

UNIVERSITY OF CALGARY

Risk and Resilience: The Role of Parent Functioning in Pediatric Chronic Pain

by

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A THESIS

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Abstract

Objective: Pediatric chronic pain is prevalent and can significantly interfere with children's physical, emotional, social, and educational functioning. Parent factors have been shown to play an important role in children's chronic pain; however, research has predominately focused on parent responses to child pain to the exclusion of parents' own functioning (i.e., their physical and mental health). The broad aim of this dissertation was to examine the association between parent functioning, specifically their own chronic pain and mental health symptoms, and child chronic pain using a multi-method approach.

Methods: Three studies were conducted. The first study was a systematic review and meta-analysis of the extant literature examining associations between parent mental health and children's chronic pain and related functioning in both clinical and community samples. The second study used daily diary data from a clinical sample of 76 youth referred to a tertiary pain program and one of their parents to examine the associations between parent chronic pain status, parent daily variability (in their anxiety, mood, protective responses, and parenting stress), and youth daily pain intensity and interference. The third study used data from 1128 mother-child dyads enrolled in a longitudinal, community-based cohort study to identify risk and resilience factors throughout childhood that moderated the intergenerational transmission of chronic pain.

Results: Poorer functioning (i.e., chronic pain and/or mental health problems) in parents was significantly associated with the presence of chronic pain in community samples of children as well as the pain-related functioning of clinical samples of children with chronic pain. Parent chronic pain and mental health symptoms were related to children's chronic pain and functioning in distinct as well as interacting ways. Several general parent and child factors were found to contribute to the association between parent functioning and child chronic pain, either increasing

or decreasing the strength of the association, including ineffective parenting practices, child optimism, and child connections with adults.

Conclusions: Parent functioning plays an important role in pediatric chronic pain, increasing children's risk for poor adaptation to chronic pain, and should be more widely considered in research and clinical interventions for pediatric chronic pain.

Keywords: intergenerational; parent; pediatric; risk; resilience; chronic pain; mental health

Preface

The manuscripts that comprise this dissertation were prepared for publication in peer-reviewed journals (see Appendix A for letters of permission). J. Beveridge was the lead contributor to the first study (Chapter 2), conceptualizing and pre-registering the study. No funding or ethical approval was required. M. Noel is the principal investigator for the Pain and Mental Health in Youth (PATH) study (data used in Chapter 3), leading the conceptualization of the study and obtaining research grants and ethics approvals (REB15-3100). J. Beveridge was involved in the design of the second phase of the PATH study. S. Tough and S. McDonald conceptualized and lead the All Our Families (AOF) study (data used in Chapter 4), obtaining research grants, ethics approvals (REB13-0868), and managing follow-up surveys. J. Beveridge contributed to the design of the latest timepoints of the AOF study with other co-authors. For all three manuscripts included in this dissertation, J. Beveridge conceptualized the research question, conducted the statistical analyses, and wrote the manuscript. All authors contributed to the intellectual content and provided critical reviews of the manuscripts.

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CHAPTER ONE: Introduction

Introduction

Pediatric chronic pain (i.e., pain that recurs or persists for longer than three months) is prevalent, affecting approximately 20% of children, and can significantly interfere with physical functioning, mental health, school participation, sleep, social life, and leisure activities (Chambers et al., 2024; Miró et al., 2023; Treede et al., 2019). Many children continue to have chronic pain into adulthood, with negative impacts on their emotional, social, educational, and occupational functioning as well as intergenerational impacts on their offspring's functioning (Murray et al., 2020; Stone et al., 2023; Walker et al., 2012). Given its prevalence and functional impact, a greater understanding of factors that contribute to pediatric chronic pain is needed.

A substantial literature has demonstrated that parents play an important role in pediatric chronic pain (Donnelly et al., 2020). The majority of this research has focused on how parents respond to their child's pain, with several pain-specific responses (e.g., catastrophic thinking, protective behaviours) implicated in the onset, severity, and maintenance of children's chronic pain (Donnelly et al., 2020; Harrison et al., 2020). However, research is increasingly recognizing the importance of parents' own functioning (i.e., their physical and mental health) for children's chronic pain and related outcomes. Several recent studies suggest that parents' chronic pain and mental health symptoms (e.g., anxiety and depressive symptoms) may be stronger predictors of poor pain-related outcomes in children than parents' pain-specific responses (Law et al., 2017; Poppert Cordts et al., 2019; Stone et al., 2018). While these recent studies have advanced this area of research, further investigation is needed to more comprehensively understand the associations between parent functioning and child chronic pain as well as factors that mediate or moderate these associations and contribute to either negative child adaptation (i.e., risk) or positive child adaptation (i.e., resilience) in the context of poor parent functioning.

The overarching aims of this dissertation were to address several gaps in the literature using a multi-method approach with both clinical and community samples. These studies aimed to identify: (1) the magnitude of associations between parent mental health and child chronic pain through a systematic review and meta-analysis; (2) the role of daily variability among parents with versus without chronic pain and its association with their child's daily chronic pain-related functioning; and (3) risk and protective factors in childhood that predict either increased or decreased risk for chronic pain in early adolescence among children of mothers with (and without) chronic pain using data from a longitudinal, community-based cohort study.

Pediatric Chronic Pain

Chronic pain, defined as persistent or recurring pain lasting longer than three months, is a common pediatric health condition, affecting approximately 20% of children and adolescents (Chambers et al., 2024; Treede et al., 2019). Chronic pain may occur as a symptom secondary to an injury, surgical procedure, or medical condition (e.g., cancer, arthritis) or as the primary health condition with no other medical or disease process better accounting for the pain (e.g., chronic migraine, fibromyalgia, functional abdominal pain). Current nosology classifies these two types of chronic pain as chronic secondary pain and chronic primary pain (Treede et al., 2019). The most common types of pain reported by children are headache, abdominal, and musculoskeletal pain; however, many children report more than one type of pain (e.g., headaches and abdominal pain) (Chambers et al., 2024; Perquin et al., 2000; Stanford et al., 2008). Pediatric chronic pain is more common among girls and rates tend to increase with age, with a peak onset in adolescence between the ages of 12 and 15 (King et al., 2011; Perquin et al., 2000; Stanford et al., 2008).

Chronic pain is a complex condition, regarded as both multifactorial and biopsychosocial (Goubert et al., 2021; Nicholas, 2022). As such, the severity and impact of chronic pain can vary

greatly between individuals. While many children are able to function well in the presence of pain, even when the pain is rated as moderate to severe intensity, a small subset (approximately 5%) report moderate to high levels of pain-related disability (Huguet & Miró, 2008). For these children, pain and related disability can significantly interfere with their ability to attend and participate in school, engage in physical and recreational activities, socialize with friends, and have a good quality sleep (Miró et al., 2023; Palermo, 2000). Mental health problems, including clinically-elevated anxiety, depressive, and post-traumatic stress disorder symptoms, are also more common among children with versus without chronic pain (Vinnall et al., 2016).

Due to its prevalence and functional impact, pediatric chronic pain is related to increased health care utilization including physician visits and hospitalizations (Hogan et al., 2016). Recent estimates suggest that the annual health care cost of chronic pain among adolescents and adults in Canada is \$7.2 billion, which is comparable to other costly diseases including cardiovascular and neuropsychiatric disorders (Hogan et al., 2016). In the United States, total incremental health care expenditures for pediatric chronic pain have been estimated at \$11.8 billion, exceeding the costs of childhood asthma and obesity (Groenewald et al., 2015). Current treatments for pediatric chronic pain align with the biopsychosocial approach and include medical and pharmacological interventions, physical therapies, and psychological therapies. Psychological interventions tend to focus on managing the child's pain, disability, and distress, and can include behavioural strategies such as relaxation training and biofeedback as well as cognitive strategies such as stress management and cognitive restructuring (Fisher et al., 2018). Parent interventions may also be included to provide parents with strategies for helping to manage their child's pain. This training often focuses on reducing parent behavioural responses that are thought to reinforce the child's pain and related disability (e.g., protective responses) (Lee et al., 2021). Unfortunately,

current psychological interventions are often ineffective, with small and short-lasting effects for improving pain and related outcomes (Fisher et al., 2018). As such, many children (i.e., up to two-thirds) continue to have chronic pain into adulthood and are at increased risk as adults for lower educational and occupational success, mental health problems including anxiety and depressive disorders, and poor social functioning (Fearon & Hotopf, 2001; Murray et al., 2020; Stone et al., 2023; Walker et al., 2012). The long-term effects of pediatric chronic pain have also recently been shown to extend into the next generation, with greater emotional problems reported in the offspring of parents who had chronic abdominal pain as children as compared to control parents (Stone et al., 2023).

Parent Factors in Pediatric Chronic Pain

Parents play a critically important role in the development and health of their children, including their experiences of pain (Palermo & Eccleston, 2009). Research over the past 30 years has identified a number of parent factors that are directly related to the onset, maintenance, and severity of children's pain and related functioning. This research has predominately focused on parent responses to their child's pain – specifically maladaptive responses – including cognitive (e.g., pain catastrophizing, fear of pain), affective (e.g., stress related to caring for a child with chronic pain), and behavioural (e.g., protective or solicitous behaviours) responses that increase children's *risk* for poor pain-related outcomes. Two meta-analyses recently synthesized this literature and found the largest correlations between parent pain catastrophizing and protective behaviours and children's pain-related disability (Donnelly et al., 2020; Harrison et al., 2020). These two parent factors were more commonly assessed than the other parent factors and effect sizes were still relatively small ($r_s \leq 0.30$). More recent research with a focus on *resilience* to pediatric chronic pain has identified adaptive cognitive and behavioural parent responses that

predict more positive pain-related outcomes for children. Specifically, greater parent acceptance of child pain, psychologically flexible parenting responses to child pain (i.e., remaining focused on the present, accepting distress, and aligning behaviour with values), and encouragement of child engagement in activities are associated with reduced pain, disability, and activity avoidance and greater psychosocial and emotional functioning for children with chronic pain (Beeckman, Hughes, et al., 2019; Beeckman, Simons, et al., 2019; Feinstein et al., 2018).

Parent functioning, including parents' own chronic pain and mental health problems, has also been positioned as an important factor contributing to children's chronic pain experience but has received less attention in the literature than parent responses (Palermo & Eccleston, 2009). The research that has been conducted suggests that poor physical and mental health in parents is significantly related to the development, maintenance, and severity of chronic pain in children. First, a substantial literature on the intergenerational transmission of chronic pain has shown that children are at significantly increased risk for developing chronic pain when one or both of their parents have chronic pain (K. S. Higgins et al., 2015). Longitudinal research with community samples has also demonstrated that poor mental health in parents, particularly elevated anxiety and depressive symptoms, in early childhood is significantly associated with the presence of child chronic pain in later childhood and adolescence (Fryer et al., 2017; Helgeland et al., 2010; Kolaitis et al., 2022; Ramchandani et al., 2006). Longitudinal, population-based studies have also demonstrated that chronic pain and/or poor mental health in parents can increase the likelihood that children's chronic pain persists throughout childhood and adolescence (Hinze et al., 2023; Ramchandani et al., 2007; Stanford et al., 2008).

Studies with clinical samples provide further evidence that parent functioning is associated with children's pain-related functioning. Specifically, parent chronic pain and related

disability and parent mental health symptoms (e.g., anxiety, depressive, and post-traumatic stress symptoms) are significantly associated with worse pain, disability, and mental health in children with chronic pain (Beveridge et al., 2018; Campo et al., 2007; Poppert Cordts et al., 2019).

Critically, poor physical and mental health is common in parents of children with chronic pain, with 50-75% of parents reporting their own chronic pain and related disability, 40% reporting clinically-elevated depressive symptoms, 62% reporting clinically-elevated anxiety symptoms, and 20% meeting clinical cut-offs for posttraumatic stress disorder (Beveridge et al., 2018; Birnie et al., 2020; Eccleston et al., 2004; Law et al., 2020; Noel et al., 2016; Stone et al., 2018).

The majority of research on parent factors in pediatric chronic pain has been cross-sectional and thus the (bi)directionality of the associations between parent functioning and child chronic pain has not been thoroughly investigated. It is likely that children's chronic pain and related disability also contribute to increased anxiety, depression, and disability among parents. Indeed, in qualitative research, parents of children with chronic pain describe struggling to adapt their life to their child's pain as well as substantial feelings of uncertainty, fear, distress, and loss (Jordan et al., 2007). Quantitative research has also assessed the impact of child chronic pain on parent functioning using the Bath Adolescent Pain – Parental Impact Questionnaire (BAP-PIQ). This cross-sectional research shows that parents endorse depression, anxiety, self-blame, and helplessness in response to having a child with pain (Jordan et al., 2008). Nonetheless, the few studies that have investigated the directionality of these associations have found that parent factors are better predictors of children's pain-related functioning than vice versa (Law et al., 2020; Neville, Griep, et al., 2020). Moreover, a prospective study that examined a cognitive-behavioural therapy for youth with chronic pain and their parents found that higher levels of

parent distress at pre-treatment predicted *less* improvement in child disability at post-treatment (Law et al., 2017).

Taken altogether, parent functioning – specifically their own chronic pain and mental health – appears to play an important role in the onset, maintenance, and severity of pediatric chronic pain. For the most part, however, the effect sizes for these associations are small and some studies do not find a significant association between parent functioning and child chronic pain (Donnelly et al., 2020; Eccleston et al., 2004; K. S. Higgins et al., 2015; K. B. Smith & Chambers, 2006). Thus, there are likely other factors or processes that contribute to their association, either increasing or decreasing the strength. Recent research has started to build more complex statistical models that include several parent factors as well as child factors to better understand how these variables interact and contribute to children’s chronic pain. Results from these studies have revealed that parent functioning is directly related to their parenting responses, such that parents with (versus without) chronic pain and/or poorer mental health report greater pain catastrophizing and protective behaviours, which are, in turn, related to worse child pain outcomes (e.g., greater disability) (Birnie et al., 2020; Fussner et al., 2018). Other studies have examined the relative contribution of various parent factors for child chronic pain. Results from some of these studies suggest that parent functioning is more strongly related to children’s pain-related functioning than parent responses. For example, one study that used structural equation modelling to develop a multifactorial model of parent factors and children’s pain-related outcomes (i.e., pain, disability, or psychological functioning) found that the best-fitting model did *not* include parent responses (i.e., protective and monitoring behaviours) and only included parent functioning (i.e., chronic pain, related disability, psychological functioning) (Poppert Cordts et al., 2019). Two other studies using similar statistical modeling procedures

(i.e., structural equation modelling and path analysis) found similar results, wherein parent behavioural responses were not directly related to adolescent chronic pain or disability when parent functioning, specifically parent chronic pain and distress, were also included in the model (Stone et al., 2018; Vowles et al., 2010). Of note, to my knowledge, no research has examined whether resilience factors mediate or moderate the association between parent functioning and children's chronic pain and/or related functioning.

While these recent findings have highlighted the important role of parent physical and mental health for children's chronic pain, the relative contributions of each of these domains of parent health remains unclear. First, some studies do not measure or control for the other, despite substantial evidence that chronic pain and mental health problems co-occur at high rates (Burke et al., 2015). Second, the results from studies that have included both are mixed, with parent chronic pain more strongly predicting child chronic pain than parent mental health in some studies, parent mental health symptoms more strongly predicting child chronic pain than parent chronic pain in other studies, or both domains similarly contributing to child chronic pain (Coenders et al., 2014; Helgeland et al., 2010; Hoftun et al., 2013; Incedon et al., 2016; Poppert Cordts et al., 2019; Ramchandani et al., 2006).

Chronic pain and mental health disorders are not only common among parents of children with chronic pain, but also among adults in the general population. Recent prevalence estimates from Canada and the United States indicate that 20-40% of adults have a chronic pain condition, with 10-15% endorsing significant pain-related disability (Dahlhamer et al., 2018; Schopflocher et al., 2011; Yong et al., 2022), 10-20% have an anxiety disorder, and 5-10% have depressive disorders (Kessler et al., 2012; Knoll & MacLennan, 2017; Yeretizian et al., 2023). As noted, chronic pain conditions and mental health disorders often co-occur; thus, many adults are likely

to present with both physical and mental health conditions. Given their prevalence, in both parents of children with chronic pain and the general population, as well as their comorbidity and impact on child outcomes, identifying and addressing chronic pain and mental health problems in parents may have important implications for the prevention and treatment of children's chronic pain and related disability. Further research is needed to better understand the associations of parent chronic pain and mental health with child chronic pain, and the factors or processes contributing to these associations, including those that strengthen the association between poor parent functioning and child chronic pain (i.e., increase risk) as well as those that buffer this association and predict positive adaptation despite exposure to poor parent functioning (i.e., promote resilience).

Theoretical Models

Social Learning

The role of parent factors in pediatric chronic pain has predominately been guided by social learning theory, which broadly posits that individuals can learn through directly observing the behaviour of others (i.e., observational learning) and/or engaging in a behaviour and having it positively or negatively reinforced by others (i.e., operant learning) (Bandura & Walters, 1977). Specific to pediatric chronic pain, social learning theory suggests that parent modeling and reinforcement of maladaptive pain responses contributes to the development, maintenance, and severity of pain problems in children. Operant learning has been used to explain the association between parent protective responses and children's pain-related disability, such that when a parent engages in protective behaviours, for example allowing their child to stay home from school due to pain, the child's pain is reinforced (and thus is more likely to continue to occur) due to the consequence of not having to go to school (Walker & Zeman, 1992). Observational

learning has been applied to the other parent factors, including pain-specific responses (e.g., catastrophic thinking about pain) and functioning (i.e., chronic pain, mental health), wherein children can learn pain behaviours (i.e., how to perceive and react to pain) through observing their parent. For example, children may learn to respond to pain with maladaptive cognitive-affective (e.g., pain catastrophizing) and behavioural (e.g., avoidance) strategies through observing their parent's use of these strategies.

Parents with chronic pain and/or mental health problems report greater use of these maladaptive strategies – specifically protective responses and pain catastrophizing – and thus the association between parent functioning and child chronic pain and disability has been suggested as due to parent modeling and reinforcement of these maladaptive coping strategies (Birnie et al., 2020; Fussner et al., 2018; Sieberg et al., 2011; Wilson & Fales, 2015). While this pathway has received empirical support (K. S. Higgins et al., 2019; Stone et al., 2018), it is likely that other processes also contribute to the association between parent factors, particularly parent functioning, and child chronic pain. As noted, to my knowledge, the potential for parents with chronic pain and/or mental health problems to model or reinforce *adaptive* coping strategies (e.g., pain acceptance) has not been empirically tested to date.

Developmental Systems

Theories from developmental science, including developmental psychopathology, ecological systems theory, resilience theory, and developmental systems theory, are particularly relevant for conceptualizing the role of parent functioning within pediatric chronic pain (Bronfenbrenner, 1981; Cicchetti, 1984; Masten, 2001; Masten et al., 2021). These theories have been applied to related areas of research, including the intergenerational transmission of mental illness and childhood exposure to maltreatment, to understand variations in human development

(Cicchetti, 2013; Goodman & Gotlib, 1999). Research in this area has found that, while children exposed to adversities are at-risk for poor developmental outcomes (e.g., mental and physical health problems), not all children display these poor outcomes and instead appear to demonstrate resilience – that is, the capacity to successfully adapt to or be protected from risk (Masten et al., 2021). These findings have led scholars to study the processes that differentiate positive from negative adaptation in the context of various risks with the goal of informing practice and policy to *reduce* problems and *promote* positive outcomes (Cicchetti & Toth, 2009).

Various scholars have integrated these theories from developmental science to develop more comprehensive models for understanding variations in development. In particular, Masten (2016, 2021) has detailed a multisystem developmental framework for resilience research that outlines the multiple levels of a system that may influence a child's adaptation to risk including genetic, neurobiological, behavioural, and environmental as well as family, community, and society. Masten (2016, 2021) highlights the reciprocal interactions that can occur between these systems and the dynamic nature of these systems. That is, a child's development depends on the function of many different systems as well as the interactions and interconnections between these systems – from the micro (e.g., the child's immune system) to the macro (e.g., resources available through the broader sociocultural context). Similarly, a child's capacity to successfully adapt to adverse or stressful situations that could interfere with their development also depends on the function and interactions between a variety of systems. Critically, children's adaptation is not only shaped by these systems but can also influence these systems. Moreover, the processes of adaptation are dynamic and developmentally-sensitive, changing over time based on the function of different systems and the developmental stage of the child.

