. It is also important to note that the experiences of parents who were unresponsive in the recruitment process for this study may significantly differ from those who consented their participation in the interview process. While it is impossible to determine the exact reasons for which the unresponsive parents chose not to participate in the study at this point, the researchers question the role avoidance of the illness reality played in the decision.

As with all qualitative research, the primary researcher’s interpretative lens must be considered. While such a factor is not necessarily a limitation of the research, variability in the interpretation of the data may present itself in future research based on other researchers’ own particular interpretation biases. Future research should be conducted in order to further elucidate the phenomena revealed in this study and in order to explore the differences in experiences with parents of children in similar but different circumstances, where parents themselves have polyposis diagnoses, for example.

**Avenues for Future Research**

This study provided insight into the experiences of parents of children diagnosed with polyposis conditions. Continued research in this domain will be key in deepening our understanding of these experiences and developing more specialized medical services. Specifically, qualitative research will afford researchers the opportunity to gain rich data in a scarcely studied domain.

The current study should be replicated using a larger sample size, with participants representing children with *de novo* and familial diagnoses, and with participants themselves diagnosed with a hereditary polyposis condition, in order to
better reflect the diversity of experiences within the population. Furthermore, future studies may consider asking eligible recruits why they have chosen not to participate in the study, as possible reasons for non-participation (apathy concerning their children's condition, strong emotions tied to their children's conditions or distrust of the medical system overall due to difficult experiences, for example) warrant reporting and possible further investigation as parents who choose not to participate in studies possibly differ from those who choose otherwise in meaningful ways.

This study's findings also reveal interesting directions researchers may wish to explore. Many of the findings remain in line with previous research however several significant and novel results were also discovered. It may be of interest to researchers and clinicians alike to further study role of gratitude in illness, high-stress contexts. The ways in which parental advocates transmit their skills to their children should also be further examined, as such behaviours play an important role in the patient experience. Moreover, applied researchers could explore the effectiveness and best means of implementation of support groups for individuals (and their families) with rare and chronic illnesses, as this was a need expressed by the parents of this study. Lastly, as patient and family-centered care utilized by physicians played an important role in the experiences of pediatric patients and their parents, deeper exploration of the specific skills demonstrated by physicians to validate the experiences of individuals affected by rare and chronic illnesses may advance overall healthcare practice. While important practical applications can be developed from my findings, further research should be conducted in order to further substantiate their validity.
Conclusions

Parents of children with polyposis conditions face many challenges in coping with their children’s health. The current study explored the experiences of parents of children with polyposis diagnoses, gaining insight into their struggles, needs, appreciation, and everyday lives. It is hoped that this study and its results will give physicians, researchers and individuals unfamiliar with polyposis experiences an opportunity to peek into the worlds of parents of children with chronic and rare conditions. It is the aim of this study that such a perspective will translate into more effective care for pediatric patients and their parents, with more focus placed on the emotional and psychological needs of parents. Furthermore, it is hoped that the medical community will further view parents as valuable resources in the care of children with polyposis conditions, with this study revealing the parents’ unique role in fostering the advocacy skills of their children.
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RUNNING HEAD: Polyposis diagnoses: Experiences of parents


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Appendix A

Demographic Questionnaire

1. Sex: Male___________ Female____________ Other_____________
2. Primary language of communication: __________________________
3. Marital status:
4. Please select your highest level of education:
   a. Less than high school
   b. High school
   c. CEGEP/Technical college
   d. University (Undergraduate degree)
   e. University (Graduate degree)
5. Do you yourself have a polyposis diagnosis? _______________
6. Number of children: ______________
   a. Number of children with polyposis diagnosis:_____________
      i. Please specify the children's ages: _______________
      ii. Please specify specific polyposis diagnosis:_____________
      iii. Please specify any other physical or mental diagnoses your child or
           have children have been given:____________________
      iv. At what age was/were your child/ren diagnosed with polyposis
          syndromes?
7. Do you have any relatives with polyposis syndromes?
   a. If yes, please specify their relation to you and their diagnosis.
_________________________________________________________________
8. Did your child receive genetic testing?
   a. If yes, have you shared the results of the tests with relatives and friends of
      the child?
9. How often does your child receive medical attention?
10. Please indicate the groups of individuals and services who are a part of your
    support system:
    a. Partner/spouse
    b. Family
    c. Friends
    d. Physicians
    e. Co-workers
    f. Psychological services
    g. Internet forums
    h. Other (Please specify):_______________________________________
Questionnaire démographique

