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Uncertainty Among Youth with Chronic Pain, their Parents, and Clinicians

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Uncertainty Among Youth with Chronic Pain, their Parents, and Clinicians

by

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Abstract

Objective: Uncertainty is inherent to the experience of chronic primary pain (CPP). Diagnostic uncertainty (DU) has been alluded to in the pediatric chronic pain literature; however, the prevalence, facets, impacts, and drivers of this phenomenon have yet to be explored. The objectives of this dissertation were to explore DU among youth with CPP, their parents, and clinicians, and how it is influenced by the clinical encounter. As well, youth and parent intolerance of uncertainty were investigated as potential risk factors in the Interpersonal Fear Avoidance Model of Pain.

Methods: Data included samples of youth with CPP seeking treatment in tertiary level care settings, their parents, and clinicians who treat youth with CPP. These samples spanned three geographical locations, including Canada, the United Kingdom, and the United States. Uncertainty was investigated using a multi-method approach. DU was explored qualitatively from the perspectives of youth, parents, and clinicians, as well as quantitatively among youth and parents. Intolerance of uncertainty was investigated quantitatively using self-report measures. Finally, interpretative phenomenological analysis was used to explore components of the clinical encounter that influence youth's and parents' experiences of DU.

Results: DU is characterized by fear that something more serious is causing youth's pain, which fuels a search for alternative diagnoses. Approximately one-third of youth with CPP and their parents experience DU, which is linked to poorer youth pain, pain-related constructs, and health-related quality of life. Clinicians also experience DU treating youth with CPP. Components of the clinical encounter, including pain explanations, validation, and messages of uncertainty, influence youth's and parents' experiences of DU, which has implications for youth's engagement in treatment. Results also suggest that parent and youth intolerance of uncertainty

contribute to increases in youth pain interference over time through increased pain catastrophizing, parent protectiveness, and youth fear of pain.

Conclusions: This dissertation characterized the multifaceted nature of DU among youth with CPP, their parents, and clinicians, and demonstrated that it is common and often debilitating. Further, parent and youth intolerance of uncertainty play important roles as risk factors in the maintenance of pediatric chronic pain over time.

Keywords: chronic pain, pediatric, youth, adolescents, children, parents, diagnostic uncertainty, intolerance of uncertainty, fear avoidance

Preface

During the completion of this dissertation, the following five manuscripts have been prepared for publication (see the Appendix for written permissions). M. Noel was involved in the conceptualization of the Pain and Mental Health in Youth (PATH) research program, as well as obtaining research grants and ethical approval. A. Neville did the majority of the writing and was the primary contributor to the research and all papers included in this thesis. All authors contributed to the intellectual content and provided critical reviews of the manuscripts.

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Chapter 5 presents a peer-reviewed non-final version of an article published in final form by Wolters Kluwer in PAIN.

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Chapter 6 presents a manuscript that is prepared for submission to a peer-reviewed journal.

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CHAPTER ONE: Introduction and Literature Review

Introduction

Pediatric chronic pain, persistent or recurrent pain lasting longer than 3 months (Merskey & Bogduk, 1994a; Treede et al., 2015), affects 11-38% of youth (King et al., 2011). Pediatric chronic pain is associated with increased odds of using specialty care, complementary and alternative medicine, and emergency care (Tumin et al., 2018). Chronic pain can have a profound impact on youth's mental health and daily functioning, including school absences, sleep disturbance, and social functioning (Palermo, 2000). Further, pediatric chronic pain impacts the entire family. Parenting a child with chronic pain is associated with stress, anxiety and depressive symptoms, which are linked to worse child pain and functioning (Eccleston, Crombez, Scotford, Clinch, & Connell, 2004; Poppert Cordts, Stone, Beveridge, Wilson, & Noel, 2019).

Typically, pain is viewed as a symptom and patients expect that an assessment of their pain by a physician will lead to investigations, a diagnosis, and ultimately treatment (Butow & Sharpe, 2013). Unfortunately, youth who experience chronic primary pain face a major challenge in terms of understanding the underlying cause of their pain. Indeed, the nature of chronic pain is inherently complex and rarely explained by a precise pathophysiological explanation. Following the onset of chronic pain, families often consult several physicians in search of answers and a cure for the pain (Eccleston & Malleson, 2003), and many children perceive the explanations provided by physicians for their pain as being false (Meldrum, 2009). This time spent investigating the child's pain in search of a diagnosis and cure, typically lasting several years, has been described as a "diagnostic vacuum", which often delays chronic pain treatment and is characterized by considerable frustration and uncertainty (Eccleston & Malleson, 2003; Schechter, 2014a).

A recent topical review has raised diagnostic uncertainty, the perception of a lack of a, or incorrect, label to explain symptoms, as a critical issue for further investigation in pediatric chronic pain (Pincus, Noel, Jordan, & Serbic, 2018). Diagnostic uncertainty has been investigated among adults with chronic pain and reported by parents of youth with chronic pain. Specifically, parents have described their struggle to accept a lack of explanation for their child's pain and an unrelenting and distressing search for a legitimate diagnosis, even after receiving a diagnosis of chronic pain (Jordan, Eccleston, & Osborn, 2007). Parents of youth with chronic pain describe being "in limbo," a perpetual state of uncertainty and suspended in a holding pattern as they attempt to manage and make sense of their child's pain and associated disability (Jordan et al., 2007). Pincus and colleagues' (2018) topical review on diagnostic uncertainty proposed that future research should seek to understand the prevalence of diagnostic uncertainty among youth with chronic pain and their parents, its influence on treatment and pain outcomes, and how it unfolds over time.

In addition to the anxiety, distress, and helplessness that diagnostic uncertainty brings, it is also associated with information processing biases. Among adults with chronic low back pain, diagnostic uncertainty is associated with recall bias for negative health related stimuli (Serbic & Pincus, 2014). Current conceptualizations of cognitive biases in pain conceive of biases as being interconnected, such that attention, interpretation, and recall influence one another (Van Ryckeghem, Noel, Sharpe, Pincus, & Van Damme, 2019). Thus, being uncertain about a chronic pain diagnosis may impact other aspects of cognitive processing, such as how individuals interpret health information, which may influence patients' communication with clinicians, patients' understanding of information provided by clinicians, and treatment adherence and outcomes. Moreover, clinicians' own uncertainty when providing a diagnosis of chronic primary

pain may play an important role in this interaction and parent and youth 'buy-in' to the diagnosis. Clinicians' own perceptions and confidence surrounding provision of a diagnosis of chronic pain in youth have yet to be investigated.

Given the lack of research on diagnostic uncertainty and uncertainty more broadly in pediatric chronic pain and its potential impact on treatment outcomes, this dissertation aims to fill critical gaps in the literature by:

1. Qualitatively characterizing the phenomenon of diagnostic uncertainty and exploring how it is experienced by youth with chronic primary pain and their parents.
2. Examining the prevalence and consequences (i.e., pain interference, pain intensity, pain-related constructs, and health-related quality of life) of diagnostic uncertainty among youth with chronic primary pain and their parents.
3. Exploring diagnostic uncertainty from the perspectives of clinicians who diagnose and treat youth with chronic primary pain.
4. Investigating, more broadly, parent and youth intolerance of uncertainty (IU) as potential risks factors involved in the experience and maintenance of pediatric chronic pain.
5. Exploring how uncertainty unfolds in the clinical encounter between youth with chronic primary pain, parents, and clinicians at their initial visit at a tertiary chronic pain clinic when a diagnosis, explanation, and treatment plan are provided.

Pediatric Chronic Pain

Chronic pain is commonly recognized as pain that is persistent or recurrent lasting longer than 3 months (Merskey & Bogduk, 1994a; Treede et al., 2015). Pediatric chronic pain is a serious developmental health concern, affecting 11-38% of youth (King et al., 2011). Although

many children are able to cope in the face of chronic pain, approximately 3-5% of children with chronic pain are severely impacted, demonstrating moderate to high levels of pain-related disability (Hechler, Dobe, & Zernikow, 2010; Huguet & Miro, 2008). Youth may develop chronic pain related to an injury or underlying health condition (e.g., juvenile rheumatoid arthritis), or chronic pain may be idiopathic and best conceived of as being the primary illness itself (e.g., headaches, complex regional pain syndrome) (Friedrichsdorf et al., 2016). Indeed, in the absence of underlying organic pathology or positive test results, many youth receive a primary diagnosis of 'chronic pain' (Eccleston & Malleson, 2003) or 'Primary Pain Disorder' (Schechter, 2014b). The most frequent pediatric chronic pain conditions are migraine headaches, abdominal pain, and musculoskeletal pain (Goodman & McGrath, 1991), and many youth report pain in multiple locations (King et al., 2011). In an effort to standardize the diagnosis and treatment for chronic pain, the International Classification of Diseases, 11th Revision (ICD-11) has recently proposed the first unified coding system for chronic pain (Smith, Fors, Korwisi, Barke, Cameron, Colvin, Richardson, Rief, Treede, & Iasp Taskforce for the Classification of Chronic Pain, 2019). This coding system differentiates between chronic pain as a disease (chronic primary pain) and chronic pain as a symptom (chronic secondary pain) (Smith, Fors, Korwisi, Barke, Cameron, Colvin, Richardson, Rief, Treede, & Iasp Taskforce for the Classification of Chronic Pain, 2019). The diagnosis of chronic primary pain is made when pain is chronic, associated with significant emotional distress and/or functional disability, and is not better accounted for by another condition (Nicholas et al., 2019)

Chronic pain has a substantial impact on the daily functioning of youth (Palermo, 2000), can lead to persistent pain problems into adulthood (Walker, Sherman, Bruehl, Garber, & Smith, 2012), and is associated with increased lifetime risk of developing internalizing mental health

disorders (anxiety, PTSD, depression) (Noel, Groenewald, Beals-Erickson, Gebert, & Palermo, 2016). Furthermore, greater pain severity is related to increased likelihood of visiting a health care professional and using pain medication (Huguet & Miro, 2008). Thus, pediatric chronic pain poses a significant cost to society. In the United States, the economic toll of pediatric chronic pain is estimated to be over $19 billion USD annually (Groenewald, Essner, Wright, Fesinmeyer, & Palermo, 2014). Pediatric chronic pain conditions are associated with $11.8 billion USD in total incremental health care expenditures annually, exceeding the costs of childhood obesity and asthma (Groenewald, Wright, & Palermo, 2015). In Canada, chronic pain among adolescents and adults is estimated to cost $7.2 billion CAD annually in health expenditures, which is comparable to the costliest diseases in Canada (Hogan, Taddio, Katz, Shah, & Krahn, 2016). However, current treatments for pediatric chronic pain are often ineffective (Fisher et al., 2018). Further, response to treatment is poorest for children with comorbid anxiety (Cunningham et al., 2013) and parents with high distress (Law et al., 2017).

Pediatric Chronic Pain and Comorbid Mental Health Conditions

In addition to interfering with youth's daily functioning, youth with chronic pain have been found to experience high rates and elevated symptoms of internalizing mental health disorders (Coffelt, Bauer, & Carroll, 2013; Vinall, Pavlova, Asmundson, Rasic, & Noel, 2016). Coffelt and colleagues (2013) found that, among a sample of youth admitted to a tertiary-level pediatric hospital with chronic pain, 28% of children experienced a co-morbid mood disorder and 18% of children experienced anxiety and panic disorders. Moreover, youth who experience chronic pain in adolescence are at greater risk of developing a lifetime internalizing mental health disorder (Noel, Groenewald, et al., 2016; Shelby et al., 2013). Notably, regardless of whether pain resolves in childhood, chronic pain increases children's vulnerability to anxiety and depressive

disorders into adulthood (Shelby et al., 2013). Anxiety symptoms are highly prevalent, affecting up to two thirds of youth with chronic pain (Dufton, Dunn, & Compas, 2009; Knook, Lijmer, Konijnenberg, Taminiau, & van Engeland, 2012). Importantly, children with clinical anxiety have also been shown to be less likely to respond to treatment for chronic pain (Cunningham et al., 2016).

The impact of chronic pain extends beyond the child to the family unit (Palermo, 2000). Parents of youth with chronic pain report anxiety, depressive symptoms, and role stress (Eccleston et al., 2004). Recent findings suggest that up to 50% of parents of children with chronic pain have pain themselves (Beveridge, Neville, Wilson, & Noel, 2018). Chronic pain in parents may confer risk of chronic pain to the child through maladaptive affective-cognitive responses such as pain catastrophizing (Stone & Wilson, 2016). There is recent evidence that parental factors such as parent chronic pain, physical function (pain interference), and psychological factors (catastrophizing, anxiety, and depressive symptoms), are interrelated and powerfully predict youth's chronic pain experiences (Poppert Cordts et al., 2019). Given that parent's own mental health symptoms have been found to exacerbate children's pain (Noel, Wilson, et al., 2016) and impede treatment response (Law et al., 2017), parental factors are critical to investigate in children's chronic pain. Moreover, understanding what drives parental distress is critically important.

Diagnostic Uncertainty

Diagnostic uncertainty has recently been defined as a subjective perception of an inability to provide an accurate explanation of the patient's health problem (Bhise et al., 2018). Diagnostic uncertainty can also relate to a perception that a label or diagnosis is incorrect or missing (Pincus et al., 2018). Indeed, many patients with chronic pain do not receive a diagnosis that precisely

explains a pathological cause of their pain (Butow & Sharpe, 2013). The etiology of chronic pain is complex and not fully understood (Linton, Flink, & Vlaeyen, 2018). Research suggests that over 40% of adult patients with chronic pain who experience diagnostic uncertainty also report receiving a clear label and explanation for their pain, suggesting beliefs that the diagnosis or label provided does not fully explain their pain (Serbic & Pincus, 2014). Among adults with chronic pain, diagnostic uncertainty is associated with poorer pain outcomes (Geisser & Roth, 1998; Serbic, Pincus, Fife-Schaw, & Dawson, 2016), emotional distress, pain catastrophizing, disability (Geisser & Roth, 1998), depression, and anxiety (Serbic et al., 2016). Moreover, individuals who perceive their pain as not appropriately labeled or legitimized may continue to pursue medical investigations, which may contribute to the economic impact of chronic pain (Newton, Southall, Raphael, Ashford, & LeMarchand, 2013) and delay chronic pain treatment.

Diagnostic uncertainty has been largely overlooked within the pediatric chronic pain literature. In a qualitative study of parents of children with chronic pain, parents reported uncertainty with regards to their child's pain condition and future, which was related to feelings of helplessness, distress, and guilt (Jordan et al., 2007). Many children experiencing chronic pain are simply given a diagnosis of 'chronic pain' (Eccleston & Malleson, 2003). In the absence of a clear explanation for their child's pain, some parents have described hoping for positive test results that would medically validate their child's pain, even if these indicated serious pathology (Jordan et al., 2007). Indeed, parents may continue to search for different diagnoses even after receiving a diagnosis of chronic pain (Jordan et al., 2007). This time spent searching for certainty or a 'right diagnosis' is likely to be a critical factor in the child's adjustment to managing pain (Eccleston & Malleson, 2003). In another study, nearly 40% of parents reported an 'unresolved' diagnostic orientation towards their child's chronic pain (i.e., not accepting or 'buying in' to the

diagnosis) (Noel, Beals-Erickson, Law, Alberts, & Palermo, 2016). This unresolved orientation has been found to be a critical aspect of a distress narrative among parents of children with chronic pain, which was associated with increased parental pain catastrophizing about child pain, specifically helplessness in the face of pain (Noel, Beals-Erickson, et al., 2016).

An emerging area of research is investigating the influence of diagnostic uncertainty on information processing biases among adults with chronic pain. Among adults with chronic low back pain, diagnostic uncertainty has been associated with recall bias for negative health-related stimuli (Serbic & Pincus, 2014). Serbic and Pincus (2014) suggest that this pattern of recall may be indicative of preoccupation with the meaning and consequences of pain. Within this study, the group with diagnostic uncertainty was also found to have higher levels of depression compared to those who were certain about their diagnosis (Serbic & Pincus, 2014). Research is needed to understand if diagnostic uncertainty is related to pain and pain-related outcomes in pediatric chronic pain populations, and to understand risk factors and mechanisms underlying these relationships.

Among youth with chronic pain themselves, one study has shown that the majority of children who participated perceived the explanations provided by physicians for their pain as false and that physicians did not "fully understand" their pain (Meldrum, 2009). Youth also perceived not being believed or taken seriously by clinicians as well as that clinicians did not know what was wrong (Meldrum, 2009). Similarly, in a qualitative study of children with medically unexplained symptoms, children and their parents expressed struggling to accept a lack of a credible diagnosis and needing an explanation for their symptoms (Moulin, Akre, Rodondi, Ambresin, & Suris, 2015). Moreover, most children reported not feeling understood, believed, or listened to by physicians (Moulin et al., 2015). This has important clinical

implications given that poor patient-clinician communication has been linked to increased likelihood of patient non-adherence to treatment (Butow & Sharpe, 2013). This points to the importance of understanding clinicians' own perceptions of pediatric chronic pain diagnoses, as well as the study of factors that may be at play during these clinical encounters when pain is discussed or a diagnosis is communicated to patients and families. How a diagnosis of chronic pain is communicated to youth and parents is likely critical to their 'buy-in,' and may be influenced by developmental factors and individual needs. A better understanding of factors influencing diagnostic uncertainty (in terms of youth, parents *and* clinicians) may help tailor how clinicians deliver diagnoses to families to achieve 'buy in', increase understanding of pain and diagnosis, and ultimately improve treatment adherence and response. If parent and/or youth diagnostic uncertainty is associated with pain and pain-related constructs, interventions to decrease diagnostic uncertainty could have far-reaching implications across pain outcomes and health care expenditures. Currently, an understanding of how diagnostic uncertainty is experienced by youth with chronic pain and their parents, as well as its antecedents and consequences, is unclear.

Clinician Uncertainty and Chronic Pain

Clinicians' own beliefs about pain as well as other characteristics of the clinician (e.g., professional status, therapeutic style, language used with clients, confidence in a diagnosis and treatment) are suggested to affect treatment (e.g., recommendations and explanations provided to patients, treatment decisions) (Main, Foster, & Buchbinder, 2010). Clinicians may continue to refer patients for further testing if they are concerned about missing a serious medical condition (Eccleston & Malleson, 2003), or believe this will reassure patients (van der Weijden, van Bokhoven, Dinant, van Hasselt, & Grol, 2002). Among a sample of adults with chronic daily

headaches, MRI brain scans were found to reduce worry about a serious cause of headaches in the short term, but this was not maintained at one year follow up (Howard et al., 2005). Indeed, despite a belief widely held by clinicians that additional testing will reassure patients (van der Weijden et al., 2002), there is limited evidence that diagnostic testing does reassure (van Ravesteijn et al., 2012).

Among adults, informational reassurance (e.g., explanations about physical causes for pain) has been shown to be more reassuring than emotional reassurance (e.g., being taken seriously) (Holt, Pincus, & Vogel, 2015). However, the effects of informational reassurance may be complicated by factors such as health anxiety, such that those with high health anxiety may experience an increase and maintenance of worry following informational reassurance (Lucock, Morley, White, & Peake, 1997). The current lack of findings supporting the use of reassurance may point to the need for experiential-based interventions to reduce fear and worry (de Jong et al., 2005). Among adults with chronic pain, other clinician attempts to provide reassurance, such as through conveying that a condition is mild, also served to increase worry (Donovan & Blake, 2000). These attempts also likely lead patients to feel that their pain is being delegitimized and disbelieved by clinicians, which has been well documented (Newton et al., 2013). In the context of medically unexplained symptoms, clinician dismissal and delegitimization of symptoms has been linked with adult patients' increased expressions of concern and requests for diagnostic tests (Dowrick, Ring, Humphris, & Salmon, 2004). In this same study, reassurance that was connected to an explanation related to the patient's concerns facilitated acceptance of the explanation, whereas explanations unrelated to the patient's concerns were found to be ineffective. Little is known about reassurance in the clinical encounter in the context of pediatric chronic pain. However, among youth undergoing acute painful procedures, parental reassurance

has been found to be associated with increased expressions of fear (McMurtry, McGrath, & Chambers, 2006). McMurtry and colleagues propose that these results may suggest that providing reassurance signals to children there is something that warrants worry. Further, children who are more worried may elicit reassurance from their caregiver. An understanding of reassurance within the context of pediatric chronic pain and the clinical encounter is needed.

Clinicians' own uncertainty may play a role in youth and parents' uncertainty about a diagnosis of chronic primary pain. The patient-clinician relationship is traditionally an authoritarian one in which the clinician holds the power of medical decision-making (Katz, 1984). Katz (1984) described that clinicians' may maintain an "aura of infallibility" and deny uncertainty in interactions with patients to uphold their role as experts in this relationship. Few studies have investigated uncertainty in the context of chronic pain care. A recent ethnographic study examined clinical encounters between clinicians (general practitioners, pain specialists, psychiatrists, nurses, physiotherapists, psychologists, occupational therapists, pharmacists and addiction specialists) and adults with chronic low back pain (Costa et al., 2022). Within this analysis, incongruencies between diagnostic testing results and pain experience were found to bring about uncertainty for patients (Costa et al., 2022). Clinicians were found to express certainty in response to patients' expressions of emotions, such as disappointment and frustration, which served only to increase patients' uncertainty rather than reassure (Costa et al., 2022). Costa and colleagues' analysis highlighted nuances in the ways clinicians attempted to manage uncertainty in the context of low back pain, including providing reassurance as well as acknowledging uncertainty. Another way clinicians attempted to reduce uncertainty was by shifting power in the clinical interaction by emphasizing the patients' control in symptom management (Costa et al., 2022). The authors conclude that there are likely consequences if

uncertainty goes unacknowledged or is denied in the clinical encounter, including erosion of trust in the patient-clinician relationship (Costa et al., 2022).

Clearly uncertainty is complex, associated with discomfort, and is experienced between clinician and patient in the context of chronic primary pain. Uncertainty among clinicians who treat *youth* with chronic primary pain is much less understood. To be fully understood, uncertainty must be investigated from multiple perspectives, including the clinician, patient, and family, and with a critical lens on the clinical encounter.

Intolerance of Uncertainty

Individual differences in how youth and parents manage uncertainty may play a role in youth's experiences of chronic pain. The way in which pain information is interpreted is widely considered to be important in the maintenance of chronic pain (Eccleston & Crombez, 1999). Youth with chronic pain typically receive differing explanations for their pain and a lack of continuity as they search for a diagnosis (Eccleston & Malleson, 2003; Nutkiewicz, 2008). Through a lens of diagnostic uncertainty, patient-clinician communication about pain or a diagnosis may become ambiguous and confusing (Pincus et al., 2018). How youth interpret this information may be critically important and influence patient 'buy-in' and certainty about a diagnosis, which may in turn impact pain treatment adherence and outcomes. Youth's interpretations of ambiguous bodily threat information may also be critical in their experience of chronic pain, with negative interpretations leading to worse pain outcomes (Heathcote, Jacobs, Eccleston, Fox, & Lau, 2017).

Intolerance of uncertainty (IU) is described as a tendency to perceive uncertain and ambiguous events as stressful and upsetting and to believe that experiencing uncertainty about the future is unfair (Laugesen, Dugas, & Bukowski, 2003). It has been defined as a "dispositional

incapacity to endure the aversive response triggered by the perceived absence of salient, key, or sufficient information sustained by the associated perception of uncertainty" (p. 31) (Carleton, 2016b). IU has been found to be a key component of excessive worry (Laugesen et al., 2003) as well as anxiety disorders (Dugas, Gagnon, Ladouceur, & Freeston, 1998), and is conceived of as a transdiagnostic risk factor for the latter. Individuals who are high in IU have been found to interpret ambiguous situations as more threatening than those who are low in IU (Dugas et al., 2005). Given that anxiety is highly prevalent among youth with chronic pain and their parents, and uncertainty is an inherent aspect of a chronic pain diagnosis, IU may be critical to the experience of pediatric chronic pain. Nevertheless, IU has never been examined in this population. While previous research in adults has examined other transdiagnostic risk factors (e.g., anxiety sensitivity) for fear-avoidance in chronic pain, IU has not been examined and may be particularly important for driving threatening pain-related cognitions, emotions, and disability in pediatric chronic pain.

Patient beliefs and appraisal of pain are important factors in pain perception (Main et al., 2010). The Interpersonal Fear Avoidance Model of Pain (IFAM) is a prominent conceptualization that posits bidirectional relationships between parent and youth psychological (e.g., pain catastrophizing) and behavioural (e.g., parent protectiveness) factors that influence and maintain youth pain and disability (Goubert & Simons, 2014; Simons, Smith, Kaczynski, & Basch, 2015). For example, a child's appraisal of their pain as threatening may fuel pain-related fear and anxiety (Goubert & Simons, 2014; Simons et al., 2015). Parental responses have been found to be critical to youth's experiences of chronic pain (Palermo & Eccleston, 2009). For example, parent protective responses, characterized by attending to pain symptoms, are associated with maladaptive child outcomes, such as increased somatic symptoms, functional

disability, and poor school attendance (Logan, Simons, & Carpino, 2012) (Claar, Simons, & Logan, 2008; Langer, Romano, Levy, Walker, & Whitehead, 2009). Thus, understanding the roles of both youth *and* parent IU as potential predisposing risk factors in the maintenance and development of chronic pain is needed.

Dissertation Study Aims

Understanding the experiences of uncertainty among youth with pediatric chronic pain, their parents, and clinicians is a critical step in improving care for youth with chronic primary pain. Despite being alluded to in the literature, the nature of uncertainty in pediatric chronic pain and its role in pain experience and pain-related outcomes is not known. Thus, the overall aim of this dissertation was to characterize the experiences of uncertainty among youth with chronic primary pain, their parents, and clinicians. Using a multi-method approach, this dissertation investigated diagnostic uncertainty in the context of pediatric chronic pain. As well, IU was investigated as a risk factor in the maintenance of pediatric chronic pain. The manuscripts that form the current dissertation address five broad aims:

The ***first study*** (Chapter 2) explored how diagnostic uncertainty is experienced by youth with chronic primary pain and their parents. Given the dearth of research on diagnostic uncertainty in pediatric chronic pain, this study took a qualitative, exploratory approach. In-depth reflexive thematic analysis of semi-structured interviews sought to understand the core components of diagnostic uncertainty from the perspectives of youth with chronic primary pain and their parents. In keeping with the qualitative thematic analysis approach, no hypotheses were made (Braun & Clarke, 2006).

The ***second study*** (Chapter 3) examined the prevalence and consequences of diagnostic uncertainty among youth with chronic pain and their parents. This was the first study to examine

the relation of parent and youth diagnostic uncertainty to pain, pain-related constructs, and health-related quality of life. The primary objectives of this study were to: (a) establish the prevalence of diagnostic uncertainty among youth presenting with chronic primary pain to a pediatric tertiary level pain clinic and their parents; and (b) determine if youth and parent diagnostic uncertainty is related to youth pain, pain-related constructs, and health-related quality of life.

The ***third study*** (Chapter 4) explored how diagnostic uncertainty is experienced by clinicians who diagnose and treat youth with chronic primary pain. Semi-structured interviews with pediatricians who treat youth who experience complex chronic pain were analysed using reflexive thematic analysis.