Several conceptual models within pediatric chronic pain have incorporated ideas from this developmental systems perspective. One of the first, an integrative model of parent and family factors in pediatric chronic pain proposed by Palermo & Chambers (2005) outlined three system levels – parent factors (e.g., protective behaviours), dyadic factors (e.g., quality of parent-child interaction) and family factors (e.g., family functioning) – that interact with one another as well as with child factors (e.g., developmental status, emotional symptoms) to influence child pain and disability. The authors highlighted the importance of integrating these different system levels to better understand the pathways through which parents may influence children’s pain. Stone & Wilson’s (2016) conceptual model for the intergenerational transmission of chronic pain also incorporated aspects of developmental systems theory, including the multiple complex interactions between genetic, neurobiological, and environmental factors that can increase children’s risk for poor adaptation to parent chronic pain. In particular, this model outlined five mechanisms through which parent chronic pain may transmit to children including genetics, alterations in early neurobiological development, pain-specific social learning, general parenting and family health, and exposure to stressful environment as well as child vulnerability factors (e.g., altered pain processing, pain coping behaviours) and other moderating factors (e.g., chronic pain in the second parent, child developmental stage) that may interact with the mechanisms to increase or decrease the child’s risk for poor outcomes. In both of these models, the system levels were limited to the child, parent, and family environment. A model by Cousins et al. (2015) outlines risk and resiliency factors at the individual and family level as well as the social and cultural level that may interact to influence children’s adaptation to chronic pain over time. Specifically, resilience resources (e.g., child optimism, adaptive family functioning, community support) are posited to promote adaptive pain outcomes by enhancing resilience mechanisms

(e.g., child and parent pain acceptance, cultural values) and minimizing risk factors (e.g., poor parent health) and mechanisms (e.g., child pain catastrophizing, parent protective behaviours) while risk factors are posited to contribute to poor pain outcomes by enhancing risk mechanisms and minimizing resilience factors and mechanisms.

These models extend the focus from specific pain behaviours within a parent-child dyad and consider various other factors and processes occurring in the larger context of a child's life. This broader focus is particularly relevant for understanding the role of parent functioning within pediatric chronic pain as poor physical and mental health in parents likely interacts with many other factors – across levels of the child's ecology – to impact children's adaptation to pain. Some potential risk factors are outlined in the chronic pain models described above, including general parenting practices (e.g., permissive, controlling, or inconsistent parenting), the parent-child relationship (e.g., high conflict, low autonomy, insecure attachment), the family environment (e.g., low cohesion, marital conflict), and the broader social context (e.g., unsafe neighbourhood, low socioeconomic status) (Palermo & Chambers, 2005; Stone & Wilson, 2016). These risk factors may contribute to the association between poor parent functioning and children's poor adaptation to pain through, not only parent modeling and reinforcement of maladaptive coping strategies, but exposure to (chronic) stress. As delineated by the extensive literature examining the effects of early life adversity on child health and development, chronic stress can alter the stress response system and cause 'wear and tear' on the body that increases the risk for physical health problems (McEwen & Stellar, 1993). Exposure to a stressful home environment can also impact children's health through psychosocial mechanisms including deficits in emotion processing and social competence (Repetti et al., 2002). Children whose parents have chronic pain and/or mental health difficulties may be exposed to more adverse or

stressful home environments, due to the impact these health conditions can have on one's physical and emotional functioning, and thus experience (chronic) stress that negatively impacts their physical and mental health.

Critically, the effects of early life stress on children's health and development can be buffered by protective factors. The resilience literature has identified a number of factors that predict positive adaptation in children exposed to a wide range of adverse or stressful situations (e.g., maltreatment, poverty, parent mental illness, war) including effective parenting practices, close relationships with adults and peers, intellectual and emotional capacities (e.g., problem-solving skills, emotion regulation), self-efficacy, hope, and engagement with effective schools and communities (Masten, 2015). Masten (2015) posits that these resilience factors are tied to fundamental adaptive systems that are the product of both biological and cultural evolution and protect human development from a variety of potential threats. For example, effective parenting practices and close relationships with capable adults are tied to the attachment system, which has been situated as a protective system that evolved to protect young children and foster emotional security and learning. Relatedly, intellectual and emotional capacities are tied to the learning, thinking, and self-regulation systems of the central nervous system, while self-efficacy is tied to the mastery motivation and related reward systems. These biological and cultural systems are interconnected and interact with one another to influence development as well as adaptation to risks (Masten, 2015). In their model, Stone & Wilson (2016) discuss several potential protective factors and resilience processes that may be relevant to disrupting the transmission of risk for chronic pain from parents to children including positive parent-child interactions and parenting practices (e.g., warmth, consistent responses to child distress), a supportive family environment, child optimism, and parent and child use of active coping strategies. Taken altogether, parent

functioning likely relates to child risk for chronic pain and related functioning through multiple pathways that can interact with other factors or processes across system levels to increase or, importantly, decrease children's risk for poor pain adaptation.

Dissertation Study Aims

The overarching aim of this dissertation was to examine the associations between parent functioning – specifically their own chronic pain and mental health symptoms – and pediatric chronic pain using a multi-method approach that included both clinical and community samples and investigated pain-specific as well as general risk and resilience factors across the child's ecology that may contribute to children's adaptation to chronic pain in the context of poor parent functioning. The manuscripts that form this dissertation address three specific aims:

The aim of the first study (Chapter 2) was to comprehensively review and meta-analyse the extant literature investigating associations between parent mental health and child chronic pain. As noted, numerous studies have shown that parent depressive, anxiety, and posttraumatic stress disorder symptoms are significantly related to child chronic pain and related outcomes, and thus may be a factor contributing to the onset and maintenance of pediatric chronic pain (Birnie et al., 2020; Campo et al., 2007; Noel et al., 2016). However, other studies have found non-significant associations between parent mental health and child chronic pain (Eccleston et al., 2004; Sieberg et al., 2011). Thus, the *magnitude* of these associations, and therefore importance of parent mental health for child chronic pain, was not well understood in the literature.

The second study (Chapter 3) used daily diary data to explore the association of parent daily variability with parent chronic pain status and youth's daily pain-related functioning. This study examined parent factors that have been shown, in studies using retrospective measures, to be more common among parents with chronic pain and relate to poor outcomes for youth with

chronic pain, including greater parent anxiety and depressive symptoms, protective responses, and parenting stress (Evans et al., 2005; Fussner et al., 2018; Stone et al., 2018; Wilson & Fales, 2015). However, this study measured parent *variability* in these factors across days to investigate whether parents with chronic pain were more variable in their functioning from day to day than parents without chronic pain, and whether this variability was related to worse pain outcomes for their children. Research with community samples has demonstrated that day-to-day variability in parent-child interactions is related to worse mental and physical health outcomes for youth (Lippold et al., 2015, 2019). However, this daily variability in parents had not been explicitly examined in the pediatric chronic pain literature.

The third study (Chapter 4) used data from a longitudinal, community-based cohort study to identify risk *and* protective factors across childhood that either increased or decreased risk for chronic pain in early adolescence among children whose parents have chronic pain as compared to children of parents without chronic pain. This study examined general (i.e., not pain-specific) factors across various system levels (child, parent, school, peers, and community). Previous research examining parent functioning within pediatric chronic pain has focused on pain-specific risk factors; however, several studies have examined more general factors. These studies have demonstrated that parents with chronic pain report more ineffective parenting practices (e.g., permissive parenting), poorer relationship quality with their children, less cohesion and more conflict in their family, fewer social supports, and lower household income (Evans et al., 2005, 2006; Wilson & Fales, 2015). Moreover, children whose parents have chronic pain report lower self-esteem, social competence, family cohesion, and insecure attachment (Evans & Keenan, 2007; Kaasbøll et al., 2015, 2018). This study aimed to extend these findings by examining protective factors that may buffer the transmission of chronic pain from mothers to children.

**CHAPTER TWO: The Association Between Parent Mental Health and Pediatric Chronic
Pain: A Systematic Review and Meta-Analysis**

Abstract

Mental health problems are common among parents of children with chronic pain and associated with worse outcomes for the child with chronic pain. However, the effect sizes of these associations between parent mental health and pediatric chronic pain vary widely across studies. The aim of this systematic review and meta-analysis was to generate pooled estimates of the (1) prevalence of mental health problems among parents of children with chronic pain and (2) associations between parent mental health and the (2a) presence of child chronic pain and (2b) functioning of children with chronic pain. Embase, MEDLINE, PsycINFO, Web of Science, and CINAHL were searched up to November 2022. Observational studies that examined symptoms or diagnoses of parent anxiety, depression, or general distress and the presence of child chronic pain and/or related functioning were included. From 32,848 records, 2 coders identified 49 studies to include in random-effects meta-analyses. The results revealed that mental health problems among parents of children with chronic pain were common (anxiety: 28.8% [95% CI 20.3-39.1]; depression: 20.0% [15.7-25.2]; general distress: 32.4% [22.7-44.0]). Poorer parent mental health was significantly associated with the presence of chronic pain (anxiety: OR = 1.91 [1.51-2.41]; depression: OR = 1.90 [1.51-2.38]; general distress: OR = 1.74 [1.47-2.05]) and worse related functioning (i.e., pain intensity, physical functioning, anxiety and depression symptoms; $r_s = 0.10-0.25$, all $p_s < .05$) in children. Moderator analyses were generally non-significant or could not be conducted due to insufficient data. Findings support the importance of addressing parent mental health in the prevention and treatment of pediatric chronic pain.

Introduction

Approximately 25% of children and adolescents (hereafter shortened to “children”) endorse having chronic primary pain (i.e., pain lasting >3 months that cannot be better accounted for by another chronic condition; Treede et al., 2019; King et al., 2011). Pediatric chronic pain can be difficult to treat, often persists into adulthood, confers risk for additional physical and mental health problems, and transmits across generations (Stone et al., 2023). In an effort to improve these outcomes, a 2020 *Lancet Child & Adolescent Health* Commission emphasized the need for a greater understanding of factors contributing to the onset, maintenance, and severity of chronic pain in children (Eccleston et al., 2021).

Research has shown that internalizing mental health disorders (e.g., depression, anxiety) are 2-3 times more common in children with (versus without) chronic pain and their parents (Vinall et al., 2016), suggesting shared vulnerabilities for these conditions within families. Critically, parent mental health problems are associated with worse outcomes for children with chronic pain, including poorer response to psychological interventions for their pain and disability (Cunningham et al., 2016; Law et al., 2017). As compared to other parent variables, however, parent mental health has received less attention in the pediatric pain literature, especially as compared to literature on other childhood chronic conditions (e.g., diabetes; Palermo & Eccleston, 2009; Piquart, 2019). Research on the role of parent factors in pediatric chronic pain tends to focus on parent *responses* to their child’s pain, including cognitive (e.g., parent catastrophizing about child’s pain), behavioural (e.g., protective responses to child’s pain), and affective (e.g., anxiety related to parenting a child with pain) responses, to the exclusion of parents’ general psychological functioning (Eccleston et al., 2021; Vinall et al., 2016). While recent meta-analyses have estimated the associations between parent responses and children’s

pain-related functioning (Donnelly et al., 2020; Harrison et al., 2020), associations between the general psychological functioning of parents and the incidence and severity of chronic pain in children have not been comprehensively reviewed or meta-analysed. Poor mental health in parents may be a risk factor for the onset and maintenance of pediatric chronic pain (McKillop & Banez, 2016) and/or a consequence of having a child with a chronic condition (Gaughan et al., 2014; Jordan et al., 2007). Moreover, although many studies report high rates of mental health problems among parents of children with chronic pain, which, in turn, are significantly associated with the child's pain-related functioning, some studies report low rates and/or non-significant associations (Birnie et al., 2020; Eccleston et al., 2004; Sieberg et al., 2011). Given these gaps in the research, a synthesis of the existing literature is needed to better understand the associations between parent general mental health and children's chronic pain and related disability.

The primary aims of this systematic review and meta-analysis were to: (1) estimate the prevalence of mental health problems among parents of children with chronic pain; (2) derive pooled effect sizes of the associations between parent mental health and the (2a) presence of child chronic pain and (2b) functioning of children with chronic pain (e.g., pain characteristics; physical, psychological functioning). A secondary aim was to examine potential moderators of these estimates, which may explain between-study variability. Moderators were selected *a priori* based on moderators identified in related theoretical and empirical literature (Goodman, 2020; Palermo & Chambers, 2005; Stone & Wilson, 2016) and included sample characteristics (child age, sex/gender, race/ethnicity, pain condition; clinical vs. community) and study design (type and timing of measurement).

Methods

Search Strategy and Selection Criteria

This study was registered with PROSPERO (CRD42020124201) and followed Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) reporting guidelines. Definitions of constructs are provided in Supplementary Table 2.1 in Appendix B. Embase, MEDLINE, PsycINFO, Web of Science, and CINAHL were searched from inception until June 21, 2019 by a librarian with experience in systematic reviews. Searches were updated in August 2021 and November 2022 by J.B. using the same search strategy, which was developed by the librarian and included terms for parent, internalizing mental health, child, and chronic pain (see Supplementary Table 2.2 in Appendix B).

Records were screened in Covidence (Veritas Health Innovation). Titles were screened by three independent coders while abstracts and full-texts were reviewed by one coder after good agreement (>80%) was achieved on a random sample (20%) of studies with another independent coder. The senior author was consulted when there was any uncertainty. Observational studies were included if they met the following criteria: (1) examined symptoms or diagnoses of anxiety disorders, depressive disorders, post-traumatic stress disorder, or general distress in parents and the presence of chronic pain in children and/or functioning of children with chronic pain; (2) assessed chronic primary pain in children aged 2-18 years; (3) published in English (Nussbaumer-Streit et al., 2020); (4) in a peer-reviewed journal. Studies were excluded if they: (1) used outdated or vague language related to mental health (e.g., “nervous problem”), subsumed mental health into a broader category (e.g., “chronic diseases”), and/or did not clearly assess parent mental health in the same manner for each participant; (2) were qualitative studies; or (3) were intervention studies that did not include a baseline assessment. Efforts to obtain all

eligible records for full-text review included use of interlibrary loans and attempts to contact authors. Reference lists of included studies were hand searched for additional records but yielded no eligible studies for inclusion.

Data Extraction

Data extraction was conducted in Research Electronic Data Capture (REDCap) by one coder after good agreement (>80%) was achieved on a random sample (20%) of studies with another independent coder. Summary data were extracted using a standardized form with entries for: (1) study characteristics; (2) effect size data; and (3) moderator variables. Child variables aligned with recommended outcomes (McGrath et al., 2008; Palermo et al., 2021) and a conceptual model (Stone & Wilson, 2016) and were grouped into the following domains: presence of chronic pain, pain characteristics (e.g., pain intensity), physical functioning (e.g., functional disability), psychological functioning (e.g., anxiety and depression symptoms), role functioning (e.g., school attendance), quality of life, sleep, and pain-related experiences (e.g., pain catastrophizing). Categorical moderator variables included type of sample (clinical vs. community), child chronic pain condition (recurrent abdominal pain [RAP], headache, fibromyalgia, vs. mixed), and measurement (self-report vs. diagnostic interview) and timing (current vs. history) of parent mental health. Continuous moderator variables included child age (mean of sample), sex/gender (% female), and race/ethnicity (% White or nationality of study country). While we recognize that % White continues to center Whiteness as the standard of comparison (Letzen et al., 2022), we used % White as included studies were most consistent in reporting % White in their samples.

Data Synthesis

When a study examined more than one relevant variable, effect sizes were examined in separate meta-analyses. When there were overlapping samples or analyses (e.g., variable assessed at multiple timepoints), the analysis with the larger sample size, more relevant to the current research question, or most similar to other analyses was included. When distinct subgroups were included in a study, means and standard deviations were combined for analyses (J. Higgins et al., 2022) while effect sizes were entered separately into analyses. When case-control studies had more than one comparison group, the group with no physical or mental health conditions was selected. Effect sizes adjusted for relevant covariates were selected over unadjusted effect sizes. Self-report data for child outcomes were chosen, when available, over parent-report. When studies reported maternal and paternal data separately, maternal data were chosen given comparability to other studies.

Study Quality Assessment

The quality of each included study was assessed using the National Institutes of Health (NIH) Quality Assessment Tools for Observational Cohort and Cross-Sectional Studies and for Case-Control Studies (NIH National Heart Lung and Blood Institute, 2021). Study quality was assessed by one coder, with good agreement (>80%) being achieved by a second independent coder on a random 20% of included studies.

Statistical Analysis

Random-effects meta-analyses were conducted in Comprehensive Meta-Analysis Software 3.0 (CMA 3.0; BioStat; Borenstein et al., 2013) when there was sufficient data (>3 studies). Random-effects models were chosen as included studies were expected to differ in their populations, methods, and thus effect sizes; as such, a model that accounts for this heterogeneity was preferred (Borenstein et al., 2009; J. Higgins et al., 2022). To estimate prevalence rates and

95% confidence intervals (CIs) of mental health problems (i.e., clinically-elevated symptoms or diagnoses) among parents of children with chronic pain (Aim 1), all event rates and sample sizes were pooled. To estimate odds ratios (ORs) and 95% CIs for the association between parent mental health (i.e., symptoms and diagnoses) and the presence of child chronic pain (Aim 2a), all extracted data (e.g., ORs with CIs; event rates, means and standard deviations, or correlations with sample sizes) from studies that compared children with versus without chronic pain were pooled. To estimate correlations (r) and 95% CIs for the associations between parent mental health (i.e., symptoms and diagnoses) and the functioning of children with chronic pain, (Aim 2b), all correlations and sample sizes were pooled.

Heterogeneity of the effect sizes was assessed with the Cochran Q test and I^2 index. Moderator analyses were conducted in CMA 3.0 when these heterogeneity statistics were significant (i.e., Q considered significant at $p < 0.10$, I^2 when $\geq 50\%$; J. Higgins et al., 2003), and there was sufficient data, to explore potential sources of between-study variability. Categorical moderators were examined using subgroup meta-analyses (clinical vs. community samples, child chronic pain conditions of RAP, headache, fibromyalgia, vs. mixed; self-report vs. diagnostic interview measures; current vs. history of parent mental health). Subgroup meta-analyses were conducted when there was at least 10 studies in the overall analysis and at least 3 studies per subgroup (Borenstein et al., 2009). Continuous moderators were examined via univariable meta-regression (child mean age, % of child sample that was female, % of child sample that was White or nationality of study country). Meta-regression analyses were conducted when there was at least 3 studies in the overall analysis (Borenstein et al., 2009). Publication bias was examined through inspection of funnel plots and the Egger test (significance = $p < 0.10$; Egger et al., 1997). Sensitivity analyses were conducted using the leave-one-out approach.

Quality of the Evidence

The quality of the evidence base for each meta-analysis was assessed with the five domains identified in the GRADE (Grading of Recommendations, Assessment, Development, and Evaluation) approach (Guyatt et al., 2011; Schünemann et al., 2013) as applied to a body of non-intervention research (Iorio et al., 2015) similar to other meta-analyses of prevalence and/or prognosis (Akl et al., 2010; Jafari et al., 2022; Papola et al., 2018; Taddio et al., 2022).

Limitations in the quality of included studies was assessed with specific items from the NIH Quality Assessment Tools that aligned with key criteria in the GRADE Handbook (Schünemann et al., 2013). Inconsistency in estimates was based on variance in point estimates, degree of overlap of confidence intervals, and statistical significance of heterogeneity statistics.

Indirectness of outcome measurement was based on the use of parent-report (vs. child-report) of child outcomes. Imprecision of results was determined from the overall sample size (based on power calculations with $\alpha = 0.05$ and $\beta = 0.80$) and range of the confidence intervals of the estimates. Publication bias was determined from the funnel plots and Egger tests. Ratings on the five domains for each analysis were decided through consensus of two coders. A rating of the overall quality of evidence for each outcome was not generated given the lack of consensus on the starting rating for evidence from observational studies (Iorio et al., 2015; Yousefifard & Shafiee, 2023).

Results

Included Studies

The search identified 32,848 records and 278 reports were retrieved for full-text review (Figure 2.1). A total of 67 reports met inclusion criteria for the systematic review and 49 reports

had data suitable for meta-analysis. Studies that met inclusion criteria but were not included in meta-analyses are detailed in Supplementary Table 2.3 (Appendix B).

Study Characteristics

Table 2.1 presents the characteristics of the 49 studies included in the meta-analysis. Most recruited clinical ($n = 32$, 65.3%) vs. community ($n = 16$, 32.7%) samples ($n = 1$ recruited both), assessed mixed pain conditions ($n = 25$, 51.0%) or RAP ($n = 18$, 36.7%) vs. headache ($n = 5$, 10.2%) or fibromyalgia ($n = 3$, 6.1%) in children, assessed current ($n = 36$, 73.5%) vs. history of ($n = 14$, 28.6%) parent mental health using self-report measures ($n = 46$, 93.9%) or interview ($n = 3$, 6.1%). Overall, children had a mean age of 12.0 years and were predominately female ($Mean = 62.6%$) and White ($Mean = 83.1%$ with 22 studies not reporting race/ethnicity statistics).