1. Sexe : Masculin _________ Féminin _________
   Autre ____________
2. Langue première de communication : _________________________
3. État civil : _________________________
4. SVP veuillez indiquer le plus haut niveau de scolarité complété :
   a. Primaire
   b. Secondaire
   c. CEGEP/Collège technique
   d. Université (premier cycle)
   e. Université (cycle supérieur)
5. Avez-vous personnellement reçu un diagnostic de polypose? __________
6. Nombre d’enfants : __________
   a. Nombre d’enfants ayant un diagnostic de polypose : __________
      i. Veuillez préciser l’âge de vos enfants: ___ ___ ___ ___
      ii. Veuillez indiquer le type de diagnostic de polypose:

      iii. Veuillez indiquer tout autre diagnostic de maladies physiques ou
            mentales que votre enfant ou vos enfants a/ont reçu :
            __________________________________________
      iv. À quel âge votre ou vos enfant(s) a-t-il/ont-ils reçu un diagnostic
          d’un syndrome de polypose? ___ ___ ___ ___
7. Est-ce qu’un membre de votre famille souffre d’un syndrome de polypose?
   a. Si oui, quel est son lien familial avec vous et quel est le type de polypose
      dont il souffre?

8. Est-ce que votre enfant a eu un test de dépistage génétique?
   a. Si oui, avez-vous partagé les résultats des tests avec des parents et des
      amis votre enfant? __________
9. À quelle fréquence votre enfant reçoit-il des soins médicaux?

10. Veuillez indiquer les groupes d’individus et les services qui font partie de votre
    réseau de soutien :
    a. Conjoint(e)/époux(se)
    b. Famille
    c. Amis
    d. Médecins
    e. Collègues de travail
    f. Services de psychologie
    g. Forums sur internet
    h. Autre (veuillez préciser) : ________________________________
Appendix B
Permission to Contact Participants Form: Cover Letter

Dear Parent,

We are contacting you on behalf of the [redacted] in our relative capacities as Chief of the Department of Genetics and Division Head of Gastroenterology, to tell you about a study currently being conducted at [redacted] by Dr. [redacted] and Dr. [redacted]'s research team. You are being invited to participate in a study taking place in the [redacted] at [redacted]. The title of the study is: The Lived Experiences of Parents of Children with a Polyposis Diagnosis.

Given the care your child has received in the [redacted], you, as a parent, have been identified as an eligible participant for this study. Your participation in this study is completely voluntary and there will be no ramifications on the quality of care your child will receive at [redacted]. This study seeks to learn about parents' experiences of their children's polyposis conditions. Your child's personal health information will not be released in the process of this research.

If you would like to know more about this study, we encourage you to read the study description on the following pages and to complete the Permission to Contact Participant Form found on the following page, or email Andrea Too, co-investigator, to indicate your interest to learn more about the study [redacted]. Once the form is completed and mailed back to us or you have indicated your interest by email, a co-investigator of the study will contact you to further describe the study to you. You will then have the opportunity to ask the co-investigator any questions you may have about participation in the study. Your participation in this study is voluntary. We thank you for your time and consideration. If you have any concerns about this research study, you can contact the Chair of the Research Ethics Board at [redacted] ext. [redacted].

Telephone: [redacted]

Telephone: [redacted]
Chers parents,


Les expériences vécues par les parents d'enfants ayant un diagnostic de polyposse.

Étant donné que votre enfant a reçu des soins du [nom des départements], vous avez été choisi, en tant que parent, comme participant admissible pour cette étude. Votre participation à cette étude est entièrement volontaire, et il n'y aura aucune conséquence sur la qualité des soins que votre enfant recevra au [nom du centre]. Cependant, cette étude cherche à en apprendre davantage sur les expériences des parents d'enfants souffrant de polyposse. Les renseignements médicaux personnels de votre enfant ne seront pas dévoilés dans le processus de cette recherche.

Si vous souhaitez en savoir plus sur cette étude, nous vous encourageons à lire la description de l'étude sur les pages suivantes et de bien vouloir compléter le formulaire d'autorisation à contacter les participants que vous trouverez sur la page suivante, ou de communiquer par courriel avec Andrea Too, co-chercheuse, afin d'indiquer votre intérêt et pour en savoir plus sur l'étude [nom de l'étude]. Une fois que vous aurez rempli et que vous nous aurez envoyé le formulaire ou que vous aurez indiqué votre intérêt par courriel, un co-chercheur de l'étude vous contactera pour vous décrire l'étude plus en détail. Vous pourrez poser vos questions au co-chercheur sur la participation à l'étude. Votre participation à cette étude est volontaire.