The ***fourth study*** (Chapter 5) examined whether parent and youth intolerance of uncertainty were risk factors to core components of the Interpersonal Fear Avoidance Model of Pain. The primary objectives of this longitudinal study were to evaluate whether: (a) youth intolerance of uncertainty at baseline would predict 3-month pain interference through youth pain catastrophizing and fear of pain; and (b) higher parent intolerance of uncertainty at baseline would predict youth 3-month pain interference through parent pain catastrophizing, parent protectiveness, and youth fear of pain.

The ***fifth study*** (Chapter 6) explored components of the clinical encounter that influence diagnostic uncertainty for youth with chronic pain and their parents over time. This study utilized interpretative phenomenological analysis to examine transcripts of the first clinical encounter when youth and parents are enrolled in a pediatric pain clinic, as well as pre- and post- semi-structured interviews with youth and parents.

CHAPTER TWO: Diagnostic Uncertainty in Youth with Chronic Pain and their Parents

Abstract

Diagnostic uncertainty, the perception of a lack of or incorrect label to explain symptoms, has been reported by parents of youth with chronic pain. This study was the first to examine diagnostic uncertainty in both youth with chronic pain and their parents using qualitative methodology. Individual, face-to-face, semi-structured interviews were conducted with twenty youth with chronic pain recruited from a pediatric chronic pain program. Independent interviews were also conducted with one of their parents. Interviews explored participants' memories and perceptions around diagnosis. In-depth thematic analysis revealed four themes: (1) The Function of a Diagnosis. Parents and youth struggled with the meaning of the diagnosis, needed further explanation for the pain, and perceived the 'right' diagnosis (i.e., one that fit with their beliefs) as justification for the pain. (2) Haunted by Something Missing. Negative test results did not provide relief or counter the belief that something serious could have been missed by clinicians. (3) The Search for an Alternative Diagnosis. A search persisted for the 'right' diagnosis, particularly when a non-pharmacological treatment plan was provided. (4) Mistrust in the Medical System. Clinician communication and perceptions of clinicians' uncertainty impacted parent and youth 'buy in' to the diagnosis. Findings suggest that many youth with chronic pain and their parents experience diagnostic uncertainty, which is integrally tied to their past experiences with the medical system. Greater understanding of diagnostic uncertainty may help tailor how clinicians deliver diagnoses to achieve 'buy in', increase understanding of pain and diagnosis, and improve treatment response.

Introduction

Pediatric chronic pain, affecting 11-38% of youth (King et al., 2011), can have a profound impact on daily functioning, and is associated with frequent school absences, sleep disturbance, emotional distress, and reduced participation in peer activities (Palermo, 2000). The impact of pediatric chronic pain extends to the broader family. For example, parenting a child with chronic pain is associated with anxiety, depressive symptoms, and parenting stress (Eccleston et al., 2004). A major challenge that children with chronic pain and their parents face is understanding the cause of the pain. Indeed, the nature of a chronic pain diagnosis is inherently complex, challenging to explain and understand, and fraught with uncertainty. The time spent investigating the child's pain in search of a diagnosis and cure has been described as a "diagnostic vacuum" and may be critical to parents' and children's pain conceptualization and treatment (Eccleston & Malleson, 2003). Among adults with chronic pain, diagnostic uncertainty, recently defined as a subjective perception of an inability to provide an accurate explanation of the patient's health problem (Bhise et al., 2018), is associated with higher levels of anxiety, depression, pain intensity (Serbic et al., 2016), and disability (Geisser & Roth, 1998). However, an understanding of how diagnostic uncertainty is experienced by youth with chronic pain and their parents is unclear.

Diagnostic uncertainty has been alluded to in the pediatric chronic pain literature and a recent review has raised it as a critical issue for further investigation in this population (Pincus et al., 2018). In a study of 53 children with chronic pain, the majority of children perceived the explanations provided by physicians for their pain as false (Meldrum, 2009). Two studies have explored diagnostic uncertainty among parents of youth with chronic pain. In a qualitative study, Jordan and colleagues (2007) described parents' struggle to accept a lack of explanation for their

child's pain and an ongoing, unrelenting search for a diagnosis that would validate the pain, even after a diagnosis of chronic pain had been provided. Uncertainty with regards to diagnosis was tied to parental guilt, helplessness and distress (Jordan et al., 2007). Parents also described a sense of being "in limbo," a state of uncertainty in which they could not move forward in daily life and felt unsure about the future (Jordan et al., 2007). In a later observational study, nearly 40% of parents reported an unresolved diagnostic orientation towards their child's chronic pain (i.e., not accepting or 'buying in' to the diagnosis) (Noel, Beals-Erickson, et al., 2016). This unresolved orientation was found to be a critical aspect of a distress narrative among parents of children with chronic pain, which in turn was associated with increased helplessness (Noel, Beals-Erickson, et al., 2016).

Given the lack of research on diagnostic uncertainty with regard to pediatric chronic pain from the perspectives of youth and their parents and its potential impact on treatment outcomes, this study employed a qualitative methodology to investigate how diagnostic uncertainty is experienced by *both* youth with chronic pain and their parents. A dyadic qualitative approach to examine this phenomenon offers an opportunity to elucidate the complex experience of diagnostic uncertainty from the unique perspectives of youth and parents that may not be conveyed through questionnaire data.

Methods

Participants

Twenty youth with chronic pain (15 female, M_{age} = 14.6 years, range = 10-18 years) and one of their parents (17 mothers) each participated in separate face-to-face interviews with a researcher. Youth and parents were recruited from an interdisciplinary, tertiary-level pediatric chronic pain program in Western Canada. This team was led by an Anesthesiologist and included

clinicians from Anesthesiology, Psychology, Nursing, Physiotherapy, and Family Therapy. At the time of the interview, all participants had been at least initially seen in the pain clinic. Youth were eligible to participate if they were 10-18 years old and reported a pain problem that had been present for at least three months (in accordance with the current definition of chronic pain endorsed by the International Association for the Study of Pain) (Merskey & Bogduk, 1994a). Exclusion criteria for youth included a diagnosis of a serious medical condition (i.e., juvenile arthritis or cancer), neurodevelopmental disorder (e.g., intellectual disability, autism spectrum disorder), and/or a serious mental health disorder (e.g., schizophrenia, psychotic disorder). Parents were eligible to participate if they were the legal guardian of the youth.

Youth were referred to and/or attending the headache (*n*=8) or complex pain (*n*=12) clinic of a tertiary-level pediatric chronic pain program. Of youth who reported on their pain duration (*n*=16), the average pain duration was 3.67 years (*SD* = 3.32 years, range = 0.5–11 years). Youth (*n*=20) reported an average pain intensity of 5.95 out of 10 (*SD* = 1.55) in the past week. During the interview, parents and youth were asked whether or not a diagnosis for the youth's pain had been provided by a clinician. Sixteen parent-youth dyads were in agreement in terms of the diagnosis that the youth had received: complex regional pain syndrome (*n*=6), headaches/migraines (*n* =2), migraines and stomach migraines (*n* =1), migraines and constipation (*n* =1), chronic pain (*n* =2), nephroptosis (*n* =1), costochondritis (*n* =1), no diagnosis (*n* =2). However, among 4 parent-youth dyads, either the parent or youth reported a diagnosis of migraines, while the other member of the dyad reported that no diagnosis had been provided (parent reported migraine diagnosis *n* =1, youth reported migraine diagnosis *n* =3). Information with regards to these diagnoses were reported by families to have been provided by a variety of clinicians/teams, including the pain clinic, emergency physicians, neurologists, general

practitioners, nurses, psychologists, and physiotherapists. Additional sociodemographic information (e.g., ethnicity, household annual income) are presented in Table 1.1. The current sample size is consistent with previous studies using thematic analysis (Sibeoni, Chambon, Pommepuy, Rappaport, & Revah-Levy, 2017; Sibeoni et al., 2018). In accordance with the views of Braun and Clarke, no attempt was made to achieve saturation within the data as this is an unhelpful concept (O'Reilly & Parker, 2012). Instead, a focus was placed on establishing a transparent process around how the data was collected, as research transparency is perceived as a marker of quality (Spencer, Ritchie, Lewis, & Dillon, 2003).

Procedure

All study procedures were approved by the institutional health research ethics board (REB). Parents were asked by a member of the pain clinic staff for permission to be contacted by the research team. A member of the research team contacted permitting parents over the telephone or via email with information about the study. A consent call was performed with parents and youth to determine eligibility and answer any questions about the study. Interested parents and youth were then emailed separate online consent and/or assent (for youth aged 10-13) forms via Research Electronic Data Capture (REDCap), a secure online data collection tool (Harris et al., 2009), to complete prior to the interview. On the day of the interview, hardcopy written, informed consent/assent was also obtained from youth and parents. Youth and parents were informed that interviews would be audio-recorded and provided permission for quotations from the interviews to be published for academic purposes.

Interviews with youth and parents were conducted separately in a private testing room at the hospital where the chronic pain program is located. Interviews were conducted in-person by members of the research team (the PI [clinical psychologist] or clinical psychology graduate

students with previous training and experience interviewing youth in clinical and research settings). The same researcher interviewed both the youth and parent of a dyad, with the youth being interviewed first and the parent immediately afterwards. Interview duration ranged between 10-50 minutes (mean duration youth interviews = 19 minutes, mean duration parent interviews = 27 minutes). Interviews were audio-recorded and transcribed verbatim by members of the research team. All identifying information was removed from the transcripts to ensure the anonymity of participants and related individuals. Youth and parents both received a $25 CAD gift card after the visit to thank them for their time.

Interview Schedule

Researchers with expertise in qualitative research methods, diagnostic uncertainty, and pediatric chronic pain (MN, AJ) developed the interview schedule (see Table 1.2). The interview schedule was designed to gain insight from the youth and parent about their 'pain journey' (i.e., when the pain started, what living with pain has been like, perceptions regarding future outcomes with regard to pain and life). One question in the interview schedule (item #4 in Table 1.2) focused specifically on the youth's and parent's experience of obtaining a diagnosis for the pain. Youth interviews focused on the experience of living with chronic pain whereas parent interviews focused on the experience of parenting a youth with chronic pain.

The interview schedule was semi-structured and comprised open-ended questions in order to allow interviewers to follow-up and expand on topics of interest as they occurred during the interviews (Mason, 2004). Specific prompts were included in the interview schedule to encourage elaboration on topics of interest (e.g., 'Tell me more about that?').

Data Analysis

Data were analyzed using reflexive thematic analysis, in accordance with Braun and Clarke's six-phase thematic analysis guide (Braun & Clarke, 2006). Specifically, familiarization with the data was achieved through repeated and active reading of the interview transcripts. Through iterative familiarization with the data, codes were generated for data items related to receiving a diagnosis and then collated to identify potential themes. Initial analyses to generate codes and identify potential themes were conducted by co-author AN. AN first reviewed all youth interviews, followed by all parent interviews. Themes were then compared across each parent-youth dyad to identify patterns that were common and differed within dyads. Throughout this process, frequent debriefing sessions were conducted with AN, JB, and MN, to discuss the development and interpretation of themes. Co-authors AJ and TP frequently reviewed the ongoing analyses and debriefed with authors AN, JB, and MN (e.g., discussion of alternative interpretations, need for further analysis) (Shenton, 2004). Potential themes were iteratively reviewed and refined to determine the final themes, and concise definitions for each theme were generated. All co-authors were involved in, and agreed upon, the analyses and interpretations, providing credibility and trustworthiness in the data and analytic interpretations (Elliott, Fischer, & Rennie, 1999; Morrow, 2005). Lastly, the data were analyzed within the identified themes, and a report was written. In keeping with the principles of an inductive thematic analysis approach, we did not count frequencies of themes. Indeed, the importance or meaningfulness of a theme does not necessarily equate to frequency or quantifiable measures (Braun & Clarke, 2006).

Data analysis was completed using QSR International's NVivo software, a computer-assisted qualitative data analysis package. Attention was paid to include quotations from various participating youth and parents for representation of all perspectives within each theme. The

consolidated criteria for reporting qualitative research (COREQ) checklist was followed to ensure a rigorous and thorough approach to establishing quality and reporting of this qualitative study (Tong, Sainsbury, & Craig, 2007).

Results

Four themes were generated from thematic analysis of the data: The Function of a Diagnosis, Haunted by Something Missing, The Search for an Alternative Diagnosis, and Mistrust in the Medical System. These themes are presented below with supporting verbatim quotes from youth and parents. Pseudonyms are used below for each participant to protect their confidentiality. In addition, we use the term 'clinicians' below to refer to all types of clinicians who may provide information to families about a chronic pain diagnosis (e.g., anesthesiologists, neurologists, rheumatologists, general practitioners, nurses, psychologists), recognizing variability within this sample and across contexts (e.g., pain teams, internationally).

1. The Function of a Diagnosis

When provided with a diagnosis for their chronic pain from a clinician, many youth and their parents struggled with the meaning of their diagnosis. For many, a label was not enough and the diagnosis provided little clarity in terms of understanding their pain. Parents and youth alike expressed needing further explanation beyond a label for their pain.

> *"Uh, they said chronic regional pain syndrome. Mm, a whole bunch of words that really tell us nothing." (Mother of Lauren)*

When youth and parents reported understanding the label provided for their pain, further explanation about the etiology and causal mechanisms underlying the diagnosis was still needed.

"I've always had anxiety surrounding my pain, cause I'm like 'I don't know why this is happening.' And I think… a lot of my anxiety is like when I don't know why something's happening I'm like 'I need to know why' and I don't know why I have migraines… I know what a migraine is but I don't know why I have them." (Isabelle, age 17)

A diagnosis of 'chronic pain' itself seemed to elicit uncertainty and was perceived by parents as a diagnosis given either in the absence of a true understanding of the cause of the pain or as a "catch all" or "default" diagnosis, which was unsatisfactory for parents.

"Um, and it was like kinda called chronic migraines and we have zero history of that in our family right… So the whole thing was just perplexing. But I understand like I think it's probably a default diagnosis." (Mother of James)

Discrepancies were found among some parent-youth dyads in how they made sense of or felt about the diagnosis. For example, while Cecilia seemed to express that the diagnosis carried little meaning and only fit "to a certain extent", her mother reported experiencing relief in having a diagnosis. For this parent, a new explanation that had not previously been provided and that no longer focused solely on common explanations for the pain (e.g., nerve pain) was reassuring and validating.

"It's just a diagnosis to me. It's just a word… it's just a word that they think is what I have… It's just a label that they put the pain I have under." (Cecilia, age 15)

"It was just a relief to have someone that actually gave you a possible answer for what had been going on all this time as opposed to, you know, 'well it's just pain, you just have to wait for this, here's some tablets off you go'… It just seemed, it was like a completely different answer as opposed to just nerves or anything else, it was just a term I hadn't heard before. But it was, I, I don't know it just, it just seemed something that fitted I suppose… You know, someone doesn't throw out a random term just for the hell of it." (Mother of Cecilia)

In the absence of a diagnosis from a clinician, some youth's pain was disbelieved by others, including parents, peers, and clinicians. Parents and youth described a diagnosis as external

validation that would justify the pain. Being disbelieved by parents was particularly salient perhaps because of the widely held belief that a parent should unconditionally believe their child. For example, the mother of Olivia described receiving a diagnosis as a "game changer" in that it allowed the father to finally believe that his daughter's pain was real.

> *"But all of a sudden there was a diagnosis and all of sudden Dad's on board and he's there. And he's supportive and which is what she needs. So the diagnosis was a bit of game changer for him. For him like oh she isn't just faking it, she isn't just a being dramatic, she isn't being a girl." (Mother of Olivia)*

Emily's mother explained how, to Emily, a diagnosis meant a physical cause or explanation that would 'prove' the validity of the pain to clinicians. For Emily, a 'chronic pain' diagnosis conveyed that her pain wasn't real and wasn't believed. Emily explained that a physical abnormality in her wrist to explain her chronic pain problem was more reassuring than a diagnosis of complex regional pain syndrome.

> *"I think she has distrust in the system and they still don't get her… even our GP, [emergency doctor], nobody gets her or trusts that it's real pain. And I think her definition of real pain may be a little bit, like she needs a physical diagnosis to justify her pain." (Mother of Emily)*

> *"They just told me I have chronic pain and I was like a bit upset, I was like excited that there was a diagnosis, but I was a bit upset because... I thought that chronic pain was like your brain... I was upset that like people didn't think I had real pain. So, I got really upset… then they told me there is still something. My wrist clicks and stuff, so there is still something in my wrist that is happening." (Emily, age 11)*

For some, uncertainty extended beyond the diagnosis to the treatment for the pain. In addition to the diagnosis not providing clarity, a non-pharmacological treatment plan (e.g. physiotherapy or psychology) fuelled further confusion, ambiguity, and uncertainty and was seen as less valid than a pharmacological intervention.

> *"It's not this, it's not this, it's not this. They're not telling you what it is. And even when they*

do tell you what it is, then you know, well the main therapy is physio which is great 'cause it's something you can do but then as soon as you hear that, you know it's something that's going to take time and then you don't really know. This is a very ambiguous thing from what I can tell to be diagnosed with." (Mother of Jada)

"And my doctors have said like maybe you have endometriosis. And I'm like well, like what the hell is that. Like how does that help me? You can't give me medication for it." (Jada, age 17)

Relief was anticipated if the 'right' diagnosis (i.e., one that fit with the parent and youth's perception of the cause of the pain and anticipated management) was discovered. Furthermore, the 'right' diagnosis was perceived as being one that required a pharmacological solution and 'silver bullet cure', which would abolish the pain entirely.

"I think if I were to finally get the right diagnosis it would be like a huge relief. Because then I would know if I can be taking medication for it to help it or if I could be doing something to help it or anything. Like then I can just make it stop. I think that's the best part about it. Like I can make it stop." (Kristen, age 17)

When treatment for a diagnosis did not lead to improvement in pain and functioning, some parents questioned their child's pain, and believed that recovery was needed to provide evidence that the diagnosis was accurate and the pain was real.

"So we're like okay, well if it's an inflamed joint, then the inflammation pills should've at least eased it down. But they did actually nothing. So that's when we were like, you know, well maybe it is in her head, maybe there is something that she's associating this with and uh, not getting enough sleep and being overdramatic and emotional over something that's you know, making a mountain out of a molehill kind of thing." (Father of Lilly)

2. Haunted by Something Missing

Uncertainty about the youth's diagnosis fuelled worry that something could have been missed and, further, that what was being missed was serious and sinister.

"It makes me kinda worried to what it might be if it could be something really severe. But it's probably not. But still that little inkling… if I have something in my brain that might not be good. Something that I might have to live with forever. Not knowing what it might be that's also in that we know that 'something's bad we just can't tell you what' kind of feeling…" (Nicole, age 14)

Thoughts and beliefs that something could be missed were intricately tied to previous experiences with clinicians and the medical system. Long periods waiting in a state of uncertainty seemed to feed a lack of acceptance of a diagnosis when it was finally provided. For some, fear and health anxiety developed while searching and waiting. In this way, delays in receiving a diagnosis and repeated failed explanations for the pain further fuelled diagnostic uncertainty.

"…like because it took so long to figure out what was wrong with me… it enabled me to like, with my anxiety and stuff, it enabled me to kind of like just create all these different like serious problems with me because like they, they missed like if they can miss something like this [recent diagnosis], what else are they missing, kind of thing? Um so like I just have a general fear of cancer. I always think I have cancer at some point in time so. That's like my first question going into a doctor. Like, 'Do I have cancer?' 'Doesn't sound like it.' 'Okay, cool…' 'Cause like with um this endometriosis, just kind of the symptoms I have been experiencing as of late are very um, synonymous with ovarian cancer." (Olivia, age 18)

When pain did not take the course that was perceived as promised by clinicians, uncertainty developed for Lilly that something had been missed. This bred frustration when time, even years, passed with little improvement in pain symptoms. In the midst of longing for a cure, youth doubted whether clinicians had done everything they could when the pain began and they wondered if something else could have been done if searched for earlier.

"I still feel slightly as if they're missing something, as if something else is wrong because they said it's going to rule itself out but they've been saying that for two years and it hasn't. So I think it's just the frustration like, I just want to be fine. I want to be able to run without feeling like I can't breathe and fainting. I want to be able to leave my house and not be in pain

constantly… So I think it was just the sake of- I was like did they catch everything they could've?" (Lilly, age 15)

For others, the fear that they could be missing the cause of the pain was fuelled by guilt and by consequences of past experiences when something *had* been missed. Although intricately tied to mistrust in the medical system, James' mother shouldered the guilt of having missed a diagnosis earlier in his childhood because she had accepted an inaccurate clinical diagnosis and had not advocated effectively for her child at that time. This past experience drove current worries that something sinister was being missed in explaining her child's chronic pain condition, a decade later. For some, when explanations from clinicians were perceived as inadequate or inaccurate, the assumed responsibility of finding a resolution for the pain shifted from clinicians to the parents themselves.

"And what if we're missing it because we missed his [reflux]- this is where it goes back to me with the guilt right- We missed that reflux that caused the damage to the kidneys so so, I'm always now really worried, like what if we're missing something that's causing this and is gonna have long term effects. 'Cause I feel like that we missed it the first time. Right- like so that's what probably my biggest worry is." (Mother of James)

In the face of negative test results, parents and youth struggled to make sense of a chronic pain diagnosis. Test results seemed to have ruled out all other possibilities, and yet this did not provide definitive proof of, or a cause for, the pain. Furthermore, living with chronic pain did not fit with their expectations of what a normal childhood should be.

"I'd go see doctors but they'd just kind of um test my blood, and I'd get CAT scans and they'd say like everything was fine. It was strange, 'cause we knew everything wasn't fine. 'Cause it's not normal to get so many headaches." (Julia, age 16)

Parents seemed to oscillate between rationalizing that all tests had been done and feeling that there must still be a physical cause for the pain that was yet to be found. For many, negative test results did not provide relief and raised more questions. Parents and youth wanted tests to 'prove' a 'physical' diagnosis, especially one that was curable, rather than tests which continued to exclude diagnoses.

> *"I know she hasn't got any kind of like actual physical structural problems because they did an MRI last summer, but I just think it has to be some kind of, either, you know, the wonders of the world of migraines that people don't understand or stress, like a bunch of little triggers… despite my lack of uh belief in catastrophic thinking, uh if you are, if somebody's fainting 10 times a day there must be some kind of physical trigger, you'd think. There needs to be a physical trigger." (Father of Nicole)*

Parents expressed tension between the risks involved in continuing to search for a cause of the pain and the need to end the search despite never definitively knowing whether something had been missed. Parents conveyed lingering questions about how long to continue testing, whether or not tests were necessary, and when the number of tests would be enough to stop the search. This was made even more difficult as parents were making decisions about their child and not themselves.

> *"I just feel like probably the biggest thing is… from a pain point of view, I feel like I really hope we're not missing something that's causing this. And yet you don't want to do a million tests, like there's risks to MRI, there's risks to CT scans, right like, so you don't want to, you don't want to be… also, you want to be sure you're being diligent but you also want to be sure you're not doing things unnecessarily so it's a very fine balance." (Mother of James)*

3. The Search for an Alternative Diagnosis

Even after youth received a diagnosis and treatment, some parents and youth continued to search for an alternative diagnosis for the pain. When clinicians were not perceived as fulfilling their role in providing the 'right' diagnosis, this led to frustration and a pursuit for another

clinician who would provide an acceptable answer, reassurance, and alternative explanations. These alternative clinicians were perceived as offering hope and promise for achieving the ultimate goal of abolishing the pain.

> *"... I had done some research and had found there is a [doctor 1] here in [City] is a specialist in endometriosis so I had asked [doctor 2] if we could get a referral... I want [Olivia] to see [doctor 1] because [she] wouldn't be satisfied until the person who is a specialist in that field tells her." (Mother of Olivia)*

Even parents and youth who perceived the medical system as being supportive reported searching for another explanation for the pain. While *both* parents and youth were in pursuit of an elusive "satisfied" result, which ultimately was a *fix* for the pain, parents took on this search as part of their parental role. In this way, finding a resolution for the pain was part of being successful in their role as a parent.

> *"You know I've said to my husband you know, 'do you think like we should take him like, maybe we should get a consult elsewhere'... As a mom I feel like we shouldn't be stopping until we can fix this headache is what I feel like." (Mother of James)*

In some cases, the youth's need for a diagnosis, a reason, and closure for the pain was so strong that it was worth undergoing invasive surgical procedures, even when the parent was unsure it would help. As Emily's mother explains below, undergoing surgery brought hopeful expectations of a final validation for her daughter that would end her search for a cause of the pain. However, for many families, a final validation was seemingly just beyond grasp, tenuous, and intangible.

> *"Well I truly hope that when she has the [surgical procedure], she'll know that everything has been done, that can possibly be done. I don't want her to have a surgical procedure. I wish she could just- but I think for her to move forward. That's her final validation." (Mother of Emily)*

Others expressed a shift from an arduous search for the cause of the pain to a focus on function. However, within dyads, this shift did not necessarily occur in unison.

> *"I think, I think early on I was always trying to find "it", you know? If we just figure this out you know, then happy pain-free ever after. Like, "the cause" and, therefore, "the solution…" And, where I'm at now… I think it's all about function and making the best of what it is, you know?" (Mother of Julia)*

> *"I'm still not really sure where I stand with [the diagnosis] 'cause it's just very confusing like different doctors saying different things." (Julia, age 16)*

4. Mistrust in the Medical System

Uncertainty and fear that something could be missing was met with varied emotions. When individuals felt that diagnoses provided by clinicians were lacking credibility, individuals perceived a lack of voice and sense of not being heard. Beyond worry about a lack of, or unsatisfactory, diagnosis, youth expressed frustration with communication with their clinicians. A disconnect between patient and clinician agendas was sensed by youth, creating a deeper divide between clinicians and youth. This ultimately impacted their trust in clinicians and the medical system as a whole.

> *"I don't want to tell people how to do their job but I felt that nobody was doing their job for me. Like they weren't paying enough attention so I was like, 'This is what it is.' And they're like, 'No it's not.' And I'm like, 'But you just think I'm pregnant so—' 'Cause every time I went in she gave me a pregnancy test which was super frustrating because I wasn't even sexually active then… But every time I went in, it was pregnancy test and I'm like, 'Last week I wasn't pregnant, this week I'm not pregnant, let's like try something else.'" (Olivia, age 18)*

Parents also noted the divide between their children and clinicians. For Olivia and her mother, a credible plan to receive the 'right' diagnosis was celebrated. The emotional intensity was about more than the relief of having a plan, it was about being listened to and validated. Importantly,

Olivia's mother anticipated that even if a negative result was found (i.e., "it's clean") the results of this procedure would lead to a subsequent plan.

> *"Again, we came out of there crying because [Olivia] was like oh my God she's awesome she heard me. That's exactly what she wanted someone to go in there and say okay if it's cleans it's clean what are we going to do about it, if it's not clean okay we're fixing it. A plan, and she was heard. I can't tell you it was like [sigh], like I'm just the adrenaline right now is just cause it's been such a long road." (Mother of Olivia)*

An explanation that didn't fully 'fit' left youth feeling dismissed, unworthy of clinicians' time, and that they had not received proper care.

> *"I do believe them that hormones are playing into it but it's not just hormones. It feels like they're just trying to find a way so that way they can see their other patients that have been waiting. They're just like 'it's hormones, goodbye.' I think that they are saying that for a reason but I don't like it because it makes me feel like I'm not worth the extra effort." (Nicole, age 14)*

Parents and youth also perceived clinicians' own uncertainty or lack of confidence in the diagnosis. Stephanie's mother particularly expressed a lack of confidence in the amount of specialty training that the clinician providing the diagnosis had received.