Study quality ratings are presented in Supplementary Tables 2.4 and 2.5 (Appendix B). Study quality among cohort and cross-sectional studies was generally fair. The most common limitations of these studies were the use of cross-sectional designs, not including sample size calculations, and not adjusting analyses for potential confounding variables. However, most studies recruited their sample from similar populations, had clear eligibility criteria, and used psychometrically-sound measures for exposure and outcome variables. Some studies were limited by not reporting the specifics of their recruitment methods including the participation rate. Study quality among case-control studies was generally fair. Common limitations included not randomly selecting participants, not including specific recruitment details or sample size calculations, and not adjusting analyses for potential confounding variables. However, the majority of these studies recruited cases and controls from similar populations, clearly differentiated cases from controls, and used psychometrically-sound measures.

Aim 1. Prevalence of Mental Health Problems among Parents of Children with Chronic Pain

Meta-analytic results for the prevalence of mental health problems in parents of children with chronic pain are displayed in Figure 2.2. Results from moderator analyses are presented in Supplementary Table 2.6 (Appendix B). Publication bias findings are presented in Supplementary Figure 2.1 (Appendix B). Results did not vary widely in the sensitivity analyses (Supplementary Table 2.7 in Appendix B).

Parent Anxiety

The pooled effect size for the prevalence of anxiety from 12 studies (Beveridge et al., 2022; Birnie et al., 2020; Buonavolontà et al., 2010; Campo et al., 2007; Cohen et al., 2010; Eccleston et al., 2004; Hodges, Kline, Barbero, & Woodruff, 1985; Ibeziako et al., 2021; Ramchandani et al., 2006; Reid et al., 1997; Sieberg et al., 2011; Wiwe Lipsker et al., 2016) was 28.8% (95% CI 20.3-39.1). There was evidence of significant between-study heterogeneity of effect sizes ($Q = 215.42, p < 0.10; I^2 = 94.89$) and thus moderator analyses were conducted. Five moderators (type of child chronic pain, timing of parent mental health, child age, sex/gender, and race/ethnicity) were examined; however, none emerged as significant.

Parent Depression

The pooled effect size for the prevalence of depression from 17 studies (19 samples) (Benjamin et al., 2020; Beveridge et al., 2022; Birnie et al., 2020; Buonavolontà et al., 2010; Campo et al., 2007; Cohen et al., 2010; Eccleston et al., 2004; Hammond et al., 2019; Hodges, Kline, Barbero, & Flanery, 1985; Ibeziako et al., 2021; Law et al., 2019; Mortimer et al., 1992; Ramchandani et al., 2006; Reid et al., 1997; Sieberg et al., 2011; Tran et al., 2021; Wiwe Lipsker et al., 2016) was 20.0% (95% CI 15.7-25.2). Heterogeneity statistics were significant ($Q =$

199.26, $p < 0.10$; $I^2 = 90.97$) and six moderators were examined (type of sample, type of child chronic pain, timing of parent mental health, child age, sex/gender, and race/ethnicity). None of the moderators examined explained the between-study variability.

Parent General Distress

The pooled effect size for the prevalence of general distress from 8 studies (Birnie et al., 2020; Campo et al., 2007; Galli et al., 2009; Helgeland et al., 2010; Ibeziako et al., 2021; Incledon et al., 2016; Schneider et al., 2019; Sieberg et al., 2011) was 32.4% (95% CI 22.7-44.0). Statistically significant heterogeneity between the studies was found ($Q = 115.14$, $p < 0.10$; $I^2 = 93.92$). There were too few studies to examine categorical moderators; thus, only the three continuous moderators (child age, sex/gender, and race/ethnicity) were examined. Child age emerged as a significant moderator ($b = -0.35$, $p = 0.03$), with parents of younger (vs. older) children having higher prevalence rates (Supplementary Figure 2.2 in Appendix B).

Quality of the Evidence

Ratings of the quality of the evidence across the GRADE criteria for the prevalence of mental health problems among parents of children with chronic pain are displayed in Table 2.2. Overall, confidence in these estimates were reduced due to limitations in the quality of included studies (e.g., use of convenience samples, small sample sizes, non-validated measures) and inconsistency in the estimates (i.e., $I^2 > 75\%$) across all analyses. There were no substantial concerns with indirectness, imprecision, or publication bias.

Aim 2a. Associations Between Parent Mental Health and Presence of Child Chronic Pain

Meta-analytic results for the associations between parent mental health and the presence of child chronic pain are displayed in Figure 2.3. Results from moderator analyses are presented in Supplementary Table 2.8 (Appendix B). Publication bias findings are presented in

Supplementary Figure 2.3 (Appendix B). Results were robust across sensitivity analyses (Supplementary Table 2.9 in Appendix B).

Parent Anxiety

The pooled effect size for the association between parent anxiety and the presence of child chronic pain from 14 studies (Buonavolontà et al., 2010; Campo et al., 2007; Czyzewski et al., 2007; Darlington et al., 2012; Garber et al., 1990; Helgeland et al., 2010; Kashikar-Zuck et al., 2008; Kaufman et al., 1997; Kolaitis et al., 2022; Liakopoulou-Kairis et al., 2002; Ramchandani et al., 2006; Reid et al., 1997; Tran et al., 2021) was significant, OR = 1.91 (95% CI 1.51-2.41). There was some evidence of significant between-study heterogeneity of effect sizes ($Q = 23.53, p < 0.10; I^2 = 44.74$) and thus moderator analyses were conducted. Six moderators were examined (type of sample, type of child chronic pain, timing of parent mental health, child age, sex/gender, and race/ethnicity) but none emerged as significant.

Parent Depression

The pooled effect size for the association between parent depression and the presence of child chronic pain from 17 studies (18 samples) (Buonavolontà et al., 2010; Campo et al., 2007; Darlington et al., 2012; Garber et al., 1990; Hammond et al., 2019; Helgeland et al., 2010; Hodges, Kline, Barbero, & Flanery, 1985; Kashikar-Zuck et al., 2008; Kolaitis et al., 2022; Liakopoulou-Kairis et al., 2002; Moore et al., 2020; Mortimer et al., 1992; Ramchandani et al., 2006; Reid et al., 1997; Robinson et al., 1990; Tran et al., 2021; Walker & Greene, 1989) was significant, OR = 1.90 (95% CI 1.51-2.38). Statistically significant heterogeneity between the studies was found ($Q = 44.91, p < 0.10; I^2 = 62.15$), and six moderators were examined (type of sample, type of child chronic pain, timing of parent mental health, child age, sex/gender, and race/ethnicity). All categorical moderators examined were significant. Specifically, stronger

associations between parent depression and presence of child chronic pain were found in studies with clinical (OR = 3.29; 95% CI 2.40-4.51) vs. community (OR = 1.41; 95% CI 1.18-1.67) samples, among children with RAP (OR = 2.80; 95% CI 1.93-4.04) vs. headache (OR = 1.26; 95% CI 0.79-2.01) or mixed conditions (OR = 1.50, 95% CI 0.96-2.33), and in studies measuring current (OR = 3.28; 95% CI 2.19-4.92) vs. history (OR = 1.53; 95% CI 1.24-1.89) of parent depression. None of the continuous moderators explained the between-study variability.

Parent General Distress

The pooled effect size for the association between parent general distress and the presence of child chronic pain from 11 studies (Brown et al., 2022; Campo et al., 2007; Feldman et al., 2010; Helgeland et al., 2010; Hinze et al., 2023; Inledon et al., 2016; Jamison & Walker, 1992; Liakopoulou-Kairis et al., 2002; Lommel et al., 2011; Mikkelsen et al., 2021; Moayedi, A & Moayedi, F, 2015) was significant, OR = 1.74 (95% CI 1.47-2.05). There was some evidence of significant between-study heterogeneity of effect sizes ($Q = 16.45, p < 0.10; I^2 = 39.21$) and thus moderator analyses were conducted. Six moderators were examined (type of sample, type of child chronic pain, timing of parent mental health, child age, sex/gender, and race/ethnicity) but none emerged as significant.

Quality of the Evidence

Ratings of the quality of the evidence across the GRADE criteria for each estimate of the association between parent mental health and the presence of child chronic pain are displayed in Table 2.2. Overall, confidence in these estimates were reduced due to limitations in the quality of included studies across all analyses (e.g., use of convenience samples, small sample sizes and/or substantial attrition, unadjusted analyses), indirectness in the estimates (i.e., use of parent-report for child outcomes) for parent depression and parent general distress, and suspected publication

bias in the analyses of parent anxiety and parent depression. There were no concerns with inconsistency or imprecision in any of the estimates.

Aim 2b. Associations Between Parent Mental Health and Functioning of Children with Chronic Pain

Meta-analytic results for the associations between parent mental health and the functioning of children with chronic pain are presented in Table 2.3. Results from moderator analyses are presented in Supplementary Table 2.10 (Appendix B). Publication bias findings are presented in Supplementary Figures 2.4, 2.5, and 2.6 (Appendix B). Results did not vary widely in the sensitivity analyses (Supplementary Table 2.11 in Appendix B).

Parent Anxiety

Data was available to estimate pooled effect sizes for associations between parent anxiety and child pain intensity (Beveridge et al., 2022; Dutta et al., 2021; Eccleston et al., 2004; Evans et al., 2010; Goubert et al., 2006; Poppert Cordts et al., 2019; Sieberg et al., 2011; Vetter et al., 2013), physical functioning (Beveridge et al., 2022; Brown et al., 2021; Cohen et al., 2010; Dutta et al., 2021; Eccleston et al., 2004; Evans et al., 2010; Goubert et al., 2006; Poppert Cordts et al., 2019; Reid et al., 1997; Sieberg et al., 2011; Vetter et al., 2013), anxiety symptoms (Brown et al., 2021; Cohen et al., 2010; Dutta et al., 2021; Eccleston et al., 2004; Poppert Cordts et al., 2019; Ramchandani et al., 2007; Soltani et al., 2022; Vetter et al., 2013), and depression symptoms (Brown et al., 2021; Cohen et al., 2010; Dutta et al., 2021; Eccleston et al., 2004; Evans et al., 2010; Poppert Cordts et al., 2019; Soltani et al., 2022; Vetter et al., 2013). Meta-analyses revealed significant effect sizes for all associations: child pain intensity $r = 0.10$ (95% CI 0.01-0.19); physical functioning $r = 0.17$ (95% CI 0.11-0.23); anxiety symptoms $r = 0.22$ (95% CI 0.15-0.28); depression symptoms $r = 0.20$ (95% CI 0.10-0.29). There was some evidence of

significant heterogeneity of effect sizes for the associations with child pain intensity ($Q = 12.52$, $p < 0.10$; $I^2 = 44.08$) and depression symptoms ($Q = 16.12$, $p < 0.10$; $I^2 = 56.58$). Only continuous moderators (child age, sex/gender, and race/ethnicity) could be examined for each association as there were too few studies to examine categorical moderators. None of the moderators explained the between-study variability.

Parent Depression

Data was available to estimate pooled effect sizes for associations between parent depression and child pain intensity (Beveridge et al., 2022; Dutta et al., 2021; Eccleston et al., 2004; Evans et al., 2010; Goubert et al., 2006; Poppert Cordts et al., 2019; Sieberg et al., 2011; Vetter et al., 2013), physical functioning (Beveridge et al., 2022; Brown et al., 2021; Cohen et al., 2010; Dutta et al., 2021; Eccleston et al., 2004; Evans et al., 2010; Goubert et al., 2006; Poppert Cordts et al., 2019; Reid et al., 1997; Sieberg et al., 2011; Vetter et al., 2013), anxiety symptoms (Brown et al., 2021; Cohen et al., 2010; Dutta et al., 2021; Eccleston et al., 2004; Poppert Cordts et al., 2019; Soltani et al., 2022; Vetter et al., 2013), and depression symptoms (Brown et al., 2021; Cohen et al., 2010; Dutta et al., 2021; Eccleston et al., 2004; Evans et al., 2010; Poppert Cordts et al., 2019; Soltani et al., 2022; Vetter et al., 2013). Meta-analyses revealed significant effect sizes for all associations: child pain intensity $r = 0.12$ (95% CI 0.05-0.18); physical functioning $r = 0.20$ (95% CI 0.15-0.26); anxiety symptoms $r = 0.23$ (95% CI 0.15-0.30); and depression symptoms $r = 0.25$ (95% CI 0.15-0.34). There was evidence of significant heterogeneity of effect sizes for the association with child depression symptoms ($Q = 17.25$, $p < 0.10$; $I^2 = 59.41$). There were too few studies to examine categorical moderators so only continuous moderators (child age, sex/gender, and race/ethnicity) were examined. None emerged as significant.

Parent General Distress

Data was available to estimate the pooled effect size for the association between parent general distress and child physical functioning (Birnie et al., 2020; Brown et al., 2022; Logan & Scharff, 2005; Wendland et al., 2010). Meta-analysis revealed a significant and positive effect size for this association $r = 0.11$ (95% CI 0.05-0.17). There was no evidence of heterogeneity and thus moderator analyses were not conducted.

Quality of the Evidence

Ratings of the quality of the evidence across the GRADE criteria for each estimate of the association between parent mental health and the functioning of children with chronic pain are displayed in Table 2.2. Overall, confidence in these estimates were reduced due to limitations in the quality of included studies across all analyses (e.g., use of convenience samples, broad eligibility criteria, small sample sizes, unadjusted analyses), imprecision in the estimates (i.e., confidence interval overlapped with no effect) between parent anxiety-child pain intensity, parent depression-child pain intensity, and parent general distress-child physical functioning, and suspected publication bias in five of the nine analyses. There were no substantial concerns in any of these analyses for inconsistency or indirectness.

Discussion

Results revealed that mental health problems, specifically anxiety, depression, and general distress, were common among parents of children with chronic primary pain. Prevalence rates were higher than the general population (20.0% vs. 5.4% for depression; 28.8% vs. 6.7% for anxiety; Steel et al., 2014) but similar to other childhood chronic medical conditions such as cancer (21% for anxiety, 28% for depression; Warmerdam et al., 2019) and diabetes (22% for depression; Chen et al., 2023). Children whose parents reported higher (vs. lower or no)

symptoms of anxiety, depression, or general distress were almost twice as likely to endorse having chronic pain. Poorer parent mental health was also significantly related to the functioning of children with chronic pain, with higher (vs. lower or no) parent anxiety and depression symptoms associated with worse child pain intensity, physical functioning, and psychological functioning. Associations were small in size, suggesting that parent mental health is one of many factors associated with children's chronic pain and functioning. However, the associations, which examined heterotypic continuity (i.e., one disorder predicting a different disorder), are similar in magnitude to meta-analyses examining homotypic continuity (i.e., continuation of same disorder) in the intergenerational transmission of depression (Goodman et al., 2011) and chronic pain (K. S. Higgins et al., 2015). The results were also similar to recent meta-analyses examining associations between parent responses and children's chronic pain intensity (r s around 0.10) and disability (r s around 0.25; Donnelly et al., 2020; Harrison et al., 2020). These findings highlight the importance of including parent mental health, which has received less attention than these other parent variables (e.g., chronic pain, parent responses), in observational and treatment research for pediatric chronic pain.

Confidence in the current estimates, as assessed by GRADE, were reduced due to limitations in the included studies. The majority of studies recruited small, convenience samples from pediatric pain clinics and did not adjust analyses for potential covariates. However, these clinical studies used psychometrically-sound measures and had well-defined eligibility criteria. While many of the community studies had large samples and prospective designs, they were limited by low participation rates, high attrition, and use of single-item measures as well as parent-report for child outcomes, which limited certainty in some estimates of the association between parent mental health and presence of child chronic pain. Publication bias was also

suspected in many analyses; thus, effect sizes for the associations between parent mental health and child chronic pain, in particular parent depression-presence of child chronic pain, may be over-estimates as studies with non-significant findings may not have been included. Similarly, the confidence intervals for smaller correlations (i.e., $r < 0.15$; e.g., parent anxiety-child pain intensity) overlapped with no effect, reducing certainty in the statistical significance of these associations. Lastly, considerable heterogeneity reduced confidence in the prevalence estimates. Although heterogeneity was identified in other analyses, the magnitude was not enough to downgrade our confidence. To explore sources of between-study variability, however, moderator analyses were conducted for these estimates. These analyses found that the prevalence of general distress in parents was higher in studies with younger (vs. older) children and the association between parent depression and presence of child chronic pain was stronger in studies with clinical (vs. community) samples, studies of children with RAP (vs. headache or mixed conditions), and studies measuring current (vs. history of) parent depression. All other moderator analyses were either non-significant, potentially because they were underpowered, or could not be conducted due to insufficient data.

Most studies included in this meta-analysis were cross-sectional, thus limiting our ability to estimate prospective associations between parent mental health and child chronic pain. However, several studies provided evidence that poor parent mental health increases risk for pediatric chronic pain, with prospective studies showing that parent mental health symptoms in early childhood (6 months to 3 years) predict the presence of child chronic pain at later ages (6-14 years; Fryer et al., 2017; Helgeland et al., 2010; Kolaitis et al., 2022; Ramchandani et al., 2006) and longitudinal studies demonstrating that poor parent mental health predicts the continuation of child pain (Hinze et al., 2023; Ramchandani et al., 2007). Research on the

intergenerational transmission of risk for psychopathology has shown that parent depression and anxiety increases child risk for poor social and emotional outcomes (Goodman et al., 2011; Lawrence et al., 2019), likely through neurobiological (e.g., disruptions to the stress response system) and psychosocial (e.g., parenting practices) processes (Goodman, 2020). These processes may also be relevant to the transmission of risk from parent mental health to child chronic pain. The high co-occurrence of chronic pain and psychiatric disorders has been explained through the multitude of risk factors shared by these conditions (e.g., serotonin availability, avoidance behaviours, catastrophizing, attention biases; Holley et al., 2016; Soltani et al., 2019; Vinall et al., 2016), which may also underlie the association between parent mental health and child chronic pain. Although much of this literature remains theoretical (Stone & Wilson, 2016; Vinall et al., 2016), emerging empirical research has started to identify psychosocial variables that mediate the association between parent mental health and child chronic pain (Brown et al., 2021; Darlington et al., 2012; Hammond et al., 2019; Neville et al., 2018). For example, one prospective study found that parent depression in early childhood predicted greater child anxiety and depression symptoms in middle childhood, which in turn predicted migraine incidence in adolescence (Hammond et al., 2019).

The presence and severity of chronic primary pain in children likely also has an impact on the psychological functioning of parents. Indeed, the emotional consequences of having a child with pain that cannot be readily attributed to underlying pathology has been supported in qualitative research (Gaughan et al., 2014; Jordan et al., 2007; Le et al., 2019; Neville et al., 2019; Ngo et al., 2023). Parents of children with chronic primary pain experience many of the same challenges as parents of children with other medical conditions (e.g., cancer, diabetes) including trying to manage the child's condition and related disability, navigating the medical

system and advocating for care, disruptions to social and occupational functioning, and financial burdens (Le et al., 2019; Ngo et al., 2023; Palermo & Eccleston, 2009). In addition, these parents must grapple with the lack of medical explanation for their child's pain. For many parents, this diagnostic uncertainty results in feeling anxious about the child's prognosis, feeling helpless or hopeless about improving the pain, feeling disbelieved and dismissed by others including healthcare providers, feeling guilty or anxious they have not done enough to find a cause (or are doing too many tests), fearing there is a serious cause that has been missed, and feeling exhausted in the search for a cause and cure (Gaughan et al., 2014; Jordan et al., 2007; Le et al., 2019; Neville et al., 2019; Ngo et al., 2023). The impact of child chronic pain on parent mental health could not be examined in the current analysis as only a few studies examined parent mental health as an outcome variable and most studies were cross-sectional. Indeed, compared to other chronic medical conditions, there has been less focus on the psychological impact that caring for a child with chronic pain has on parents, particularly in the quantitative literature (Palermo & Eccleston, 2009; Pinqart, 2019).

The current findings highlight considerations for future research. Overall, more research is needed to increase the robustness of the current estimates and add nuance to the complex role that parent mental health plays in pediatric chronic pain. First, studies should recruit larger samples, adjust analyses for potential confounders, and pre-register their methods to improve study quality and reduce publication bias. Future research should focus on including more fathers, boys, and gender diverse youth and adopting antiracism practices (Letzen et al., 2022) to reduce gender- and race-based disparities in pain assessment, treatment, and research. Many child outcomes (e.g., pain duration, quality of life, sleep) and parent posttraumatic stress disorder could not be examined as too few studies provided the necessary data. Research on these less

studied, yet important, variables is needed. Lastly, more prospective research is needed to examine (1) risk and protective factors mediating the association between parent mental health and child chronic pain and (2) the bidirectionality of dyadic associations between parent and child mental health and chronic pain.