Merci pour votre temps et votre attention. Si vous avez des préoccupations ou des questions au sujet de cette étude, vous pouvez joindre le président du Comité d'éthique de la recherche au : [nom de l'adresse].

Téléphone: [nom du numéro de téléphone]

________________________________________
Gail Graham, MD, FRCPC, chef du [nom du département]
Programme régional de génétique de l'Est de l'Ontario
401 chemin Smyth, Ottawa, ON, K1H 8L1
Téléphone: 613 737-7600, poste 2620

________________________________________
David R. Mack, MD, FRCPC
Professeur de pédiatrie et chef de division de gastroentérologie pédiatrique, d'hépatologie et de nutrition
Centre hospitalier pour enfants de l'est de l'Ontario
401 chemin Smyth, Ottawa, ON, K1H 8L1
Téléphone: 613 737-7600, poste 2516
Appendix C:

Permission to Contact Participants Form: Study Description

Title: The lived experiences of parents of children with polyposis diagnoses

Investigators: [Redacted]

Co-Investigators: [Redacted]

Dr. André Samson
Andrea Too, BA

Description:
As part of the recruitment process for a study exploring the experiences of parents of children with polyposis diagnoses, we would like your permission to contact you to further explain the objectives and procedures of this study. You are being invited to participate in this study since you have a child who has sought treatment at the Children's Hospital of Eastern Ontario for a polyposis condition.

The primary goal of this study is to better understand the experiences of parents of children with polyposis diagnoses. Parents of children with either clinical or molecular diagnoses of Peutz-Jeghers Syndrome or Juvenile Polyposis are eligible to participate in this study. Participants in this study will be asked to participate in a 60-minute interview and to complete a demographic questionnaire. Participation in this study will not affect your child's current or future care at the Children's Hospital of Eastern Ontario. Participation is voluntary and you may withdraw from the study at any time with no ramifications.

Signing this form only indicates that you would like to be contacted in the future to learn more about this study. It does not endorse your participation in the study. If you agree to be contacted to learn more about this study, we ask that you complete and sign the attached form. Please mail the form back to the principal investigator using the addressed and stamped envelope. If we receive the form with consent to be contacted to hear more about this study, we will telephone or email you with information about the study and answer any questions that you may have.

You also have the option to email one of the study's co-investigators, Andrea Too, at [Redacted] to learn more about the study. With this information, you can make a decision about your participation in the study.

If we do not hear back from you within 21 days (3 weeks), a reminder letter will be mailed to you to see whether you are interested in participating in the study or not.

This research project has received the approval of the [Redacted] Research Ethics Board. If you would like more information about the ethical nature of this study or your rights as a participant in this study, please contact the [Redacted] Research Ethics Board at [Redacted] ext. [Redacted].
Any questions regarding this research study can be directed to:
Andrea Too (Co-Investigator)
Telephone number: [redacted]
Email address: [redacted]

Permission to Contact Participants Form
Title: The lived experiences of parents of children with polyposis diagnoses

Investigators:

Co-Investigators

Andre Samson
Andrea Too, B.A.

☐ I would like to be contacted by telephone/email to learn more about this study.

☐ I would NOT like to be contacted by telephone to learn more about this study.

Name: ________________________________

If you have indicated you would like to learn more about this study, please indicate your name, contact information and the best time to reach you.

Name: ________________________________
Telephone number: ________________________________
Day: ( ) ________________________________
Evening ( ) ________________________________
Email address: ________________________________

Please indicate the most convenient time for you to be contacted by placing an X:

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Signature ______________________ Name (Please Print) ___________________ Date __________________

*** Please return this form by mailing it in the [redacted]-addressed stamped envelope when complete.

Thank you for your time and consideration.
Formulaire d’autorisation à contacter les participants: Description de l’étude

Titre: Les expériences vécues par les parents d’enfants ayant un diagnostic de polyposose.