> *"Initially I was not super accepting of [the CRPS diagnosis]. It seemed like it was 'kind of but we're not sure so', um was the feeling... And some of that you get because you end up with residents who end up with, who are, who are gracious enough to say, "We don't know." Right? So they go get an emerg doc and I've had enough experience with some of these guys and they're not specialists." (Mother of Stephanie)*

Clinician communication affected not only patient 'buy in' to the diagnosis but also patient confidence in the clinicians' ability to help or provide an accurate diagnosis.

> *"I didn't really know what it was. Like I had never really heard of it before and he was like um, and the doctor, he was like 'You're the first person that I've ever diagnosed with this.'...*

So um I was just like this doesn't really make any sense. …But um, yeah, I was just, I didn't really know what to think of it. Like, I don't know, I was just kinda confused." (Jada, age 15)

Perceived credibility of the treatment team was critically important for some families in reducing their diagnostic uncertainty. Jada reported a better understanding of the diagnosis after meeting with an interdisciplinary pain team who "knew more about [the diagnosis]". Whereas Jada expressed acceptance of the diagnosis, Catherine on the other hand, expressed continued doubts in clinician's understanding of her condition. For her, she perceived that clinicians were uncertain in the diagnosis themselves.

"I think they're missing stuff. Or they don't know what it is and they're just trying to diagnose it." (Catherine, age 11)

Even when trust in clinicians was expressed, an air of uncertainty lingered. While expressing a sense of hope that a solution would be found, unanswered questions undermined parents' ability to fully trust their child's clinicians.

"I mean I trust the radiologist has some ideas on what it is– I guess… I have that little inkling of, 'okay well, why is his bloodwork off too?' Like, 'could it be something more?' But I trust that they're gonna figure… I trust they're gonna figure it out…" (Mother of Samantha)

Discussion

This is the first study to explore the experience of diagnostic uncertainty in youth with pediatric chronic pain and their parents. In-depth thematic analysis of interview data with youth and parents resulted in the generation of four themes, which illustrate the complex nature of a chronic pain diagnosis and the experience of diagnostic uncertainty. Across the themes, which were labelled, The Function of a Diagnosis, Haunted by Something Missing, The Search for an Alternative Diagnosis, and Mistrust in the Medical System, parents' and youth's struggle to

accept and understand a chronic pain diagnosis was met with frustration, disempowerment, feeling unheard and alone, and mistrust in the medical system.

Parents and youth expressed a lack of clear understanding of the diagnosis provided. For many, a diagnosis of chronic pain was not a satisfying result of medical investigations and parents and youth were left wanting a biological/physical explanation for the pain. For some youth, a diagnosis of 'chronic pain' conveyed that clinicians did not understand the pain or did not perceive the pain as being 'real.' A diagnosis from a clinician that 'fit' was a 'game changer' and was critical for others, particularly parents, to 'buy in' to the authenticity of the pain. Without a diagnosis that was perceived as credible, however, youth felt invalidated, which is commonly reported among patients with chronic pain (Carter, 2002; Kool, van Middendorp, Boeije, & Geenen, 2009). In a qualitative study of children with medically unexplained symptoms, children and their parents expressed struggling to accept a lack of a credible diagnosis and needing an explanation for their symptoms (Moulin et al., 2015). Moreover, most children reported not feeling understood, believed, or listened to by physicians (Moulin et al., 2015). Although validation has not been shown to lead to reductions in pain, invalidation during a pain tolerance task with healthy adults has been linked to a decrease in positive affect and increased worry (Linton, Boersma, Vangronsveld, & Fruzzetti, 2012). Future research is needed to examine the impact of validation on other components of pain (e.g., disability) and in chronic pain populations (Edmond & Keefe, 2015). Indeed, validation was a core objective in parents' and youth's search for a diagnosis. Validation of pain (i.e., communicating that pain-related thoughts and feelings are understandable and legitimate) may impact patients' emotional state (i.e., increase/maintain positive affect) which could reduce pain (Edmond & Keefe, 2015) and possibly increase orientation to and acceptance of a chronic pain diagnosis.

Uncertainty extended beyond diagnosis to treatment. Parents and youth expressed holding beliefs that the 'right' diagnosis would lead to a pharmacological treatment, which would 'fix' the pain. This same belief was found in reverse throughout parent and youth interviews, such that a reduction in, or eradication of, pain was seen as proof that the 'right' diagnosis had been found. The objective of receiving a diagnosis that would lead to a treatment that would make the child pain-free, has been previously reported by parents of children with functional abdominal pain (Brodwall, Glavin, & Lagerlov, 2018). This belief that a 'right' diagnosis would lead to a cure for the pain demonstrates a desire that is incompatible with current chronic pain treatments, which prioritizes improving function, often with the expectation/hope that this will lead to improvements in pain (Friedrichsdorf et al., 2016; Lynch-Jordan et al., 2014).

Parents and youth expressed uncertainty and concerns that a serious health condition (e.g., cancer) could have been missed, which fuelled an unrelenting search for the 'right' diagnosis and cause of the pain. For many, negative test results did not provide relief but rather fuelled diagnostic uncertainty while parents and youth attempted to make sense of the pain in the absence of objective evidence. Despite a widely held belief by clinicians that additional testing will reassure patients (van der Weijden et al., 2002), there is limited evidence that diagnostic testing reassures patients (van Ravesteijn et al., 2012). Among adults, informational reassurance (e.g., explanations about physical causes for pain) has been shown to be more reassuring than emotional reassurance (e.g., being taken seriously) (Holt et al., 2015). This is important given the economic toll pediatric chronic pain has on families and society (Groenewald et al., 2014). The impact of pain education on diagnostic uncertainty in youth and their parents may be a particularly fruitful area for future research (Robins, Perron, Heathcote, & Simons, 2016).

Diagnostic uncertainty seemed to vary in intensity across participants. While some individuals expressed acceptance of the diagnosis, they continued to pursue other clinicians and additional diagnostic tests in a quest for answers and to exhaust other possible explanations. Parents found themselves caught between two opposing notions of being a good parent (i.e., protecting their child from potential harm of invasive procedures vs. not ending the search until the pain was 'fixed'), which was distressing. This is consistent with previous literature on parents' experience of anguish when they are not able to 'make things better' for their child with pain (Jordan et al., 2007). Enhanced understanding about how parental diagnostic uncertainty influences children's uncertainty, adjustment, and adherence to treatment, and pain outcomes, is important for future research. This qualitative analysis demonstrated that parents and youth are not always in agreement in terms of their pain conceptualization and 'buy-in'. What this lack of concordance means for children's outcomes is currently unknown.

How pain is conceptualized has been shown to be important for pain knowledge and catastrophizing, although its impact on disability is unclear (Gallagher, McAuley, & Moseley, 2013). Moreover, how a diagnosis of chronic pain is communicated to youth and families is likely critical to their 'buy-in,' and is likely influenced by developmental factors and individual needs. A better understanding of diagnostic uncertainty, between and within parent-youth dyads, may help tailor how clinicians deliver diagnoses to families to achieve 'buy in', increase understanding of pain and diagnosis, and ultimately improve treatment engagement and response. Youth also reported sensing uncertainty in clinicians providing the diagnosis and misalignment in agendas between themselves and clinicians. Frustration has previously been reported by children with chronic pain during medical encounters when they feel they have something to contribute but their narratives and beliefs are not elicited (Carter, 2002). In the

current sample, misalignment in patient-clinician communication and expectations was tied to diagnostic uncertainty and (mis)trust in the medical system. However, alignment between the orientation of patients (and caregivers) and their clinicians (e.g., if the patient is uncertain *and* perceives that the clinician is also uncertain about the pain) may also play a role in acceptance of a diagnosis and fuel diagnostic uncertainty. Past experiences of medical encounters and a learning history of incorrect diagnoses from clinicians fuelled uncertainty, demonstrating the importance of memories of previous medical care in influencing the experience of diagnostic uncertainty.

This research should be interpreted in light of limitations. The current data are from the perspectives of youth and parents and thus, the information families were provided with regards to the youth's diagnosis is unknown. Future research should investigate how aspects of the clinician-patient encounter (e.g., type of clinician, provision of pain education) when a diagnosis is provided, influence parents' and youth's memories of the encounter, their perceptions of the diagnosis, and the impact of communication on diagnostic uncertainty. In addition, interviews were conducted at a single time point; thus, this study provided a snapshot of parents and youth at a particular time in their pain journey. It is likely that perceptions of diagnostic uncertainty are dynamic over time and are influenced by experiences and developmental factors. Future research might consider contacting families following the provision of a diagnosis to explore their interactions with clinicians and identify potential ways to reduce uncertainty. Finally, parent data in this study largely reflect maternal accounts of diagnostic uncertainty due to the low number of fathers (n=3) who participated. Furthermore, participants in the current sample predominantly reported high socioeconomic status, thus these accounts may not be representative of families

from lower socioeconomic status who may face additional challenges (e.g., stigma, lack of resources) affecting diagnostic uncertainty.

This study highlights diagnostic uncertainty as a core feature of pediatric chronic pain. Given that untreated chronic pain in childhood can lead to persistent pain problems (Walker et al., 2012) and internalizing mental health disorders (Noel, Groenewald, et al., 2016) into adulthood, investigation of diagnostic uncertainty and its impact on treatment engagement and response in this population is warranted. Illness uncertainty in pediatric chronic illness has been linked to worse psychological functioning and illness-related distress (Szulczewski, Mullins, Bidwell, Eddington, & Pai, 2017). Among adults with chronic pain, diagnostic uncertainty is associated with poorer pain outcomes (Geisser & Roth, 1998; Serbic et al., 2016). Further research is needed to understand factors that influence diagnostic uncertainty in youth and their parents, as well as the impact of diagnostic uncertainty on chronic pain and treatment outcomes over time. Research suggests that over 40% of adult patients with chronic pain (Serbic & Pincus, 2014) and parents of youth with chronic pain experience diagnostic uncertainty (Noel, Beals-Erickson, et al., 2016). Our data suggest this is also frequently experienced by youth themselves. Nevertheless, the prevalence of diagnostic uncertainty in pediatric chronic pain populations should be established. Pincus and colleagues (Pincus et al., 2018) provide suggestions on how diagnostic uncertainty can be quantified in future research. This study suggests that there may be differences in how diagnostic uncertainty is perceived by parents and youth. Given that parental beliefs can impact children's medically unexplained symptoms (Eminson, 2007), parents' perceived diagnostic uncertainty warrants further investigation. If parent and/or youth diagnostic uncertainty is associated with response to pain treatment, interventions to decrease diagnostic

uncertainty could have far-reaching implications across pain outcomes and health care expenditures.

Table 1.1

Sociodemographic Characteristics of the Sample

	N	**Mean**	**Range**
Youth's age, years	20	14.6	10-18
Parent's age, years	18[a]	46.8	33-55
Relationship to youth			
Biological Mother	17		
Biological Father	3		
Youth's ethnicity (%)			
White (Caucasian)	18		
Latin American	1		
Do not want to answer	1		
Household income			
<$10,000 to $29,999	1		
$30,000 to $59,999	1		
$60,000 to $89,999	1		
More than $90,000	15		
Do not want to answer	2		
Marital Status			
Married	14		
Single	1		
Separated/divorced	3		
Widowed	1		

Common-law	1

[a]Data only reported on 18 parents as two parents did not report their age

Table 1.2
Semi-structured Interview Schedule

Interview Questions
1. Where did this all start?
2. Where are you now in your journey?
3. Where do you see this going?
4. What do you remember about getting the diagnosis and what do you make of that?

CHAPTER THREE: Diagnostic Uncertainty in Pediatric Chronic Pain: Nature, Prevalence, and Consequences

Abstract

Introduction: Diagnostic uncertainty (DU), which is the perception that a label or explanation for a patient's health problem is missing or inaccurate, has been linked to distress, anxiety, and difficulty coping among adults with pain. This study examined the prevalence of DU among youth with chronic pain and their parents and the relation of parent and youth DU with youth pain, pain-related constructs, and health-related quality of life. **Methods:** Participants included 174 youth with chronic pain (M_{age}=14.28 years; 73% female) and one of their parents (91% mothers) recruited from a tertiary-level pediatric chronic pain program in Canada. Youth and parent DU was assessed using a brief measure of three empirically-derived yes/no questions regarding whether the youth and parent had received a clear diagnosis/explanation for their/their child's pain and whether they believed there was something else happening with their/their child's pain which doctors had not yet found. Youth reported on their pain intensity, pain interference, pain catastrophizing, fear of pain, and health-related quality of life. **Results:** 31% of youth and 28% of parents experienced DU. 70% of parents and youth were in agreement regarding their experience of DU. Youth DU was linked to higher youth catastrophic thinking about their pain. Parent DU was linked to greater youth pain interference and intensity and lower youth health-related quality of life. **Discussion:** DU is experienced by nearly a third of youth with chronic pain and their parents and is linked to worse youth pain, pain catastrophizing, and health-related quality of life.

Introduction

Pediatric chronic pain affects 11-38% of youth (King et al., 2011), with approximately 3-5% reporting significant disability (Hechler et al., 2010; Huguet & Miro, 2008). In the absence of underlying organic pathology or positive test results, many youth receive a primary diagnosis of 'chronic pain' (Eccleston & Malleson, 2003) or 'Primary Pain Disorder' (Schechter, 2014b). As a consequence, youth and their parents may experience uncertainty regarding the diagnosis, prognosis, and treatment (Neville, Jordan, Beveridge, Pincus, & Noel, 2019). Diagnostic uncertainty (DU), the perception that a label or explanation for a patient's health problem is missing or inaccurate, is associated with higher levels of anxiety, depression, pain intensity (Serbic et al., 2016), and disability (Geisser & Roth, 1998) among adults with chronic primary pain. Research suggests that over 40% of adults with chronic low back pain experience DU (Serbic & Pincus, 2014). However, the prevalence and impact of DU in the context of pediatric chronic primary pain is relatively unknown.

Nearly 40% of parents of youth with chronic primary pain report not accepting or 'buying in' to their child's diagnosis (Noel, Beals-Erickson, et al., 2016), suggesting that the prevalence of DU among parents of youth with chronic pain may be consistent with adult literature. In a qualitative examination of DU among 20 youth with chronic primary pain and their parents, DU was characterized by a relentless search for the 'right' diagnosis, which may limit acceptance of a diagnosis and delay or prevent engagement in treatment (Neville et al., 2019). A recent topical review proposed that research should examine the prevalence of DU among youth with chronic pain and their parents, and its influence on treatment and pain outcomes (Pincus et al., 2018). A brief measure of DU was developed for adults with chronic pain (Pincus et al., 2018; Serbic & Pincus, 2013, 2014), but has never been used among youth with chronic pain and their parents.

This study aimed to 1) describe the prevalence and nature of DU among youth with chronic pain and their parents, 2) compare youth pain (intensity and interference), pain-related constructs (pain catastrophizing, fear of pain) and health-related quality of life (HRQoL) in youth with chronic pain who do versus do not report DU and in parents of youth who do versus do not report DU. We hypothesized that prevalence rates of DU would parallel adult literature and that youth and parent DU would be related to worse youth pain, pain-related constructs, and HRQoL.

Methods

The current cross-sectional survey is part of the Pain and Mental Health in Youth study (PATH). The current aims are distinct from previously published papers on this study (Neville, Griep, et al., 2020; Neville et al., 2019; Neville, Kopala-Sibley, et al., 2021; Pavlova et al., 2020). The study was approved by the institutional health Research Ethics Board. Participants included 174 youth with chronic pain (M_{age}=14.28 years; 73% female, range=10-18 years) and one of their parents (91% mothers) recruited from a tertiary-level pediatric chronic pain program in Canada. Youth were enrolled from headache (67%), complex pain (28%), or abdominal pain (5%) clinics. Youth were eligible for the study if they were between 10-18 years of age and identified as having chronic pain (i.e., pain ≥ 3 months) without an underlying disease (e.g., cancer) (Merskey & Bogduk, 1994b). Youth with a diagnosis of a severe cognitive disability or developmental disorder were excluded. Parents and youth consented and completed self-report questionnaires using Research Electronic Data Capture (REDCap) (Harris et al., 2009).

Measures

Parent and youth diagnostic uncertainty. DU was assessed using a brief measure consisting of three empirically derived categorial questions, which were constructed from a study

investigating DU among adults with low back pain (Serbic & Pincus, 2013, 2014). This is the first study to use this measure since it was adjusted for use with any chronic pain condition (Pincus et al., 2018).Youth and parents responded yes/no to 3 questions presented in Table 2.1. Research has demonstrated that individuals with chronic pain often simultaneously report having received a clear diagnosis, while believing that something else is happening with their pain (Serbic et al., 2016).Thus, question 3 is particularly informative in terms of DU and has been linked to adverse outcomes among adults with chronic pain (Pincus et al., 2018; Serbic & Pincus, 2014; Serbic et al., 2016). DU was therefore operationalized as youth and parents who responded 'yes' to question 3. If youth or parents responded yes to questions 1 or 2, they were asked whether or not they agreed with the diagnosis or explanation provided to them.

Youth pain. Youth reported on their pain characteristics using the valid and reliable Pain Questionnaire (Palermo, Witherspoon, Valenzuela, & Drotar, 2004). Youth reported their average pain intensity in the past seven days using a validated 11-point numeric rating scale (0="no pain", 10="worst pain possible") (Castarlenas, Jensen, von Baeyer, & Miro, 2017; von Baeyer et al., 2009).Youth completed the Pain Interference Subscale of the Patient-Reported Outcomes Measurement Information System (PROMIS) Pediatric Profile-25 (Irwin et al., 2010; Kashikar-Zuck et al., 2016; Neville, Soltani, Pavlova, & Noel, 2018; Noel et al., 2018; Noel, Wilson, et al., 2016).

Youth pain-related constructs. The Pain Catastrophizing Scale-Child version (PCS-C) (Crombez et al., 2003) was used to assess catastrophic thoughts about child pain. The Fear of Pain Questionnaire, child report (FOPQ-C), was used to assess pain-related fears and avoidance behaviours (Simons, Sieberg, Carpino, Logan, & Berde, 2011).

Youth health-related quality of life. HRQoL was assessed using the Pediatric Quality of Life Short Form (PedsQL-SF) (Chan, Mangione-Smith, Burwinkle, Rosen, & Varni, 2005).

Results

Tables 2.2 and 2.3 present sample characteristics and key variable associations. Girls reported significantly higher pain interference than boys ($p<.05$). Older youth reported higher pain interference ($r=0.17$, $p<.05$), fear of pain ($r=0.24$, $p<.01$), and lower HRQoL ($r=-0.29$, $p<.01$) than younger youth.

Participants' responses to the DU questions are summarized in Table 2.4. Nearly a third of youth (31%) and parents (28%) believed something else was happening with their/their child's pain which the doctors had not yet found. 21% of youth and 17% of parents who reported that they had received a clear diagnosis also reported thinking something else was happening with their/their child's pain. 70% of parents and youth concurred regarding their DU (i.e., both responded yes or no to question 3); 16% of parents and 14% of youth reported being uncertain while their child or parent was certain, respectively.

Table 2.5 presents descriptive statistics and results of ANCOVA analyses. Controlling for youth age and gender, youth who experienced DU reported significantly higher pain catastrophizing than youth who did not experience DU. Youth whose parents experienced DU reported significantly greater pain interference, greater pain intensity, and lower HRQoL than youth whose parents did not experience DU.

Discussion

This is the first quantitative investigation of DU among a large sample of youth with chronic pain and their parents. Nearly a third of youth with chronic pain and their parents

experienced DU. Consistent with findings among adults with low back pain (Serbic & Pincus, 2014), a substantial minority of youth and parents experienced DU despite reporting that they received a clear diagnosis for their pain. Serbic and Pincus postulated that such findings may indicate beliefs that a diagnosis may be correct but does not fully capture the cause of the pain (Serbic & Pincus, 2014). In the current study, most parents and youth who reported receiving a diagnosis also reported *agreeing* with that diagnosis. Yet, many families, who are imbedded in a tertiary level chronic pain program for primary pain disorders, continue to believe something undiagnosed is causing the pain. This finding alludes to the perception that the diagnosis provided did not fully explain the youth's pain and what was lacking was the core etiology. This has important implications for clinical encounters when a chronic pain diagnosis is provided, suggesting that the diagnosis itself is not sufficient to allay fears and beliefs that something unknown is causing the pain. Qualitative research with youth with chronic pain and their parents has revealed that many parents and youth express needing further explanation for their pain (Jordan et al., 2007; Neville et al., 2019). Indeed, a chronic pain diagnosis is often perceived as a diagnosis of exclusion and to not reflect a true understanding of the cause of the pain (Neville et al., 2019). Further research is needed to understand how clinicians could tailor their explanation of a chronic pain diagnosis in a way that would reduce DU.

This study is the first to investigate the relation of youth and parent DU with youth's pain, pain-related constructs, and HRQoL. Whereas youth DU was linked to higher youth catastrophic thinking about their pain, it was *parent* DU that was linked to greater youth pain interference and intensity and lower HRQoL. In line with the interpersonal fear avoidance model of pain, uncertainty may fuel parental distress and protective responses around child pain, which may facilitate youth avoidance behaviours and influence youth pain interference (Neville,

Kopala-Sibley, et al., 2020; Simons et al., 2015). Further longitudinal research is needed to understand how DU unfolds over time, its temporal relations with outcomes, and what factors during diagnosis and treatment influence DU. Moreover, the direction of the association between youth pain and parent DU remains unknown—that is, whether having a child with worse pain exacerbates parent DU or vice versa.

A limitation of the current study is the dichotomous nature of the DU questions, which may not capture the varying intensity and nuances of DU (Neville et al., 2019). DU may fluctuate over time depending on where youth are in their pain and treatment journey, which was not assessed in the current study. Further, the current data is based on self-report; thus, the information provided to families regarding diagnosis is unknown. Future research should examine how DU differs based on chronic pain diagnosis and explanation (e.g., etiological versus primary pain). Nevertheless, our findings demonstrate that DU is a common experience among youth with chronic pain and their parents and is linked to worse pain, pain-related constructs, and HRQoL.

Table 2.1
Brief measure of perceived diagnostic uncertainty

Questions
Youth
1. I have been given a clear label/diagnosis for my pain
2. I have been given a clear explanation about why I have pain
3. I think there is something else happening with my pain, which the doctors have not found out about yet.
Parent
1. My child has been given a clear label/diagnosis for their pain
2. My child has been given a clear explanation about why they have pain
3. I think there is something else happening with my child's pain, which the doctors have not found out about yet.

Table 2.2
Socio-demographic and pain characteristics of the sample

Sample Characteristics	*N* = 174
Child age (*M* years, *SD*)	14.28(2.22)
Child gender (% female)	73.0
Parent gender (% female)	91.3
Child's Ethnicity (%)	
White	79.3
Two or more ethnicities	8.0
Other	4.2
Black	1.7
Arab/West Asian	1.7
South Asian	1.7
Aboriginal	1.1
Latin American	1.1
Filipino	0.6
Did not want to answer	0.6
Annual household income (%)	
<$10,000-$29,999	5.7
$30,000-$59,999	10.3
$60,000-$89,999	12.1
>$90,000	57.5
Did not want to answer	9.2
No income reported	5.2
Pain locations (%)	
Multiple locations reported	43.7
Head	72.4
Other	24.7
Muscles and joints	23.0
Stomach	20.1
Legs	12.6
Chest	10.9
Child pain intensity (*M* out of 10, *SD*)	5.53(1.84)
Child pain duration (*M* years, *SD*)	3.20(3.11)

Table 2.3
Correlations among variables of interest

Variable	1	2	3	4	5
(1) Youth pain intensity	1	0.50‡	0.39‡	0.33‡	-0.39‡
(2) Youth Pain interference		1	0.48‡	0.50‡	-0.60*
(3) Youth pain catastrophizing			1	0.77‡	-0.60‡
(4) Youth fear of pain				1	-0.65‡
(5) Youth quality of life					1

Note. ‡ $p < .01$; * $p < .05$

Table 2.4
Participant responses to diagnostic uncertainty questions

	Yes *n*(%)		No *n*(%)		Total *n*	
	Youth	Parents	Youth	Parents	Youth	Parents
I/My child have/has been given a clear label/diagnosis for my/their pain	72(42.6)	79(46.2)	97(57.4)	92(53.8)	169	171
I agree with this label/diagnosis	70(97.2)	76(96.2)	2(2.8)	3(3.8)	72	79
I/My child have/has been given a clear explanation about why I/they have pain	42(24.9)	73(42.9)	127(75.1)	97(57.1)	169	170
I agree with this explanation	39(92.9)	68(94.4)	3(7.1)	4(5.6)	42	72
I think there is something else happening with my/my child's pain, which the doctors have not found out about yet.	51(30.5)	47(27.5)	116(69.5)	124(72.5)	167	171

Table 2.5

ANCOVA results controlling for youth age and gender

Variable	*N*	Uncertain *M (SD)*	Certain *M (SD)*	*df*	*F*	*P*
Parent DU						
Youth pain interference	165	57.83 (9.07)	54.06 (8.90)	1,161	4.23	.04
Youth pain intensity	165	6.09 (1.55)	5.27 (1.86)	1,161	5.81	.02
Youth Catastrophizing	164	21.67 (12.44)	18.01 (12.08)	1,160	2.38	.13
Youth fear of pain	160	35.91 (22.65)	28.50 (19.13)	1,156	3.48	.06
Youth health-related quality of life	164	57.86 (19.04)	68.14 (19.10)	1,160	7.44	.01
Youth DU						
Youth pain interference	163	56.04 (9.11)	54.49 (9.16)	1,159	.88	.35
Youth pain intensity	163	5.90 (1.68)	5.30 (1.86)	1,159	3.81	.05
Youth Catastrophizing	162	22.60 (12.50)	17.42 (11.70)	1,158	6.06	.02
Youth fear of pain	159	34.90 (18.87)	28.40 (20.55)	1,155	3.45	.07
Youth health-related quality of life	162	61.25 (19.56)	67.32 (19.01)	1,158	3.37	.07

CHAPTER FOUR: ‘Drawing a Line in the Sand’: Physician Diagnostic Uncertainty in Paediatric Chronic Pain

Abstract

Background: Diagnostic uncertainty is the subjective perception of an inability to provide an accurate explanation of the patient's health problem or that a label is missing or incorrect. While recently explored in youth with chronic pain and families, this is the first study to investigate diagnostic uncertainty from the perspectives of physicians. **Methods:** Individual, semi-structured interviews were conducted with sixteen pediatricians who assess and/or treat youth who experience complex chronic pain. Interviews explored pediatricians' perceptions, beliefs, and confidence regarding the assessment and management of chronic pain in youth and how they manage uncertainty regarding the diagnosis. Interviews were analysed using inductive reflexive thematic analysis. **Results:** Analyses generated one prominent theme: 'drawing a line in the sand'. Within this theme, physicians discussed uncertainty as inherent to their role treating youth with chronic pain. The metaphor of 'drawing a line in the sand' was used to describe a process of identifying a point at which physicians no longer sought a new diagnosis for the child's pain or continued diagnostic investigations. This line was influenced by numerous factors, which are highlighted through four subthemes: physician training, experience, and mentorship; individual patient and family factors; perceived reassurance of diagnostic investigations; and the broader social context and implications. **Conclusions:** How physicians manage diagnostic uncertainty must be understood, as it is likely to critically impact how a diagnosis of chronic pain is communicated, the diagnostic investigations undertaken, the wait time to receiving a diagnosis, and ultimately youth's pain experiences.