This future research can help inform interventions that break the intergenerational continuity of health problems, effectively support parents, and improve child outcomes. Parent interventions for pediatric chronic pain predominately focus on parenting behaviours, with little focus on parents' own health (Lee et al., 2021). However, addressing mental health may be critical for improving the limited effectiveness of current psychological interventions for pediatric chronic pain (Eccleston et al., 2021; Vinall et al., 2016), with emerging research showing that interventions targeting parent distress have downstream effects on children's pain-related functioning (Palermo et al., 2016). The current findings, which demonstrate the prevalence of parent mental health problems and their association with child functioning, further corroborate the importance of addressing parent mental health in the treatment of pediatric chronic pain.

Strengths of this study include the comprehensive search strategy, examination of multiple aspects of child chronic pain (i.e., presence, functioning), inclusion of moderator analyses, and quality assessment of each included study and meta-analytic finding. Limitations include the correlational nature of the results, which limits causal inferences, the low number of studies in some analyses, which limits the robustness of some results, the lower quality of some included studies, which increases risk of bias (e.g., unadjusted analyses) and reduces confidence in the current estimates, and the number of analyses conducted, which may have inflated the risk for type I errors.

Conclusions

Mental health problems, including anxiety, depression, and general distress, are common among parents of children with chronic pain, and higher (vs. lower or no) symptoms of anxiety, depression, and general distress in parents are significantly associated with the presence of chronic pain and worse functioning in children. Limitations in the quality of included studies, publication bias, and heterogeneity reduced confidence in the current findings. Further research is needed to better understand the role of parent mental health in pediatric chronic pain.

Table 2.1*Characteristics of Studies Included in Meta-analysis*

Study	Study characteristics				Child characteristics					Parent characteristics			
	Country	Setting	Data type ^a	N ^b	Age, M	% female	% White or nationality of study country	Chronic pain condition	Pain variables measured	Age, M	Included parent, % mothers ^c	Mental health variables measured ^d	Timing, measure type
Clinical samples with cohort/cross-sectional design													
Benjamin, 2020	USA	Outpatient pain day program	C	268	16.10	72.0	95.1	Mixed	Prev	48.65	Either, 90.3	DEP	Cur, SR
Beveridge, 2022 ^e	Canada	Outpatient pain clinics	L	192	14.38	75.5	81.8	Mixed	Char ⁱ , Phys ⁱ	44.89	Either, 92.2	ANX, DEP, PTSD	Cur, SR
Birnie, 2020	USA	Outpatient pain clinic	C	448	14.57	73.0	60.7	Mixed	Phys ⁱ , Exp ⁱ	45.86	Either, 90.0	ANX, DEP, DIS	Cur, SR
Cohen, 2010	UK	Outpatient rheumatology clinic and pain day program	C	102	14.80	74.6	99.1	Mixed	Phys ⁱ , Exp ⁱ , Psys ^c , Role ⁱ	43.80	Either, 94.1	ANX, DEP	Cur, SR
Dutta, 2021	USA	Outpatient GI clinics	C	93	11.68	60.2	91.4	RAP	Char ⁱ , Phys ⁱ , Exp ⁱ , Psys ^c	NR	Either, 89.2	ANX, DEP	Cur, SR
Eccleston, 2004	UK	Outpatient pain service	C	75	14.45	71.3	NR	Mixed	Char ⁱ , Phys ⁱ , Exp ⁱ , Psys ^c	NR	Either, NR	ANX, DEP	Cur, SR
Evans, 2010	USA	Outpatient pain clinic	C	179	14.34	71.5	69	Mixed	Char ^{i,j} , Phys ⁱ , Psys ^c	NR	Only mothers	ANX, DEP	Cur, SR

Galli, 2009	Italy	Outpatient headache clinic	C	198	M: 10.82, F: 11.13	54.0	NR	Headache	Prev	M: 40.77, F: 43.70	Both	DIS	Cur, IN
Goubert, 2006	UK	Outpatient rheumatology and pain clinics	C	107	14.58	80.4	NR	Mixed	Char ⁱ , Phys ^{i,j} , Role ⁱ	M: 42.82, F: 49.50	Either, 95.3	ANX, DEP	Cur, SR
Ibeziako, 2021	USA	Inpatient for SSRDs	C	213	13.92	77.5	80.3	Mixed	Prev	NR	Both	ANX, DEP, DIS	His, NR
Law, 2019 ^f	USA	Outpatient neurology clinic and community ads	C	239	14.85	66.8	82.7	Headache	Prev	44.00	Either, 94.0	DEP	Cur, SR
Logan, 2005	USA	Outpatient and primary care clinics	C	70	12.1	67.9	80.8	Mixed	Char ⁱ , Phys ⁱ	NR	Both	DIS	Cur, SR
Poppert-Cordts, 2019	Canada	Outpatient pain program	C	146	12.97	68.5	NR	Mixed	Char ⁱ , Phys ⁱ , Exp ⁱ , Psyc ⁱ	NR	Either, 90.4	ANX, DEP	Cur, SR
Schneider, 2019	Switzerland	Outpatient pain clinic	C	135	13.95	80.0	NR	Mixed	Prev	NR	Both	DIS	His, SR
Sieberg, 2011	USA	Outpatient pain clinic	C	157	13.7	87	92	Mixed	Char ⁱ , Phys ⁱ	NR	Either, 94	ANX, DEP, DIS	Cur, SR
Soltani, 2022 ^e	Canada	Outpatient pain program	C	156	14.27	71.8	85.9	Mixed	Phys ⁱ , Psyc ⁱ	NR	Either, 90.3	ANX, DEP, PTSD	Cur, SR

Tran, 2021	France	Previous patients from GI units	C	88	15.0	44.1	NR	RAP	Presence ^k , Psyc ⁱ	NR	Either, 79.6	ANX, DEP	Cur, SR
Vetter, 2013	USA	Outpatient pain clinic	C	99	13.2	71	81	Mixed	Char ⁱ , Phys ⁱ , Exp ⁱ , Psyc ⁱ	NR	Either, 94	ANX, DEP	Cur, SR
Wendland, 2010	USA	Outpatient GI clinic	C	100	11.72	65	79	RAP	Char ⁱ , Phys ⁱ , Psyc ⁱ	NR	Only mothers	DIS	Cur, SR
Wiwe Lipsker, 2016	Sweden	Outpatient pain clinic	C	263	14.0	72	NR	Mixed	Prev	44.4	Either, 81	ANX, DEP	Cur, SR
Clinical samples with case-control design													
Buonavolonta, 2010	Italy	-	C	-	-	-	-	RAP	Presence ^k	NR	Both	ANX, DEP, DIS	Cur, SR
Cases	-	Outpatient pediatric clinic	-	103	7.71	50.4	NR	-	-	-	-	-	-
Controls	-	Primary care clinic	-	65	7.48	55.3	NR	-	-	-	-	-	-
Campo, 2007	USA	Primary care clinics	C	-	-	60.0	84.4	RAP	Presence ^k	-	Only mothers	ANX, DEP, DIS	His, IN & Cur, SR
Cases	-	-	-	59	11.3	-	-	-	-	39.5	-	-	-
Controls	-	-	-	76	12.1	-	-	-	-	42.3	-	-	-
Czyzewski, 2007	USA	Health care network and outpatient GI practices	C	-	8.6	-	-	RAP	Presence ^k	NR	Only mothers	ANX	Cur, SR

Cases	-	-	-	82	-	74.4	61.0	-	-	-	-	-	-
Controls	-	-	-	38	-	55.3	63.2	-	-	-	-	-	-
Garber, 1990	USA	Outpatient clinics	C	-	NR	-	-	RAP	Presence ^k	NR	NR	ANX, DEP	Cur, SR
Cases	-	-	-	13	-	NR	NR	-	-	-	-	-	-
Controls ^g	-	-	-	16	-	NR	NR	-	-	-	-	-	-
Hodges, 1985a ^e	USA	-	C	-	-	-	-	RAP	Presence ^k	NR	Both	ANX	Cur, SR
Cases	-	Outpatient GI service	-	30	M: 10, F: 11	63.3	NR	-	-	-	-	-	-
Controls ^g	-	Community	-	42	10	47.6	NR	-	-	-	-	-	-
Hodges, 1985b ^e	USA	-	C	-	-	-	-	RAP	Presence ^k	NR	Both	DEP	Cur, SR
Cases	-	Outpatient GI service	-	25	M: 10, F: 11	63.3	NR	-	-	-	-	-	-
Controls ^g	-	Community	-	42	10	47.6	NR	-	-	-	-	-	-
Kashikar-Zuck, 2008	USA	-	C	-	-	-	-	Fibro	Presence ^k	NR	Only mothers	ANX, DEP	Cur, SR
Cases	-	Outpatient rheumatology clinic	-	47	14.85	89.4	87.2	-	Phys ⁱ	-	-	-	-
Controls	-	Classrooms of cases	-	46	15.02	91.3	87.0	-	-	-	-	-	-
Kaufman, 1997	USA	Outpatient GI clinic	C	-	-	-	-	RAP	Presence ^k	NR	Only mothers	ANX	Cur, SR
Cases	-	-	-	24	13.8	62	96	-	-	-	-	-	-
Controls ^g	-	-	-	19	14.1	63	100	-	-	-	-	-	-

Liakopoulou-Kairis, 2002	Greece	-	C	-	-	-	-	Mixed	Presence ^k	-	Only mothers	ANX, DEP, DIS	Cur, SR
Cases	-	Outpatient clinics	-	69	10.0	62.3	NR	-	-	36.2	-	-	-
Controls	-	University hospital	-	60	9.6	43.3	NR	-	-	39.1	-	-	-
Moayedi, 2015	Iran	NR	C	-	11.52	-	-	RAP	Presence ^k	NR	Only mothers	DIS	Cur, SR
Cases	-	-	-	38	-	65.8	NR	-	-	-	-	-	-
Controls ^g	-	-	-	42	-	52.4	NR	-	-	-	-	-	-
Reid, 1997	Canada	-	C	-	-	-	-	Fibro	Presence ^k	-	NR	ANX, DEP	Cur, SR
Cases	-	Outpatient rheumatology clinic	-	15	14.5	86.7	NR	-	Phys ⁺⁺ , Role ^l	41.0	-	-	-
Controls ^g	-	Community and friends	-	15	14.6	86.7	NR	-	-	44.0	-	-	-
Robinson, 1990	UK	-	C	-	9.4	-	NR	RAP	Presence ^k	NR	Both	DEP	His, SR
Cases ^g	-	Outpatient pediatric clinic	-	40	-	52.5	-	-	-	-	-	-	-
Controls ^g	-	Local authority health records	-	40	-	52.5	-	-	-	-	-	-	-
Walker, 1989	USA	Outpatient clinics	C	-	NR	-	-	RAP	Presence ^k	NR	Both	ANX, DEP	Cur, SR
Cases	-	-	-	41	-	46.3	97.6	-	-	-	-	-	-

Controls ^g	-	-	-	41	-	46.3	92.7	-	-	-	-	-	-
Community samples with cohort/cross-sectional design													
Brown, 2021 ^e	Germany	Secondary schools	C	182	12.7	51	97	Mixed	Phys ⁱ , Psyc ⁱ	NR	Only mothers	ANX, DEP	Cur, SR
Brown, 2022 ^e	Germany	Secondary schools	C	1450	12.68	50	96.5	Mixed	Presence ⁱ , Char ⁱ , Phys ⁱ , Psyc ⁱ	NR	Either, 82	DIS	Cur, SR
Darlington, 2012	Netherlands	Primary schools	L	2093	13.6	51.3	89.4	Mixed	Presence ⁱ	40.45	Either, 95.6	ANX, DEP	His, SR
Feldman, 2010	USA and Puerto Rico	Representative sample of households of Puerto Rican background	C	2491	9.2	49.1	0	RAP and Headache ^b	Presence ^j	NR	Either, 96.3	DIS	His, SR
Hammond, 2019 ^f	Canada	Canadian households	L	2313	14.5	51.1	NR	Headache	Presence ^j	NR	Either, 90.6	DEP	His, SR
Helgeland, 2010	Norway	Child health clinics	L	456	14	56	> 95	RAP	Presence ⁱ	30.5	Only mothers	ANX, DEP, DIS	His, SR
Hinze, 2023	UK	Representative sample of children in Great Britain	C	7977	10.54	48.5	86.8	Mixed	Presence ^j	NR	Either, NR	DIS	Cur, SR
Inledon, 2016	Australia	Representative sample of Australian children	L	3821	12.4	49.1	NR	Mixed	Presence ^j	NR	Only mothers	DIS	His, SR

Jamison, 1992	USA	Adult pain clinic	C	42	11.69	38.1	93	Mixed	Presence ^j	37.81	Either, NR	DIS	Cur, SR
Kolaitis, 2022	Netherlands	Routine prenatal visit	L	3962	6	50	NR	Mixed	Presence ^j	M: 31.75, F: 33.91	Both	ANX, DEP	His, SR
Lommel, 2011	USA	Adolescent psychiatric hospital	C	55	NR	100	NR	Fibro	Presence ^k	42.0	Only mothers	DIS	Cur, SR
Mikkelsen, 2021	Norway	Secondary schools	C	508	14.0	55.3	NR	Mixed	Presence ⁱ	45.2	Either, 77.4	DIS	Cur, SR
Moore, 2020	USA	State birth records	L	50 parents, 100 children	6	51	60	Mixed	Presence ^j	36.56	Only mothers	DEP	His, SR
Mortimer, 1992	UK	Age sex register	C	1083	NR	NR	NR	RAP and Headache ^b	Presence ^k	NR	Only mothers	DEP	His, IN
Ramchandani, 2006 ^e	UK	Community, and routine prenatal visits	L	8272	6.75	48.5	96.2	RAP	Presence ^j	NR	Both	ANX, DEP	His, SR
Ramchandani, 2007 ^e	UK	Community, and routine prenatal visits	L	860	7	NR	NR	RAP	Presence ^j , Psyc ^k , Role ^j	NR	Only mothers	ANX	His, SR

Abbreviations. ANX, anxiety; C, cross-sectional; Char, pain characteristics; Cur, current symptoms assessed; DEP, depression; Exp, pain-related experiences; F, Female; Fibro, fibromyalgia; GI, gastroenterology or gastrointestinal; DIS, general distress; His, history of symptoms/diagnosis assessed; IN, diagnostic interview; L, longitudinal; M, Male; NR, not reported; Phys, physical functioning; Presence, presence of chronic pain; Prev, examined prevalence of parent mental health problems; Psyc, psychological functioning;

PTSD, posttraumatic stress disorder; QoL, quality of life; RAP, recurrent abdominal pain; Role, role functioning; SR, self-report measure; SSRDs, somatic symptom and related disorders

Note. ^a Based on data extracted for meta-analysis (e.g., only cross-sectional data extracted from longitudinal data). Law (2019), Soltani (2020), and Hinze (2023) also have longitudinal data relevant to the research question.

^b Based on data relevant to research question; Ns vary slightly between variables/analyses

^c Of the 49 studies, 21 (42.9%) assessed one parent, with the majority being mothers (*Mean*=89.8%); 16 (32.7%) studies assessed only mothers; 10 (20.4%) studies included both parents; and 2 (4.1%) were not clearly reported

^d 30 (61.2%) studies assessed parent anxiety; 33 (67.3%) studies assessed parent depression; 19 (38.8%) studies assessed parent general distress

^e Have overlapping samples with other reports but distinct data (Beveridge, 2022 & Soltani, 2022; Hodges, 1985a & Hodges, 1985b; Brown, 2021 & Brown, 2022; Ramchandani, 2006 & Ramchandani, 2007)

^f Have distinct cohorts that were analysed separately in meta-analyses. Specifically, Hammond (2019) examined participants reporting headache separately from those reporting migraine and Law (2019) reported participants recruited from a clinical setting (*n* = 157) separately from those recruited from community settings (*n* = 82)

^g Other case or control groups were included in the original study but not included in the current meta-analysis

^h Only one pain condition examined as groups were not mutually exclusive (Feldman (2010) = RAP examined; Mortimer (1992) = Headache examined)

ⁱ Child self-report

^j Parent-report

^k Clinician/researcher diagnosed

^l Other (e.g., school records)

Table 2.2*Quality of the Evidence for Each Meta-analysis Based on GRADE Criteria*

	Limitations	Inconsistency	Indirectness	Imprecision	Publication bias
Aim 1. Prevalence of mental health problems among parents of children with chronic pain					
Parent anxiety	Serious ¹	Serious ⁵	Not serious	Not serious	Undetected
Parent depression	Serious ¹	Serious ⁵	Not serious	Not serious	Undetected
Parent general distress	Serious ²	Serious ⁵	Not serious	Not serious	Undetected
Aim 2a. Associations between parent mental health and presence of child chronic pain					
Parent anxiety	Serious ³	Not serious	Not serious	Not serious	Suspected ⁸
Parent depression	Serious ³	Not serious	Serious ⁶	Not serious	Strongly suspected ⁹
Parent general distress	Serious ³	Not serious	Serious ⁶	Not serious	Undetected
Aim 2b. Associations between parent mental health and functioning of children with chronic pain					
Parent anxiety – child pain intensity	Serious ⁴	Not serious	Not serious	Serious ⁷	Suspected ⁸
Parent anxiety – child physical functioning	Serious ⁴	Not serious	Not serious	Not serious	Undetected
Parent anxiety – child anxiety symptoms	Serious ⁴	Not serious	Not serious	Not serious	Suspected ⁸
Parent anxiety – child depression symptoms	Serious ⁴	Not serious	Not serious	Not serious	Undetected
Parent depression – child pain intensity	Serious ⁴	Not serious	Not serious	Serious ⁷	Suspected ⁸
Parent depression – child physical functioning	Serious ⁴	Not serious	Not serious	Not serious	Suspected ⁸

Parent depression – child anxiety symptoms	Serious ⁴	Not serious	Not serious	Not serious	Undetected
Parent depression – child depression symptoms	Serious ⁴	Not serious	Not serious	Not serious	Undetected
Parent general distress – child physical functioning	Serious ⁴	Not serious	Not serious	Serious ⁷	Suspected ⁸

Abbreviations. GRADE, Grading of Recommendations Assessment, Development, and Evaluation.

Note.

¹ Predominately included convenience samples from pediatric pain clinics; several studies with small sample sizes ($n < 100$); several studies used non-validated measures for assessing parent mental health

² Several studies included convenience samples from pediatric pain clinics; one study with small sample size ($n < 100$); several studies used non-validated measures for assessing parent mental health

³ Case-control studies included convenience samples and had small sample sizes; many prospective cohort studies had $> 20\%$ attrition; many studies did not adjust analyses for covariates; several studies used non-validated measures for assessing parent mental health

⁴ Predominately included convenience samples from pediatric pain clinics with broad eligibility criteria; generally small sample sizes ($n \cong 50-150$); effect sizes did not adjust analyses for covariates

⁵ Wide variance of point estimates across studies, minimal or no overlap of confidence intervals, and considerable heterogeneity identified in statistical tests (i.e., $I^2 > 75\%$)

⁶ More than 20% of included studies used parent-report (vs. child-report) of child outcomes

⁷ Confidence interval overlaps with no effect (i.e., $r = 0.05$)

⁸ Asymmetry detected in the funnel plot

⁹ Asymmetry detected in the funnel plot and the Egger test was significant ($p < 0.10$)

Table 2.3

Pooled Effect Sizes (r) for the Correlations Between Parent Mental Health and the Functioning of Children with Chronic Pain

Parent mental health variable	Child functioning variable	k	N	r	95% CI	Q	I^2
Anxiety							
	Pain intensity	8	890	0.10	0.01, 0.19	12.52	44.08
	Physical functioning	11	1200	0.17	0.11, 0.23	11.29	11.45
	Anxiety symptoms	8	1698	0.22	0.15, 0.28	4.52	0.00
	Depression symptoms	8	924	0.20	0.10, 0.29	16.12	56.58
Depression							
	Pain intensity	8	890	0.12	0.05, 0.18	5.55	0.00
	Physical functioning	11	1200	0.20	0.15, 0.26	10.70	6.55
	Anxiety symptoms	7	838	0.23	0.15, 0.30	8.36	28.23
	Depression symptoms	8	924	0.25	0.15, 0.34	17.25	59.41
General distress							
	Physical functioning	4	1072	0.11	0.05, 0.17	2.73	0.00

Abbreviations. CI, confidence interval; k, studies

Note. Significant correlations ($p < .05$ for r statistics, $p < .10$ for Q statistics, $I^2 \geq 50\%$) bolded.