Chercheurs:                   Co-chercheurs

Dr Janice Barkey, MD
Mindy Finkelstein, MSW
Cathy Gilpin, MSc, CCGC
Dr Eva Tomiak, MD
Dr André Samson, PhD
Andrea Too, BA

Description:
Dans le cadre du processus de recrutement pour une étude examinant les expériences des parents d’enfants ayant reçu un diagnostic de polyposose, nous aimions avoir l’autorisation de vous contacter pour vous expliquer plus en détail les objectifs et les modalités de cette étude. Vous êtes invité à participer à cette étude puisque vous avez un enfant atteint de polyposose ayant reçu des traitements au centre hospitalier pour enfants de l’est de l’Ontario.

L’objectif principal de cette étude est de mieux comprendre les expériences des parents d’enfants atteints de polyposose. Les parents d’enfants ayant reçu soit un diagnostic clinique ou moléculaire pour le syndrome de Peutz-Jeghers ou de polypose juvénile sont admissibles pour participer à cette étude. Les participants à cette étude seront invités à participer à une entrevue de 60 minutes et à remplir un questionnaire démographique. La participation à cette étude n’affectera en aucun cas les soins actuels ou futurs de votre enfant au CHEO. Votre participation est volontaire, et vous pouvez arrêter de participer à l’étude à tout moment, et ce, sans aucune conséquence.

Si vous signez ce formulaire, cela indique seulement que vous souhaitez être contacté dans le futur pour en savoir plus sur cette étude. Il ne cautionne pas votre participation à l’étude. Si vous acceptez d’être contacté pour en savoir plus sur cette étude, nous vous demandons de bien vouloir remplir et signer le formulaire ci-joint. S’il vous plaît, envoyez le formulaire au chercheur principal en utilisant l’enveloppe adressée et affranchie. Si nous recevons le formulaire avec votre consentement à être contacté pour en savoir plus sur cette étude, nous vous téléphonerons ou vous enverrons des informations sur l’étude et nous répondrons à vos questions.

Vous pouvez aussi envoyer un courriel à l’un des co-chercheurs de l’étude, Andrea Too, (atoo086@uottawa.ca) pour en savoir plus au sujet de l’étude. Après avoir eu ces informations, vous pourrez prendre une décision éclairée quant à votre participation à l’étude.
Si nous ne recevons pas de réponse de votre part d’ici 21 jours (3 semaines), une lettre de rappel vous sera envoyée afin de savoir si vous êtes intéressé à participer à l’étude ou non.

Ce projet de recherche a reçu l’approbation du Comité d’éthique de recherche du CHEO. Si vous souhaitez avoir plus d’informations sur le caractère éthique de cette étude ou à propos de vos droits en tant que participant à cette étude, veuillez s’il vous plaît contacter le Comité d’éthique de la recherche du CHEO au poste 3272.

Toute question concernant cette étude peuvent être adressées à : Andrea Too (co-chercheuse)
Téléphone : 613 737-7600
Courriel: atoo086@uottawa.ca
FORMULAIRE D'AUTORISATION À CONTACTER LES PARTICIPANTS :
Titre : Les expériences vécues par les parents d'enfants ayant un diagnostic de polypose.

Chercheurs: 
Andrea Too, B.A.

Co-chercheurs 
Dr André Samson, PhD

☐ Je veux être contacté par téléphone ou courriel pour en savoir plus sur cette étude.

☐ Je NE veux PAS être contacté par téléphone ou courriel pour en savoir plus sur cette étude.

Nom :

Si vous avez indiqué que vous souhaitez en savoir plus sur cette étude, s'il vous plaît indiquer votre nom, vos coordonnées et le meilleur moment pour vous joindre.

Nom : __________________________________________

Téléphone :

Jour : (___) ________________________________

Soir : (___) ________________________________

Courriel : ________________________________

Veuillez s'il vous plaît indiquer le meilleur pour vous joindre en inscrivant un X :

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Signature __________________ Nom (en caractères d'imprimerie) __________ Date ______

*** Veuillez s'il vous plaît retourner le formulaire par la poste au CHEO dans l'enveloppe affranchie, une fois le formulaire complété.

Merci pour votre temps et votre attention.
You have been invited to participate in a research study being conducted by Dr. Janice Barkey from the CHEO Division of Pediatric Gastroenterology, Hepatology and Nutrition, Dr. Eva Tomiak from the CHEO Department of Genetics, Dr. André Samson from the Faculty of Education at the University of Ottawa and Andrea Too, a Master’s student in the Educational Counselling program at the University of Ottawa.

Purpose of this research study
The purpose of this study is to better understand the experiences of parents of children with polyposis diagnoses. Specifically, the researcher would like to better understand experiences of psychological adjustment, navigation of the medical domain, regular illness management practices, and experiences that parents of children with polyposis syndromes have found to be particularly meaningful once their child was diagnosed with a polyposis condition.