Introduction

Many patients with chronic pain do not receive a diagnosis that precisely explains a pathological cause of their pain (Butow & Sharpe, 2013; Serbic & Pincus, 2013). Over 40% of adults with chronic pain experience diagnostic uncertainty (DU), the perception that a label or explanation for a patient's health problem is missing or inaccurate (Serbic & Pincus, 2013), which is associated with elevated levels of anxiety, depression, pain intensity (Serbic et al., 2016), and disability (Geisser & Roth, 1998). Individuals who perceive their pain as not appropriately labeled or legitimized, experience emotional distress and may continue to pursue medical investigations, which contribute to increasing economic burden (Newton et al., 2013).

DU has been identified as a critical area of research in pediatric chronic pain (Pincus et al., 2018). In a qualitative study, youth with chronic pain and their parents expressed needing further explanations for their pain beyond a diagnostic label, and an unrelenting search for an alternative diagnosis that many believed had been missed by clinicians (Neville et al., 2019). Notably, parents and youth perceived that clinicians appeared uncertain, which influenced their confidence in the clinician's understanding of their condition, the diagnosis, and the treatment plan. Thus, DU may have critical implications for pediatric chronic pain outcomes and treatment engagement, and is integrally tied to clinical encounters youth and parents experience.

Although clinicians regularly experience DU, defined as the subjective perception of an inability to provide an accurate explanation of the patient's health problem (Bhise et al., 2018), clinicians experience unique challenges caring for patients with chronic pain. Clinicians who manage adult chronic pain have reported feeling unsuccessful as physicians and in their ability to effectively manage their patients' chronic pain problem (Matthias et al., 2010). In the context of *pediatric* chronic pain, uncertainty has been studied among clinicians in terms of assessing pain

in children with profound cognitive impairment, with clinicians managing uncertainty through seeking mentorship and discussion with colleagues (Carter, Simons, Bray, & Arnott, 2016). The implications of uncertainty among clinicians are enormous at both individual (e.g., unnecessary and invasive medical investigations) and societal (e.g., increased healthcare costs and wait times) levels (Allison et al., 1998; Bhise et al., 2018; Kassirer, 1989; Schneider et al., 2010; Simpkin & Schwartzstein, 2016).

The experiences of DU for youth with chronic pain and their parents are likely linked to how clinicians themselves experience DU. Youth have been found to consult an average of 4 physicians prior to referral to a chronic pain program, with the majority of referrals made by pediatricians (Cucchiaro, Schwartz, Hutchason, & Ornelas, 2017). Low agreement among pediatricians regarding primary cause and diagnostic approach to treating children with unexplained chronic pain suggests that pediatricians experience considerable uncertainty (Konijnenberg et al., 2004). Understanding DU from the perspectives of pediatricians is critical as they are often the first physicians youth consult when they develop a pain problem. The aim of the current study was to qualitatively explore pediatricians' own perceptions, beliefs, and confidence with regard to the assessment and management of chronic pain in youth.

Methods

Study Design

Adopting both a relativist epistemology and phenomenological approach, this study sought to gain an understanding of how pediatricians understand pediatric chronic pain, how they manage uncertainty regarding the diagnosis of chronic pain in the absence of underlying pathology, and how this diagnosis is delivered to and received by families. In brief, relativism refers to ideas which propose that "our experiences, moral judgments, claims to knowledge, and

so on can be understood only relative to something else such as particular languages and particular social and cultural practices", and the lack of a single universal truth (p750) (Smith, 2008), whilst phenomenology refers to a focus on the exploration of experience and meanings (Davidsen, 2013). This study adopted a qualitative semi-structured interview design to allow physicians the flexibility to describe their own personal perspectives and experiences (Mason, 2004). To ensure a rigorous approach to this qualitative analysis, the consolidated criteria for reporting qualitative research (COREQ) checklist were followed (Tong et al., 2007).

Participants

Sixteen pediatricians who assess and/or treat youth who experience complex chronic pain were recruited through established networks, contacts, and healthcare organizations in the United Kingdom (UK). Sociodemographic information of participants is presented in Table 3.1. Pediatricians were eligible to participate in this study if they treated children and adolescents who experience chronic pain in a UK based healthcare setting. Participants were excluded from the study if they had never treated a child or adolescent with pain that had lasted for three months or longer (i.e., chronic pain) and/or were unable to speak and understand English. Eligibility for this study did not require physicians to have any specific training in pain medicine. Pediatricians, rather than pediatric pain specialists, were purposively sampled as they are the most likely physicians to encounter youth early in their pain problem and prior to their enrollment in a tertiary level pediatric chronic pain program.

As research transparency is perceived as an indicator of quality (Spencer et al., 2003), a focus was placed on being thorough and transparent in our appraisal and reporting of the sample size and research procedure. In keeping with this methodological approach, no attempt was made to achieve saturation (O'Reilly & Parker, 2012). Braun and Clarke contend that there are an

endless number of possible interpretations of a data set, suggesting the impossibility of generating all possible interpretations (Braun & Clarke, 2019b). Rather, specific characteristics of this study, including the study aims, the quality and richness of the data, and the researchers' skills in qualitative methods, are recommended in determining sample size and served as a guide in the current study (Vasileiou, Barnett, Thorpe, & Young, 2018). We deliberately recruited a small sample of pediatricians who had experience treating youth with chronic pain in the UK. This is fitting with the theoretically flexible approach of inductive reflexive thematic analysis (Braun & Clarke, 2006). Given the depth of the analysis and richness of the data collected by co-author A.J., who is an expert in the field and an experienced and skilled qualitative researcher, the current sample size met the aims of the study and was methodologically appropriate for the analytical approach (Braun & Clarke, 2013).

Procedure

All study procedures were approved by the Psychology Research Ethics Committee at the University of Bath (ref 17-212). Invitations to take part in the study were circulated through professional paediatric networks. Snowballing was also used through encouraging interested participants to send emails to colleagues. All recruitment notices contained the contact information of co-author A.J., who screened all interested participants for eligibility. Study procedures were verbally reviewed with the physician prior to participation and verbal informed consent was obtained. Interested participants were then emailed a consent form via Qualtrics online survey software (Qualtrics, Version 2019). Following enrollment and consent to participate in the study, participants completed a demographic questionnaire online via Qualtrics. Semi-structured interviews were conducted over the phone by co-author A.J., who did not know any of the participants personally. Interview duration ranged between 18-45 minutes (mean

duration = 29 minutes). Interviews were audio-recorded and transcribed verbatim. To ensure the anonymity of participants and related individuals, all identifying information was removed from the transcripts and participant numbers were used. Participants received a £20 Amazon voucher to thank them for their time.

Measures

Demographic Characteristics. Pediatricians reported on their gender, years of training, job title, medical specialty, and the clinical setting in which they worked at the time of participating in the study.

Interview Schedule

The interview schedule (Table 3.2) focused on exploring physician beliefs and confidence surrounding the diagnoses that they provide to youth with chronic pain and their families. In particular, physicians were asked to discuss how the diagnosis is offered, their confidence in the accuracy of the diagnosis, how they manage uncertainty regarding the diagnosis in the absence of underlying disease/pathology, and how they perceive that the diagnosis is received and understood by the child and their parent(s). The interview schedule was semi-structured and comprised open-ended questions and prompts in order to allow the interviewer to follow-up and expand on topics of interest as they occurred during the interviews (Mason, 2004).

Data Analysis

Data analysis was completed using QSR International's NVivo 12 qualitative data analysis software, a computer-assisted qualitative data analysis package ("NVivo qualitative data analysis software Version 12," 2018). Data were analyzed using inductive reflexive thematic

analysis, in accordance with Braun and Clarke's (Braun & Clarke, 2006, 2019a) six-phase thematic analysis guide. This is a theoretically flexible method of qualitative data analysis that is not limited to a single epistemological view (Nowell, Norris, White, & Moules, 2017) and allows for a detailed and complex account of qualitative data (Braun & Clarke, 2006). Inductive thematic analysis is a "bottom-up" approach aiming to identify patterns of meaning from data (Braun & Clarke, 2006, 2019a). In the reflexive thematic analysis approach, themes are conceptualized as meaning-based patterns (Braun & Clarke, 2006, 2019a). Regarding specific steps taken, following transcription of the data and verification of the transcriptions, A.N. and A.J. read and re-read the transcripts in order to familiarize themselves with the data. Through an iterative process, the data were thoroughly and comprehensively coded by A.N. and A.J. Codes were then collated to identify potential themes. In accordance with Braun & Clarke (Braun & Clarke, 2006), analysis extended beyond description and the data were interpreted for meaning. To address issues around quality of qualitative research, themes and interpretations were iteratively reviewed and refined through discussions with all co-authors, ensuring credibility and trustworthiness in the data and analytic interpretations (Elliott et al., 1999; Morrow, 2005). Definitions of each theme were generated, paying close attention to the specifics of each theme, the relation of each theme with others, as well as the overall story of the data (Braun & Clarke, 2006). Lastly, data were analysed within each theme. Refinement of themes led to the generation of one robust theme within the data with four subsequent sub-themes. Example quotations of this theme and corresponding sub-themes were selected from a variety of participants for the final written report.

Results

A single, complex, prominent theme was generated from the thematic analysis, which was labeled ‘drawing a line in the sand’. Within this single theme, four subthemes were generated including: physician training, experience, and mentorship; individual patient and family factors; perceived reassurance of diagnostic investigations; and the broader social context and implications. Below, this theme and subthemes are presented along with supporting quotations from a range of participants. Identifying information has been removed from quotations and physician numbers have been used to protect the confidentiality of participants.

Drawing a line in the sand

The experience of uncertainty and how to manage it were salient features of physician accounts, with participants describing this as inherent to their role of treating youth with chronic pain. A key aspect of managing DU for physicians related to their perceived need to be able to identify a point at which they no longer sought a new diagnosis for the child’s pain. In particular, the identification of a defining point at which the decision is made to cease all diagnostic testing as part of this wider process was described as being akin to “drawing a line in the sand”. This suggests a clear limit beyond which investigations would proceed any further. However, in reality, determining where and when this line should be drawn was far more complex than the metaphor implies. Despite the acknowledgment of needing to draw a line, the decision to do so was complicated by numerous factors including aspects of the individual case, as well as physicians’ *own* need for reassurance. Further, even when this line in the sand had been drawn, it was not as definitive as one might first imagine. This is illustrated by Physician 4 below who explained that even when this line is drawn, some physicians may continue to follow

the patient ‘just in case’ an underlying ‘cause’ had indeed been missed, even when they perceived no sinister cause for the child’s pain.

> *And so, what I will do is set the line in the sand and then lead forward, make sure the diagnosis is there, and then keep seeing the child just in case I've missed something, and also to manage their pain. (Physician 4, woman, 17 years qualified)*

Physician training, experience, and mentorship

Physicians’ ability to sit with uncertainty and cease diagnostic testing was influenced by a variety of factors, such as the specific nature of their medical training. Interestingly, physicians described how their work treating youth with chronic pain was the antithesis of the focus of their clinical training. Consequently, physicians discussed how their medical training had focused on finding a diagnosis and “fixing” a problem, which conflicted with their current role of managing children’s chronic pain where a clear underlying ‘cause’ and subsequent ‘cure’ was often lacking. In particular, physicians reported difficulties with adjusting to the requirements of chronic pain treatment, which were perceived to be based on principles that were in opposition with their lengthy clinical diagnostic training.

> *I think because it's not what we're trained to do, is it, as clinicians we are trained to find a problem, sort it out, give the medication or whatever it is, the treatment, and everybody's happy, everybody moves on, problem solved and that's it. But with chronic pain … you can't do that, and so it's a completely different scenario, a different situation, and you just have to accept that sometimes there is no cause and there is no answer in terms of why, and whatever the situation is that you're faced with, that's the situation that you've got to deal with and try to improve. (Physician 1, woman, 28 years qualified)*

Due to the perceived incongruence between managing DU in pediatric chronic pain and previous medical training, physicians described needing to adopt a new understanding of their role and identity as a physician. As part of this reconfigurement, they described how they needed to reformulate their own definition of success from that of symptom reduction to encouraging adaptive functioning in their patients. To achieve this, physicians described re-

examining their entire aim and philosophy of care, how they approach this with families, and their role in supporting patients. These processes resulted in physicians actively redefining success as no longer being contingent on symptom management.

> *Well, for me it's not about getting them [patients] better, it's about allowing them to thrive as a person despite them having their symptoms. So, I'm not expecting to get anyone better, but I am expecting them to go through school, get their qualifications, do what they want to do in life. But I don't tend to rely on symptoms to know that I've done a good job. (Physician 2, woman, 25 years qualified)*

The 'line in the sand' was influenced by a multitude of factors, with one including the extent of physician experience associated with assessing and managing children with idiopathic chronic pain. Experience occurred 'on the job' and learning typically involved interacting with other more experienced colleagues in the form of regular meetings and case conferences. Consequently, with increasing experience, physicians noticed themselves becoming more confident in treating youth with chronic pain and in turn, in reducing the number of diagnostic tests that they ordered. Whilst such results are inherently positive, this developmental trajectory in physicians' learning and comfort with risk means that there are clear timepoints during which many families are likely exposed to repeated, and possibly unnecessary, diagnostic testing. Moreover, Physician 16's use of the word 'brave' below implies that this learning process is endured in the face of physician fears and conflicts with their previous training that underscored the importance of caution and certainty in the diagnostic process. This process of learning may also be linked to physicians' earlier conceptualizations of chronic pain as a symptom without an underlying cause rather than the disease in and of itself.

> *My colleagues have been doing it a little bit longer, but we've got braver over time. So, I was already moderately brave about the headache, I don't do a lot of scans compared to other colleagues who don't do headache much. But we've noticed with this more difficult group of patients actually, we're also getting braver and doing fewer tests as time goes by, because I think you get a reflective memory for how to manage it. (Physician 16, woman, 26 years qualified)*

Physicians described that without an adequate mentorship and support system, such as is characteristic of a multidisciplinary team, they often felt isolated. Lack of experience and colleague support led some physicians to manage DU through continuing to see families past their 'the line in the sand'. Some physicians recognized the potential devastating impact this could have on youth and their families.

> *I think if you're not in a multidisciplinary team with these cases, it's very isolating… I think your gut feeling is to keep bringing them [youth with chronic pain] back, because you can't solve their problem but you don't know what else to do. And what I'm trying to make my colleagues realise is that perhaps just keeping on seeing them is possibly one of the worst things you can do. But I can totally see where they're coming from, it's exactly what I would have done a few years ago before we started working in this way." (Physician 14, woman, 24 years qualified)*

At times, managing uncertainty took an emotional toll on physicians. Strategies to manage uncertainty included sharing feelings of uncertainty with fellow physicians, although this process did not fully alleviate the uncertainty they experienced. Rather, it was described as a means to reduce the discomfort associated with experiencing uncertainty and to increase physicians' sense of comfort with managing a particular level of risk. Many physicians sought mentorship from more senior clinicians, who were more experienced both in terms of employment duration as a clinician and in treating patients with chronic pain specifically, as a way to ease uncertainty. Particularly for less experienced physicians, support from colleagues appeared to serve as an unofficial training experience and reassurance that they were not missing a more sinister cause for the pain.

> *Yeah, there's been weekends where I've been really thinking, oh my goodness, have I missed something, what if it's this, what if it's that, I've never come across this before… So how do I cope with that? I basically seek mentorship from a senior… So, I would go down and I would sit down and say, look, I'm really worried about this patient, I'm pretty sure that is chronic pain or medically unexplained, but I'm just worried I could be missing something, and go through the story. And on the whole, she's always been really reassuring and will say this*

> *definitely fits this picture, I've seen patients exactly like this. (Physician 10, woman, 16 years qualified)*

Individual patient and family factors

Explanations of physicians' willingness or ability to sit with uncertainty were complex and multi-factorial. Physician 3, below, describes how patient differences and individual thresholds were some reasons offered by physicians with regard to explaining their ability to sit within a particular level of uncertainty. For example, individual thresholds for severity of symptoms influenced whether a physician might seek out another colleague to share their uncertainty.

> *I'm very comfortable as a general pediatrician to sit with uncertainty. For me it's just about your threshold and the severity of the symptom. I suppose if the symptoms are within a realm that I'm comfortable with, then I'm happy to sit with uncertainty, if the symptoms are clearly worse then I'm going to struggle, because it's all about how severe the problem is, I think. I mean, I'm happy to sit with uncertainty, but I have my limit. (Physician 3, man, 14 years qualified)*

Memory and experience were important for both physicians and patients, influencing individuals' perceptions of where the 'line in the sand' could be drawn in terms of no longer seeking a medical explanation for the child's pain. Physicians recognized that youth and parents were influenced by their own previous medical experiences, including interactions with clinicians along that journey. The physicians' sense of patient and family beliefs, 'buy-in', and past medical experiences (e.g., a family member diagnosed with a brain tumor, experiencing a misdiagnosis) led them to continue investigations *beyond* the point of actually needing further diagnostic information. Thus, the ability and willingness of physicians to 'draw a line in the sand' was also influenced at a more individual level, with characteristics and beliefs of each youth and their family influencing physicians' decisions about when to halt ordering diagnostic tests to identify a 'cause' for the pain.

And, unfortunately, the way the family are and their belief in you and how the consultation has gone, does affect probably what investigations you order. So, if they're dead set on the fact that this is definitely cancer, that might push you towards doing more investigation, for example. (Physician 8, woman, 15 years qualified)

Physicians also recognized that the previous experiences that patients had before arriving to their clinic influenced both the patients' alignment with the physician as well as their understanding and acceptance of the diagnosis. However, physicians observed that this alliance was hampered for other patients and families in situations where many clinicians had been seen and multiple explanations of pain had been previously provided. Indeed, this was seen as resulting in mistrust in the medical system. In such situations, this mistrust due to past experience invariably affected 'buy in' and alignment with physicians. Misalignment of agendas, such as when families continue to believe that an underlying cause for the pain has yet to be found, was seen as a major barrier to moving forward with treatment. When a discrepancy in views occurs, how this is negotiated may have implications on patient acceptance of a diagnosis, treatment, and general trust in the medical system.

And I think it depends what their experience has been before, because I think if you get someone quite early on in the process then you are more likely to get a family that are going to be amenable to your way of thinking about it and buy into that explanation of chronic pain and not carry on thinking that they need investigations into that. But if you get somebody that's had this problem for quite a long time and they've had lots of different opinions from different doctors, and that's really difficult and then they lose trust and you've completely lost them then. (Physician 7, woman, 17 years qualified)

Additionally, this once considered definitive 'line in the sand' shifted according to physician perceptions of the severity of fears and anxieties held by individual families and the importance that the physician ascribed to managing these fears and anxieties.

But for some families they need a little bit more than that, and so they've got specific, focused concern and so in fact the risk-benefit is doing investigation to allay their anxiety around that particular thing as a line in the sand. (Physician 4, woman, 17 years qualified)

Perceived reassurance of diagnostic investigations

Physicians described, at times, adopting a cost-benefit analytic approach, such that they occasionally used diagnostic tests to curtail further, more invasive procedures. In an endeavor to prevent potential serious and unnecessary damage to a child, such as undergoing surgery, physicians reported ordering further diagnostic testing for the sole means of assuaging family or patient worries of a sinister cause of the pain. Yet, ordering further investigations was reported to be contradictory to physicians' professional judgment and held the explicit expectation that nothing would be found. Indeed, this induced considerable discomfort in physicians. However, when benefits were perceived to be possible, such as reassuring the family and preventing invasive procedures, discomfort with acting against clinical judgment was perceived to be the more comfortable option and could be described as the lesser of two evils.

> *And then occasionally you meet people that are a little bit more entrenched in their views. So sometimes I have to in those situations done extra scans and things like that, like MRIs, just to say, look, there really is nothing, you can see there, we're not going to go and open you up to look at your tummy, you know this has to stop here sort of thing. (Physician 7, woman, 17 years qualified)*

Physicians perceived families to be reassured by diagnostic testing and the return of test results that indicate the absence of pathology. As expressed by Physician 15 below, 'normal' or negative test results were believed by some physicians to reduce anxiety in children and families with regard to reinforcing the idea that the pain was not caused by something more serious.

> *It can actually be reassuring to have a normal test of some sort because it takes that worry [that something more sinister can explain the pain] away. It doesn't necessarily always cure the pain but it takes that worry away. (Physician 15, man, 24 years qualified)*

In direct opposition to the perception of the reassuring and necessary nature of diagnostic investigations for families, a small number of physicians recognized the potentially harmful

nature of unnecessary diagnostic tests for families in terms of exacerbating existing levels of patient anxiety and uncertainty about the cause of the pain.

> *I am aware that ordering investigations is not always reassuring for the patients, in fact it can make them feel that actually you do think there's something wrong, and then there's more anxiety leading up to the tests. And then when you've got the result it's sort of are you sure, you obviously thought this was really bad because you had to order this. On the other hand, you have to sort of do it on a patient-by-patient basis. (Physician 8, woman, 15 years qualified)*

Furthermore, some physicians strongly reported that such tests were a necessary component to achieving family 'buy-in' to a chronic pain diagnosis. Until all possibilities had been ruled out, family beliefs about other diagnoses would remain a barrier to patient-physician communication and hamper any progress in terms of chronic pain treatment. This suggests that extraneous testing was seen by some as a necessary step in the direction towards initiating chronic pain treatment, providing a stark contrast with other physicians who worked hard to avoid such testing in their practice. This might also reflect differences in physician perceived self-efficacy in their ability to generate patient buy-in through effective communication.

> *My personal view is that there's no point in trying to counsel them in the management of chronic pain until you've ruled everything else out, because they're just hanging on that diagnosis and won't really listen to you. (Physician 2, woman, 25 years qualified)*

The broader social context and implications

The decision of whether to conduct further investigations for potential 'causes' for the pain involved balancing a number of complex factors such as the 'need' for investigation and/or reassurance, the invasiveness of a test, and the broader societal implications, such as costs to the medical system. Physicians described following a stepped care approach according to the nature and invasiveness of a test. At times, physicians compromised with families in terms of deciding where to draw their 'line in the sand' with regard to deciding to stop ordering investigative tests. This was often presented as a compromise between the physicians' desires to stop testing, costs

to the family and healthcare system, and individual desires of the child and their family. Often ordering medical tests were used by physicians as a tool to align with families, even when they believed that these tests were medically unnecessary.

> *I think tummy aches, again might do a blood test and a urine test, and in some cases might go as far as an ultrasound scan, on the basis that it's not invasive, non-painful, relatively cheap thing to do and often is one that you can easily draw a line under with the families. I try really hard to avoid more expensive, more invasive imaging if I possibly can… I guess in my mind I'm balancing up not wanting to propagate the medical belief, but also only considering something ... because I think sometimes we do order tests even though we know that they're going to be normal, so you are using the test as a way of drawing a line in what you're going to do… (Physician 14, woman, 24 years qualified)*

Interestingly, some physicians perceived that unnecessary medical investigations could be financially beneficial for the health care system if they prevented families from continuing to present for further more costly investigations.

> *You could argue, yeah, don't do an MRI, that costs the health service, but if it stops that person keep presenting and is also a saving as well, if it stops loads of medicines being prescribed, then there's a saving. So, I suppose it's like balancing that up in your mind. (Physician 7, woman, 17 years qualified)*

Some physicians felt reassured by the number of clinicians that patients had previously seen and the amount of investigations completed, prior to the youth being in their care. Previous consultations with colleagues served to dilute this sense of individual responsibility with regard to managing a young person with idiopathic chronic pain. Physician 10 below acknowledges the wider social context in which uncertainty is experienced and managed.

> *...because they've seen so many other people before me, generally rheumatologists in [city] or neurosurgeons and had every scan under the sun, I feel really reassured by that and it's quite clear and barn door [obvious] to me, I think, that there doesn't need to be any worry about missing something and it's chronic pain. (Physician 10, woman, 16 years qualified)*

Physicians' experiences and beliefs about uncertainty and how this might affect the patient-physician relationship influenced the extent to which physicians shared their uncertainty

with families. For some, sharing uncertainty with families was considered to be risky and to undermine their competence as physicians with families. Others felt confident sharing their uncertainty with families, claiming that sharing reduced the power differential between physician, patient, and family. For these physicians, uniting with the family in the face of uncertainty was perceived to demonstrate humanity and increase the therapeutic alliance between the physician and family.

> *I think people often worry when they're new to this [managing children with unexplained pain] and with the new consultants, that if you demonstrate uncertainty that you undermine a family's confidence in you. But I don't think that's the case, I think that what happens is with uncertainty I think they respond to you as a human being and as a professional and they respect the honesty, and they don't feel patronised, I think, by that, I think they feel more like it's a team effort. (Physician 11, woman, 19 years qualified)*

Discussion

This study explored physicians' experience of DU regarding the assessment and management of chronic pain in youth. Through the theme 'drawing a line in the sand', physicians described how they manage uncertainty in their role, and the complex factors involved in their decisions of whether to continue or cease diagnostic testing. Even after a 'line in the sand' had been drawn, this point after which no further investigations would be sought was likely to shift depending on multiple factors, which were highlighted through four subthemes: physician training, experience, and mentorship; individual patient and family factors; perceived reassurance of diagnostic investigations; and the broader social context and implications.

Degree of experience and stage of training were key dimensions of physicians' experience of uncertainty. Many physicians reported receiving no formal training in chronic pain and described all of their learning as occurring 'on the job'. This highlights important gaps in

pain education, which has been found to be inadequate both in the UK and internationally (Bhatia, Brennan, Abrahams, & Gilder, 2008; Briggs et al., 2015; Hurley-Wallace, Wood, Franck, Howard, & Liossi, 2019; Shipton et al., 2018). Physicians discussed incongruence between their medical training, which focused on diagnosing and "fixing" a problem, and their current role in managing chronic pain in youth, which required supporting adaptive functioning despite pain. A lack of training was evident through physicians' descriptions of having to redefine their aim of care and turn to more experienced colleagues to learn about managing chronic pain. However, more experienced physicians may not necessarily be more knowledgeable regarding chronic pain and may have received training at a time when pain was even less understood. If physicians were not part of a multidisciplinary team, which could provide this type of mentorship, they described feeling isolated in managing DU and also continuing to see families past the 'the line in the sand'. A lack of training in pediatric chronic pain likely drives physicians' perceptions of its etiology. Physicians expressed not subscribing to the notion and conceptualization of chronic pain as being the disease in and of itself. The communication of this belief to families may contribute to uncertainty among youth and their parents (Neville et al., 2019).

While some physicians perceived sharing their uncertainty as being beneficial, research shows that this belief is not shared by families. Indeed, perceptions of clinician uncertainty can fuel DU in youth and parents, and can lead to mistrust in clinicians and the medical system (Neville et al., 2019). Physician intolerance of uncertainty has been found to be positively associated with diagnostic action (Schneider et al., 2010). In the current study, physician experience was noted to influence their actions; physicians described becoming "braver" over time and ordering fewer tests. This parallels findings in other areas of medicine. In a recent

vignette study, more experienced emergency department physicians reported less risk averse decision making (ordering fewer tests, offering fewer treatments, and making fewer referrals) (Lawton et al., 2019). Unfortunately, the consequence of this 'on the job' learning and gradual development of tolerance of uncertainty, is that youth are repeatedly exposed to potentially unnecessary and harmful diagnostic testing (e.g., MRI, ultrasound, x-ray, endoscopy). A retrospective study of medical records of youth seen in a chronic pain clinic found that 15% of youth underwent surgery due to their pain problem prior to referral to the pain clinic, which resulted in no change to their pain (Cucchiaro et al., 2017).