Figure 2.1

PRISMA Flow Chart

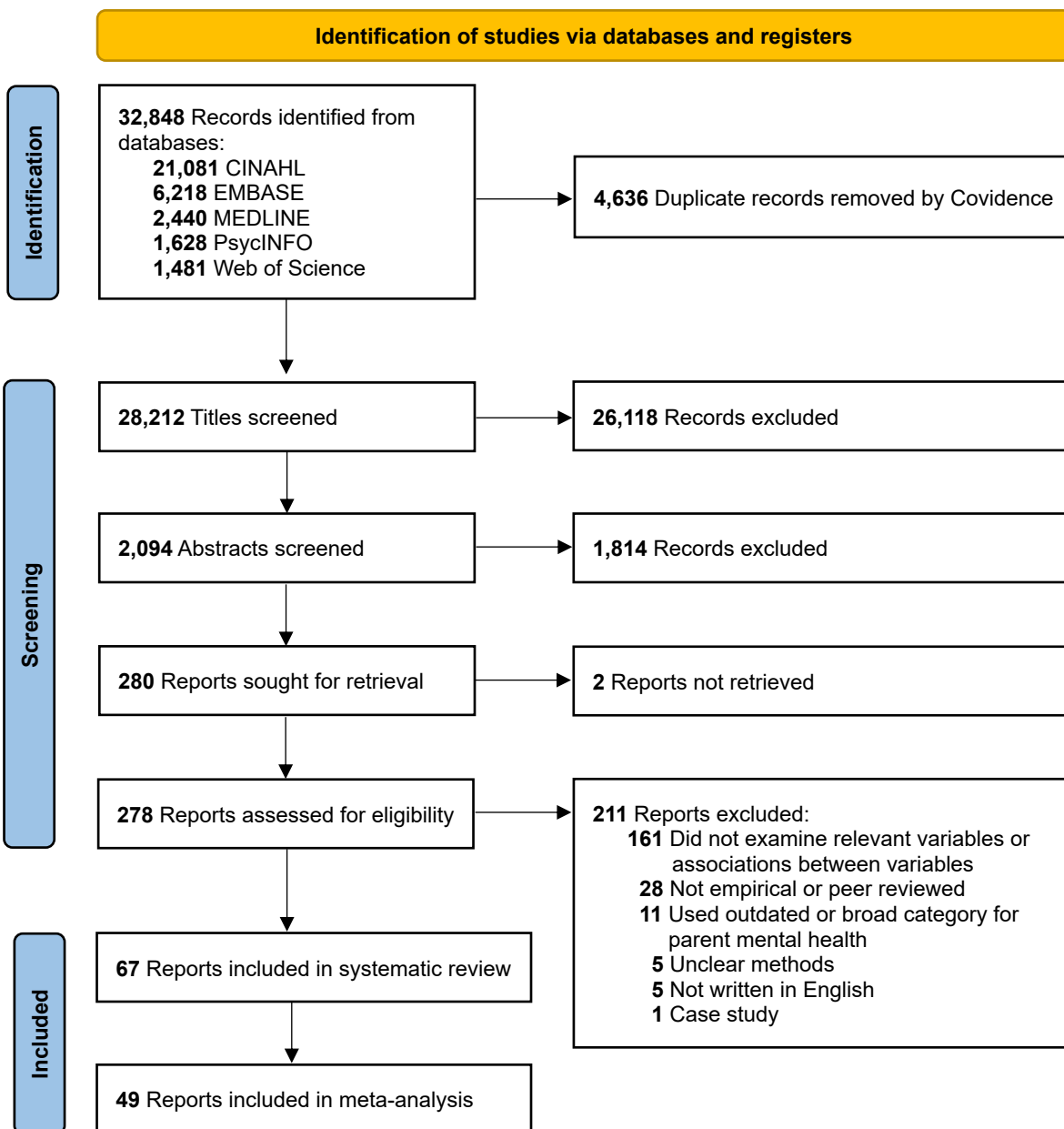
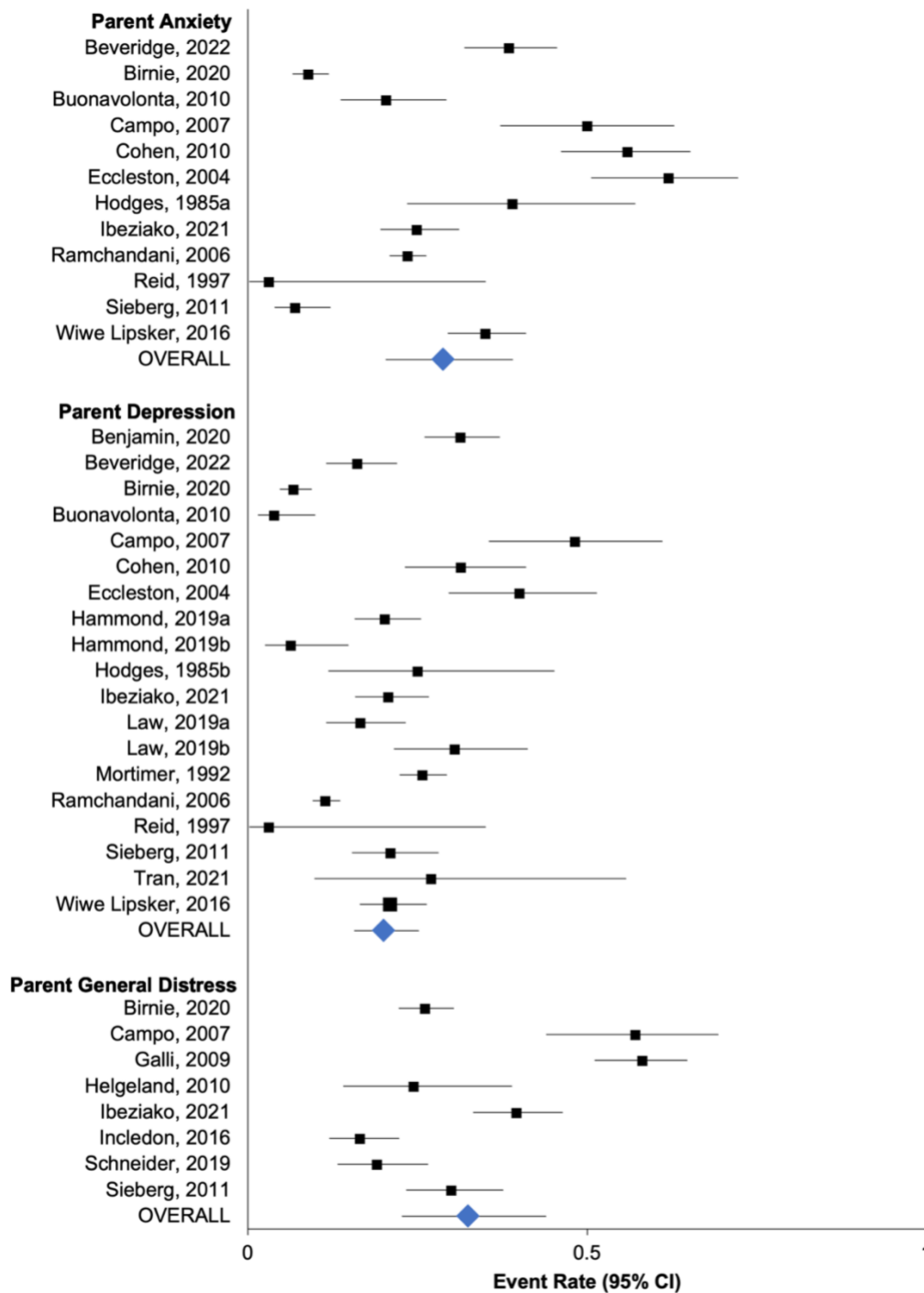


Figure 2.2

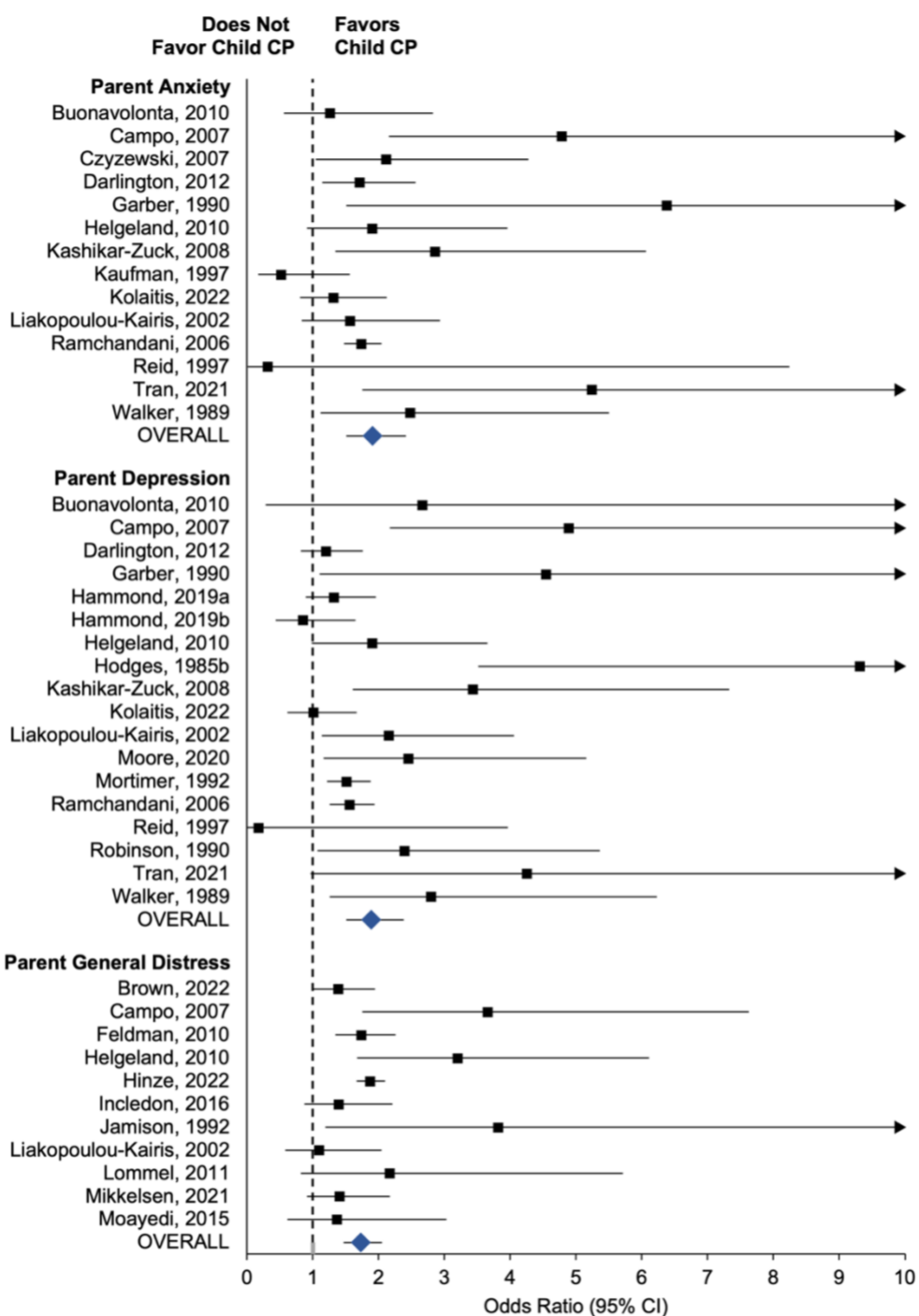
Prevalence Rates (%) of Mental Health Problems Among Parents of Children with Chronic Pain



Abbreviations. CI, confidence interval.

Figure 2.3

Associations Between Parent Mental Health and the Presence of Child Chronic Pain



Abbreviations. CI, confidence interval; CP, chronic pain

**CHAPTER THREE: Parent Anxiety, Depression, Protective Responses, and Parenting
Stress in the Context of Parent and Child Chronic Pain: A Daily Diary Study of Parent
Variability**

Abstract

Parents with (versus without) chronic pain report poorer psychosocial functioning (e.g., worse mental health, parenting difficulties), which has been linked to poorer child outcomes (e.g., child pain). However, emerging research suggests that individuals vary in their functioning from day-to-day, particularly those with chronic pain. This study used daily diaries to compare parents with (versus without) chronic pain on variability in their anxiety, mood, protective responses, and parenting stress. We also examined parent chronic pain status as a moderator of the associations between parent variability and youth daily pain and interference. Participants were 76 youth with chronic pain ($M_{\text{age}} = 14.26$; 71.1% female) and one of their parents (89.5% mothers; $n = 38$ or 50.0% endorsing chronic pain). Parents and youth completed self-report questionnaires and seven days of diaries. Parent variability was calculated to reflect the frequency and size of day-to-day changes. Multilevel models revealed that parents with (versus without) chronic pain were significantly more variable in their parenting stress, but not in their anxiety, mood, or protective responses. Contrary to hypotheses, parent variability was *not* significantly related to youth daily pain intensity or interference and parent chronic pain did *not* moderate any associations. Instead, mean levels of parent anxiety, protective responses, and parenting stress across the week significantly predicted youth daily pain interference. Findings suggest that while variability was observed among parents (with and without chronic pain) of youth with chronic pain, it did not significantly predict youth's daily pain-related functioning. Further research is needed to confirm these initial findings.

Introduction

Parent chronic pain increases risk for poor child outcomes including child pain, disability, and social-emotional difficulties (K. S. Higgins et al., 2015). Several mechanisms are posited to underlie this intergenerational transmission of risk including differences in general and pain-specific distress and parenting behaviours (Stone & Wilson, 2016). Indeed, research has shown that parents with chronic pain endorse greater anxiety and depressive symptoms, respond with more protective behaviours to child pain, and report greater difficulties with parenting tasks, on average, than parents without chronic pain (Evans et al., 2005; Fussner et al., 2018; Stone et al., 2018; Wilson & Fales, 2015). These differences are related to worse functioning in their children (Beveridge et al., 2024; Donnelly et al., 2020; Harrison et al., 2020). However, this research has relied on retrospective measures that assess parents' typical feelings or behaviours. Emerging research using daily diary methodology in pediatric chronic pain has shown parents' psychosocial functioning can vary greatly from day-to-day (Beeckman, Simons, et al., 2019; Neville, Griep, et al., 2020). Moreover, this within-person variability in parent functioning has shown different associations with adolescents' pain-related outcomes than parents' mean levels of functioning (Beeckman, Simons, et al., 2019).

Parents with chronic pain may have greater variability in their day-to-day functioning than parents without chronic pain. Daily diary studies have demonstrated that adults with versus without chronic pain are more variable in their daily mood, and aspects of their pain condition (e.g., severity, disability) are related to this variability (Gerhart et al., 2018; Rost et al., 2016, 2021). In qualitative studies, parents with chronic pain described having daily fluctuations in their pain, which impacted their mood and parenting and increased parenting stress (e.g., feeling frustrated they were unable to fulfill their parenting role; Barlow et al., 1999; Wilson & Fales,

2015). For example, one mother with chronic pain explained “some days I just can’t do anything. And it drains you – physically and emotionally” (Evans & de Souza, 2008). Parents perceived their pain-related variability to negatively impact their children, with another mother explaining she “could be happy one second and the next minute be really foul, and [my son’s] thinking, “What have I done?”” (Evans & de Souza, 2008).

Greater parent variability may negatively impact children’s pain-related functioning through parent modeling of maladaptive responses to mental and physical health symptoms or through less predictable parent behaviours and home environments, which could decrease child coping abilities (e.g., emotion regulation) and increase child stress and pain (Goodman & Gotlib, 1999; Manczak et al., 2017, 2019; Stone & Wilson, 2016). Previous research with non-pain community samples has shown that greater day-to-day variability in parent-youth interactions (e.g., positive statements between parent and youth, parent knowledge of the youth’s activities) is associated with poor youth outcomes including more physical health complaints, above and beyond the mean level of these variables (Lippold et al., 2015, 2017). However, a daily diary study examining associations between maternal distress, harsh parenting, and youth distress among mothers with versus without chronic pain found a weaker association between mother and youth anger, despite mothers with chronic pain endorsing greater anger (Downey et al., 1999). The authors posited that the children of mothers with chronic pain may be less affected by their mother’s anger because they can attribute it to her pain condition as opposed to something the youth has done. Thus, parents with chronic pain may be more variable in their day-to-day functioning than parents without chronic pain, but this variability may not significantly affect their children.

Parent variability in the context of parent chronic pain and child chronic pain has not yet been examined in a quantitative study. This initial study aimed to: (1) compare parents with versus without chronic pain on variability in anxiety, mood, protective responses, and parenting stress; and (2) examine associations between this parent variability and the daily pain experiences of youth with chronic pain. We hypothesized that: (1) parents with chronic pain would display greater variability in their anxiety, mood, protective responding, and parenting stress than parents without chronic pain; and (2) greater parent variability would be significantly related to worse youth pain intensity and interference, with weaker associations for dyads where the parent also has chronic pain.

Methods

This cohort study used data from a larger study, the Pain and Mental Health in Youth (PATH) study, which was designed to examine cognitive, behavioural, neurobiological, and social factors underlying the co-occurrence of mental health disorders and pediatric chronic pain. The PATH study collected data at three timepoints (i.e., baseline, one-month follow-up, and three-month follow-up) using a variety of methods including questionnaires, daily diaries, actigraphy, laboratory-based tasks, and semi-structured interviews. This study used observational data from questionnaires and daily diary surveys administered at the baseline timepoint of the second phase of the PATH study. The second phase expanded on the original design by adding measures including those used in the current study (i.e., daily parenting stress). Participants enrolled in the PATH study between January 2017 and December 2018 completed the first phase while participants enrolled after January 2019 completed the second phase. All study procedures were approved by the institutional Conjoint Health Research Ethics Board (REB15-3100).

The current aims to examine parent variability in the context of parent and child chronic pain are distinct from those of the PATH study as well as previously published studies that used questionnaire data from the PATH study. These studies examined parent and child experiences of diagnostic uncertainty, intolerance of uncertainty, adverse childhood experiences, trauma and pain traumatization, sleep, and attentional biases (Beveridge et al., 2020, 2021, 2022; Christensen et al., 2021; Janssen et al., 2022; Maunder et al., 2022; Nelson et al., 2021; Neville et al., 2019, 2021; Neville, Jordan, et al., 2020; Pavlova et al., 2020; Soltani et al., 2020, 2022). A previously published study used daily diary data from the first phase of the PATH study to examine pain catastrophizing as a moderator of the daily associations between parent mood and protective responses and child pain (Neville, Griep, et al., 2020). The current study used daily diary data collected during the second phase of the PATH study; this data has not previously been published.

Participants

Youth with chronic pain and one of their parents were recruited through a tertiary, outpatient pain program at a pediatric hospital in Western Canada. Clinic staff obtained consent from parents of patients to be contacted by the research team. The research team also received access to a list of families who had previously received care in the pain program as well as participants from a clinical outcomes study that had consented to be contacted for future research studies. Potential participants were contacted via email or telephone and interested parent-child dyads were screened for eligibility over the phone. Youth were eligible to participate if they were between 10-18 years old and reported having chronic pain (i.e., pain that persists or recurs for \geq 3 months; Treede et al., 2015) at the time of recruitment that was not associated with an underlying disease (e.g., juvenile idiopathic arthritis). Youth were not eligible to participate if

they were unable to complete the study measures, because they were not able to read/speak English or access the internet, or had any of the following: severe cognitive impairment or developmental disorder, schizophrenia spectrum or other psychotic disorder, or presence of a serious chronic health and/or life-threatening condition (e.g., cancer). Parents were eligible to participate if they were the legal guardian of the youth and could complete the study measures.

Participants were recruited and data were collected between January 2019 and March 2020. Recruitment was halted due to the COVID-19 pandemic and thus the current study used a convenience sample of participants recruited and enrolled before the onset of the pandemic. In total, 148 families were contacted about participating in the second phase of the PATH study. Eighteen did not meet eligibility criteria, 50 either declined participation or could not be reached after initial contact to be enrolled, and 80 were enrolled. One family withdrew at the baseline stage and one could not be reached after enrollment. Two parent-child dyads completed fewer than five of the seven daily diary surveys so were excluded from the current analyses. The final sample consisted of 76 parent-child dyads. This sample size meets basic recommendations for multilevel regression models to ensure adequate power and non-biased estimates (Maas & Hox, 2005; McNeish, 2017).

Procedure

Informed consent was obtained verbally over the phone and through online consent forms. Youth who were 14 years or older provided their own informed consent while youth who were younger than 14 years provided informed assent. Parents provided informed consent for their own participation and their child's when aged younger than 14 years. All consent forms and study measures were administered and completed online through Research Electronic Data Capture (REDCap), a secure online data collection site (Harris et al., 2009, 2019). Youth and

parents each received a CAD\$20 gift card for their participation in this timepoint of the PATH study. Only procedures and measures relevant to the current study are described.