Your participation in this study is completely voluntary, with no ramifications, either beneficial or detrimental, to your child’s medical care at the CHEO. You are free to withdraw from this study at any time.

Procedures
If you choose to participate in this study, one of the study’s investigators, Master’s student Andrea Too, will meet you at [blank] to begin the interview process. During the interview process, the investigator will ask you a series of questions regarding your experiences as a parent of a child with a polyposis syndrome. The format of the interview process will include open-ended questions such as, “Please describe for me the context surrounding your child’s polyposis diagnosis.” If there are any questions you feel uncomfortable answering, you are not required to do so. It is estimated that the interview process will take 1 hour to complete. At the end of the interview, you will be given the opportunity to discuss the process with the researcher and pose any questions you have. You will also be given the primary researcher’s contact information and that of her supervisor, should you have any questions or comments once the interview has been completed. Interviews will be audio recorded and transcribed for data analyses purposes. Transcriptions will not include your name or any other identifying information. Transcripts will be safely kept on a password-protected computer to which only the primary researcher and her supervisor will have access.

Prior to the interview process, you will be asked to complete a questionnaire regarding your demographic variables. It is expected the completion of this form will take approximately 10 minutes. This form will ask you demographic questions about yourself, your family, and your child’s diagnosis.

Are there any risks to participating in the research?
There are no social or physical risks associated with participation in this study. It is possible that some questions in the interview may elicit negative memories, thoughts or feelings; but you are free to refrain from answering such questions. If needed, the researcher can provide you with information regarding contacting our gastroenterology social worker, [redacted], should you wish to discuss your feelings further.

**Benefits**
You may experience indirect benefits by participating in this study. Firstly, you may find that sharing your experiences to be cathartic and emotionally beneficial. Secondly, your participation in this research will hopefully enable the primary researcher to contribute to the research literature on polyposis syndromes. This research will present important themes in the lives of this study’s participants, ultimately helping to better prepare medical doctors and clinicians to work with parents of children with polyposis syndromes.

**Withdrawling from the study**
You are completely free to withdraw your participation from this study at any time, with no consequences. Should you feel at any point during the research process that your psychological wellbeing is compromised, you are encouraged to notify the researcher and withdraw from the study.

**Compensation**
Parking or public transportation costs related to participation in the study will be covered.

**Limits of Confidentiality**
All identifying information that you provide to the researcher will be removed from all final reports related to the study. Your name will be coded using a pseudonym in all transcripts. All identifying information will remain confidential. Only in cases where it is mandated by law or by the courts will your personal information be revealed. Audio recordings and transcripts will be securely kept in a locked cabinet in the primary researcher’s office for a maximum of 7 years to allow time for the data analyses and publication processes. After 5 years, all files relating to this study will be destroyed. You will be given a copy of the Consent Form to keep for your files. Should you be interested in obtaining the final results of this study, please notify the primary investigator.

By signing this consent form, you hold the research investigators to their professional duties and responsibilities and ensure that your legal rights are upheld as a participant. If a circumstance arises in which you suffer an injury as a result of your participation in this study, general legal conventions will be applied.

The [redacted] Research Ethics Board has approved the procedures of this study. The [redacted] Research Ethics Board works to protect the rights of participants in research studies. If you would like more information concerning the ethical standards of this research
process, please feel free to contact the Ethics Review Board at ext.

Questions
Questions about participation in this study can be directed to Master’s student Andrea Too by telephone at or by email at The principal investigators of this study can be reached with the following contact information:

Consent
I have read the above statements and any questions I may have concerning the research have been answered. I am voluntarily agreeing to participate in this study. I have received a copy of this three-page consent form.

  o Yes, I agree to be contacted for further follow-up questions.
  o No, I do not agree to be contacted for further follow-up questions.

Name of Participant (Please print): ________________________________
Signature of Participant: _________________________________________
Date: __________________________________________________________

Name of person obtaining consent: ________________________________
Signature of person obtaining consent: _____________________________
Date: __________________________________________________________
Informations du participant et formulaire de consentement

Les expériences vécues par les parents d’enfants ayant reçu un diagnostic de polypose

Vous avez été invité à participer à une étude de recherche menée par la Dr Janice Barkey de la division de gastro-entérologie pédiatrique du CHEO, en hépatologie et nutrition (Département de pédiatrie), la Dr Eva Tomiak du Département de génétique du CHEO, le Dr André Samson de la Faculté d’éducation de l’Université d’Ottawa et Andrea Too, étudiante à la maîtrise du programme en counselling éducationnel de l’Université d’Ottawa.