The 'line in the sand' was also influenced by physicians' perceptions of individual parent and youth characteristics, including severity of symptoms. Perceptions of parent and youth DU influenced physicians' decision making. The nature of idiopathic chronic pain in terms of its limited improvement of symptom severity, lack of underlying pathology, and use of non-pharmacological treatments (Fisher et al., 2018; Friedrichsdorf et al., 2016; Lynch-Jordan et al., 2014), make certainty difficult to attain. Many physicians believed that diagnostic tests would serve to reassure and assuage family anxiety that a sinister cause of the pain had been missed, even if the pediatrician expected tests would not indicate pathology. Physicians described taking a stepped care approach to investigations, according to the nature, invasiveness of a test, potential for harm, and need to align with the family. Nevertheless, unnecessary testing increases healthcare expenditures and carries risks of false positives and iatrogenic injury (Simpkin & Schwartzstein, 2016). Further, continuous negative test results and prolonged time with a lack of definitive answers may lead to increased psychological issues and distress for youth with chronic pain and their families, and exacerbate their uncertainty and fears that something is still being missed (Neville et al., 2019). Thus, where many pediatricians reported ordering further tests to

alleviate uncertainty around a diagnosis of idiopathic chronic pain, this may have the unintended opposite effect. Understanding the impact of unnecessary diagnostic testing is also important given that patients' wellbeing declines while waiting for chronic pain treatment (Lynch et al., 2008; Palermo et al., 2019). Only a minority of physicians acknowledged that unnecessary diagnostic investigations may cause psychological harm, pointing to the need for improved pain education for pediatricians. Physicians acknowledged the impact that their patients' past experiences with the medical system (e.g., misdiagnosis) had on their ability to 'buy in' to a chronic pain diagnosis. The number and nature of the explanations for pain that patients and families were previously provided were seen to have a negative influence on their current diagnostic orientation. This is in line with previous literature showing DU is fueled by time spent waiting for a diagnosis, previous interactions with clinicians, and past experiences when a diagnosis had been missed (Neville et al., 2019).

In terms of further diagnostic testing, the lengths that physicians were willing to go to reflected their perceptions of a critical need to align with families. Misalignment in agendas between the physician and family was seen as a crucial barrier in treatment. Physicians' beliefs in, and reliance on, the 'reassurance' of tests may reflect a lack of self-efficacy and training in the communication and delivery of a diagnosis to patients, particularly of idiopathic chronic pain. How physicians explain chronic pain (e.g., pain neuroscience education), and tailor this explanation to the developmental stage of the child, has been identified as a critical area of future research (Robins et al., 2016) and may have an important impact on alignment of physician and patient agendas. The oral testimonies of patients with chronic pain revealed that a breakdown in communication between patients and clinicians led youth to perceive that clinicians did not

attend to their lived experiences, which resulted in them doubting that their clinicians were able to help (Nutkiewicz, 2008).

The current study should be viewed in light of limitations. Results are based on interviews with pediatricians in the UK and may differ geographically. While not expressed in the current interviews, in certain contexts, the 'line in the sand' described in these interviews may also be influenced by physician fears of litigation and socio-cultural and political factors. This should be explored in future research. Due to a recruitment process wherein pediatricians self-selected in to the study, the current sample likely represents pediatricians who have a particular interest in chronic pain and findings may thus even underrepresent the uncertainty physicians experience in assessing and managing pediatric idiopathic chronic pain. We are unable to report on additional context regarding the healthcare settings (e.g., typical pattern of care, how long physicians typically follow patients) of participants. Nevertheless, these physicians reported little to no formal training in pediatric chronic pain and are the most likely clinicians to encounter youth in the early stages of their pain problem (Cucchiaro et al., 2017). This is important, as these interactions leave a lasting impact on families and set the stage for DU at the point of entry into a chronic pain program (Neville et al., 2019). Future research is needed to understand the experience of diagnostic uncertainty among pediatric pain specialists and the potential impact of pain education on DU among physicians and other healthcare professionals. Notably, the current findings emphasize a need for improved pain education for pediatricians, access to multidisciplinary pain teams and formalised mentorship schemes.

The experience of DU is inherent to the role of pediatric chronic pain provider and invariably influences how DU is experienced by youth and their parents (Neville et al., 2019). Given the widespread problem and economic toll of pediatric chronic pain (Groenewald et al.,

2014) and potential consequences of delayed treatment (Lynch et al., 2008), understanding the factors involved in DU for youth with chronic pain, their parents, and clinicians is crucial. These physician accounts showed that, in the face of DU, physicians order more tests to reassure *themselves*, in addition to youth and their families. In this way, the management of DU may lead to potentially unnecessary and harmful diagnostic investigations in youth and longer wait times for a diagnosis, which can have devasting consequences for youth's health, trust in the medical system, and pain outcomes (Lynch et al., 2008; Neville et al., 2019). Thus, understanding DU among clinicians has critical implications for harm reduction. Recent development of a new classification system for chronic pain as part of the Classification of Diseases (ICD-11) offers opportunity for consistency in language and internationally recognized labels for children's pain. Future research is needed to examine optimal ways in which physicians can deliver a diagnosis and communicate with youth and their parents about chronic pain. This messaging is likely influenced by physicians' own uncertainty and invariably influences DU of patients and families.

Table 3.1
Sociodemographic Characteristics of the Sample

Physician	Gender	Number of Years qualified as a physician	Clinical Setting
1	Woman	28	Rheumatology
2	Woman	25	Chronic pain
3	Man	14	General pediatrics
4	Woman	17	General pediatrics
5	Man	23	Epilepsy
6	Woman	17	General pediatrics
7	Woman	17	Diabetes and metabolic disorders
8	Woman	15	Rheumatology
9	Woman	24	General pediatrics, neonates
10	Woman	16	General pediatrics
11	Woman	19	General pediatrics
12	Woman	19	General pediatrics
13	Woman	15	Community
14	Woman	24	Pain
15	Man	24	General Pediatrics
16	Woman	26	General Pediatrics and pediatric Neurology

Table 3.2
Semi-Structured Interview Schedule

Question number	Question and prompts
1	Can you tell us a bit about any training you have received about treating individuals with chronic pain? Prompts: a. What about pediatric pain specifically?
2	Can you please tell us about your experiences of treating children with chronic pain? Prompts: a. What kinds of patients have you treated? b. Can you walk us through the process of seeing a child with chronic pain and providing a diagnosis? c. How have these patients and their families managed the diagnosis and treatment?
3	Can you please tell us about what happens once a child with chronic pain arrives in your clinic at an initial assessment appointment? Prompts: a. Can you tell us about any referrals that you might make at this point? b. Can you tell us about any tests that you might administer?
4	How do you sit with uncertainty regarding diagnosing children with idiopathic chronic pain? Prompts: a. What is it like to diagnose a child with chronic pain in the absence of a test result? b. How does this experience fit with your training and understanding of what it means to be a physician?
5	How do you explain chronic pain to pediatric patients and their parents? Prompts: a. Why do you choose to explain pain in that manner?
6	What happens when you provide the diagnosis and it is not believed or accepted by children and/or parents? Prompts: a. How do you negotiate the ‘disagreement’?
7	Is there anything else that we have not talked about that you think is important for us to understand about clinician uncertainty in pediatric chronic pain?

CHAPTER FIVE: A Longitudinal Examination of the Interpersonal Fear Avoidance Model of Pain: The Role of Intolerance of Uncertainty

Abstract

Youth with chronic pain and their parents face uncertainty regarding their diagnosis, treatment, and prognosis. Given the uncertain nature of chronic pain, and high comorbidity of anxiety among youth, intolerance of uncertainty (IU) may be critical to the experience of pediatric chronic pain. This study longitudinally examined major tenets of the Interpersonal Fear Avoidance Model of Pain, and included parent and youth IU as key factors in the model. Participants included 152 youth with chronic pain (M_{age}=14.23 years; 72% female) and their parents (93% female). At baseline, parents and youth reported on their IU and catastrophic thinking about youth pain; youth reported on their fear of pain, pain intensity, and pain interference; and parents reported on their protective responses to child pain. Youth reported on their pain interference three months later. Cross-lagged panel models, controlling for baseline pain interference, showed that greater parent IU predicted greater parent pain catastrophizing which, in turn, predicted greater parent protectiveness, greater youth fear of pain, and subsequently greater youth 3-month pain interference. Youth IU had a significant indirect effect on 3-month pain interference via youth pain catastrophizing and fear of pain. The results suggest that parent and youth IU contribute to increases in youth pain interference over time via increased pain catastrophizing, parent protectiveness, and youth fear of pain. Thus, parent and youth IU play important roles as risk factors in the maintenance of pediatric chronic pain over time and may be important targets for intervention.

Introduction

The experience of pediatric chronic pain is marked by uncertainty. For youth with chronic pain and their parents, uncertainty regarding a chronic pain diagnosis and treatment may fuel beliefs that something serious was missed by clinicians and a search for the 'right' diagnosis (Neville et al., 2019). Meanwhile, parents of youth with chronic pain report living in a state of uncertainty regarding their child's condition and prognosis, their parental role, and the future, making it difficult to move forward with daily life (Jordan et al., 2007).

Intolerance of uncertainty (IU) is defined as a "dispositional incapacity to endure the aversive response triggered by the perceived absence of salient, key, or sufficient information, and sustained by the associated perception of uncertainty" (p. 31) (Carleton, 2016b). IU is a key component of excessive worry (Laugesen et al., 2003) and anxiety disorders in general (Carleton, 2016b; Dugas et al., 1998), and is conceived of as a transdiagnostic dispositional risk factor for the development and maintenance of fear and anxiety-related psychopathology (Carleton, 2016b). At the core of IU is a dispositional fear of the unknown (Carleton, 2016a), not unlike the fear many parents and youth report in their search to know the cause of their pain and whether it could be something more sinister (Neville et al., 2019). Individuals high, versus low, in IU interpret ambiguous situations as more threatening (Dugas et al., 2005), and IU has been postulated as directly and indirectly contributing to chronic pain (Asmundson & Carleton, 2008). Thus, youth high in IU may interpret the ambiguity of their pain experience as more threatening. Given the uncertain nature of pediatric chronic pain and the high comorbidity of anxiety among this population, IU may be critical to the experience of pediatric chronic pain. Nevertheless, IU has never been examined in this population.

Previous research has examined a variety of putative risk factors for fear-avoidance in chronic pain (e.g., anxiety sensitivity) (Asmundson & Taylor, 1996; Cappucci & Simons, 2015; Martin, McGrath, Brown, & Katz, 2007). IU may be particularly important for eliciting threatening pain-related cognitions, emotions, and disability in pediatric chronic pain. The Interpersonal Fear Avoidance Model of Pain (IFAM) is a prominent conceptualization illustrating bidirectional relationships between parent and youth psychological and behavioral responses that influence youth pain disability/interference over time (Goubert & Simons, 2014; Simons et al., 2015). Specifically, the model posits that when confronted with pain, youth may engage in catastrophic thinking that, in turn, drives pain-related fears and avoidance behaviour, ultimately resulting in pain disability/declines in functioning. Given that parental responses are critical to youth's experience of chronic pain, the model also illustrates the influence of parent pain catastrophizing, pain-related fears, and behavioral (e.g., protectiveness) responses on youth's pain-related cognitions, fears, behaviour, and disability (Goubert & Simons, 2014; Simons et al., 2015).

The current study is the first to longitudinally examine the roles of parent and youth IU as risk factors to core components of the IFAM (Goubert & Simons, 2014; Simons et al., 2015). Specifically, we predicted that, in a longitudinal study of youth chronic pain, 1) youth IU at baseline would predict 3-month pain interference through youth pain catastrophizing and fear of pain, and 2) higher parent IU at baseline would predict 3-month pain interference through parent pain catastrophizing, parent protectiveness, and youth fear of pain.

Methods

The current analysis is part of the Pain and Mental Health in Youth study (PATH), a larger Canadian longitudinal program of research investigating internalizing mental health

disorders in youth with chronic pain. The aims of the broader study are to examine a myriad of neurobiological, cognitive, behavioral, and social factors underlying co-occurring mental health symptoms and chronic pain. The current study was designed with unique and specific goals to examine parent and youth IU in the IFAM; thus, the current goals are distinct from the previously published papers pertaining to sleep disturbances, diagnostic uncertainty, and daily parent-child relationships (Neville, Griep, et al., 2020; Neville et al., 2019; Pavlova et al., 2020).

Participants

Participants included 152 youth with chronic pain and one of their parents recruited from a tertiary-level pediatric chronic pain program. Youth were eligible for the study if they were between 10-18 years of age and identified by a health care provider as having chronic pain (i.e., pain ≥ 3 months (Merskey & Bogduk, 1994b) without an underlying disease (e.g., cancer). Exclusion criteria for both the youth and parent included being unable to read/speak English, diagnosis of a severe cognitive disability, developmental disorder, schizophrenia spectrum or other psychotic disorders, or inability to access the internet.

Procedure

All study procedures were approved by the institutional health Research Ethics Board. Parents were first approached by a member of the pain clinic staff for permission to be contacted by the research team. Parents and youth were then contacted by a member of the research team to assess eligibility, provide further information about the study, and obtain informed consent. Parents and youth consented or assented using an online consent (or assent) form and completed baseline questionnaires using Research Electronic Data Capture (REDCap), a secure online data collection tool (Harris et al., 2009). Youth who were at least 14 years of age provided their

consent to participate and signed a consent form. Youth who were below the age of 14 years were asked to provide their assent and signed an assent form. All participating parents provided their consent to participate. Additionally, for youth below the age of 14, parents provided consent for their child to participate in the study.

At the baseline assessment and 3-month follow-up, parents and youth completed psychometrically sound self-report questionnaires using REDCap (Harris et al., 2009). Specifically, at baseline, parents reported on their own and their children's socio-demographic characteristics, as well as their own IU, catastrophic thinking about their child's pain, and protective responses to their child's pain. Youth completed measures assessing their IU, catastrophic thinking about their pain, and fear of pain, as well as pain intensity and interference. At 3-month follow-up, youth reported on their pain interference. Parents and youth each received a $25 gift card for each assessment in which they participated.

Measures

Socio-demographic Characteristics. Parents reported on their own and their child's age, gender, and race, as well as their annual household income.

Youth Pain Characteristics. Youth completed a commonly used pain questionnaire to measure child pain characteristics (Palermo et al., 2004). Specifically, youth reported the location of their body where they experience the most pain as well as how long their pain has been present in years and months. Pain frequency in the past seven days was rated on a 5-point Likert scale ranging from "not at all" to "daily." Youth reported their average pain intensity in the past seven days using a validated 11-point numeric rating scale (0 = "no pain", 10 = "worst pain possible") (Castarlenas et al., 2017; von Baeyer et al., 2009).

Youth Pain Interference. Pain interference was assessed using the Pain Interference Subscale of the Patient-Reported Outcomes Measurement Information System (PROMIS) Pediatric Profile-25 (Irwin et al., 2010). The 4-item subscale was rated using a 5-point Likert scale from "never" to "almost always". A total score of pain interference is obtained by summing responses. This total is subsequently transformed into standardized T-scores used for analyses. In the current sample, internal consistency for pain interference was good (α =.82 at baseline; α =.85 at follow-up).

Parent and Youth Intolerance of Uncertainty. Parents completed the 12-item Intolerance of Uncertainty Scale (IUS-12) (Carleton, Norton, & Asmundson, 2007) and youth completed the 12-item Intolerance of Uncertainty Scale-Revised (IUS-R) (Walker, Birrell, Rogers, Leekman, & Freeston, 2010). Both scales measure individual reactions to uncertain and ambiguous situations and the future (Carleton et al., 2007; Walker et al., 2010). On the IUS-12, each item (e.g., "I can't stand being taken by surprise") is rated on a 5-point Likert scale from "Not at all characteristic of me," to "Entirely characteristic of me". On the IUS-R, each item (e.g., "I can't stand it when things happen suddenly") is rated on a 5-point Likert scale from "Not at all like me" to "Entirely like me". On both scales, a higher total score indicates a higher level of IU. The IUS-12 has shown excellent internal consistency (Carleton et al., 2007). In the current sample, internal consistencies for the IUS-12 (α =.94) and IUS-R (α =.94) were both excellent.

Parent and Youth Pain Catastrophizing. The Pain Catastrophizing Scale-Child version (PCS-C) (Crombez et al., 2003) and the Pain Catastrophizing Scale-Parent version (PCS-P) (Goubert, Eccleston, Vervoort, Jordan, & Crombez, 2006) were used to assess catastrophic thoughts about child pain. The PCS-C and PCS-P capture trait catastrophizing. Each scale contains 13 items rated on a 5-point Likert scale (anchors 0 = "not at all," 4 = "extremely").

Items are summed to produce a total score. Lower scores indicate a lower level of pain catastrophizing. The PCS-C describes what a child might feel and think when they experience pain (e.g., "When I am in pain, I become afraid that the pain will get worse"). The PCS-P assesses how a parent might feel when his/her child is in pain (e.g., "When my child is in pain, I become afraid that the pain will get worse"). The PCS-C and the PCS-P have been previously validated in children with chronic pain and their parents and have demonstrated good validity and reliability (Crombez et al., 2003; Goubert et al., 2006). Parent and child trait catastrophizing have been linked to poorer child functioning and increased pain (Crombez et al., 2003; Guite, McCue, Sherker, Sherry, & Rose, 2011; Logan et al., 2012; Lynch-Jordan, Kashikar-Zuck, Szabova, & Goldschneider, 2013; Pielech et al., 2014). Internal consistency for youth pain catastrophizing ($\alpha = .94$) and parent pain catastrophizing ($\alpha = .90$) was excellent in this sample.

Parent Protective Responses to Youth Pain. Parents reported on their responses to their child's pain using the Protect scale of the Adult Responses to Children's Symptoms (ARCS) (Van Slyke & Walker, 2006). Using a 5-point scale ranging from "never" to "always," parents reported on the frequency with which they engage in various behaviors (e.g., "Give your child special privileges") when their child has pain. The ARCS has been shown to be reliable and valid in samples of youth with chronic pain (Noel, Alberts, et al., 2016). The current study used the ARCS scoring for a combined child and adolescent sample of youth with chronic pain, which uses 13 items (Noel et al., 2015). Responses are averaged to provide a Protect score, with higher scores indicative of greater parent protective responses. The ARCS protect scale had good internal consistency ($\alpha = .90$) in the current sample.

Youth Fear of Pain. The Fear of Pain Questionnaire, child report (FOPQ-C), is a 24-item self-report inventory used to assess pain-related fears and avoidance behaviors (Simons et

al., 2011). The FOPQ-C has two subscales: Fear of Pain and Avoidance of Activities. Each item is scored from 0 (“strongly disagree”) to 4 (“strongly agree”). Total scores are calculated by summing items. Higher scores indicate higher levels of pain-related fear and avoidance. The FOPQ-C is both highly reliable and has been validated for use in the pediatric chronic pain population (Simons et al., 2011). The Fear of Pain subscale (α=.94), Avoidance of Activities subscale (α=.91), and total FOPQ-C (α=.95) had excellent internal consistencies in the current sample.

Statistical Analyses

Descriptive statistics and correlational analyses were conducted using the Statistical Package for the Social Sciences (SPSS), version 24 (IBM Corp., 2016). Structural Equation Modeling was carried out using AMOS 25.0 (Arbuckle, 2014). Subsequent analyses consisted of cross-lagged panel models carried out using AMOS 25.0. The current sample size exceeds that required on the basis of a priori statistical power calculations. Power analyses with .80 power (α = .05) suggest a sample of 144 would provide 80% power to detect a small to medium effect size (f^2 = .11) (Soper, 2020). Analyses were initially adjusted for age and gender. In the initial model (Figure 1.1), baseline parent IU predicts baseline parent pain catastrophizing which, in turn, predicts baseline parent protective responses. The three variables, as well as baseline youth pain interference and intensity and youth’s IU, then predict youth’s baseline pain catastrophizing. All six variables then predict youth’s fear of pain which, in turn, predicts 3-month pain interference, which is also predicted by baseline pain interference. By adjusting for baseline levels of pain interference on 3-month pain interference, the model can be used to examine whether baseline variables predict change over time in pain interference. Most variables in the current study were interrelated; however, demonstrating the *incremental* predictive utility of each variable relative

to each other variable is important for understanding the unique role of each variable in our model. Accordingly, we initially included all possible paths to ensure that paths that were not the focus of the current study, but that were potentially statistically significant, were retained in the final model (Haynes & Lench, 2003).

Measures of goodness of fit included chi-square, ratio of chi-square to degrees of freedom, comparative fit index (CFI), and root-mean-square error of approximation (RMSEA). Generally, a $\chi 2/df$ less than 2 (Carmines & McIver, 1981), CFI values greater than .90 (Hoyle, 1995), and a RMSEA of less than .08 with a non-significant *p* of close fit (pClose) (Kline, 1998) indicate acceptable fit. AMOS cannot estimate indirect effects in the presence of missing data. Accordingly, missing data were singly imputed via regression imputation with Full Information Maximum Likelihood (FIML) estimation. Estabrook and Neale (2013) note that FIML most accurately estimates individual scores with missing data relative to other methods such as mean imputation. The FIML approach is generally acknowledged to be preferable to other methods for dealing with missing data, such as listwise deletion or mean imputation, as these latter approaches are more likely to yield biased estimates (Little & Rubin, 1989; Muthén, Kaplan, & Hollis, 1987; Schafer & Graham, 2002). A bias-corrected bootstrapping procedure was used to estimate confidence intervals of indirect effects. Bootstrapping is a non-parametric method based on resampling (5,000 in the current study) with replacement (Bollen & Stine, 1990; Shrout & Bolger, 2002). We present 95% confidence intervals as well as parameter estimates associated with the indirect effect. Specifically, we examine indirect effects of baseline parent and youth pain catastrophizing and IU, as well as parent protectiveness, on three-month youth pain interference via each baseline variable (see Figure 1.1). For clarity, in Figure 1.2, the paths included in the mediation analyses are bolded. On the basis of Wald tests, and in the interest of

parsimony, non-significant paths were removed in order beginning with the least significant paths (Preacher, 2006; Streiner, 2006). Following the deletion of non-significant paths, chi-square difference tests were conducted to ensure the trimmed model did not fit the data significantly worse as compared to the full model. Full details of this model trimming are available upon request.

Results

Missing Data

Variables had a range of 0.7% - 15.1% missing data (youth IU = .7%; youth pain intensity = 2.6%; baseline youth pain interference = 2.6%; youth catastrophizing = 3.3%; parent protective responses = 3.9%; parent IU = 3.9%; parent catastrophizing = 5.9%; youth fear of pain = 10.5%; follow-up pain interference = 15.1%). Estimation of missing data using FIML is only valid when data are missing at random (i.e., no systematic biases affect which participants have missing data). Little's (Little, 1988) Missing Completely at Random (MCAR) test confirmed that missingness was unrelated to any variable in our study: $\chi^2(236) = 258.85$, $p = .15$. Data can thus be viewed as missing at random for analyses.

Descriptive Statistics

Participants included 152 youth with chronic pain (M_{age}=14.23 years; 72.4% female, range=10-18 years) and one of their parents (93% female). Youth were enrolled from headache (65%), complex pain (34%), or abdominal pain (1%) clinics. Youth reported experiencing the most pain in their head (71.1%), muscles and joints (26.3%), stomach (17.8%), legs (15.1%), and chest (11.8%), or other location (e.g., mouth) (27%). At baseline, youth reported an average pain intensity level in the past week of 5.59 out of 10 (SD = 1.86) and an average pain duration of

3.21 years (*SD* = 3.17). Forty-eight percent (*n* = 73) of youth reported that they had experienced pain daily over the past week. Sociodemographic data are presented in Table 4.1.

Table 4.2 reports the means and standard deviations of the baseline and 3-month follow-up key variables. Correlations among key variables are reported in Table 4.3. Females reported significantly greater 3-month pain interference than males ($p < .05$). Older youth reported higher levels of pain interference ($r = .17$, $p < .05$), IU ($r = .17$, $p < .05$), and fear of pain ($r = .22$, $p < .01$).

Structural Equation Modelling

Our initial model (Figure 1.1) produced a good fit to the data: $\chi^2(3) = 6.033$, $p = .11$, $\chi 2/df = 2.011$, CFI = .992, RMSEA = .082, pClose = .224. The two FOPQ-C subscales (Fear of Pain and Avoidance of Activities) were highly correlated ($r = .65$, $p < .01$), and comprised the total Fear of Pain Scale score. When these subscales were tested separately, neither showed significant effects on 3-month pain interference; however, when tested as a total score, a significant effect on 3-month pain interference was found. This suggests that each subscale is likely explaining overlapping portions of variance in 3-month pain interference and, therefore, cancel out the effect of each other. Therefore, the total score of the FOPQ-C was used to capture both aspects of Fear of Pain: fear and avoidance. Youth age and gender were not significantly related to any outcome and were dropped from the model. Parent IU was not correlated with youth's pain interference or intensity, while parent pain catastrophizing was not correlated with youth pain intensity, interference, or IU. Parent protectiveness was only correlated with youth pain intensity. All non-significant correlations were trimmed from the model. Regarding regression paths, parent IU, parent pain catastrophizing, and parent protectiveness were not significantly related to youth pain catastrophizing. Youth pain intensity, parent pain

catastrophizing, and parent IU were also not related to youth's fear of pain. Finally, parent IU, parent pain catastrophizing, parent protectiveness, youth pain intensity, and youth IU were not related to 3-month youth pain interference. Therefore, these paths were removed from the model.

The trimmed model (Figure 1.2) showed an excellent fit to the data: $\chi^2(20) = 21.62$, $p = .36$, $\chi 2/df = 1.08$, CFI > .99, RMSEA = .023, pClose = .746. Greater parent IU predicted greater parent pain catastrophizing which, in turn, predicted greater parent protectiveness, and subsequently greater youth fear of pain. Youth pain intensity and pain interference as well as IU independently (i.e., controlling for each other) predicted increased youth pain catastrophizing. Youth pain catastrophizing, IU, and pain interference independently predicted youth fear of pain. Pain interference showed moderate rank-order stability. Over and above baseline pain interference, youth fear of pain predicted increases over time in youth pain interference. This suggests that youth's fear of pain may contribute to increases in pain interference over time, and that parent factors are linked to youth fear of pain.

A significant indirect effect was found from greater baseline pain intensity to greater 3-month pain interference via greater pain catastrophizing followed by fear of pain ($b = .027$, 95% CI [.010, .057], $p = .012$). A significant indirect effect also was found from greater baseline pain interference to greater 3-month pain interference via youth pain catastrophizing followed by youth fear of pain ($b = .100$, 95% CI [.06, .159], $p < .009$). Finally, a significant indirect effect was found for youth IU on 3-month pain interference via youth pain catastrophizing and fear of pain ($b = .132$, 95% CI [.078, .198], $p < .009$). These results suggest that youth pain intensity and IU may contribute to increases in pain interference over time via increased pain catastrophizing and fear of pain. They also suggest that pain interference maintains itself over time via pain catastrophizing and fear of pain.