Once enrolled, parents and youth were emailed links to the baseline questionnaire measures for each to complete independently. Daily diary surveys were sent to parents and youth via email or text message, depending on parent and youth preference, every evening at 6pm for seven consecutive days. Parents and youth were instructed to complete the survey before bed using their own web-enabled device. Although all data in the current study was collected at the baseline timepoint of the PATH study, for ease herein, the questionnaire measures are referred to as “baseline” measures while the daily diary measures are referred to as “daily” measures.

Baseline Questionnaire Measures

Demographic Characteristics

Parents provided demographic information on their own age, gender, racial/ethnic identity, marital status, education, employment status, and annual household income as well as their child’s age, gender, and racial/ethnic identity.

Parent and Youth Pain

Parent chronic pain status was assessed with a yes/no item that asked parents about the presence of pain for at least 3 months in a row, which aligns with the current definition of chronic pain (Treede et al., 2015). This single yes/no item is consistent with previous research assessing the presence of chronic pain in parents in both clinical and community samples (Birnie et al., 2020; Brown et al., 2021, 2022; Clementi et al., 2019; Wiwe Lipsker et al., 2018).

Youth and parents who endorsed chronic pain completed self-report items on their pain characteristics for descriptive purposes. Primary pain locations were assessed with a single item that asked about the parts of their body where they experienced the most aches or pains in the

past week from a checklist of six options (stomach, head, muscles and joints, legs, chest, other). Pain frequency was assessed with a single item that asked how often they had pain in the past week on a 5-point scale (0 = not at all to 4 = daily). Daily pain duration was measured with a single item that asked how long their pain usually lasts on a 4-point scale (0 = less than 1 hour to 3 = all day). Total pain duration was assessed with a single item that asked respondents to report how long they have had pain in years and months. Parents also reported on whether they have received treatment for their own pain and the type (e.g., physical therapy, medication), and the number of school days their child had missed in the past 3 months because of the child's pain.

Youth and parents who endorsed chronic pain also self-reported on their pain intensity and pain interference. Parent scores on these measures were used for descriptive purposes while youth scores were included as covariates in the main analyses. Pain intensity was measured using a validated and reliable 11-point Numerical Rating Scale (0 = no pain to 10 = worst pain possible) that asked about usual pain in the past seven days (Castarlenas et al., 2017; Safikhani et al., 2018; von Baeyer et al., 2009). Pain interference was assessed with the Patient Reported Outcomes Measurement Information System (PROMIS) Pain Interference - Short Form for Adult or Pediatric samples. The 4-item Adult self-report measure asks how much pain interfered with daily activities such as working around the home or participating in social activities in the past seven days on a 5-point scale (1 = not at all to 5 = very much). The 4-item Pediatric self-report measure asks how much pain interfered with daily activities such as walking or paying attention in the past seven days on a 5-point scale (1 = never to 5 = almost always). A total score was obtained by summing the ratings for each item, with higher scores indicating greater pain interference. These measures were developed by the National Institutes of Health using item response theory and have demonstrated reliability and validity in adult and pediatric chronic pain

populations (Amtmann et al., 2010; Kashikar-Zuck et al., 2016). They demonstrated good to excellent internal consistency in the current study (Adult: $a = 0.96$; Pediatric: $a = 0.81$).

Parent Anxiety and Depressive Symptoms

Parent baseline anxiety and depressive symptoms were assessed via self-report with the 14-item Hospital Anxiety and Depression Scale (HADS) (Zigmond & Snaith, 1983). Items ask about anxiety or depressive symptoms experienced in the past week on a 4-point scale, with each item having different anchors (range: 0-3). Total scores for the Anxiety subscale (HADS-A) and the Depression subscale (HADS-D) were obtained by summing the ratings of the relevant 7 items for each subscale (range: 0-21). Higher scores indicate greater anxiety or depressive symptoms, with a score of 8 or above on each subscale suggested as indicating clinically-significant symptoms (Bjelland et al., 2002). In this study, we used the clinical cut-off to characterize the sample and the continuous score of each subscale for the main analyses. The HADS has demonstrated good reliability and validity in patient (e.g., primary care, cancer) and community populations (Bjelland et al., 2002). Both subscales showed good internal consistency in the current study (HADS-A: $a = 0.81$; HADS-D: $a = 0.82$).

Daily Diary Surveys

Parent Anxiety and Mood

Parent daily anxiety was self-reported with a single item that asked parents to rate how anxious/nervous they felt “today” on an 11-point Numerical Rating Scale (NRS; 0 = not at all anxious/nervous to 10 = extremely anxious/nervous). Parent daily mood was self-reported with a single item that asked parents to rate their mood “today” on an 11-point Numerical Rating Scale (NRS; 0 = extremely negative mood to 10 = extremely positive mood). These single items were chosen to reduce participant burden and facilitate completion of the daily diaries as research has

shown that diaries with fewer items have greater compliance (Morren et al., 2009). Previous research using similar single items for measuring daily anxiety or daily mood demonstrated significant convergence between the single items and standard questionnaire measures of anxiety and depression (i.e., the Beck Anxiety Inventory and the Patient Health Questionnaire-9, respectively; Aguilera et al., 2015; Starr & Davila, 2012).

Parent Protective Responses

Parent daily protective responses to their child's pain were assessed with the Protect subscale from the Adult Responses to Children's Symptoms (ARCS) with a pain-specific stem (i.e., "when your child had pain..."; Van Slyke & Walker, 2006). This subscale was adapted for the daily diary surveys to ask about "today." Specifically, parents were asked to rate how often they engaged in protective responses when their child had pain "today" on a 5-point scale (0 = never to 4 = always). Based on research that identified different factor structures of the ARCS for caregivers of children versus adolescents with chronic pain (Noel et al., 2015), different item sets of the Protect subscale were administered to parents of youth aged 10-12 years (11 items) and parents of youth aged 13-18 years (6 items). These item sets were identified in the developmental analysis as providing the most factorially valid structure of the Protect subscale for each age group (Noel et al., 2015). Total scores for both item sets were computed by summing responses to each item and calculating the mean. Higher scores indicate more protective responses. The ARCS and this developmentally-tailored scoring have been used in previous daily diary research with samples of youth with chronic primary and secondary pain (Connelly et al., 2017; Neville, Griep, et al., 2020). In this study, item sets generally had good internal consistency across diaries (Child: mean $\alpha = 0.83$, range = 0.24-0.97; Adolescent: mean $\alpha = 0.78$, range = 0.59-0.93).

Parent Parenting Stress

Parent daily parenting stress was assessed via self-report with the 7-item Parental Stress scale (Pearlin & Schooler, 1978). This scale asks respondents to think of their experiences as a parent and rate the extent to which they feel various emotions (e.g., frustrated, worried, unhappy) on a 4-point scale (1 = not at all to 4 = very). This scale was adapted for the daily diaries to ask about “today.” Total scores were obtained by summing responses to each item (range: 7-28), with higher scores indicating higher parenting stress that day. This scale has shown good reliability (Breslau et al., 1982; Meltzer & Mindell, 2007; Pearlin & Schooler, 1978) and has previously been used in a sample of mothers of children with health conditions (Breslau et al., 1982). In this study, the scale had excellent internal consistency across diaries (mean $\alpha = 0.93$, range = 0.92-0.96).

Youth Pain Intensity and Pain Interference

Youth daily pain intensity and pain interference were assessed with the same youth self-report measures of pain intensity (11-point NRS) and pain interference (4-item PROMIS Pediatric Short Form) that were administered in the baseline questionnaire. However, similar to previous research (Beeckman, Simons, et al., 2019; Neville, Griep, et al., 2020; Rost et al., 2016, 2021), each measure was modified for the daily diaries to ask about “today.” The pain interference measure had good internal consistency across diaries in the current study (mean $\alpha = 0.83$, range = 0.80-0.86).

Parent Daily Variability

Parent variability across the daily diaries was calculated with the square of successive differences (SSDs). For each of the daily parent variables, a set of SSD scores was derived by subtracting each parent’s score on a given day (x_{i+1}) from their score on the preceding day (x_i) and then squaring each of these successive difference scores. Higher SSD scores indicate greater

parent variability. SSD scores were calculated from all pairs of adjacent observations and thus a series of six SSD scores were possible for each parent variable. Based on recommendations (Jahng et al., 2008), these SSD scores were used in the multilevel models to account for the nested structure of the daily diary data.

$$\text{SSD} = (x_{i+1} - x_i)^2$$

Mean SSDs (MSSD) were also calculated for descriptive analyses when there were at least three SSD scores for a parent, consistent with previous research (Li & Lansford, 2018).

$$\text{MSSD} = \frac{1}{N-1} \sum_{i=1}^{N-1} (x_{i+1} - x_i)^2$$

The SSD/MSSD has been recommended over other indices of intraindividual variability (e.g., intraindividual standard deviation) because it incorporates both the frequency and size of changes over the daily diary period and is robust to systematic trends in time series data (Jahng et al., 2008; Mun et al., 2019). In line with previous research, for analyses where SSD/MSSD scores were the outcome variable (descriptive and first set of models), SSD/MSSD scores were log-transformed to adjust for skewness of the distributions, with zero values replaced with half of the smallest non-zero value in the distribution (Koval et al., 2013; Mun et al., 2019; Rost et al., 2016, 2021).

Statistical Analyses

Descriptive statistics, independent samples *t*-tests, correlation analyses, and multilevel models were conducted using SPSS (version 28; IBM Statistics, Armonk, NY, USA). Descriptive statistics characterized the sample, independent samples *t*-tests compared scores on study variables across parent chronic pain status, and correlation analyses examined bivariate associations between the key study variables. Continuous data are presented as mean +/- standard

deviation (SD) and count data are presented as number (percentage). Multilevel modeling was used for the main analyses to account for the non-independence of repeated measurements from the daily dairies (Level 1) nested within each parent-youth dyad (Level 2).

Separate sets of multilevel random intercept models were conducted to address the two study aims (described below). Across models, Level 1 variables included SSD scores for parent daily variability in anxiety, mood, protective responses, and parenting stress and ratings of youth daily pain intensity and pain interference. All Level 1 predictors were continuous and entered grand mean-centered (Enders & Tofighi, 2007). Level 2 variables included baseline measures of parent and youth covariates, parent chronic pain status, and mean levels of parent daily anxiety, mood, protective responses, and parenting stress (aggregated across the daily dairies). All continuous Level 2 predictors were entered grand-mean centered while categorical variables of parent chronic pain status (0 = no chronic pain; 1 = yes chronic pain) and youth gender (0 = male or other; 1 = female) were dummy coded and entered uncentered (Nezlek, 2012). A model building procedure was used wherein unconditional models were first run to derive the intraclass correlation coefficient and confirm use of multilevel modeling. Key study variables were then added and retained. Potential covariates were then added but only retained when they significantly improved model fit (Nezlek, 2012), as determined by the -2 Log Likelihood difference test. Multilevel models were estimated using full information maximum likelihood to allow for model comparison. Assumptions of multilevel models were explored using histograms, boxplots, and model parameters. Due to the novelty of this study, analyses were considered exploratory and the significance level was set at $p < 0.05$. Missing data was handled within the analyses, as multilevel models using full information maximum likelihood allow for all available

information to be used when estimating the model and generating parameters and thus missing data does not need to be addressed outside of the analyses (Field, 2018).

Aim 1: Parent Daily Variability as a Function of Parent Chronic Pain Status

In these models, parent chronic pain status was entered as the Level 2 predictor and parent SSD scores were entered as the Level 1 outcome. Separate models were conducted for each outcome variable, resulting in four models. The mean level of the parent daily variable was included in each model to ensure results reflected variability across days versus higher or lower mean levels of the parent variable (Rost et al., 2016, 2021). Parent baseline anxiety and depressive symptoms were examined as Level 2 covariates to account for the potential role of mental health in variability (Lippold et al., 2019).

Aim 2: Associations Between Parent Daily Variability and Youth Daily Pain and Interference with Parent Chronic Pain Status as a Moderator

In these models, parent SSD scores were entered in the first step as the Level 1 predictor and youth daily pain intensity or interference were entered as the Level 1 outcome. Separate models were conducted for each predictor and outcome variable, resulting in eight models. Similar to previous research (Kukk & Akkermann, 2017), the first day of youth daily pain intensity or interference ratings were omitted to make the data compatible with the set of parent SSD scores. The mean level of the parent daily variable was also entered in the first step as a Level 2 covariate, to ensure the effect was specific to variability versus mean levels (Rost et al., 2016, 2021). Parent chronic pain status (Level 2) and its interaction with the parent SSD score were then entered to examine the moderating role of parent chronic pain status. Youth gender, age, and baseline pain intensity and interference, and parent baseline anxiety and depressive symptoms were also examined as Level 2 covariates.

Results

Data Preparation

Less than 5% of data were missing from the baseline questionnaire measures (parent chronic pain status: 0%; HADS: 0%; youth pain intensity: 1.3%; youth pain interference: 2.6%). Of a total of 532 possible daily diary observations (one observation for 7 days among 76 dyads), less than 10% of data were missing (parent anxiety: 2.8%; parent mood: 2.4%; parent protective responses: 6.6%; parent parenting stress: 4.5%; youth pain intensity: 5.3%; youth pain interference: 7.0%). Overall, parents completed an average of 6.83 diaries ($SD = 0.44$; range: 5-7), with 85.5% completing all 7 diaries. Youth completed an average of 6.66 diaries ($SD = 0.64$; range: 3-7), with 71.1% completing all 7 diaries. The number of diaries completed by parents was not significantly correlated with any of the key parent study variables (all $ps > 0.05$). The number of diaries completed by youth was significantly correlated with youth gender ($r = 0.34$, $p = 0.003$), such that girls tended to complete more days of diaries than boys, but no other key youth study variables (all $ps > 0.05$). All assumptions of multilevel models were met.

Participant Characteristics

Demographic and pain characteristics of the sample are presented in Table 3.1. Parents ranged in age from 36 to 64 years old, and predominately identified as mothers (89.5%) and White (81.6%). Most reported working in a part- or full-time job (79.0%) and had an annual household income greater than \$90,000 (56.6%). Half of parents ($n = 38$; 50.0%) reported having chronic pain. Parents with versus without chronic pain were largely similar across demographic variables, except more parents with chronic pain reported an annual household income $< \$90,000$ (42.1% vs. 15.8%) and met cut-offs for clinically-significant anxiety (44.7% vs. 18.4%) and depressive (15.8% vs. 2.6%) symptoms. Parents reported having chronic pain for, on average,

12.44 years ($SD = 12.11$; range = 5 months to 41 years). Most parents with chronic pain reported more than one pain location ($M = 1.90$; $SD = 1.01$) and having received treatment for their pain, with physical therapy, medication, and acupuncture being the most common.

Youth ranged in age from 10 to 17 years old and predominately identified as female (71.1%) and White (75.0%). Youth reported an average pain duration of 3.05 years ($SD = 2.76$; range = 3 months to 12 years and 8 months). Many youth reported more than one pain location ($M = 1.79$; $SD = 1.11$) and having pain at least 4 times per week (52.7%) that lasts at least half of the day (52.6%). Youth whose parents also had chronic pain reported, on average, a longer pain duration (3.23 vs. 2.87 years) and more pain locations (57.9% vs. 34.2% reporting more than one location) than youth whose parents did not endorse chronic pain. Table 3.2 reports the descriptive statistics of the study variables, grouped by parent chronic pain status, and Table 3.3 displays the correlations between the baseline and daily diary variables.

Multilevel Models

Unconditional models indicated that approximately 20% (19.7 to 22.0%) of the total variance for parent daily anxiety, mood, protective responses, and parenting stress and approximately 70% (69.5 to 70.4%) of the total variance for youth daily pain intensity and pain interference was attributable to between-person differences. Intercepts also varied significantly (all $ps < 0.001$) in all unconditional models; thus, the use of multilevel models was warranted.

Aim 1: Parent Daily Variability as a Function of Parent Chronic Pain Status

Results of final models are displayed in Table 3.4. Model fit was not significantly improved with the inclusion of parent chronic pain status in the first step of model building for parent daily anxiety variability, mood variability, or protective responses variability ($ps = 0.077$ to 0.096), but was significantly improved for parent daily parenting stress variability, $\chi^2(1) =$

5.76, $p = 0.016$. Across all models, fit was significantly improved with the addition of the mean level of the parent daily variable (i.e., anxiety, mood, protective responses, or parenting stress) in the second step (all $ps < 0.001$). Parent baseline anxiety symptoms did not significantly contribute to models and thus was not retained in any final model. Parent baseline depressive symptoms significantly improved model fit, and thus was retained in the final models, for parent daily anxiety variability and daily parenting stress variability.

Parent daily anxiety variability was significantly predicted by parent mean daily anxiety and parent baseline depressive symptoms, but not parent chronic pain status, such that parents with greater mean levels of daily anxiety and lower-than-average depressive symptoms had greater daily anxiety variability than parents with higher-than-average depressive symptoms. Parent daily variability in mood and protective responses were both significantly predicted by the respective daily mean levels (i.e., lower mood and more protective responses), but not parent chronic pain status. Parent daily parenting stress variability was significantly predicted by parent chronic pain status, parent mean daily parenting stress, and parent baseline depressive symptoms, such that parents with chronic pain, greater means levels of parenting stress, and lower-than-average depressive symptoms had greater variability in their daily parenting stress.

Aim 2: Associations Between Parent Daily Variability and Youth Daily Pain and Interference with Parent Chronic Pain Status as a Moderator

Predicting Youth Daily Pain Intensity. Across all four models, the addition of parent daily variability (in anxiety, mood, protective responses, or parenting stress) and mean levels of these variables in the first step significantly improved model fit from the unconditional models, but neither variable significantly predicted youth daily pain intensity. Model fit did not significantly improve with the addition of parent chronic pain status, the interaction between

parent daily variability and parent chronic pain status, or parent baseline anxiety or depressive symptoms in any of the four models. However, youth gender, age, and baseline pain intensity significantly contributed to all models; thus, these covariates were retained in all four final models. Results of the final models of parent daily variability predicting youth daily pain intensity are displayed in Table 3.5. Overall, across all final models, youth daily pain intensity was significantly predicted by youth gender, age, and baseline pain intensity (while adjusting for parent daily variability and daily means of anxiety, mood, protective responses, or parenting stress, parent chronic pain status and its interaction with parent daily variability).

Predicting Youth Daily Pain Interference. Across all four models, the addition of parent daily variability (in anxiety, mood, protective responses, or parenting stress) and mean levels of these variables significantly improved model fit from the unconditional models. Parent *mean* levels of anxiety, protective responses, and parenting stress significantly predicted youth daily pain interference when added in the first step of their respective models. However, parent daily *variability* was not significantly related to youth daily pain interference when added in the first step of any model. The addition of parent chronic pain status and the interaction between parent daily variability (in anxiety, mood, protective responses, or parenting stress) and parent chronic pain status did not significantly improve any model fit. Youth gender, youth age, and parent baseline anxiety symptoms were also not significantly related to youth daily pain interference in any model and thus were not retained. Youth baseline pain interference significantly contributed to all models and was retained in all final models. Parent baseline depressive symptoms significantly contributed to the model with parent daily mood variability, but no other model, and thus was retained in the parent daily mood variability final model.

Results of all final models of parent daily variability predicting youth daily pain interference are displayed in Table 3.6.

In the final model with parent daily anxiety variability, youth daily pain interference was significantly predicted by parent mean daily anxiety and youth baseline pain interference. In the final model with parent daily mood variability, youth daily pain interference was significantly predicted by parent baseline depressive symptoms and youth baseline pain interference. In the final model with parent daily protective responses variability, youth daily pain interference was significantly predicted by parent mean daily protective responses and youth baseline pain interference. In the final model with parent daily parenting stress variability, youth daily pain interference was significantly predicted by parent mean daily parenting stress and youth baseline pain interference.

Discussion

Intraindividual variability in adults with chronic pain has been examined in such domains as mood (e.g., negative affect) and pain (e.g., intensity, interference). These studies show that adults with versus without chronic pain have more variability, which is significantly associated with poorer functioning (Gerhart et al., 2018; Ravyts et al., 2019; Rost et al., 2016, 2021; Wesolowicz et al., 2021). The current study extended this literature to parenting variables, the parent-child dyad, and youth chronic pain. Specifically, we used daily diaries to examine parents' day-to-day variability and its associations with parent chronic pain status and the daily pain experiences of youth with chronic pain. In our sample, 50% of parents endorsed chronic pain and findings were largely inconsistent with hypotheses. The first set of multilevel models revealed that, while controlling for mean levels, parents with versus without chronic pain were significantly more variable in their parenting stress across the week, but not in their anxiety,

mood, or protective responses. In the second set of multilevel models, parent variability in anxiety, mood, protective responses, or parenting stress was *not* significantly related to youth daily pain intensity or interference. Moreover, parent chronic pain status did *not* moderate associations between parent variability and youth daily pain. Instead, mean levels of parent anxiety, protective responses, and parenting stress across the week significantly predicted youth daily pain interference.