But de cette étude de recherche

Le but de cette étude est de mieux comprendre les expériences des parents d’enfants ayant reçu un diagnostic de polypose. Plus précisément, la recherche cherche à mieux comprendre les expériences touchant à l’adaptation psychologique, la navigation dans le domaine médical, les pratiques régulières de gestion de la maladie, et les expériences que les parents d’enfants atteints de syndromes de polypose ont trouvé particulièrement significatives une fois qu’ils ont appris que leur enfant était atteint de cette maladie.

Votre participation à cette étude est entièrement volontaire, sans ramifications, bénéfiques ou néfastes, à l’égard des soins médicaux que reçoit votre enfant au CHEO. Vous êtes libre de vous retirer de cette étude à tout moment.

Procédures

Si vous choisissez de participer à cette étude, l’une des intervieweuses de l’étude, Andrea Too, étudiante à la maîtrise, vous rencontrera au CHEO pour commencer le processus d’entrevue. Pendant le processus d’entrevue, on vous posera une série de questions concernant vos expériences en tant que parent d’un enfant ayant syndrome de polypose. Le format du processus d’entrevue comprendra des questions ouvertes telles que : « Veuillez s’il vous plaît me décrire le contexte entourant le diagnostic de polypose de votre enfant. » S’il y a des questions avec lesquelles vous ne vous sentez pas à l’aise, vous n’êtes pas obligé d’y répondre. On estime que l’entrevue durera à peu près une heure. À la fin de l’entrevue, vous aurez l’occasion de discuter du processus avec le chercheur et de poser toutes les questions que vous pourriez avoir. On vous donnera également les coordonnées du chercheur principal et celles de son superviseur, si vous aviez des questions ou des commentaires, une fois que l’interview aura été réalisée. Les interviews seront enregistrées en format audio et transcrits à des fins d’analyse des données. Les transcriptions ne comprendront pas votre nom ou toute autre information permettant de vous identifier. Les transcriptions seront conservées en toute sécurité sur un ordinateur protégé par un mot de passe auquel seul le chercheur principal et son superviseur auront accès.

Avant le processus d’entrevue, vous serez invité à remplir un questionnaire concernant vos renseignements démographiques. Il est prévu que ce formulaire prendra une dizaine de minutes à remplir. Les questions de ce formulaire porteront sur vous, votre famille et le diagnostic de votre enfant.
Y a-t-il des risques à participer à la recherche?

Il n’y a pas de risques sociaux ou physiques liés à votre participation à cette étude. Il est possible que certaines questions de l’entrevue puissent faire ressurgir des souvenirs, des pensées ou des sentiments négatifs; mais vous êtes libres de ne pas répondre à ces questions. Si nécessaire, le chercheur peut vous fournir des informations afin de pouvoir contacter notre travailleuse sociale en gastro-entérologie, [membre de l’équipe], si vous souhaitez discuter plus en détails de ce que vous ressentez.

Avantages

Vous pourriez retirer des bénéfices indirects en participant à cette étude. Tout d’abord, vous pourriez constater que le fait de partager vos expériences peut vous faire du bien et être bénéfique émotionnellement. Deuxièmement, votre participation à cette recherche, nous l’espérons, permettra au chercheur principal de contribuer à la littérature dans la recherche sur les syndromes de polypose. Cette recherche permettra de présenter des thèmes importants dans la vie des participants de cette étude, et en fin de compte, à aider à mieux préparer les médecins et les cliniciens dans leur travail avec les parents d’enfants atteints de syndromes de polypose.

Retrait de l’étude

Vous êtes complètement libre de mettre fin à votre participation à cette étude, à tout moment et sans conséquences. Si vous vous sentez à un moment, pendant le processus de recherche, que votre bien-être psychologique est compromis, vous êtes encouragés à en informer le chercheur et à mettre fin à votre participation à cette étude.

Compensation

Les frais de stationnement ou de transport en commun liés à la participation à l’étude seront couverts.