Discussion

The current study is the first to longitudinally examine the IFAM (Simons et al., 2015) and include parent and youth IU as putative risk factors in the model. The current results supported the hypothesis that greater parent IU predicts greater parent pain catastrophizing, which in turn predicts greater parent protectiveness and youth fear of pain (i.e., fear and avoidance behaviors), and subsequently 3-month youth pain interference. Youth IU at baseline also influenced 3-month youth pain interference via indirect effects involving youth pain catastrophizing and fear of pain. Consistent with the IFAM, greater baseline pain intensity also predicted greater 3-month pain interference through greater pain catastrophizing and fear of pain. Thus, while tested among youth whose pain was already chronic, the results suggest that following a painful experience (pain intensity), the worsening of chronic pain begins with a cognitive appraisal of the pain as threatening (catastrophizing), followed by increased fear of pain (a fear response and avoidance behaviors), which can be expected to facilitate increased pain interference over time. Furthermore, per previous research and theory (Asmundson & Carleton, 2008; Carleton, 2016b), IU appears to serve as a risk factor in the current model; specifically, youth who were high in IU tended to appraise their pain with greater catastrophizing. The current results are consistent with previous research identifying IU as a transdiagnostic risk factor across various mood and anxiety disorders (Boswell, Thompson-Hollands, Farchione, & Barlow, 2013; Carleton, 2016b; Shihata, McEvoy, Mullan, & Carleton, 2016).

In research with adults, high IU has been implicated in generalized anxiety disorder (Khawaja & McMahon, 2011; Koerner, Dugas, Koerner, Dugas, & Dugas, 2008), social anxiety disorder (Boelen & Reijntjes, 2009), panic disorder and agoraphobia (Boelen & Carleton, 2012;

McEvoy & Mahoney, 2012), obsessive-compulsive disorder (Lind & Boschen, 2009), PTSD (Fetzner, Horswill, Boelen, & Carleton, 2013), and depression (Gentes & Ruscio, 2011). Comparatively less research has examined the construct of IU in children and adolescents; however, the extant research to date echoes the findings in adults. For example, high IU has been implicated in social anxiety in adolescents (Boelen, Vrinssen, & van Tulder, 2010). IU has also been found to be positively associated with worry in children (Fialko, Bolton, & Perrin, 2012; Kertz & Woodruff-Borden, 2013) and adolescents (Thielsch, Andor, & Ehring, 2015). Furthermore, high levels of IU have been shown to differentiate clinically significant levels of anxiety in children (Comer et al., 2009; Laugesen et al., 2003). An investigation into the associations between IU and health anxiety found significant positive associations between IU, anxiety sensitivity, and a range of anxiety symptoms (e.g., panic, separation anxiety, social anxiety, generalized anxiety, physical injury fears), further supporting the transdiagnostic role of IU in anxiety in youth (Wright, Lebell, & Carleton, 2016).

The current study is the first to introduce and demonstrate the role of IU in the experience of pediatric chronic pain. In the current study, youth IU was significantly correlated with youth pain interference at baseline and 3-month follow-up. Previous research has shown that high IU is associated with higher self-reported pain severity in unpredictable, acute pain experiences (Bélanger et al., 2017). In addition to its influence on acute pain, IU has particular implications for individuals affected by chronic pain, where the etiology of pain is often unknown, and where uncertainty is therefore especially salient. The current results suggest that IU may influence pain-related outcomes, in part by influencing other key pain-related constructs, such as pain catastrophizing. Individuals who are high, versus low, in IU have been shown to interpret ambiguous situations as more threatening (Dugas et al., 2005). Thus, for youth who are high in

IU, pain, which is often uncertain and ambiguous, may be perceived as more threatening. Catastrophizing, in turn, is associated with increased pain and pain-related dysfunction (Cunningham et al., 2014; Tran et al., 2015). The results underscore the importance of continued research designed to improve our understanding of how IU may influence the chronic pain experience in order to inform the refinement of treatment, perhaps emphasizing reducing IU in impacted youth.

With respect to our understanding of fear-avoidance models of chronic pain, comprehensive reviews of IU and relevant theoretical models have suggested that IU can be distinguished from other fear-related constructs such as anxiety sensitivity and fear of pain, in that the latter constructs are characterised by fear in response to a particular stimulus or object (Asmundson & Carleton, 2008; Carleton, 2016a, 2016b). In contrast, fear experienced as a consequence of IU stems from the absence of stimuli or information (Carleton, 2016b). Moreover, previous research has found that the associations between IU and emotional disorders persist even when controlling for other dispositional risk factors such as anxiety sensitivity, neuroticism, and negative affect, pointing to the unique role of IU in psychopathology (Boelen & Reijntjes, 2009; McEvoy & Mahoney, 2011) and, by extension, theoretical models that seek to explain the intersection of psychopathology and chronic pain.

In the current study, greater parent IU predicted greater parent pain catastrophizing and protectiveness, which, in turn, predicted greater youth fear of pain (fear and avoidance behaviors) and greater 3-month youth pain interference. The current results are consistent with recent investigations of the IFAM (Simons et al., 2015). Specifically, Simons and colleagues' (Simons et al., 2015) found that parent behavior (parent protectiveness, minimization responses, and avoidance) directly influenced child avoidance behavior, which subsequently predicted child

disability. Parent distress (fear and catastrophizing) also indirectly influenced child avoidance behavior through parent behavior (Simons et al., 2015). The current study did not investigate parent pain-related fear, through which the IFAM posits parent pain catastrophizing influences protective behavior. Parent pain-related fear should be included in future longitudinal examinations of the IFAM. Further, inconsistent with the IFAM, parent pain catastrophizing was not significantly related to youth pain catastrophizing in the current study. In several recent studies parent protectiveness assessed at one point in time was not associated with youth chronic pain outcomes (Law et al., 2017; Poppert Cordts et al., 2019; Van der Veek et al., 2012). Rather than directly impacting youth pain outcomes, our results suggest that, consistent with the IFAM, parent protective responses may influence youth behaviors, which subsequently influence youth pain. Recent evidence also points to the need to assess parent behaviors using ecological momentary assessment. Increased parent protective responses measured daily across 7 days has predicted higher youth pain unpleasantness, pain interference, and pain intensity (Neville, Griep, et al., 2020). Parent protective responses, which have been found to be dynamic and variable from day to day (Neville, Griep, et al., 2020), may not be adequately captured by the static measure used in the current study.

The current study has limitations that also provide directions for future research. First, although the analytical model is in line with the IFAM and pain interference was assessed longitudinally, most variables were assessed at a single time point. Thus, strong conclusions about the directions of effects cannot be drawn. Indeed, as proposed in this model, some effects are likely bidirectional. Prospective longitudinal analyses are needed to provide an ideal test of the current model. In order to prospectively examine the chronification of pain, future research should investigate first onset pain complaints or surgery/injury models to investigate how early

factors influence the transition from acute to chronic pain. Furthermore, the current study investigated pain interference rather than functional disability, which is the core outcome proposed in the IFAM. The current measure of pain interference was chosen as a pain-specific patient-oriented measure (DeWalt, Rothrock, Yount, Stone, & Group, 2007), which has been used extensively in the pediatric chronic pain literature (Neville et al., 2018; Noel et al., 2018; Noel, Wilson, et al., 2016). Also, as stated previously, the FOPQ-C includes both fear of pain and avoidance behavior subscales. Based on the high correlations between these subscales, fear of pain and avoidance behavior subscales were aggregated into a single total fear of pain scale rather than assessed separately as the IFAM proposes. Finally, consistent with studies of tertiary pediatric chronic pain programs, the current sample was predominantly white and of high socioeconomic status, and most parent participants were mothers. Future research should also investigate how fear avoidance constructs may differ between youth with comorbid and non-comorbid pain conditions.

The current results highlight several potential targets of intervention for youth with chronic pain. In support of cognitive behavioral therapy that is already used for youth with chronic pain (Fisher et al., 2018), youth cognitions (catastrophizing) and behavior (avoidance) were identified as key components in the maintenance of pediatric chronic pain. Given that youth IU was found to be a risk factor in the current model, other transdiagnostic cognitive-behavioral treatments targeting IU may also be warranted. Among adults, IU has been targeted in transdiagnostic treatment (Boswell et al., 2013) using the Unified Protocol for the Transdiagnostic Treatment of Emotional Disorders (UP) (Ellard, Fairholme, Boisseau, Farchione, & Barlow, 2010; Farchione et al., 2012). Among individuals with anxiety disorders (generalized anxiety disorder, social anxiety, obsessive compulsive disorder, panic disorder,

posttraumatic stress disorder), Boswell and colleagues found a decrease in IU over the course of UP treatment (Boswell et al., 2013). Moreover, greater reductions in IU were associated with reduced anxiety and depression symptom severity (Boswell et al., 2013). The Unified Protocol for Transdiagnostic Treatment of Emotional Disorders in Children and Adolescents (Ehrenreich-May et al., 2017) may offer a new treatment option for youth with chronic pain. The current results and the high comorbidity of anxiety disorders among youth with chronic pain (Coffelt et al., 2013; Noel, Groenewald, et al., 2016) suggest that targeting underlying transdiagnostic factors, such as IU, in treatment may be warranted and effective.

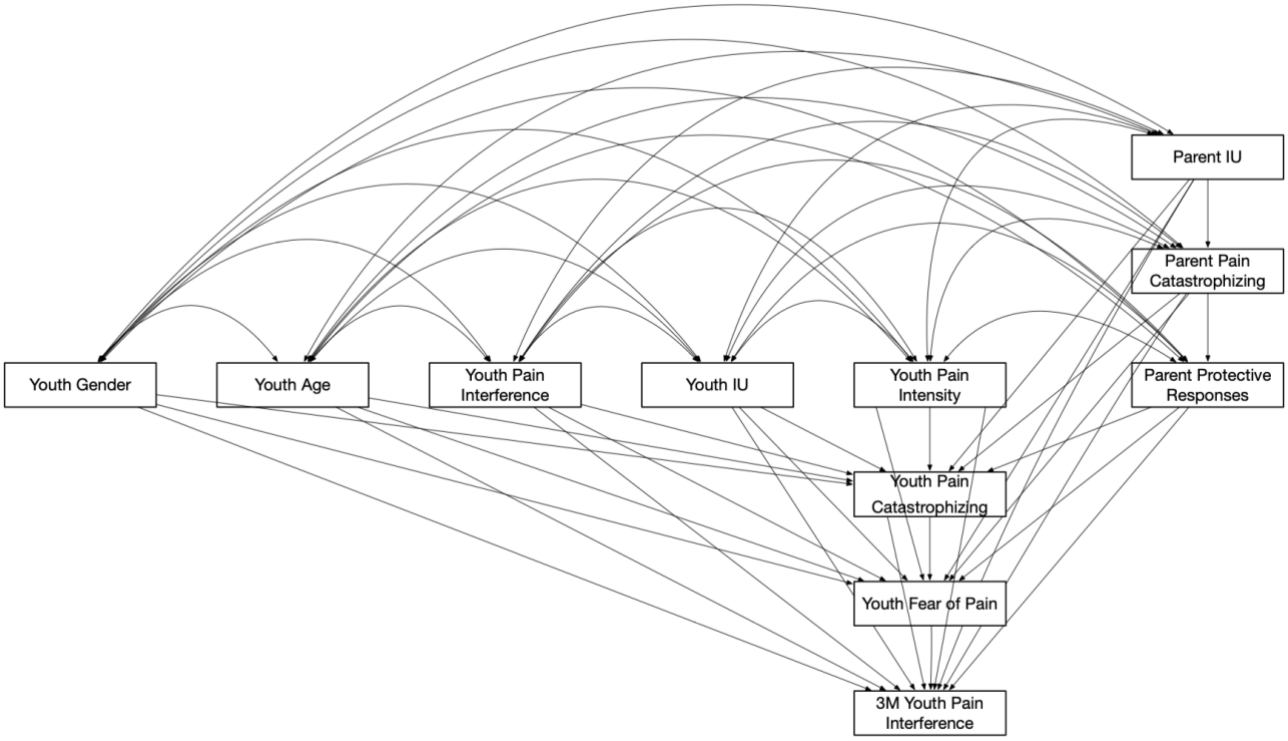

Figure 1.1 Nontrimmed model of parent and youth intolerance of uncertainty (IU) as risk factors in the Interpersonal Fear Avoidance Model of Pain. For ease of interpretability, error variances are not included in the figure. 3M pain interference was assessed at 3-month follow-up.

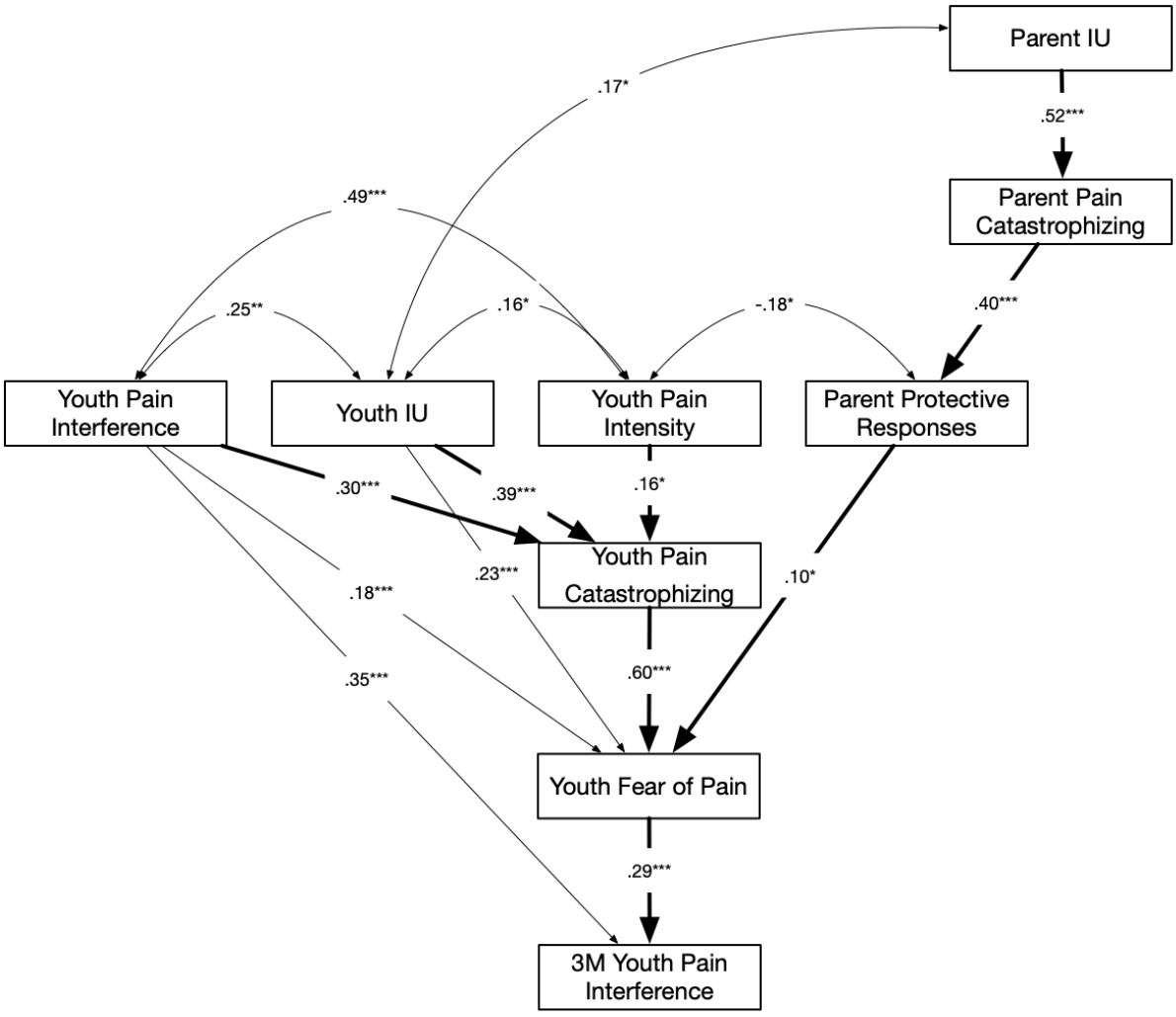

Figure 1.2. Trimmed model of parent and youth IU as risk factors in the Interpersonal Fear Avoidance Model of Pain. For ease of interpretability, error variances are not included in the figure. 3M pain interference was assessed at 3-month follow-up.

*$P < .05$; **$P < .01$; ***$P < .001$

Table 4.1

Socio-Demographic Characteristics of the Sample

Socio-Demographics	N=152
Youth mean age (SD), years	14.23 (2.25)
Youth gender (% female)	72.4
Parent gender (% female)	92.7
Relationship to the youth (%)	
Biological parent	97.3
Adoptive parent	2.0
Relative	.7
Youth race (%)	
White (Caucasian)	81.6
Two or more ethnicities	5.9
Other	3.9
Black	2.0
Aboriginal	1.3
Arab/West Asian	1.3
South Asian	1.3
Latin American	1.3
Filipino	0.7
Do not want to answer	0.7
Household income (%)	
<$10,000 - $29,999	4.6

$30,000 - $59,999	11.2
$60,000 - $89,999	12.5
More than $90,000	57.2
Do not want to answer	14.5

Table 4.2

Descriptive Statistics for Youth and Parent Variables

Variable	*M (SD)*	*N*
Baseline		
Youth pain intensity (0-10)	5.59(1.86)	148
Youth pain interference (PROMIS), T-score	55.24(9.16)	148
Youth IU (IUS-R)	25.43(11.09)	151
Parent IU (IUS)	22.61(9.28)	146
Youth Catastrophizing, (PCS-C), total	18.67(12.10)	147
Parent Catastrophizing, (PCS-P) total	13.92(8.41)	143
Youth fear of pain	30.90(20.57)	136
Parent protective responses (ARCS protect scale), mean	1.39(.73)	146
3-Month Follow-up		
Youth pain interference (PROMIS), T-score	52.80(9.60)	129

Note. Youth pain interference possible T-score range = 36.7-74; IUS and IUS-R possible score range = 12-60; PCS-P and PCS-C possible total score range = 0-52; Youth fear of pain possible score range = 0-96; ARCS Protect Scale possible score range = 0

Table 4.3

Correlations Among Variables of Interest

Variable	1	2	3	4	5	6	7	8	9
1. Youth pain interference	-	.48***	.50***	.25**	.49***	.50***	.05	.17*	-.03
2. 3M youth pain interference		-	.25**	.25**	.39***	.43***	.15	.12	-.13
3. Youth pain intensity			-	.14	.37***	.32***	-.06	.10	-.17*
4. Youth IU				-	.49***	.53***	.18*	.02	.05
5. Youth Catastrophizing, (PCS-C), total					-	.77***	.15	.11	-.02
6. Youth fear of pain						-	.16	.13	.08
7. Parent IU							-	.51***	.34***
8. Parent Catastrophizing, (PCS-P) total								-	.37***
9. Parent protective responses (ARCS protect scale), mean									-

Note. *$P<.05$; **$P<.01$, ***$P<.001$. 3M pain interference was assessed at 3-month follow-up.

CHAPTER SIX: How the Clinical Encounter Shapes Diagnostic Uncertainty in Pediatric Chronic Pain

Abstract

Approximately one third of youth with chronic primary pain and their parents in a tertiary care pediatric pain clinic report diagnostic uncertainty, which is associated with poorer child pain outcomes. Uncertainty among youth and parents regarding a pediatric chronic pain diagnosis may fuel a search for an alternative diagnosis and is intricately tied to experiences of clinical encounters along the pain care journey. This is the first study to investigate components of the clinical encounter that influence diagnostic uncertainty among youth with chronic pain and their parents. Twenty-three youth and at least one of their parents who presented for an initial visit at a tertiary pediatric pain clinic with chronic primary pain participated. Initial intake visits were videotaped, and youth and parents participated in semi-structured interviews prior to, and several months following, their intake appointment. Transcripts of clinical encounters and pre- and post-interviews were analysed using interpretative phenomenological analysis. Analyses generated four themes: 1) Diagnostic uncertainty is a social phenomenon; 2) (In)validation of pain, the journey, and diagnostic uncertainty; 3) The (missing) link between origin story & pain explanation; and 4) The fragility of certainty. These themes illustrate that youth's and parents' experiences of diagnostic uncertainty are complex, dynamic, and shaped within clinical encounters. The actions taken and explanations provided by clinicians in the clinical encounter could heighten or lower diagnostic uncertainty. Clinician communication, including validation and invalidation, messages of certainty and uncertainty, elicitation of youth's and parents' pain origin stories, and delivery of a pain explanation, influence diagnostic uncertainty and could be targets for assessment, training, and intervention.

Introduction

Pediatric chronic pain affects 11-38% of youth (King et al., 2011). While the cause of pain is clear for some, many youth experience chronic primary pain without an obvious explanation or evidence of an underlying pathophysiological etiology, leading to experiences of uncertainty. Parent and youth diagnostic uncertainty, the perception that a label or explanation for a patient's health problem is missing or inaccurate, has been identified as a core issue in pediatric chronic pain requiring investigation (Pincus et al., 2018). Empirical data shows that diagnostic uncertainty is prevalent, affecting over a third of youth and parents, and is linked to worse youth pain, emotional wellbeing, and health-related quality of life (Neville, Jordan, et al., 2020; Tanna et al., 2020). Findings from these studies highlight the importance of understanding diagnostic uncertainty among parents in addition to children. Importantly, some youth and parents report diagnostic uncertainty despite also reporting they have received a clear diagnosis for their pain (Neville et al., 2019; Tanna et al., 2020). Qualitative investigations among youth and their parents have found that a diagnosis of chronic pain is often not sufficient to alleviate fears that there is a more serious and sinister cause of the pain not yet found (Jordan et al., 2007; Neville et al., 2019). For youth and parents who experience uncertainty related to a diagnosis of chronic pain, a relentless search for alternative diagnoses often ensues, which may include invasive medical procedures and can impede recovery (Jordan et al., 2007; Neville et al., 2019). The search for a diagnosis and explanation that fits for the youth and parent can often result in youth perceptions that physicians are unable to help them, contributing to youth's feelings of loneliness and fear (Nutkiewicz, 2008). Research is only beginning to understand the consequences of diagnostic uncertainty for youth with chronic primary pain.

Clinicians also experience diagnostic uncertainty in their role treating youth with chronic primary pain (Neville, Noel, Clinch, Pincus, & Jordan, 2021). A qualitative investigation of pediatricians who assess and treat youth with chronic primary pain demonstrated that pediatricians experience uncertainty regarding when to cease diagnostic testing, which is influenced by factors such as beliefs about reassurance and perceptions of the families' own uncertainty (Neville, Noel, et al., 2021). Diagnostic uncertainty, by definition and through previous qualitative investigations of children's and parents' lived experiences, develops in the social context of family-clinician interactions through which a label, diagnosis, or explanation for pain is offered.

Clearly, the clinical encounter plays an important role in the experience of diagnostic uncertainty. However, research to date has relied on the retrospective reports of youth, parents, and clinicians. Research examining how clinical encounters are described has been important in advancing this area of research, however, direct observations of what transpires within this social interaction have yet to be investigated. The aim of the current study was to qualitatively explore components of the clinical encounter that may contribute to the experience of diagnostic uncertainty for youth with chronic primary pain and their parents. Idiographic research exploring the personal experiences of children and parents as they navigate pain care is needed to develop an understanding of how diagnostic uncertainty unfolds over time and is influenced by chronic pain care. This study investigated the initial clinical encounter when youth and parents enroll in a tertiary level pain clinic with the aim to elucidate the elements of this interaction that shape diagnostic uncertainty. Further, the current study drew on in-depth, semi-structured interviews conducted before and several months after this clinical encounter to better understand youth's and parents' experiences of diagnostic uncertainty as related to this clinical interaction.

Methods

Study Design

This study employed interpretative phenomenological analysis (IPA) to gain a detailed understanding of how diagnostic uncertainty is shaped by the clinical encounter when a diagnosis of pediatric chronic primary pain is provided (Smith, 1996). This methodology was chosen due to its phenomenological, hermeneutic, and idiographic theoretical underpinnings. This study adopted a semi-structured interview design to elicit parent and youth narratives before and after receiving a diagnosis of chronic primary pain at a tertiary university-based pediatric pain clinic. The clinical encounter attended by the parent(s) and youth was also video and audio recorded and transcribed. A phenomenological approach to this data allowed for detailed examination of parents' and youth's own understandings of their lived experiences related to their chronic pain care.

Patient Engagement

This project used a patient-oriented and patient-partnered approach, engaging two people with lived experience as collaborators and equal members of the research team (Canadian Institutes of Health Research; Manafo, Petermann, Vandall-Walker, & Mason-Lai, 2018). Patient partners were engaged in refining the interpretation and presentation of themes, as well as co-authorship of the current publication. Themes were reviewed with patient partners to ensure knowledge translation was sensitive, relevant, and meaningful to people with lived experience.

Participants

The sample consisted of 23 youth ages 10-17 years (14 female; *M*age = 13.2 years) and at least one of their parents (21 biological mothers, 1 biological father, 2 adoptive mothers, 1 adoptive father) who presented to a tertiary university-based pediatric pain clinic in the United

States with a chronic primary pain problem between March 2003 and March 2006. The majority of parent interviews were completed with the biological mother. Two father-mother dyads participated together (one biological and one adoptive). Youth pain diagnoses included: 56.5% headaches, 34.8% myofascial pain (of any body part excluding headaches), 30.4% abdominal pain, 8.7% complex regional pain syndrome. Thirty percent of youth reported more than one pain type. The mean duration of pain prior to the clinic visit was 53.3 months (range = 3-144 months). Sixty-five percent of youth in this sample had consulted five or more different doctors for pain relief prior to the current pain clinic visit. Additional sociodemographic information are presented in Table 5.1.

The current qualitative sample was part of a larger multi-method study of 179 youth from this clinical population (Bursch, Tsao, Meldrum, & Zeltzer, 2006; Clemente, 2009; Clemente, Lee, & Heritage, 2008; Clemente, 2012; Meldrum, 2008, 2009; Nutkiewicz, 2008; Tsao et al., 2005; Tsao, Meldrum, Kim, Jacob, & Zeltzer, 2007; Tsao, Meldrum, Kim, & Zeltzer, 2007). Of the 179 youth who participated in the larger study, 28 parent-child dyads participated in pre- and post-intake interviews and consented to their interviews being stored in the UCLA Library Special Collections for future research. Of those, full transcript data (i.e., pre-interview, clinical encounter, and post-interview) were available for 26 youth. Three participants were removed from the current analysis due to diagnosis of another serious medical condition or neurodevelopmental disorder.

Procedure

All study procedures were approved by the institutional health research ethics board. At the time of booking the first clinic appointment, parents were asked if they were willing to be contacted about a research study. A member of the research team contacted permitting parents

and children to discuss the study and obtain verbal consent to participate. At the time of the first interview, the study was reviewed with parents and children and written informed consent and assent was obtained. Consenting children and parents participated in a 30-90 minute, semi-structured interview at their home one month prior to their first clinic visit. Follow-up interviews were scheduled 6-12 months post- clinic intake. Parent and child interviews were conducted separately by trained graduate students. The semi-structured interview guide discussed the onset and course of the pain problem, resulting difficulties in functioning, relationships with friends and family, experiences with doctors and pain treatments, and hopes and expectations for the future. Pre- and post-intake interviews and clinical encounters were audio and videotaped and subsequently transcribed verbatim by an experienced transcriptionist. The initial clinic visits were not transcribed in full. For example, the physical exam was not transcribed. The current analysis of the clinical encounter included the opening, diagnosis, and treatment portions of the clinic visit. These segments were considered critical in this analysis given the core elements of diagnostic uncertainty gleaned from previous research, such as the pain explanation youth and parents receive and recommendations for further investigations (Neville et al., 2019; Neville, Noel, et al., 2021).