These initial findings contribute to a growing body of research using daily diaries to examine the functioning of families with chronic pain in their everyday lives (Bolger et al., 2003). While this previous research has found greater within- versus between- person variability (Beeckman, Simons, et al., 2019; Connelly et al., 2017; Neville, Griep, et al., 2020), these studies did not explicitly examine parent variability and its association with youth pain. The amount of within-person variance in parent factors in our sample (about 80%) was consistent with previous research (Beeckman, Simons, et al., 2019; Connelly et al., 2017; Neville, Griep, et al., 2020). However, parent variability was not significantly associated with youth daily pain. Instead, youth daily pain interference was predicted by parents' mean levels of anxiety, protective responding, and parenting stress. These findings align with a previous daily diary study, which found increased use of parent protective responses predicted increased child pain interference (Beeckman, Simons, et al., 2019; Connelly et al., 2017), and extend the literature to parent variables that have not previously been examined in daily dairies (i.e., anxiety, parenting stress). The finding that parent factors were not significantly related to youth daily pain intensity aligns with meta-analytic findings that parent factors are not as strongly related to youth's pain intensity as their pain-related disability (Donnelly et al., 2020; Harrison et al., 2020).

While previous research with community-based samples has shown that parent variability is linked to poorer functioning in youth, these studies examined variability in more dyadic domains than those currently assessed (except for protective responses; Lippold et al., 2015, 2017). Youth may be more sensitive to variability in parent behaviour when those behaviours are directed towards them. There may also be developmental differences, with adolescents potentially less affected by day-to-day variability in parent behaviour than younger children as they develop greater autonomy (Palermo et al., 2014). The current study also examined youth pain, which may be less dynamic on a day-to-day basis than other variables such as mood. Indeed, the current sample of treatment-seeking youth had long-standing chronic pain (3+ years) that was relatively stable over the one week period, as indicated by the amount of variance attributable to between-person differences. Future research should extend investigation to other child variables (e.g., emotional functioning) and populations (e.g., acute pain, younger children).

Parents with chronic pain were not significantly more variable in their anxiety, mood, or protective responses than parents without chronic pain, but did experience greater variability in parenting stress. These findings may alleviate worries expressed by parents with chronic pain in qualitative studies that the variable nature of their pain causes them to be more inconsistent than other parents, with negative impacts on their children (Barlow et al., 1999; Evans & de Souza, 2008; Wilson & Fales, 2015). Our findings suggest *all* parents demonstrate variability *and* this variability is not significantly associated with children's daily pain. Thus, even when parents, with or without chronic pain, have 'bad days' with greater anxiety, lower mood, more protective responses, or greater parenting stress, it may not significantly impact children's physical functioning. However, further research is needed to confirm these findings. Parents with chronic pain did endorse greater mean levels of daily anxiety, mood, protective responses, and parenting

stress (at $p < 0.05$ or trend levels) as well as greater variability in parenting stress. These differences may be due to personal challenges as well as systemic and social challenges (e.g., invalidation, diagnostic uncertainty, stigmatization, poor social support) that can contribute to (toxic) stress for those with chronic pain (Nelson et al., 2022).

Parent mental health was found to play a bigger role in youth's daily pain-related functioning than parent chronic pain. This finding aligns with recent research demonstrating the important role of parent mental health in pediatric chronic pain (Beveridge et al., 2024; Donnelly et al., 2020; Fussner et al., 2018; Law et al., 2017; Poppert Cordts et al., 2019). Current conceptual models (Asmundson et al., 2012) and empirical research stemming from these models (Harrison et al., 2020; Neville et al., 2021) tend to focus on parent distress and fear/anxiety *in relation to child pain* (e.g., catastrophizing about child pain) without consideration for parent's general distress such as anxiety, depression, or parenting stress. While the role of parent responses to child pain has been explained through social learning processes (e.g., operant conditioning), the pathways from parent general distress to child pain are less clear and may occur more indirectly, for example through a stressful home environment (Palermo & Chambers, 2005). The associations are also likely bidirectional, with poorer child functioning increasing parent distress (Connelly et al., 2017). Further research examining these dyadic processes, including mechanisms underlying their association, is needed.

Current psychological interventions for parents whose children have chronic pain focus on parent responses to child pain, with limited effectiveness (Fisher et al., 2018; Lee et al., 2021). This study adds to increasing evidence that parents' physical and psychological functioning should also be considered (Law et al., 2017; Poppert Cordts et al., 2019). For example, interventions that teach skills (e.g., problem-solving, mindfulness) that parents can

apply to their own difficulties as well as their child's pain have demonstrated effectiveness in improving both parent and child outcomes (Kemani et al., 2018; Palermo et al., 2016). Our findings suggest that parent interventions should focus on strategies that reduce parents' mean levels of distress (versus variability). However, further longitudinal research is needed to investigate the role of parent variability in children's response to treatment.

Strengths of this study include the use of diaries to capture the daily functioning of parents and youth, the use of multilevel models for the nested structure of the data, and the examination of general parent distress. The following limitations should also be considered. First, findings are specific to a clinical sample of youth with chronic primary pain, and one of their parents, and thus may not generalize to other child populations (e.g., acute pain) or address family dynamics (e.g., second parent, siblings). Additionally, parent chronic pain was assessed with a single item that may not have yielded a representative sample of adults with chronic pain. Moreover, as common in studies recruiting from tertiary pain clinics in North America (Birnie et al., 2020; Gibler et al., 2019; Nelson et al., 2018; Palermo et al., 2019; Ruskin et al., 2014), the sample predominately identified as female, White, and higher socioeconomic status. This perpetuates the lack of representation of minoritized and marginalized communities in pain research (Letzen et al., 2022). Future research should consider extending these findings to more diverse samples and the whole family (e.g., fathers). Second, the smaller sample size and sampling period limited our ability to conduct more sophisticated and time-lagged analyses. Thus, as done in previous research with similar sample sizes (Beckman, Simons, et al., 2019; Rost et al., 2016, 2021), multiple models tested each hypothesis. We did not correct for multiple testing as hypotheses were generated *a priori*, models were not rerun in different subsamples, and this was not a confirmatory study (Althouse, 2016; Bender & Lange, 2001; Perneger, 1998).

Nonetheless, findings are considered cross-sectional and may have been under-powered or had inflated type I errors. Further research with a larger sample and longer study period is needed to confirm our initial findings. Third, to increase compliance, diaries were completed once a day and included several single-item measures. Future research using ecological momentary assessment could refine the current study by examining within-day means and variability. Lastly, this study examined parent variability in domains of negative valence. However, parent variability could also reflect adaptive flexibility that is responsive to the child (Beeckman, Simons, et al., 2019). Emerging research on resilience factors within pediatric chronic pain has found that parent psychological flexibility, which can be defined as the ability to behave according to personal values in the presence of distress (S. C. Hayes et al., 2006), is associated with better child functioning (Beeckman, Hughes, et al., 2019; Cousins, Kalapurakkal, et al., 2015; Lee et al., 2020; McCracken & Gauntlett-Gilbert, 2011; Timmers et al., 2019). Further research using daily diaries to identify factors associated with improved daily functioning is needed to refine our understanding of processes contributing to more positive outcomes.

This was the first study to examine day-to-day variability among parents of youth with chronic pain including its association with parents' chronic pain status and youth's daily pain experiences. Results suggest *all* parents demonstrate variability in their anxiety, mood, protective responses, and parenting stress across the week; however, parents with chronic pain may have greater variability in their parenting stress than parents without chronic pain. Regardless, parent variability was *not* significantly associated with youth's daily pain intensity or interference. Instead, overall levels of parent daily anxiety, protective responses, and parenting stress across the week were significant predictors of youth daily pain interference. These initial findings may have important implications for clinical practice within pediatric chronic pain, including

addressing parent distress in general (versus in relation to the child's pain) and validating that 'bad days' happen with potentially little impact on the child's pain-related functioning.

Table 3.1*Demographic and Pain Characteristics of Parents and Youth*

	Total sample (<i>N</i> = 76)	Parents with chronic pain (<i>n</i> = 38)	Parents without chronic pain (<i>n</i> = 38)
	<i>M</i> ± <i>SD</i> or <i>n</i> (%)		
Parent characteristics			
Age, years	45.53 ± 5.39	45.84 ± 6.10	45.21 ± 4.64
Gender			
Female	68 (89.5)	36 (94.7)	32 (84.2)
Male	8 (10.5)	2 (5.3)	6 (15.8)
Racial/ethnic identity			
White	62 (81.6)	30 (78.9)	32 (84.2)
Bi- or multi- racial	8 (10.5)	6 (15.8)	2 (5.3)
Arab/West Asian	1 (1.3)	0 (0.0)	1 (2.6)
Filipino	1 (1.3)	0 (0.0)	1 (2.6)
Indigenous	1 (1.3)	0 (0.0)	1 (2.6)
Latin American	1 (1.3)	1 (2.6)	0 (0.0)
South Asian	1 (1.3)	0 (0.0)	1 (2.6)
Other	1 (1.3)	1 (2.6)	0 (0.0)
Marital status			
Married or common-law	61 (80.3)	32 (84.2)	29 (76.3)
Separated, divorced, or widowed	13 (17.1)	5 (13.2)	8 (21.0)
Single	2 (2.6)	1 (2.6)	1 (2.6)
Annual household income, CAD			

≤ \$29,999	5 (6.6)	4 (10.5)	1 (2.6)
\$30,000 – \$59,999	6 (7.9)	4 (10.5)	2 (5.3)
\$60,000 – \$89,999	11 (14.5)	8 (21.1)	3 (7.9)
≥ \$90,000	43 (56.6)	16 (42.1)	27 (71.1)
Did not respond	11 (14.5)	6 (15.8)	5 (13.1)
Education level			
High school or less	11 (14.5)	7 (18.4)	4 (10.5)
Vocational school or some college	20 (26.3)	10 (26.3)	10 (26.3)
College or Bachelor's degree	38 (50.0)	15 (39.5)	23 (60.5)
Graduate or professional degree	7 (9.2)	6 (15.8)	1 (2.6)
Employment status			
Full-time	37 (48.7)	18 (47.4)	19 (50.0)
Part-time	23 (30.3)	10 (26.3)	13 (34.2)
Not working	15 (19.7)	9 (23.7)	6 (15.8)
Did not respond	1 (1.3)	1 (2.6)	0 (0.0)
Clinically-significant anxiety symptoms	24 (31.6)	17 (44.7)	7 (18.4)
Clinically-significant depressive symptoms	7 (9.2)	6 (15.8)	1 (2.6)
Pain locations			
Head	-	20 (52.6)	-
Muscles and joints	-	19 (50.0)	-
Leg(s)	-	9 (23.7)	-
Stomach	-	5 (13.2)	-
Chest	-	3 (7.9)	-
Other	-	16 (42.1)	-
Two or more locations	-	24 (63.2)	-

Pain frequency			
Not at all	-	0 (0.0)	-
1 time per week	-	5 (13.2)	-
2 to 3 times per week	-	12 (31.6)	-
4 to 6 times per week	-	3 (7.9)	-
Daily	-	18 (47.4)	-
Daily pain duration			
Less than 1 hour	-	2 (5.3)	-
A few hours	-	12 (31.6)	-
Half of the day	-	8 (21.1)	-
All day	-	15 (39.5)	-
Pain intensity, 0-10	-	4.97 ± 2.20	-
Pain interference, 0-20	-	9.71 ± 4.55	-
Treatment received for pain			
Physical therapy	-	16 (42.1)	-
Medication	-	16 (42.1)	-
Acupuncture	-	16 (42.1)	-
Massage therapy	-	14 (36.8)	-
Chiropractic	-	8 (21.1)	-
Psychological therapy	-	3 (7.9)	-
Other	-	7 (18.4)	-
Youth characteristics			
Age, years	14.26 ± 2.13	14.29 ± 2.17	14.24 ± 2.11
Gender			
Female	54 (71.1)	28 (73.7)	26 (68.4)

Male	19 (25.0)	8 (21.1)	11 (28.9)
Other	3 (3.9)	2 (5.3)	1 (2.6)
Racial/ethnic identity			
White	57 (75.0)	25 (65.8)	32 (84.2)
Bi- or multi- racial	10 (13.2)	8 (21.1)	2 (5.3)
Arab/West Asian	1 (1.3)	0 (0.0)	1 (2.6)
Black	1 (1.3)	1 (2.6)	0 (0.0)
Filipino	1 (1.3)	0 (0.0)	1 (2.6)
Indigenous	1 (1.3)	0 (0.0)	1 (2.6)
Latin American	1 (1.3)	1 (2.6)	0 (0.0)
South Asian	1 (1.3)	0 (0.0)	1 (2.6)
Other	3 (3.9)	3 (7.9)	0 (0.0)
Total pain duration, years	3.05 ± 2.76	3.23 ± 2.95	2.87 ± 2.58
Pain locations			
Head	62 (81.6)	31 (81.6)	31 (81.6)
Muscles and joints	19 (25.0)	11 (28.9)	8 (21.1)
Stomach	14 (18.4)	10 (26.3)	4 (10.5)
Chest	11 (14.5)	7 (18.4)	4 (10.5)
Leg(s)	10 (13.2)	4 (10.5)	6 (15.8)
Other	20 (26.3)	11 (28.9)	9 (23.7)
Two or more locations	35 (46.1)	22 (57.9)	13 (34.2)
Pain frequency			
Not at all	2 (2.6)	0 (0.0)	2 (5.3)
1 time per week	8 (10.5)	6 (15.8)	2 (5.3)
2 to 3 times per week	26 (34.2)	12 (31.6)	14 (36.8)

4 to 6 times per week	11 (14.5)	5 (13.2)	6 (15.8)
Daily	29 (38.2)	15 (39.5)	14 (36.8)
Daily pain duration			
Less than 1 hour	16 (21.1)	6 (15.8)	10 (26.3)
A few hours	20 (26.3)	12 (31.6)	8 (21.1)
Half of the day	14 (18.4)	5 (13.2)	9 (23.7)
All day	26 (34.2)	15 (39.5)	11 (28.9)
School missed in past 3 months, days	8.46 ± 14.49	8.45 ± 16.03	8.47 ± 13.04

Table 3.2*Descriptive Statistics for Baseline and Daily Variables by Parent Chronic Pain Status*

	Parents with chronic pain (<i>n</i> = 38)	Parents without chronic pain (<i>n</i> = 38)	
Variable	M (SD)	M (SD)	Difference test
Baseline measures			
Parent anxiety symptoms	6.47 (3.69)	5.11 (3.24)	<i>t</i> (74) = 1.72, <i>p</i> = 0.090
Parent depressive symptoms	4.08 (2.95)	2.29 (3.29)	<i>t</i>(74) = 2.50, <i>p</i> = 0.015
Youth pain intensity	5.46 (1.86)	5.42 (1.80)	<i>t</i> (73) = 0.09, <i>p</i> = 0.928
Youth pain interference	11.00 (4.18)	10.92 (4.32)	<i>t</i> (72) = 0.08, <i>p</i> = 0.937
Daily measures			
Parent anxiety variability, MSSD	0.65 (1.62)	-0.01 (1.67)	<i>t</i> (74) = 1.75, <i>p</i> = 0.084
Parent mood variability, MSSD	0.50 (1.41)	0.25 (1.17)	<i>t</i> (74) = 0.84, <i>p</i> = 0.404
Parent protective responses variability, MSSD	-3.40 (1.80)	-3.75 (2.07)	<i>t</i> (72) = 0.77, <i>p</i> = 0.446
Parenting stress variability, MSSD	1.60 (1.59)	0.90 (1.93)	<i>t</i> (74) = 1.74, <i>p</i> = 0.086
Parent mean anxiety	1.91 (1.59)	1.14 (1.34)	<i>t</i>(74) = 2.28, <i>p</i> = 0.026
Parent mean mood	6.52 (1.45)	7.36 (1.27)	<i>t</i>(74) = -2.68, <i>p</i> = 0.009
Parent mean protective responses	0.33 (0.46)	0.16 (0.24)	<i>t</i> (72) = 1.98, <i>p</i> = 0.052
Parent mean parenting stress	10.98 (4.56)	9.28 (3.11)	<i>t</i> (74) = 1.90, <i>p</i> = 0.062
Youth mean pain intensity	4.38 (2.45)	4.34 (2.14)	<i>t</i> (73) = 0.08, <i>p</i> = 0.937
Youth mean pain interference	8.46 (3.87)	8.12 (3.43)	<i>t</i> (72) = 0.40, <i>p</i> = 0.694

Note. MSSD scores are log-transformed to adjust for positive skewness; mean daily scores are aggregated over the seven daily diaries; significantly different values are bolded

Abbreviations. MSSD, mean square successive difference

Table 3.3*Bivariate Correlations Between Study Variables*

Variable	2	3	4	5	6	7	8	9	10	11	12	13	14	15
Baseline														
measures														
1. Parent chronic pain status	0.20	0.28*	0.01	0.01	0.20	0.10	0.09	0.20	0.26*	-0.30**	0.23	0.22	0.01	0.05
2. Parent anxiety symptoms	-	0.62**	0.08	0.13	0.40**	0.11	0.21	0.29*	0.60**	-0.42**	0.25*	0.55**	0.14	0.18
3. Parent depressive symptoms		-	0.02	0.19	0.29*	0.27*	0.18	0.26*	0.53**	-0.56**	0.42**	0.71**	-0.02	0.28*
4. Youth pain intensity			-	0.47**	0.17	-0.06	0.19	0.15	0.21	-0.18	0.10	0.18	0.48**	0.35**
5. Youth pain interference				-	0.09	0.16	0.11	0.13	0.24*	-0.15	0.18	0.15	0.39**	0.61**
Daily measures														

6. Parent anxiety variability, MSSD	-	0.17	0.12	0.47**	0.69**	-0.31**	0.12	0.31**	-0.06	0.08
7. Parent mood variability, MSSD		-	-0.06	0.23*	0.27*	-0.48**	0.01	0.32**	0.06	0.17
8. Parent protective responses variability, MSSD			-	0.18	0.18	-0.17	0.60**	0.20	0.17	0.15
9. Parenting stress variability, MSSD				-	0.43**	-0.25*	0.16	0.55**	0.06	0.25*
10. Parent mean anxiety					-	-0.46**	0.25*	0.56**	0.16	0.25*
11. Parent mean mood						-	-0.19	-0.54**	-0.12	-0.10
12. Parent mean protective responses							-	0.44**	0.08	0.32**

13. Parent mean parenting stress	-	0.06	0.25*
14. Youth mean pain intensity	-		0.54**
15. Youth mean pain interference			-

Note. Mean Square Successive Difference (MSSD) scores are log-transformed to adjust for positive skewness; mean daily scores are aggregated over the seven daily diaries; * $p < 0.05$, ** $p < 0.01$

Table 3.4

Results of Final Multilevel Models Examining Parent Chronic Pain Status as a Predictor of Parent Daily Variability (SSDs)

Outcome	Predictor	B	SE	t	p
Parent anxiety variability					
	Intercept	0.22	0.08	2.83	0.006
	Parent chronic pain status	0.05	0.11	0.44	0.663
	Parent mean daily anxiety	0.41	0.04	9.52	< 0.001
	Parent baseline depressive symptoms	-0.04	0.02	-2.04	0.044
Parent mood variability					
	Intercept	0.22	0.09	2.34	0.022
	Parent chronic pain status	0.06	0.14	0.43	0.666
	Parent mean daily mood	-0.23	0.05	-4.65	< 0.001
Parent protective responses variability					
	Intercept	-4.38	0.14	-30.83	< 0.001
	Parent chronic pain status	0.15	0.20	0.74	0.460
	Parent mean daily protective responses	1.96	0.28	6.98	< 0.001
Parenting stress variability					
	Intercept	0.64	0.12	5.42	< 0.001
	Parent chronic pain status	0.38	0.17	2.23	0.029
	Parent mean daily parenting stress	0.20	0.03	6.62	< 0.001
	Parent baseline depressive symptoms	-0.10	0.04	-2.75	0.007

Note. Significant values are bolded.