Limites de la confidentialité

Toutes les informations d’identification que vous fournissez au chercheur seront retirées de tous les rapports finaux relatifs à l’étude. Votre nom sera codé en utilisant un pseudonyme dans toutes les transcriptions. Tous les renseignements personnels resteront confidentiels. Vos renseignements personnels ne seront révélés que dans les cas où ce serait mandaté par la loi ou par les tribunaux. Les enregistrements audio et les transcriptions seront conservés en toute sécurité dans une armoire verrouillée dans le bureau du chercheur principal pour un maximum de 7 ans afin d’avoir le temps d’analyser les données et en raison des processus de publication. Après 5 ans, tous les dossiers relatifs à cette étude seront détruits. Vous recevrez une copie du formulaire de consentement que vous pourrez conserver pour vos dossiers. Si vous êtes intéressés à obtenir les résultats définitifs de cette étude, veuillez s’il vous plaît en avertir le chercheur principal.
En signant ce formulaire de consentement, vous tenez les enquêteurs de la recherche responsables de leurs devoirs et responsabilités professionnelles, ainsi qu’à veiller à ce que vos droits soient respectés en tant que participant. S’il survenait une situation dans laquelle vous souffriez d’une blessure pendant votre participation à cette étude, les conventions juridiques générales seraient mises en application. Le Comité d'éthique de recherche du [ ] a approuvé les procédures de cette étude. Le Comité d’éthique de recherche du [ ] travaille à protéger les droits des participants à des études de recherche. Si vous souhaitez plus d’informations concernant les normes éthique de ce processus de recherche, vous pouvez contacter le Comité d’éthique du [ ] au [ ] poste [ ].

Des questions

Toutes questions au sujet de la participation à cette étude peuvent être adressées à l’étudiante de Maîtrise, Andrea Too, par téléphone au [ ] ou par courriel à [ ]. Les chercheurs principaux de cette étude peuvent être joints aux coordonnées suivantes :

[ ] [ ] Dr André Samson

J’ai lu les déclarations ci-dessus et on a bien répondu à toutes les questions que je pourrais avoir concernant la recherche. J’accepte volontairement de participer à cette étude. J’ai reçu un exemplaire de ce formulaire de consentement de trois pages.

Oui, j’accepte qu’on me contacte pour d’autres questions de suivi.

Non, je n’accepte pas qu’on me contacte pour d’autres questions de suivi.

Nom du participant (en caractères d’imprimerie) : ____________________________

Signature du participant : ____________________________

Date : ____________________________

Nom de la personne qui obtient le consentement : ____________________________

Signature de l’obtention du consentement de la personne : ____________________________

Date: ____________________________
Appendix E

Interview Protocol

To be verbalized to participant by interviewer. As part of a study on the experiences of parents of children with polyposis syndromes, I would like to ask you about your experiences. Please describe your experiences with as much detail as possible and to the best of your ability. There are no ‘wrong’ answers and no judgment will be placed. If a question makes you feel uncomfortable, please let me know and that question will be skipped. I would like to remind you that you are free to quit the study at any point during the interview with no repercussions from your doctors or the researchers. Would you like us to clarify anything before I begin?

1. When was your child diagnosed with Peutz-Jeghers Syndrome/Juvenile Polyposis Syndrome?
2. Please describe for me the diagnosis process, the context surrounding it. What aspect of the diagnosis process sticks out most in your mind?
3. Please describe if and how your child’s diagnosis has impacted your life. How has the experience impacted:
   - You emotionally?
   - You psychologically?
   - The way you interact with friends, family and/or your partner?
   - The way you approach your child’s health?
   - The way you perceive your child’s current and future health?
4. Please describe your experience in obtaining medical care for your child.
   - How did the medical community approach your child’s condition?
   - What was your experience in seeking information about your child’s condition?
   - How did the process make you feel?
5. How has having a child with Peutz-Jeghers syndrome/Juvenile Polyposis Syndrome affected your future family planning decisions?
   - Has having a child with Juvenile Polyposis Syndromes/ Peutz-Jeghers Syndromes affected your decision to have more children?
6. **(If applicable)** How has your experience with Peutz-Jeghers syndrome/ Juvenile Polyposis syndrome impacted the way you guide your child through the experience?
   - Do you provide lessons from your own experiences?
   - Do you consciously make an effort to model the ways in which you hope your child will cope with the illness?

7. What advice would you give other parents of children with polyposis experiences?

8. Would you say you have adapted to the illness? If yes, how? If no, why not?
   - What has impeded or facilitated this adaptation?

9. Since your child’s diagnosis, what has changed for you as a parent? As a person?
   - What has remained the same?

10. What has been the evolution of your experience from diagnosis until present time?
    - How have your emotions and thoughts towards the condition changed?
    - How have the challenges surrounding the condition changed?