Data Analysis

Data were analysed using interpretative phenomenological analysis in accordance with the analytic process outlined by Smith, Flowers, and Larkin (Smith, 1996; Smith, Flowers, & Larkin, 2009). IPA methodology allowed for a detailed examination of each clinical encounter and how this experience was influenced by youth's and parents' experiences of events of the past (elicited at the pre-interview) and impacted events of the future (elicited at the post-interview).

Interview transcripts were analysed case by case beginning with parent and child pre-intake interviews, followed by analysis of the clinical encounter, and finally post-intake parent and child interviews. Detailed notes of each interview and clinical encounter transcripts were collated in a chart depicting a narrative account of each child's journey from pre- to post-interview, including initial interpretations, developing themes, and quotations supporting the preliminary interpretations. In essence, the three time points (pre-intake, intake, and post-intake) were strung together into a narrative for each individual case [i.e., the youth and their parent(s)], while maintaining the youth and parent as separate individuals with their own perspectives and lived experiences. The aim of this stage of the analysis was to understand the content and complexities of each individual case, differences and similarities in the parent and child accounts and shifts across timepoints. For example, an aspect of this analysis focused on how beliefs about pain shifted from the pre- to post-interview. The clinical encounter was the central point of this analysis, with the aim of identifying important aspects of this clinical interaction that may have influenced the experience of diagnostic uncertainty among parents and youth. Pre- and post-interviews were analysed with a focus on understanding the youth's and parent's experiences and actions prior to and following the clinical encounter, as well as the degree of diagnostic uncertainty being experienced by youth and parents. Detailed notes were taken about what was significant and interesting within each individual case, including descriptive (e.g., key content, events, and responses), linguistic (e.g., particular use of language, tense, and metaphor), and conceptual (e.g., researcher interpretations and reflections on the data) comments (Smith et al., 2009). Themes developed from the first cases informed the analysis of subsequent cases. In keeping with the iterative process of IPA, as new themes were generated in later transcripts, earlier transcripts were reviewed. Latter stages of the analysis focused on identifying connections

between themes and patterns across cases. Clusters of themes were then organized into a table of superordinate themes.

Analyses were conducted by co-author A.N. Throughout the analysis, frequent debriefing sessions were conducted with co-authors M.N., I.C., M.M., and L.Z. who were familiar with the data (Shenton, 2004). Co-authors A.J., K.W., and J. D. C. reviewed the ongoing analyses and provided comments to A.N. (e.g., alternative interpretations). In the final written report, efforts were made to include quotes from a variety of participants while also illustrating the nuances of particular cases within each theme (Smith et al., 2009). Yardley's characteristics of sensitivity to context, commitment and rigour, transparency and coherence, and impact and importance, were carefully considered as markers of quality throughout the analysis (Yardley, 2000).

Results

An interpretative phenomenological analysis of the data generated four themes, which were: 1) Diagnostic uncertainty is a social phenomenon; 2) (In)validation of pain, the journey, and diagnostic uncertainty; 3) The (missing) link between origin story & pain explanation; and 4) The fragility of certainty. We will illustrate that diagnostic uncertainty is a dynamic process through the personal accounts of youth with chronic pain and their parents obtained prior to and following intake appointments at a tertiary level chronic pain clinic and from observations within the initial clinical encounter. Importantly, we will show that diagnostic uncertainty develops over time and throughout the entire journey to find a diagnosis, explanation, and treatment for chronic pain. This demonstrates the need to view social interactions within the healthcare system through a critical lens. Themes are presented with supporting verbatim quotes from youth, parents, and pain clinic physicians. Pseudonyms are used throughout to protect participant

confidentiality. Efforts were made to present excerpts from each point in time (pre-intake, intake, and post-intake) for individual cases (parent-child dyads) when possible.

1. Diagnostic uncertainty is a social phenomenon

Almost universally, youth and parents reported a lengthy journey with the medical system prior to being seen in the pain clinic. Youth and parents described this journey as being characterized by a lack of validation and understanding by clinicians, by stigma, and by conflicting explanations for their pain. These early interactions with clinicians in the child's pain care journey played a critical role in youth's and parents' responses to the diagnosis of chronic pain, sometimes received many years later. Below, Lowell describes how conflicting explanations throughout his pain care journey impeded his ability to trust in clinicians' explanations and led to lost hope and faith in the medical system. Notably, following intake at the chronic pain clinic, Lowell continued to pursue alternative explanations and treatments for his pain.

"I've been to a lot of doctors. Each one has said something different, and that's why it's so confusing… After the first three doctors and they'd say something different, I figured 'oh they're probably right'. But after a while it got old. I'm like 'oh they're wrong. It's just a waste of time'." (Lowell, age 16, back pain, pre-intake)

Often embedded within medical encounters throughout their journey were messages of uncertainty and contradiction – clinicians devalued and invalidated the pain, yet simultaneously addressed it as a sign of a more serious condition requiring further investigation.

"First [the pediatrician] was kind of like, 'Nah, you don't hurt that bad.' But I knew he was concerned because he sent [Damien] over to the neurologist that very day, so we went from one appointment to the next." (Damien's mother, pre-intake)

Youth and parents regularly described this type of interaction in which clinicians used language and actions that invalidated the child's pain. In this passage, Damien's mother specifically articulates the contradictory concern conveyed by the physician ordering further tests

and referring him to a specialist. Following this interaction, although Damien's mother described that the x-rays taken did not demonstrate pathology, she worried that Damien's pain "could be life threatening" and continued to search for alternative explanations: these are two core aspects of diagnostic uncertainty.

When youth finally arrived at the tertiary chronic pain clinic, the actions taken, and the explanations provided, by clinicians in the initial encounter could either heighten or lower diagnostic uncertainty. Messages of uncertainty embedded in clinical discussions strengthened youth's and parents' beliefs that an as yet unidentified cause for the child's pain could and would eventually be found. The following excerpt demonstrates how language in the clinical encounter can convey messages that the 'holy grail' of the cause for the child's pain would be found in the next clinical investigation and importantly, that infinite tests should be conducted until such an explanation was identified.

"There's like hundreds of tests. I'm sure that [the other physicians have] done like ten specific ones but maybe they didn't do the eleventh one […] Maybe it's something else. Maybe it's the bacterial overgrowth. Maybe it's still a parasite. So then we'll do these other tests, which is specifically for that parasite I'm worried about." (Physician speaking to Amber at intake)

In addition to ordering further tests, the physician here has implied that there are an unpredictable number of tests that *may* hold answers for Amber's pain and has possibly reinforced Amber's and her mother's conviction that they must continue to search for a cure until one is found. The language used by the clinician, such as the repeated use of the word "maybe", introduces doubt, uncertainty and worry that there could be a more sinister cause of the pain. This suggestion is particularly worrisome for Amber and her mother, given the already held beliefs and fears that they expressed in anticipation of this clinical encounter, which included possibilities of a cause of pain that could prove fatal for Amber.

"*I'm scared of finding out that what I have like could kill me." (Amber, age 10, abdominal pain pre-intake)*

"I just worry about there might be something that's worse than acid reflux. I mean, what do we do with that? Like maybe cancer or something. We don't know." (Amber's mother, pre-intake)

Following this clinical encounter, in which the clinician expressed further uncertainty, fuelling a situation in which diagnostic uncertainty was already developing, both Amber and her mother expressed continued uncertainty about the cause of the pain.

"That's my main concern, and that's my biggest worry, that it could be life threatening and they just don't catch it." (Amber's mother, post-intake)

Conversely, in other encounters, the language used by clinicians sought to ease uncertainty. As demonstrated in the excerpt from Nina's intake appointment below, the clinician's messages that pain in and of itself was the diagnosis, and that the ongoing pain did not equate to bodily harm, assuaged the family's uncertainty.

"I bet it was a hairline fracture initially. Which then you know healed. Then you have this area that's like primed to go. So all you have to do is give a knock to those nerves and they're tchoo! Off and running […] [Moving is] not harming your body…it's not damaging you. It's the pain signaling system […] It's not scary, it's just miserable to live with. And it's affecting your life." (Physician speaking to Nina at intake).

These messages of certainty and validation were particularly effective for Nina, since they were also tied to an explanation that fit for her in terms of her own understanding of her pain as originating from a possible fracture in her foot. However, messages of certainty were not effective or beneficial in terms of reducing diagnostic uncertainty in every case. Indeed, statements of encouragement and certainty such as, *"I have yet to see a kid who hasn't gotten better"* (Physician to Damien), while suggesting hope for pain relief, also opened the opportunity for doubt for youth and parents when the child and/or parent noted no improvements in the child's experience of pain. In other words, if their pain did not change for the better within a

short time frame, or if their pain worsened, youth and parents began to think perhaps this was not, after all, the appropriate clinic, diagnosis, and treatment for them.

Perhaps the most devastating consequences of diagnostic uncertainty experienced by youth and parents were abandonment, mistrust, and lack of support resulting from conflict in the clinical relationship. In Stella's case below, one physician terminated her care due to the family's continued search for alternative diagnoses and treatment from other clinicians. Stella's clinical encounter at the tertiary pain clinic followed seventeen previous spinal tap procedures consequent to a pseudotumor diagnosis, which proved ineffective in managing her chronic pain. The family's pain journey was one of developing mistrust in medical professionals, punctuated by the repeated invasiveness of the spinal taps.

"I told [the pain clinic physician] about [what the neurologist said], and she got very angry that I listened to [the neurologist] instead of her. She just said that she wouldn't be Stella's doctor anymore. She was upset because I was taking Stella to see other doctors. I said, 'I can't believe you can't understand the need for Stella to have a neurologist. These are headaches. Of course she has a neurologist. She has a pseudotumor'." (Stella's mother, post-intake)

Stella and her mother were unable to buy-in to the chronic pain diagnosis offered by the tertiary clinic, especially when the diagnosis was presented as unrelated to the pseudotumor diagnosis that had become the focus of their quest. Stella explains post-intake: "*We're still looking into if anything else is causing it…and so far, we haven't found anything."* Diagnostic uncertainty can prevent adherence to clinician recommendations and delay effective treatment for chronic pain, as described below by Stella's mother.

"Everybody says that to her- 'stop taking Vicodin'. I don't want her taking Vicodin. But nobody is giving her anything else, except deal with it […] it would be different if somebody had some kind of answers, something I could look up on the Internet and go, okay, that's why this is happening. Diagnosis X." (Stella's mother, post-intake)

Stella's journey demonstrates the lengths youth and parents are willing to go and the procedures and treatments they turn to in the process of finding a cause that fits for their pain.

2. (In)validation of pain, the journey, and diagnostic uncertainty

Validation and invalidation were critical aspects of each clinical encounter prior to enrollment at the pain clinic and at the initial pain clinic visit, particularly in terms of diagnostic uncertainty. Prior to arrival at the pain clinic, many youth reported consultations with multiple clinicians and an accompanying sense of invalidation of their pain. Such invalidation typically had eroded their trust and confidence in clinicians as these encounters continued.

"One doctor said it was my attitude ... I didn't let out enough emotions […] [Doctors] always think they are right, but they aren't all the time and they don't listen to the patient." (Cameron, age 14, headaches, pre-intake)

In Cameron's intake at the pain clinic, the physician elicited his understanding of previous explanations of stress-based pain offered by clinicians, which were explanations of stress, and his own beliefs; he responded: *"I think that part of [the headaches] could be because of my stress, but I don't think all of it is."* His response articulates the core of diagnostic uncertainty – the sense that doctors have not yet found the real cause of the pain. Although the physician in this encounter conveyed to Cameron *"your pain is real"*, they also trivialized his emotional experience by responding: *"It's not worth getting upset over"*. At the post-intake interview, Cameron shared the following:

"None of [the previous physicians] helped me. They made me worse. They always listen to my parents. None of them help me […] [the pain clinic physician] seemed a little more, 'I know what to do and I know how to fix it,' but I don't know if she does […] I don't know what is going on, but I'm scared." (Cameron, 14, headaches, post-intake)

Youth and parents often perceived the message that the *"pain is real"* as legitimization of the pain. However, as shown above, this was insufficient in terms of repairing broken relationships between youth and clinicians as the belief that they had been invalidated was so deeply entrenched in the experiences of youth. Cameron's experiences of conflicting

explanations, ineffective treatments, and invalidation by clinicians ultimately led him to believe that clinicians were aligned against him. When the clinicians did not validate the severity of his pain and his worries about a serious, as yet undiscovered, cause, his uncertainty persisted and his trust in the healthcare system further dissolved. The invalidation that he experienced from physicians denied him the opportunity to feel listened to, validated, and understood, which eroded his confidence in the clinicians' ability to help him. This, ultimately, led to him feeling disempowered and fearful.

In contrast, while Barbara (age 12, knee and abdominal pain) similarly described uncertainty and frustration as a result of a lengthy "*doctor merry-go-round"* prior to intake at the pain clinic, her experience there differed from Cameron's. The below excerpt demonstrates the physician's response after eliciting Barbara's pain journey.

"You've tried many many things. You've had many many tests. And you've had many many diagnoses along the way… I'm sorry the path has been so difficult to get to […] Certainly there's been a quest to find a cure here. That takes up a lot of time, energy, and effort. It's gotta be exhausting […] Did you need to go through the entire workup that you did? Yes, you did […] We had to make sure that nothing life threatening was going on." (Physician speaking to Barbara at intake)

Barbara's expression of her personal pain journey experience was met with validation that went beyond addressing the pain. The physician's response validated the necessity of Barbara's previous medical experiences, the emotional impact of her pain care journey, her fear that something life threatening was causing her pain, and her quest for a cure. In doing so, the physician validated the core components of diagnostic uncertainty. This understanding demonstrated by the physician was critical for Barbara's and her mother's eventual trust and buy-in to the physician's diagnosis.

"We were relieved because her problem actually was identified by a doctor. [The doctor] said, 'You're one of us. We've seen lots of patients like this before.' So all of a sudden we had a diagnosis, where we really hadn't had one before." (Barbara's mother, post-intake)

Barbara's mother's statement that they had not had a diagnosis before is noteworthy, given the lengthy journey that she and her daughter simultaneously described, which included receiving multiple diagnoses. What Barbara's mother is explaining here is that they now had a diagnosis that fit Barbara's pain experience and beliefs. These youth's and parents' experiences demonstrate that consistent validation in the clinical encounter is critical to ameliorate diagnostic uncertainty.

3. The (missing) link between origin story & pain explanation

Although children and parents might not understand the cause of the pain, most were able to narrate an origin story for their pain in their pre-intake interview. The clinicians at the tertiary pain clinic elicited this narrative by saying, "Tell me your story." For some, like Virginia, the origin was an identifiable physical injury or incident.

"I was walking my dog. She saw another dog. She ran towards it, and I went up in the air, just whap, down. I really went off my feet […] [The fall] might have triggered [the pain]." (Virginia, age 10, myofascial pain, pre-intake)

Virginia's experience encapsulates how the child's origin story, using their own language, could be interwoven with the pain explanation. After several x-rays, MRIs, and physicians seen without a test result indicative of pathology, Virginia was referred to the tertiary pain clinic, where she told her story and received the following explanation for her pain:

"You got pulled and fell on your face. Do I think your whole pain system got turned on then? Absolutely. Do I think the problem here is your brain learned really really well how to have the pain switch turned on and forgot how to turn it off? Absolutely." (Physician speaking to Virginia at intake)

The opportunity to narrate their pain origin stories, and to hear an explanation that linked these to their pain experiences allowed youth like Virginia to make sense of their experience and embrace the new explanation. Also noteworthy in the above passage, is the repeated use to the

word “absolutely”, which conveys certainty in the pain explanation and validation that the pain is real. Although Virginia was still experiencing pain at the time of her post-intake interview, she stated that she felt more in control of her pain. Her mother recalled that *“[the pain clinic explanation] was very clear and obvious and understandable.”*

In other cases, the link between origin story and pain explanation was less clear or lacking altogether. This was the case for Courtney, whose origin story included a sudden onset of headaches and dizziness, followed by the discovery of a cyst in her brain. When they were enrolled in the pain clinic, Courtney had undergone several invasive surgeries, which had not resulted in pain relief, and she and her mother were uncertain about the underlying cause of Courtney’s pain:

“It’s just it’s like the unknown stage. If you have something then you can cure it, but now you just don’t- you don’t know what it is but you know there’s something there.” (Courtney, age 14, headaches, pre-intake)

The explanation Courtney received at the tertiary pain clinic was that her pain was a learned response, with no critical discussion relating this explanation to her pain origin story:

“You like to do things really well I noticed. Kinda perfectly. You learned how to do pain really well. You’re good at pain,” (Physician speaking to Courtney at intake)

Courtney and her family therefore continued to search for “something physical” that would explain her pain and lead to a cure; and she continued to repeatedly undergo invasive procedures, including major surgeries. Courtney’s experience also highlights the dynamic aspect of diagnostic uncertainty: after each surgery, she experienced reduced pain and she and her mother often felt relief and increased certainty. However, after some time had passed, the pain, and the uncertainty, always returned.

“[No migraines] went for 4 or 5 months [after surgery]. The longest I’ve ever been. Like ‘cause all these [surgeries] happened within like a month each. So then we had the headaches over the

summer and I kinda just flipped out […] So they got an MRI of my head and the cyst was fine. I'm like, thank god. Okay. What's wrong?" (Courtney, age 14, headaches, pre-intake)

Helen's case illustrates the critical importance of language in the pain explanation and its relation to diagnostic uncertainty. Her origin story began with a fractured clavicle. At the tertiary pain clinic, the physician explained to Helen and her father that, although there was an original injury, the "meaning" of the pain had now changed. This explanation differentiated between the pain she experienced at the time of the original injury (acute pain) and the pain she was continuing to experience (chronic pain):

"Back when you had your fracture […] your circuit got turned on by something that was legitimately a problem because there was a bone coming way too close to that nerve. It turned on the circuit and the circuit never turned off." (Physician speaking to Helen at intake)

Helen's father's description of the significance of this pain explanation to him in terms of understanding the pain highlights the importance of language. Whereas previous physicians had used the term "nerve damage," leading Helen's father to believe she had endured a physical and permanent tear, this new explanation was explained in terms of function.

"Before we went to [the pain clinic] everybody just washed their hands of her and said there is nothing else we can do. What's nerve damage? What are you talking about nerve damage? I was thinking nerves were permanently torn or something. You know physical damage […] [The pain clinic doctor] explained that the problem- what nerve damage means is the nerves are telling Helen's brain that she is still injured. So they have to teach her nerves to tell the brain that she is healed. That made sense." (Helen's father, post-intake)

This explanation, however, did not appear to shift Helen's *own* understanding of her pain as being solely physiological:

"[My physiotherapist] found out that there is liquid of fluid in my left shoulder and that is what is causing some of the pain and so we are working on breaking that up." (Helen, age 16, migraines, post-intake)

4. The fragility of certainty

Even when each part of the clinical encounter appeared to go 'right', any recurrence, change, increased intensity or simple persistence of the pain exacerbated diagnostic uncertainty for both youth and parents. As Lowell's mother demonstrates below in her post-intake interview, diagnostic uncertainty was integrally tied to pain intensity and functioning.

> *Interviewer: "What led you to continue to seek out other answers?"*
> *Mother: "Because he wasn't getting better. He wasn't getting better."*

Our findings describe uncertainty as a dynamic and cyclical process greatly influenced by its social context. The experience of Damien below, beautifully depicts the fragile nature of certainty and the social context within which certainty and uncertainty are co-constructed. After their initial confidence in the diagnosis Damien and his mother had received at the tertiary pain clinic and following months of improvement, a new injury occurred and his pain suddenly worsened. At this critical juncture in Damien's pain journey, doubt resurfaced in a conversation with his pediatrician. The nature of diagnostic certainty is precarious and difficult to maintain.

"Still, you think, what if it's not [reflex sympathetic dystrophy]? What if we've still missed something? Even though everything we've read said it's this, we still have- like his pediatrician, a month ago, said, 'Are [pain clinic physicians] sure that's what it is?' […] Sometimes I just wish that there were more tests that they could do to prove- it's almost like, let's call it RSD because we don't really know for sure what it is." (Damien's mother, post-intake)

Ashley describes a similar experience. Her pain originated with a broken ankle during dance class; it radiated to her hip and prompted fears of some serious problem with her bones. After some improvement following her visit to the tertiary pain clinic, the hip pain recurred when she returned to dance classes and her diagnostic uncertainty resurfaced.

"In September it was just like a constant, but exaggerated uh not that sharp but it was just like this whole area was in excruciating pain. […] We just wanted to make sure it was nothing with the bones. […] I went to my orthopedist, did an x-ray, did an MRI and uh it showed that I had an overstretched tendon that wraps from like back here all the way around to the front […] and then I went to [physician at the pain clinic]." (Ashley, age 14, hip pain, post-intake)

Ashley's fears of permanent bone damage and need for tests to alleviate the anxiety are intricately linked to her pain journey. However, Ashley also returned to the pain clinic for support following her briefly resumed search for a cause of her pain, demonstrating the importance of a strong patient-clinician relationship, which has the power to mitigate diagnostic uncertainty.

As described through Ashley and Damien's experiences, the link between diagnostic uncertainty and pain improvement is complex. Other youth and parents expressed reduced uncertainty following the clinical encounter when their pain *did* improve. Nina stated that her worries about her pain *"had probably decreased"* after her visit to the tertiary clinic, *"because [the pain] is going away."* For her mother, Nina's complex regional pain syndrome seemed elusive:

"It's like dealing with a ghost, this condition… It's not like a broken leg, where they can fix you."(Nina's mother, pre-intake)

Nina's mother's use of the description "ghost" alludes to a pain condition that is always present and seen by some but not by all. Like a ghost, Nina's a pain condition seemed unexplained, disbelieved, and difficult to prove and understand. For Nina's mother, seeing Nina's pain improve in response to treatment reduced her experience of diagnostic uncertainty.

"From talking with [the pain clinic physician] and her reactions to Nina's positive reactions to-when [Nina] was able to get herself out of the pain event, made me understand more that it is about her. It is about Nina and what she has to learn how to do, and of all the medication in the world isn't going to do it. […] you can get on the other side of this, where six months ago I was really hoping we could, but I didn't know that we could." (Nina's mother, post-intake)

Discussion

This qualitative study sought to understand components of the clinical encounter that influence diagnostic uncertainty for youth with chronic pain and their parents. Four themes were

generated from examination of the initial clinical encounter when youth and parents were enrolled in a tertiary level pain clinic, as well as pre- and post-intake interviews. These themes were labelled: 1) Diagnostic uncertainty is a social phenomenon; 2) (In)validation of pain, the journey, and diagnostic uncertainty; 3) The (missing) link between origin story & pain explanation; and 4) The fragility of certainty. These themes demonstrate that youth's and parents' experiences of diagnostic uncertainty are complex, dynamic, and shaped within clinical encounters along the chronic pain care journey.

Just as chronic pain transpires over a course of time, so too does diagnostic uncertainty. The current findings suggest that diagnostic uncertainty occurs as a relational and evolving process that is co-constructed between youth, parents, and clinicians in every clinical encounter. As such, diagnostic uncertainty is often present before youth and parents are enrolled in care at a tertiary level chronic pain clinic. The current analysis replicates previous findings demonstrating that youth's and parents' journeys with chronic pain are characterized by invalidation, stigma, contradicting explanations for their pain, quests to find an explanation that fits, and mistrust in the medical system (Jordan et al., 2007; Neville, Jordan, et al., 2020; Wakefield, Belamkar, Litt, Puhl, & Zempsky, 2022; Wakefield, Zempsky, Puhl, & Litt, 2018). Youth's and parents' experiences along their, often lengthy, journeys prior to arriving at the pain clinic were critical to their responses in the current pain clinic interactions. Messages from clinicians were found to shape youth's and parents' beliefs about pain and search for a cause, which are core elements of diagnostic uncertainty; This included messages of uncertainty that were conveyed through language as well as through behaviours, such as referring to specialists and ordering diagnostic tests. Particularly when youth and parents already held beliefs that something serious could be causing their pain, the language and behaviours of clinicians that conveyed this could be true

served to reinforce diagnostic uncertainty rather than provide relief. Of equal importance were findings pertaining to messages of certainty. Clinicians conveying complete certainty that youth were the right fit for the chronic pain clinic, was not enough for youth and parents to buy-in to a diagnosis, and even resulted in fueling diagnostic uncertainty if youth's pain did not improve. Particularly, when youth had a learned history of clinician certainty *without* pain relief, statements of certainty and expertise from physicians were not enough to quell fears that something else was being missed and led youth and parents to further doubt clinicians. Alternatively, when the provided pain explanation fit for families, youth and parents perceived these messages of certainty as comforting and perceived clinicians as competent. Contradictory messages from clinicians, such as devaluing of pain and the youth's narrative followed by recommendations for further investigations, led to confusion for youth and parents and perceptions that their pain was not well understood. This is important given that patient-clinician communication is linked to patient adherence to pain treatment (Butow & Sharpe, 2013). Pain communication is highly complex and must be understood in a broader social context (Hadjistavropoulos et al., 2011). The meaningfulness of words is dependent on how they are both shared (e.g., verbal and nonverbal communication) and understood (e.g., interpreted and perceived) by the sender and receiver (Hadjistavropoulos et al., 2011). Further, patient-clinician interactions in a pediatric context often include parents, and thus, family dynamics. In the current analysis, some youth perceived the clinician and parents as aligned against them. Future research should seek to understand diagnostic uncertainty in a developmental and familial framework.

A critical factor in interactions with clinicians was validation, communication that an individual's experiences are understandable and legitimate (Linehan, 1997). Youth and parents had often experienced considerable invalidation of their pain prior to arriving at the tertiary level

clinic. These experiences of invalidation eroded youth's trust in physicians. The concept of validation is not well understood in pediatric chronic pain. In a recent topical review, Edmond & Keefe discussed validation as a continuum of levels with different types of responses providing differing 'doses' of validation (Edmond & Keefe, 2015). This may be related to the current findings demonstrating that statements such as, "Your pain is real", intending to legitimize the pain, were perceived by some youth and parents as validating. However, for other youth, these statements did little to repair their previous experiences of invalidation. This highlights the need for a detailed understanding of the nuances of validation in this population, including how it is expressed and received. In addition to understanding the timing and level of validation, the content of what is being validated is important. For example, in the current study, while physicians often validated the suffering and impact of pain on youth's functioning, in some clinical encounters the physician also provided validation that went beyond the pain and expressed understanding of their pain care journey and diagnostic uncertainty itself. These families perceived having received an explanation that made sense in their clinical encounter, suggesting that validation and acknowledgment of the worry that something serious could be being missed may be important in reducing diagnostic uncertainty. Indeed, an investigation of the clinical encounter among adults with chronic pain suggests that denial of uncertainty in the clinical encounter may have negative consequences in the patient-clinician relationship (Costa et al., 2022). Further, among adults with chronic pain receiving care at a pain rehabilitation clinic, individuals who experienced low validation and high invalidation reported higher pain interference and negative affectivity following treatment (Edlund, 2017). Validation and invalidation have been shown to enhance and disrupt emotion regulation, respectively, which may impact patient-clinician communication (Shenk, 2011). While research suggests that

validation skills training may improve patient satisfaction (Linton, Flink, Nilsson, & Edlund, 2017), it is not often specifically taught to clinicians. The impact of validation on emotion regulation, the clinician-patient relationship, and youth's and parents' processing of pain explanations in this population are important areas for future research.

The current findings suggest that youth's previous experiences and memories of clinical encounters, and own understanding of their pain, are important factors influencing pain communication within this clinical interaction. Emerging research is shedding light on the importance of pain education (Leake et al., 2019; Leake, Moseley, Stanton, O'Hagan, & Heathcote, 2021; Pate, Heathcote, Simons, Leake, & Moseley, 2020). Our findings suggest that this education will need to be targeted and specific to individual youth and families. Multiple factors within the clinical encounter are likely to influence whether youth and parents update their beliefs about pain when provided with new pain explanations, such as whether an explanation is linked to the youth's and parent's pain origin story. The conveying of expertise, a label, and an explanation for the pain was not enough for families to take up a new explanation for their pain, particularly after a pain care journey characterized by differing pain explanations. The new pain explanation needed to be corroborated with the patients' own personal experiences and in particular, their narrative for where the pain began. The clinical skill of interweaving youth's and parents' pain origin stories into the pain explanation helped youth and parents further understand the pain. Unfortunately, patient's narratives are often devalued in pain care or not elicited at all (Nutkiewicz, 2008). Eliciting children's personal narratives may allow physicians to provide a meaningful explanation in the context of their lived experience (Nutkiewicz, 2008). Relatedly, language within these pain explanations and broader clinical encounter was critical. The words parents and youth heard along their pain journey, such as

"damage" and "maybe", had implications in terms of their distress, their search for a cause, and their beliefs about pain. Research is needed to understand how clinician language can help youth manage uncertainty and make sense of their pain.

Many youth in the current study underwent several invasive procedures and treatments in pursuit of a cause for and relief of their pain, making the acceptance of a pain explanation unrelated to the diagnosis and treatments they had been pursuing inconceivable. A mega-ethnography of qualitative studies of adults with chronic pain documented the immense vulnerability individuals experience in deciding to end the quest for a cure, particularly after prolonged investment in this search; the decision involves a perceived loss of self-credibility (Toye, Seers, Hannink, & Barker, 2017). Toye and colleagues described that chronic pain healing journeys require validation of pain *and* the self (Toye, Belton, Hannink, Seers, & Barker, 2021). The authors draw conclusions about the importance of validating patients both through valuing their stories and providing meaningful explanations (Toye et al., 2021). In the current analysis, providing youth and parents the opportunity to narrate their pain journeys in the clinical encounter offered opportunities for both these types of validation, which served to reduce diagnostic uncertainty. The ability to validate the journey and link the pain origin story to the pain explanation, requires elicitation of the story in the clinical encounter.

Diagnostic uncertainty was found to be intricately tied to youth's functioning. When youth's pain and functioning improved following their clinical encounter at the pain clinic, youth were more likely to experience reduced diagnostic uncertainty. However, diagnostic certainty was found to be precarious and difficult to maintain, suggesting that diagnostic uncertainty should be assessed overtime and addressing it in treatment is likely to be an iterative process. In line with previous research, following intake to the chronic pain clinic, youth who continued to

experience diagnostic uncertainty also continued to pursue further investigations and alternative diagnoses for their pain (Neville et al., 2019).

These findings should be viewed considering limitations. Firstly, the current study examined the first clinical encounter when youth and parents were enrolled in a pain clinic between 2003 and 2006. Of course, the provision of a diagnosis often does not occur in one instance at a tertiary level pain clinic. It is an ongoing process that begins with the first clinicians seen by the child. As such, the process of a clinical encounter is invariably impacted by those that came before it. Research has yet to explore how diagnostic uncertainty unfolds in tertiary level care and its influence on youth's readiness for and responsiveness to pain treatment. While research has shown that diagnostic uncertainty is linked to worse pain, emotional wellbeing, and quality of life for youth, longitudinal research is needed to understand how diagnostic uncertainty drives these negative consequences and influences chronic pain treatment (Neville, Jordan, et al., 2020). Further, recent advancements in chronic pain care, such as the new ICD-11 diagnostic coding for chronic primary pain, are hopeful progressions towards reducing the differing labels and explanations provided to youth along their journeys and increasing evidence-based treatment pathways for chronic pain. Secondly, the current study data was composed of written transcripts of interviews and clinical encounters. Future research should investigate how other components of language (e.g., tone of voice) and nonverbal behaviour (e.g., silence, body language, facial expressions) play roles in the shaping of diagnostic uncertainty. Most notably, diagnostic uncertainty was gleaned from interviews before and after the clinical interaction at the pain clinic, rather than the clinical encounter itself. It was not possible to ascertain if diagnostic uncertainty was discussed in other clinical interactions at the pain clinic or elsewhere. However, in the clinical encounters in this analysis, diagnostic uncertainty itself was not specifically

assessed by physicians and was rarely explicitly expressed by the child or parent in the clinical encounter after an explanation was provided. Future research should consider that a history of conflictual and invalidating interactions within the medical system are likely to impact youth's disclosure of information in clinical interactions. The current study suggests that youth's and parents' pain origin stories, diagnostic uncertainty, and previous experiences in clinical encounters, should be assessed. Diagnostic uncertainty is experienced relationally and transactionally, within and between individuals. As such, future research might consider the use of sequential analysis and observational coding methods to understand the nuances and impacts of various, and often subtle, expressions of uncertainty.

Importantly, clinician-patient relationships are situated within broader systems and contexts, including organizational, cultural, societal, and political. Diagnostic uncertainty is likely influenced by pervasive societal beliefs about pain as a biomedical problem, a signal for harm, and a symptom rather than the primary condition. Marginalized youth and parents may experience more stressful pain journeys and be particularly vulnerable to pain-related toxic stress due to higher rates of pain dismissal, discrimination, and undertreatment of pain (Nelson et al., 2022). Given that the majority of youth and parents in this sample were white and female, the current analysis does not reflect the experiences of youth and parents from minority and marginalized groups who experience additional barriers, health disparities and discrimination (Hartford, Blume, Barry, Hauser Chatterjee, & Law, 2022; Mulchan, Wakefield, & Santos, 2021), which are likely to compound the mistrust and experiences of invalidation described in the current analysis. This is highly relevant given that diagnostic uncertainty is a relational phenomenon that is intricately connected with trust in the medical system. A deep understanding of how experiences of diagnostic uncertainty intersect with race, ethnicity, gender,

socioeconomic status, broader systems and barriers to care, and power relations, is critically needed.

Parents' simultaneous hope and dread that investigations for chronic pain would indicate serious pathology, as evidenced throughout the current analysis and as has been previously described (Jordan et al., 2007), conveys the distress of diagnostic uncertainty. The current study identifies key drivers of diagnostic uncertainty in the clinical encounter that could inform future research and clinical practice. Of note, this analysis suggests that diagnostic uncertainty may be likely to persist among some youth with chronic primary pain unless clinicians are able to validate and connect with, not just youth's pain, but also the youth's and parents' narratives of their pain journeys. To do so requires that clinicians *elicit* and *listen* to youth's and parents' pain origin stories, as well as validate and explain.

Table 5.1.

Sociodemographic Characteristics of the Sample

	N	**Mean**	**Range**
Youth's age, years	23	13.2	10-17
Parent's age, years[a]	25	46.5	33-56
Relationship to youth			
Biological Mother	21		
Biological Father	1		
Adoptive Mother	1		
Adoptive Father	1		
Youth's ethnicity (%)			
White (Caucasian)	16		
Latino	4		
American Indian	1		
Two or more ethnicities	1		
Declined to answer	1		
Parent Education Level[b]			
Some college	9		
College degree	4		
Post-graduate degree	9		
Parent Marital Status[b]			
Married	19		
Separated/divorced	2		

Single	1

[a]Two youth participated with two of their parents
[b]Three parents missing from analysis

CHAPTER SEVEN: Synthesis and Discussion

Summary of Research Findings and Concluding Remarks

Pediatric chronic pain affects one in five youth, is debilitating to youth and their families, and poses a large economic burden to society ($19 billion US/year) (Groenewald et al., 2014; King et al., 2011). Current treatments for pediatric chronic pain are often ineffective (Fisher et al., 2018) and the majority of adolescents with chronic pain continue to have pain and mental health issues into adulthood (Shelby et al., 2013; Vinall et al., 2016; Walker et al., 2012). While some youth may develop chronic pain related to an injury or underlying health condition (e.g., juvenile rheumatoid arthritis), chronic pain is often the primary illness itself (e.g., headaches, complex regional pain syndrome) (Friedrichsdorf et al., 2016). The International Classification of Diseases, 11th Revision (ICD-11) now differentiates between chronic pain as a disease (chronic primary pain) and chronic pain as a symptom (chronic secondary pain) (Smith, Fors, Korwisi, Barke, Cameron, Colvin, Richardson, Rief, Treede, & Pain, 2019). However, the etiology of chronic primary pain is complex and many chronic pain patients do not receive a diagnosis that precisely explains a pathological cause of their pain (Butow & Sharpe, 2013). Thus, a major challenge that children with chronic pain and their parents face is understanding the cause of the pain and, as such, the nature of a chronic primary pain diagnosis is fraught with uncertainty.

Diagnostic uncertainty, the perception of a lack of a, or incorrect, label to explain symptoms, has been put forth as a critical area for research in pediatric chronic pain with potential implications for treatment (Pincus et al., 2018). Research among adults with chronic pain demonstrates an impact of diagnostic uncertainty on pain prognosis and adjustment (Geisser & Roth, 1998; Serbic et al., 2016). Importantly, given that this is a social phenomenon, research on diagnostic uncertainty among youth with chronic pain warrants parallel study of parents,

clinicians, and the healthcare systems youth and parents are required to navigate during their care, including clinical encounters. Clinicians' own uncertainty is likely to play a role in youth and parents' uncertainty about a diagnosis of chronic primary pain. As such, the primary aim of this dissertation was to examine the experiences of diagnostic uncertainty among youth with chronic primary pain, their parents, and clinicians, as well as how diagnostic uncertainty unfolds in the clinical encounter.

Individual differences in youth's and parents' ability to tolerate uncertainty more broadly may play a role in pediatric chronic pain. Intolerance of uncertainty is conceived of as a transdiagnostic dispositional risk factor for the development and maintenance of fear and anxiety-related psychopathology (Carleton, 2012). Given the uncertain nature of pediatric chronic pain and the high comorbidity of anxiety among this population, intolerance of uncertainty may be critical to the experience of pediatric chronic pain. Thus, this dissertation also examined the role of youth and parent intolerance of uncertainty as risk factors in the maintenance of pediatric chronic pain.

The first study (Chapter 2) explored how diagnostic uncertainty is experienced by youth with chronic primary pain and their parents and identified core components of this phenomenon. Reflexive thematic analysis revealed four themes: The function of a diagnosis, Haunted by something missing, The search for an alternative diagnosis, and Mistrust in the medical system. These themes illustrate that many youth with chronic primary pain and their parents struggle with the meaning of their diagnosis and continue to search for alternative diagnoses to explain the cause of their pain. Findings suggested that diagnostic uncertainty is intricately tied to youth's and parents' experiences in clinical encounters and impacts youth's and parents' trust in

clinicians. Youth also reported sensing uncertainty in clinicians providing the diagnosis and clinician communication, which impacted parent and youth 'buy in' to the diagnosis.

The second study (Chapter 3) examined the prevalence and consequences of diagnostic uncertainty among youth with chronic primary pain and their parents. Data from 174 youth with chronic primary pain and their parents demonstrated that diagnostic uncertainty is prevalent, affecting 31% of youth and 28% of parents. Further, youth who experience diagnostic uncertainty report significantly higher pain catastrophizing than youth who do not experience diagnostic uncertainty. Parent diagnostic uncertainty is also linked to greater youth pain interference and intensity and lower youth health-related quality of life. Further, 21% percent of youth and 17% of parents who reported that they had received a *clear diagnosis* for their or their child's pain, respectively, also reported thinking something else was happening that doctors had not yet found. Thus, a substantial minority of youth and parents experience diagnostic uncertainty despite having received a diagnosis for their pain. This has important implications for clinical encounters when a chronic pain diagnosis is provided, suggesting that the diagnosis itself is not sufficient to ease youth's and parents' fears about an unknown cause of their pain. This research was the first quantitative investigation of the phenomenon of diagnostic uncertainty in pediatric chronic pain and the first to link parent and youth diagnostic uncertainty to youth outcomes.

The third study (Chapter 4) qualitatively explored how diagnostic uncertainty is experienced by clinicians who diagnose and treat youth with chronic primary pain. Reflexive thematic analysis of semi-structured interviews with pediatricians who treat youth who experience complex chronic pain revealed one prominent theme: Drawing a line in the sand. This theme depicted clinician's own challenges with uncertainty, particularly around deciding when to cease

diagnostic testing. This decision was influenced by several factors depicted through four subthemes: physician training, experience and mentorship; individual patient and family factors; perceived reassurance of diagnostic investigations; and the broader social context and implications.

The fourth study (Chapter 5) examined parent and youth intolerance of uncertainty as risk factors to major tenets of the Interpersonal Fear Avoidance Model of Pain. Participants were 152 youth with chronic primary pain and their parents. Results demonstrated that greater parent intolerance of uncertainty predicted greater parent pain catastrophizing, which, in turn, predicted greater parent protectiveness, greater youth fear of pain, and subsequently greater youth 3-month pain interference. Youth intolerance of uncertainty had a significant indirect effect on 3-month pain interference through youth pain catastrophizing and fear of pain. These results suggest that parent and youth intolerance of uncertainty play important roles as risk factors in the maintenance of pediatric chronic pain over time.

The fifth study (Chapter 6) explored components of the clinical encounter that influence diagnostic uncertainty for youth with chronic pain and their parents over time. Interpretative phenomenological analysis of transcripts of the first clinical encounter when youth and parents were enrolled in a pediatric pain clinic, as well as pre- and post- semi-structured interviews with youth and parents, generated four themes: 1) Diagnostic uncertainty is a social phenomenon; 2) (In)validation of pain, the journey, and diagnostic uncertainty; 3) The (missing) link between origin story & pain explanation; and 4) The fragility of certainty. These themes illustrate that diagnostic uncertainty is a dynamic process and is co-constructed in the clinical encounters that youth and parents experience throughout their pain care.

Overall, these studies demonstrate that uncertainty is a common experience among youth with chronic primary pain and their parents, which is linked to children's pain and functioning. Uncertainty regarding a chronic pain diagnosis is distressing and may fuel an unrelenting search for the 'right' diagnosis, that can impede recovery. Further, diagnostic uncertainty is experienced by each individual in the clinical encounter, namely youth, parents, and clinicians. Indeed, diagnostic uncertainty is a relational process that is co-constructed in the clinical encounter. As such, aspects of the clinical encounter influence diagnostic uncertainty, including validation, messages of certainty and uncertainty, as well as how clinicians draw connections between the youth's and parent's pain origin stories and the pain explanation. Finally, youth's and parents' general ability to manage uncertainty (i.e., intolerance of uncertainty) play important roles in the maintenance of pediatric chronic pain.

This body of work has several strengths, including its multi-method approach, incorporating longitudinally collected quantitative and qualitative data. While quantitative analyses included relatively large sample sizes and gold-standard self-report measures, qualitative analyses allowed for in-depth exploration of the lived experience of this complex phenomenon from the perspectives youth, parents, and clinicians themselves.

Limitations and Future Directions

First and foremost, the majority of youth and parents in the study samples included in this dissertation were white and of high socioeconomic status. Further, the majority of parents in these samples were mothers, married, and had college or university level education. While these demographics are representative of the current state of the literature in pediatric chronic pain, and pediatric tertiary level chronic pain clinics (Evans, Taub, Tsao, Meldrum, & Zeltzer, 2010; King et al., 2011), they do not represent the experiences of youth and parents from minoritized

and marginalized groups who are likely to experience additional barriers, health disparities and discrimination (Hartford et al., 2022; Mulchan et al., 2021). This is critical, given that interpersonal and societal factors, including healthcare mistreatment and inaccessibility, as well as diagnostic uncertainty itself, are implicated in models of toxic stress and the maintenance of pediatric chronic pain, particularly for minoritized and marginalized youth (Nelson et al., 2022).

This dissertation included samples from differing geographical locations (Canada, the United Kingdom, the United States). Although challenges regarding pediatric pain management and pain education for healthcare professionals exist worldwide (Hurley-Wallace et al., 2019), differing cultures and health care systems could be important factors impacting these results. Across geographical locations, the current dissertation demonstrates that the clinical encounters youth and parents experience along their pain care journeys are often characterized by differing explanations for their pain, confusing messages, stigma, and invalidation, which fuel uncertainty, distress, and ultimately impact their trust in the healthcare system and pain care. The consequences of diagnostic uncertainty cannot be underestimated. In addition to the impact of diagnostic uncertainty on pain and pain related-constructs, youth who experience diagnostic uncertainty often undergo invasive procedures during their and their clinicians' search for answers, which puts them at risk of iatrogenic injury and delays chronic pain treatment. Further, these youth and parents experience conflict in the patient-clinician relationship that may result in the abandonment, mistrust, and lack of support for their pain. Pediatricians themselves, also report grappling with uncertainty in this context of treating youth. These findings highlight the need for improved pain education among clinicians internationally (Bhatia et al., 2008; Briggs et al., 2015; Hurley-Wallace et al., 2019; Shipton et al., 2018).

Future research should investigate how other aspects of the clinician-patient encounter (e.g., type of clinician, provision of pain education) impact diagnostic uncertainty. The complex nature of uncertainty in this population, the critical need for validation, and the implications of pain narratives, provides further support for the role of psychology in pediatric chronic pain treatment. Indeed, multidisciplinary care for chronic pain typically takes a '3P' approach, comprising of pharmacological, physiological, and psychological strategies (Rastogi & McCarthy, 2018). The current findings implicating youth cognitions (catastrophizing) and behavior (avoidance) as key components in the maintenance of pediatric chronic pain, support cognitive behavioral therapy already commonly used for youth with chronic pain (Fisher et al., 2018). However, uncertainty is not necessarily addressed in these treatment protocols. The current findings suggest that other transdiagnostic cognitive-behavioral treatments that target intolerance of uncertainty may be warranted for some youth (Boswell et al., 2013; Ellard et al., 2010; Farchione et al., 2012). Mounting evidence, including this dissertation, suggests that parent's own journeys and interpretations of their child's pain are also critical to assess and understand. In the current research, youth's and parents' narratives were found to be important sources of information in the clinical encounter. Youth's *and* parents' narratives may have important utility in chronic pain treatment, and particularly in terms of diagnostic uncertainty, given that narratives are co-constructed between patients and clinicians over the course of clinical encounters (Mattingly, 1994). When youth's and parents' pain origin stories were incorporated into the pain explanation, this enhanced youth's and parent's understanding of the pain and reduced diagnostic uncertainty. Further, youth's expression of their pain journey provided the opportunity for their experiences to be validated by clinicians.

The concept of validation is not well understood in pediatric chronic pain, however, it is likely nuanced (Edmond & Keefe, 2015). Innovative observational coding of validation and invalidation, including verbal and nonverbal behaviour, could be applied to video-taped patient-clinician interactions (Ashton-James et al., 2017; Shenk, 2014). These methods could also be used to understand how clinicians respond to uncertainty in the clinical encounter. Of equal importance will be to measure *perceived* validation and invalidation from youth themselves (Edlund, 2017). Further research should seek to develop an understanding of the impact of validation on information processing and what is remembered from the clinical encounter. Regarding the pain explanation itself, pain neuroscience education is an emerging area of research (Leake et al., 2019; Leake et al., 2021; Pate et al., 2020). How pain is conceptualized has been shown to be important for improving pain knowledge and catastrophizing, although its impact on disability is still unclear (Gallagher et al., 2013). Understanding the impact of pain education on diagnostic uncertainty in youth and their parents is a critical area for future research (Robins et al., 2016). Finally, future research should investigate whether interventions to improve parent and youth diagnostic certainty could influence youth's pain outcomes.

Conclusion

Given the widespread problem and economic toll of pediatric chronic pain, and the potential deleterious consequences of delayed treatment, understanding the nature of diagnostic uncertainty for youth with chronic pain, their parents, and clinicians is crucial. A better understanding of diagnostic uncertainty will help clinicians address uncertainty in chronic pain treatment, tailor the delivery of diagnoses to families to increase understanding of pain and diagnosis, and ultimately improve treatment engagement and response. This research is also an important step towards understanding intolerance of uncertainty in pediatric chronic pain, its

impact, and its potential as a modifiable intervention target impacting parent and youth adjustment to a chronic pain diagnosis and treatment.

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Appendix

Wednesday, July 6, 2022 at 19:46:21 Eastern Daylight Time

Subject: RE: Thesis permission [ref:_00Dd0dixc._5003w1dEhwd:ref]
Date: Wednesday, July 6, 2022 at 11:45:50 AM Eastern Daylight Saving Time
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Subject: Re: Copyright Permission [JPAIN_2018_209] [220702-003937]
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Alex Neville <[redacted]> Mon, Jul 4, 2022 at 11:22 AM
To: Daniel Kopala Sibley <[redacted], Sabine Soltani <[redacted]>, Gordon Asmundson <[redacted] Abbie Jordan <[redacted], "R. Nicholas Carleton" <[redacted] Keith Yeates <[redacted]>, Fiona Schulte <[redacted]>, Melanie Noel <[redacted] Clinch, Jacqui" <[redacted]x>, Tamar Pincus <[redacted]>, Cara Gail Nania <[redacted], Jaimie Beveridge <[redacted]

Dear co-authors,

Several manuscripts you have contributed to will form the basis of my dissertation, which I plan to defend in August 2022. As you know, I am preparing a manuscript-style dissertation in which the individual manuscripts will be included as chapters in the thesis document. I am required by the Faculty of Graduate Studies to document that all co-authors have provided permission for the manuscripts to be included in my dissertation document.

Please reply to this email within the next week indicating whether you give permission for me to include the following manuscripts in my doctoral dissertation (as applicable):

Neville, A., Kopala-Sibley, D. C., Soltani, S., Asmundson, G. J. G., Jordan, A., Carleton, R. N., Yeates, K. O., Schulte, F., & Noel M. (2021). A longitudinal examination of the interpersonal fear avoidance model of pain: The role of intolerance of uncertainty. *PAIN.* 162(1):152-160. doi: 10.1097/j.pain.0000000000002009.

Neville, A., Noel, M., Clinch, J., Pincus, T., Jordan, A. (2021). 'Drawing a line in the sand': Clinician diagnostic uncertainty in pediatric chronic pain. *European Journal of Pain.* 25(2), 430-441. doi: 10.1002/ejp.1682

Neville, A., Jordan, A., Pincus, T., Nania, C., Schulte, F., Yeates K.O., Noel, M. (2020). Diagnostic uncertainty in pediatric chronic pain: Nature, prevalence, and consequences. *PAIN Reports.* 5(6), e871. doi: 10.1097/PR9.0000000000000871.

Neville, A., Jordan, A., Beveridge, J. K., Pincus, T., & Noel, M. (2019). Diagnostic uncertainty in youth with chronic pain and their parents. *The Journal of Pain.* 20(9), 1080-1090. doi: 10.1016/j.jpain.2019.03.004

Upon completion, my dissertation will be added to the institutional repository at the University of Calgary and the Library and Archives Canada. Here are the links for further information about where the materials will be published: 1) University of Calgary Theses Repository - The Vault (http://theses.ucalgary.ca/), and 2) Library and Archives Canada (hhttp://collectionscanada.gc.ca/obj/s4/f2/frm-nl59-2-e.pdf).

I will also obtain the appropriate copyright permissions from the journals.

Thank you,
Alex

Nicholas Carleton <[redacted]> Mon, Jul 4, 2022 at 11:24 AM
To: Alex Neville <[redacted]>

Hi Alex,

You do indeed have my permission - good luck with your next steps. Please let me know if and how else I can help in the interim - cheers!! Nick

Dr. R. Nicholas Carleton, Ph.D., R.D. Psych.

Keith Yeates <[redacted]>
To: Alex Neville <[redacted]>

Mon, Jul 4, 2022 at 11:24 AM

Hi Alex,

You have my permission.

Keith

Melanie Noel <[redacted]>
To: Alex Neville <[redacted]>

Mon, Jul 4, 2022 at 12:04 PM

You have my FULL permission :)

Gordon Asmundson <[redacted]>
To: [redacted]
Cc: [redacted]

Mon, Jul 4, 2022 at 12:14 PM

Permission granted.

Sent from my iPhone

Abbie Jordan <[redacted]>
To: Alex Neville <n[redacted]>

Mon, Jul 4, 2022 at 12:23 PM

Hi Alex,

Fine with me, good luck!
Abbie

Dr Abbie Jordan
Reader
Department of Psychology and Centre of Pain Research
University of Bath

Tamar Pincus <[redacted]> Mon, Jul 4, 2022 at 12:39 PM
To: Alex Neville <[redacted]>

Permission granted, and good luck!
Tamar

Fiona Schulte <[redacted]> Mon, Jul 4, 2022 at 12:41 PM
To: [redacted], Alex Neville <[redacted]>

Permission granted!

Fiona Schulte, Ph.D, R.Psych.

Jaimie Beveridge <[redacted]> Mon, Jul 4, 2022 at 12:55 PM
To: [redacted], Alex Neville

You have my permission! Very exciting 😊

Cara Gail Nania <[redacted]> Mon, Jul 4, 2022 at 1:41 PM
To: [redacted]a>

Cc: [redacted], Alex Neville <[redacted]>

Of course :)

Sabine Soltani <[redacted]> Mon, Jul 4, 2022 at 2:40 PM
To: [redacted]om>
Cc: F[redacted] Alex Neville
[redacted]

You absolutely have my permission :)

Sabine

—

Sabine Soltani, Ph.D., R.Psych. (she/her)

Abbie Jordan <[redacted]>
To: Alex Neville <[redacted]>

And yes of course re including in your dissertation.

[redacted]

Best wishes

Abbie

Marcia Meldrum <[redacted]> Tue, Jul 26, 2022 at 10:08 AM
To: Alex Neville <[redacted]>

[redacted] You have my permission to include the manuscript in your dissertation, of course.

Marcia L. Meldrum, PhD

Melanie Noel <[redacted]> Tue, Jul 26, 2022 at 5:02 PM
To: Alex Neville <[redacted]>
[redacted]

Hi Alex,

[redacted]

And of course you have my permission to include this ms in your incredible dissertation.

[redacted]

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With gratitude,

Melanie

Ignasi Clemente pesudo <[redacted]> Tue, Jul 26, 2022 at 5:33 PM
Cc: Alex Neville <[redacted]>

Hi Alex,
[redacted] I give my permission to include this manuscript in your dissertation, and what a magnificent dissertation it will be.

[redacted]

[redacted]

Thank you and congratulations on all fronts!!!

Ignasi

Ignasi Clemente PhD

Katelyn Watson <[redacted]> Wed, Jul 27, 2022 at 1:12 PM
To: Alex Neville <[redacted]>

Hi Alex,

So many exciting things!

[redacted] you absolutely have my permission to include this in your dissertation. [redacted]

Take care,
Katelyn

Lonnie Zeltzer <[redacted]>
To: Alex Neville <n[redacted]>

Hi Alex and you certainly have my permission for a dissertation manuscript based on the study. [redacted]

Lonnie

Lonnie Zeltzer, MD