11. What strengths or lessons do you take away from this experience if any?

12. What would you have liked me to ask, that I did not ask?

**Interviewer will summarize the answers of the participant.**

13. Have I summarized your experiences well?

14. Would you like to add anything?

15. Would you be willing to answer any future questions should we need to clarify any aspects of this interview?
**Protocole d’entrevue**

*Le texte de l’entrevue doit être transmis en paroles par l’intervieweur.* Dans le cadre d’une étude sur les expériences des parents d’enfants atteints de syndromes de polypose, j’aimerais vous interroger sur vos expériences. S’il vous plaît décrivez vos expériences avec autant de détails que possible et au meilleur de vos connaissances. Il n’y a pas de « mauvaises » réponses et aucun jugement ne sera émis à l’égard de vos réponses. Si vous n’êtes pas à l’aise de répondre à une question, veuillez s’il vous plaît me le mentionner et cette question ne vous sera pas posée. Je voudrais vous rappeler que vous êtes libre d’arrêter de participer à l’étude à tout moment au cours de l’entrevue, et ce, sans avoir à craindre des répercussions de la part de vos médecins ou chercheurs. Aimeriez-vous nous clarifier quoi que ce soit avant que je commence?

1. Quand votre enfant a-t-il reçu le diagnostic du syndrome de Peutz-Jeghers / de polype juvénile?
2. Veuillez s’il vous plaît me décrire quel a été le processus du diagnostic et le contexte qui l’a entouré. Quel aspect du processus de diagnostic vous vient le plus à l’esprit?
3. Veuillez me décrire à quel point et comment le diagnostic de votre enfant a eu une influence sur votre vie. Quel a été l’impact de cette expérience:
   - sur vous émotionnellement?
   - sur vous psychologiquement?
   - sur la façon dont vous avez interacté avec vos amis, votre famille ou votre partenaire?
   - sur la façon dont vous abordez ce qui touche à la santé de votre enfant?
   - sur la façon dont vous percevez la santé actuelle et future de votre enfant?
4. Quelle a été votre expérience en ce qui concerne l’obtention de soins médicaux pour votre enfant.
   - Comment la communauté médicale a-t-elle abordé l’état de votre enfant?
   - Quelle a été votre expérience en ce qui concerne la recherche d’informations sur l’état de votre enfant?
   - Quels sentiments avez-vous éprouvé durant ce processus?
5. À quel point le fait d’avoir un enfant atteint du syndrome de Peutz-Jeghers / de polype juvénile a-t-il affecté vos décisions futures quant à la planification familiale?
   - Est-ce que le fait d’avoir un enfant atteint du syndrome de Peutz-Jeghers / de polype juvénile a affecté votre décision d’avoir plus d’enfants?

6. **(Le cas échéant)** Comment votre expérience avec le syndrome de syndrome de Peutz-Jeghers / de polype juvénile a eu un impact sur la façon dont vous guidez votre enfant à travers cette expérience?
   - Retenez-vous des leçons de vos propres expériences?
   - Avez-vous consciemment fait un effort pour modéliser les façons dont vous souhaiteriez que votre enfant fasse face à la maladie?

7. Quels conseils donneriez-vous aux parents d’enfants atteints de polypose?

8. Diriez-vous que vous vous êtes adapté à la maladie? Si oui, comment? Si non, pourquoi?
   - Qu’est-ce qui a entravé ou facilité cette adaptation?

9. Depuis le diagnostic de votre enfant, qu’est-ce qui a changé pour vous en tant que parent? En tant que personne? Qu’est-ce qui est resté pareil?

10. Quelle a été l’évolution de votre expérience en partant du moment du diagnostic jusqu’à aujourd’hui?
   - À quel point vos émotions et vos pensées à l’égard de cette condition sont-elles changé?
   - Comment les défis entourant cette condition ont-ils changé?

11. Quels sont les points forts ou les leçons que vous retenez de cette expérience, le cas échéant?

12. Qu’est-ce que vous auriez aimé que je vous demande, et que je ne vous ai pas demandé?

**L’intervieweur résumera les réponses du participant.**

13. Ai-je bien résumé vos expériences?

14. Aimeriez-vous ajouter quelque chose?

15. Seriez-vous prêt(e) à répondre à des questions dans le futur si nous avons besoin de clarifier certains aspects de cette interview?