Table 3.5

Results of Final Multilevel Models Examining Parent Daily Variability (SSDs) as a Predictor of Youth Daily Pain Intensity

Predictors	B	SE	t	p
Model: Parent anxiety variability				
Intercept	3.66	0.47	7.71	< 0.001
Parent anxiety variability	-0.01	0.01	-0.48	0.635
Parent mean daily anxiety	0.21	0.16	1.31	0.195
Parent chronic pain status	-0.29	0.47	-0.62	0.537
Parent anxiety variability * parent chronic pain status	0.02	0.02	1.02	0.309
Youth gender	1.30	0.51	2.58	0.012
Youth age	0.26	0.11	2.47	0.016
Youth baseline pain intensity	0.45	0.13	3.42	0.001
Model: Parent mood variability				
Intercept	3.64	0.47	7.73	< 0.001
Parent mood variability	0.02	0.02	1.51	0.133
Parent mean daily mood	-0.14	0.17	-0.82	0.416
Parent chronic pain status	-0.23	0.47	-0.49	0.624
Parent mood variability * parent chronic pain status	-0.003	0.03	-0.14	0.892
Youth gender	1.31	0.51	2.59	0.012
Youth age	0.26	0.11	2.44	0.017
Youth baseline pain intensity	0.47	0.13	3.62	< 0.001
Model: Parent protective responses variability				
Intercept	3.53	0.49	7.25	< 0.001
Parent protective responses variability	-0.02	0.10	-0.20	0.840

Parent mean daily protective responses	1.10	0.66	1.66	0.102
Parent chronic pain status	-0.28	0.48	-0.58	0.562
Parent protective responses variability * parent chronic pain status	0.08	0.14	0.53	0.595
Youth gender	1.44	0.53	2.74	0.008
Youth age	0.30	0.11	2.70	0.009
Youth baseline pain intensity	0.46	0.14	3.27	0.002
Model: Parenting stress variability				
Intercept	3.59	0.47	7.61	< 0.001
Parenting stress variability	-0.01	0.004	-1.41	0.159
Parent mean daily parenting stress	0.08	0.06	1.27	0.207
Parent chronic pain status	-0.27	0.46	-0.57	0.569
Parenting stress variability * parent chronic pain status	0.004	0.01	0.79	0.433
Youth gender	1.32	0.50	2.61	0.011
Youth age	0.29	0.11	2.69	0.009
Youth baseline pain intensity	0.46	0.13	3.53	< 0.001

Note. Significant values are bolded.

Table 3.6

Results of Final Multilevel Models Examining Parent Daily Variability (SSDs) as a Predictor of Youth Daily Pain Interference

Predictors	B	SE	t	p
Model: Parent anxiety variability				
Intercept	8.32	0.51	16.37	< 0.001
Parent anxiety variability	-0.02	0.02	-0.95	0.344
Parent mean daily anxiety	0.54	0.25	2.12	0.037
Parent chronic pain status	-0.10	0.73	-0.14	0.888
Parent anxiety variability * parent chronic pain status	0.01	0.03	0.32	0.746
Youth baseline pain interference	0.51	0.09	5.78	< 0.001
Model: Parent mood variability				
Intercept	8.33	0.52	16.13	< 0.001
Parent mood variability	-0.01	0.02	-0.53	0.599
Parent mean daily mood	0.19	0.31	0.61	0.545
Parent chronic pain status	-0.09	0.75	-0.12	0.908
Parent mood variability * parent chronic pain status	-0.03	0.04	-0.76	0.448
Youth baseline pain interference	0.52	0.09	5.93	< 0.001
Parent baseline depressive symptoms	0.29	0.14	2.09	0.040
Model: Parent protective responses variability				
Intercept	8.39	0.51	16.56	< 0.001
Parent protective responses variability	0.03	0.16	0.20	0.840
Parent mean daily protective responses	2.58	1.04	2.48	0.015
Parent chronic pain status	-0.13	0.72	-0.19	0.853

Parent protective responses variability * parent chronic pain				
status	-0.002	0.22	-0.01	0.991
Youth baseline pain interference	0.51	0.09	5.91	< 0.001
Model: Parenting stress variability				
Intercept	8.35	0.50	16.82	< 0.001
Parenting stress variability	-0.01	0.01	-0.93	0.353
Parent mean daily parenting stress	0.26	0.10	2.62	0.010
Parent chronic pain status	-0.14	0.71	-0.20	0.845
Parenting stress variability * parent chronic pain status	-0.003	0.01	-0.41	0.685
Youth baseline pain interference	0.52	0.08	6.15	< 0.001

Note. Significant values are bolded.

**CHAPTER FOUR: Identifying Risk and Protective Factors in the Transmission of Chronic
Pain from Mothers to Children: A Multi-Method Longitudinal Cohort Study**

Abstract

Chronic pain is common and can significantly impact the individual and their family. Indeed, children have an increased risk of developing chronic pain when one or both of their parents have it. However, many children of parents with chronic pain *do not* report pain problems. The aim of this longitudinal study was to identify factors throughout childhood that either increased or decreased the risk for chronic pain among children of mothers with and without chronic pain. Participants were 1128 mother-child dyads from a community-based cohort study. Mothers self-reported on their chronic pain when their child was 5 and 8 years old and on potential risk and protective factors (anxiety and depressive symptoms, parenting practices, social support, coping, optimism) at various timepoints between child ages 8 and 11. Children self-reported on their chronic pain at age 13 and on potential protective factors (optimism, connections with adults and peers, community engagement) at age 12. Logistic regression analyses demonstrated that children had an increased risk for chronic pain at age 13 when their mothers reported both chronic pain and greater anxiety symptoms or ineffective parenting practices earlier in childhood. No protective factors moderated the association between mother-child chronic pain; however, greater child optimism and connections with adults at age 12 decreased the risk of chronic pain for all children, regardless of maternal chronic pain status. These findings highlight factors that can be targeted in prevention efforts to mitigate the risk of chronic pain, but future research is needed to explore additional protective factors.

Introduction

Chronic pain is prevalent in both children and adults, occurring in approximately 20% of the general population (Chambers et al., 2024; Schopflocher et al., 2011; Yong et al., 2022). It can have significant impacts on the physical, emotional, social, educational, and occupational functioning of individuals (Miró et al., 2023; Yong et al., 2022). There are also intergenerational consequences, wherein children of parents with chronic pain are at increased risk for developing their own pain and disability, mental health problems, and social difficulties compared to children of parents without chronic pain (K. S. Higgins et al., 2015). Emerging research investigating mechanisms underlying this transmission of risk has shown that psychosocial factors, including parent and child mental health (e.g., anxiety, depression) and general and pain-specific parenting practices (e.g., authoritarian parenting, protective responses to child pain) mediate the association between parent and child chronic pain (Birnie et al., 2020; Brown et al., 2021; Evans et al., 2006; K. S. Higgins et al., 2019; Stone et al., 2018; Stone & Walker, 2016). However, some children of parents with chronic pain do not display poor outcomes (Borge & Nordhagen, 2000; K. S. Higgins et al., 2015; Hoftun et al., 2013; K. B. Smith & Chambers, 2006; Wilson et al., 2020), suggesting there are factors promoting resilience – that is, the capacity to be protected from or adapt to risks (Masten et al., 2021). To our knowledge, a longitudinal examination of both risk and protective factors that moderate the association between parent and child chronic pain has not been conducted.

The broader literature on pediatric pain has predominately focused on identifying risk factors that predict the development and maintenance of pain and disability in children. This research has demonstrated that factors across the child's ecology, including individual (e.g., difficult temperament, maladaptive coping strategies such as avoidance), family (e.g., parent

distress, poor family functioning), and peer (e.g., bullying) factors, interact and increase risk for pain and disability (Donnelly et al., 2020; Fisher et al., 2024; McKillop & Banez, 2016; C. M. Sinclair & Feeney, 2016). More recently, the need for identifying factors that promote positive outcomes for children with chronic pain has been highlighted, with a conceptual model outlining child (e.g., optimism, self-esteem), family/social (e.g., positive family interactions, social support), and cultural (e.g., community support) resources that may contribute to children functioning well despite their pain and additional risks (e.g., poor parent health; Cousins, Kalapurakkel, et al., 2015; Goubert & Trompetter, 2017; Sturgeon & Zautra, 2010). Empirical research with clinical samples of youth seeking tertiary treatment for their chronic pain has confirmed the protective effects of several factors for children's pain-related functioning. For example, general factors including greater child optimism – the generalized expectancy for positive or favourable outcomes (Scheier & Carver, 1985) – and high quality peer relationships as well as pain-specific factors (e.g., pain acceptance, pain self-efficacy) are related to better outcomes for youth with chronic pain (Beeckman, Hughes, et al., 2019; Beeckman, Simons, et al., 2019; Connelly et al., 2014; Cousins, Cohen, et al., 2015; Feinstein et al., 2018; Lee et al., 2020; Ross et al., 2018; Walker et al., 2008).

Research examining factors that protect against the *development* of chronic pain in children, particularly longitudinal studies that include both parent and child report, are limited. The literature on developmental psychopathology and resilience, which study variations in child development and processes contributing to either negative or positive adaptation in the context of risk, are particularly well-suited to inform this research (Cicchetti, 1984; Masten, 2001). This literature has identified individual, interpersonal, and environmental factors that are associated with successful child adaptation to a variety of risks (e.g., maltreatment, poverty, war) as well as

adaptive systems (e.g., attachment system, central nervous system, education system) that are the product of biological and cultural evolution and contribute to positive adaptation (Masten, 2015). This literature highlights the need to consider factors at various levels of the family ecology (e.g., child, parent, community) and to differentiate factors that predict positive outcomes for children exposed to risk (i.e., protective factors) from factors predicting positive outcomes for any child, regardless of their risk exposure (i.e., promotive factors; Masten et al., 2021).

The Current Study

The aim of the current research was to identify risk and protective factors that moderate the intergenerational transmission of chronic pain using multi-wave and multi-informant data from a community-based cohort of mother-child dyads. Given the lack of research on resilience to pediatric chronic pain, protective factors were prioritized. We hypothesized that (1) children exposed to maternal chronic pain in childhood (ages 5 and 8) would be more likely to report their own chronic pain in early adolescence (age 13) and (2) risk factors, including greater maternal anxiety and depressive symptoms and ineffective parenting, would increase the strength of the association while protective factors, including greater maternal positive parenting, social support, optimism, and active coping, and greater child optimism, connections with adults and peers, and community engagement, would decrease the strength of the association between maternal and child chronic pain.

Methods

Participants and Procedure

This study used data from an ongoing, longitudinal pregnancy cohort, the All Our Families (AOF) study, which was designed to examine the determinants of maternal and child health outcomes and health care use. Study procedures were approved by the institutional

research ethics board at the University of Calgary (REB13-0868). Methods and sample characteristics have been described in detail elsewhere (McDonald et al., 2013; Tough et al., 2017). In brief, the AOF study recruited women during pregnancy from health care offices, laboratory offices, and community advertisements in Calgary, Alberta, Canada between May 2008 and December 2010. Women were eligible if they were (1) less than 25 weeks pregnant, (2) at least 18 years old, (3) receiving prenatal care in Calgary, and (4) able to complete survey measures in English. A total of 4,011 women were assessed and 3,387 were deemed eligible and enrolled (84% recruitment rate).

Prior to participation, written consent was obtained from all mothers. Mothers completed a baseline survey during pregnancy (< 25 weeks) and, to this date, have completed nine follow-up surveys at key developmental timepoints including 34-36 weeks pregnant (Q2), and at 4 months (Q3), 1 year (Q4), 2 years (Q5), 3 years (Q6), 5 years (Q7), 8 years (Q8), and 13 years (Q9) after delivery. Mothers were also invited to complete three abbreviated surveys during the COVID-19 pandemic (CV1, CV2, CV3) between Q8 and Q9 that included measures specific to the impact of the pandemic on their family as well as general measures of functioning and well-being. Starting with the COVID-19 surveys, mothers provided consent for their child to be invited to participate and complete self-report measures. Thus, from Q1 to Q8, mothers provided data on their own and their child's functioning whereas, from CV1 to Q9, children provided data on their own functioning. Sample sizes vary across timepoints, with response rates ranging from 65% to 99%. Consistent with other longitudinal studies during the COVID-19 pandemic (Kauhanen et al., 2023), completion rates were lower for the COVID-related surveys (e.g., 56% of eligible mothers completed CV2 and 88% of children whose mother consented to their participation completed CV3).

The current research used mother and child self-report survey data from Q1, Q7, Q8, CV2, CV3, and Q9 (see Figure 4.1). Mother-child dyads were included in the current research if they provided data for the maternal and child pain variables ($n = 1128$). The included sample of mothers were significantly more likely to be partnered ($\chi^2 = 15.98, p < 0.001$) and had higher levels of education ($\chi^2 = 30.19, p < 0.001$) and household income ($\chi^2 = 23.23, p < 0.001$) at baseline than mothers not retained (i.e., lost to follow-up or no pain variable data). Mothers did not differ on age or racial identity. Overall, the majority of mothers in this study were white (80%), married or common-law (94%), had at least some post-secondary education (86%), and reported an annual household income \geq \$80,000 (82%). The majority of children identified as male or female (93%) and white (76%). Sociodemographic characteristics of the included sample are displayed in Table 4.1.

Measures

Maternal Chronic Pain

Maternal chronic pain status was measured at Q7 and Q8 (child ages 5 and 8) with two single items that asked mothers whether they have chronic pain or chronic headaches (yes/no). Mothers who endorsed chronic pain or chronic headaches at both Q7 and Q8 were coded as having chronic pain.

Child Chronic Pain

Child pain was measured at Q9 (child age 13) with several self-report items. Child pain frequency was assessed with a single item that asked children to rate how often they had aches or pain in the past month on a 5-point scale (0 = not at all to 4 = everyday). Children who reported no pain at all in the past month did not complete the remaining pain items. Child pain intensity was measured using a validated and reliable 11-point Numerical Rating Scale (0 = no pain to 10

= worst pain possible) that asked how much hurt they usually have when they have aches or pains (Castarlenas et al., 2017; Safikhani et al., 2018; von Baeyer et al., 2009). Child pain chronicity was assessed with a single item that asked if they have had aches or pain for at least three months in a row (yes/no), which aligns with the current definition of chronic pain (Treede et al., 2019). Consistent with previous research assessing child chronic pain in population-based samples (Darlington et al., 2012; Perquin et al., 2000), children were coded as having chronic pain if they endorsed: pain frequency of at least 2 to 3 times per month, pain intensity of at least 5 out of 10, and having pain for at least three months in a row. Child pain locations were also assessed for descriptive purposes with a single item that asked children to select the parts of their body where they experienced the most aches or pains in the past month from a checklist of six options (stomach, head, muscles and joints, legs, chest, other).

Moderators

Maternal Anxiety Symptoms. The short-form of the state scale from the Spielberger State-Trait Anxiety Inventory (SSAI-SF; Marteau & Bekker, 1992) was completed by mothers at Q8 (child age 8). The six items of the SSAI-SF assess both the presence (e.g., “I feel tense”) and absence (e.g., “I feel content”) of anxiety on a four-point scale (1 = not at all to 4 = very much so). Anxiety-absent items were reverse-coded and then all items were summed to create a total score, with higher scores indicating greater anxiety symptoms (range: 6-24). This scale has shown acceptable reliability, validity, and correlations with the full 20-item scale in samples of students, pregnant women, and parents of infants with normal and abnormal screening tests (Marteau & Bekker, 1992; Tluczek et al., 2009). Cronbach’s alpha in this sample was $a = 0.86$.

Maternal Depressive Symptoms. Mothers completed the Center for Epidemiologic Studies Depression Scale (CES-D; Radloff, 1977) at Q8 (child age 8). The CES-D is comprised

of 20 items that assess symptoms of depression during the past week on a four-point scale (0 = rarely or none of the time to 3 = most or all of the time). Total scores were obtained by summing all items, such that higher scores indicated greater depressive symptoms (range: 0-60). The CES-D was developed for use in general population surveys and has demonstrated strong psychometric properties including good reliability, concurrent and construct validity, and diagnostic accuracy across community and clinical samples (Park & Yu, 2021; Radloff, 1977; Vilagut et al., 2016). Cronbach's alpha in the current sample was $\alpha = 0.92$.

Maternal Parenting. Mothers completed two subscales of the National Longitudinal Survey of Children and Youth (NLSCY) Parenting scales at Q8 (child age 8). The 7-item Ineffective/Hostile Parenting subscale asks respondents to rate how often they used ineffective parenting practices such as getting angry when punishing their child, repeatedly disciplining their child for the same thing, and changing their discipline based on their mood. The 5-item Positive Interaction subscale asks respondents to rate how often they have positive interactions with their child such as laughing, playing, or doing something special together. Items are rated on a 5-point scale (0 = never to 4 = many times a day/all the time) and summed to create a total score for each subscale, with higher scores indicating more ineffective (range: 0-28) or positive (range: 0-20) parenting. This scale was developed by an expert advisory group for the NLSCY and demonstrated validity in a factor analysis as well as acceptable reliability in the original sample. Both subscales had acceptable internal consistency in the current study (ineffective/hostile: $\alpha = 0.79$; positive interactions: $\alpha = 0.78$).

Maternal Social Support. The NLSCY Social Support scale was completed by mothers at Q8 (child age 8). This 8-item scale assesses the level of support that respondents feel they have from friends, family members, and members of the community (e.g., "I have family and

friends who help me feel safe, secure and happy”). Items are both positively and negatively worded and rated on a 4-point scale (0 = strongly agree to 3 = strongly disagree). Items were recoded and summed to create a total score (range: 0-24), with higher scores indicating greater social support. This measure was developed for the NLSCY by an expert advisory group using previously established measures of social support, and demonstrated acceptable reliability in the study sample. In the current sample, it had excellent internal consistency ($\alpha = 0.90$).

Maternal Optimism. The Life Orientation Test-Revised (LOT-R; Scheier et al., 1994) measured maternal optimism at CV2 (child age 11). The LOT-R is comprised of six items that measure aspects of optimism (e.g., “In uncertain times, I usually expect the best”) as well as four filler items that are not included in the total score. Items are rated on a 5-point scale (0 = I agree a lot to 4 = I disagree a lot) and summed to create a total score, with higher scores indicating higher optimism (range: 0-24). The LOT-R has demonstrated acceptable internal consistency, temporal stability, and validity with other measures in the literature (Scheier et al., 1994). In the current sample, Cronbach’s alpha demonstrated good internal consistency ($\alpha = 0.86$).

Maternal Active Coping. The Brief Resilient Coping Scale (BRCS; V. G. Sinclair & Wallston, 2004) was completed by mothers at CV2 (child age 11). The BRCS consists of four items that assess tendencies to cope with stress in a highly adaptive manner. Respondents rate each item on a five-point scale (1 = describes me not at all to 5 = describes me very well). Total scores were obtained by summing the items, with higher scores reflecting more active coping behaviours (range: 4-20). The BRCS was developed in samples of adults with rheumatoid arthritis, and has demonstrated acceptable reliability and validity in both clinical and population-based samples (Kocalevent et al., 2017; V. G. Sinclair & Wallston, 2004). Cronbach’s alpha in the current sample was acceptable ($\alpha = 0.77$).

Child Optimism. The 3-item Optimism subscale of the Middle Years Development Instrument (MDI; Schonert-Reichl et al., 2013) was administered as a self-report measure of child optimism at CV3 (child age 12). This subscale asks respondents to rate how much they agree with various statements (e.g., “I believe more good things than bad things will happen to me”) on a 5-point scale (1 = disagree a lot to 5 = agree a lot). Items are summed to create a total score (range: 3-15), with higher scores indicating greater optimism. The MDI has shown strong psychometric properties including acceptable reliability and convergent and discriminant validity in population-based samples of school-aged children (Gregory et al., 2019; Schonert-Reichl et al., 2013). In the current sample, the Optimism subscale had good reliability ($\alpha = 0.84$).

Child Connections with Adults and Peers. Children self-reported their sense of support and belonging with adults at home, at school, in the community, and with peers at CV3 (child age 12) using the subscales for each of these domains from the MDI (Schonert-Reichl et al., 2013). Each subscale is comprised of three items that asks children to rate how much they agree with various statements (e.g., “In my home, there is a parent or another adult who I can talk to about my problems” and “When I am with other kids my age, I feel like I belong”) on a 4-point scale (1 = not at all true to 4 = very much true). Items for each subscale were summed to create total scores (range: 3-12), with higher scores indicating greater connections with adults or peers. The subscales showed acceptable to excellent reliability in the current sample (adults at home: $\alpha = 0.77$; adults at school: $\alpha = 0.86$; adults in the community: $\alpha = 0.95$; peers: $\alpha = 0.84$).

Child Engagement with Community. Children self-reported whether they had taken part in five different recreational activities (i.e., individual or team sports, music or art classes, community programs, or outdoor activities such as hiking) in the past two weeks (yes/no) at CV3

(child age 12). A summed score was calculated across all items with higher scores representing greater engagement in community activities (range: 0-5).

Sociodemographic Characteristics

Sociodemographic information was collected from mothers and children across various timepoints. Mothers reported on their age (Q1), racial/ethnic identity (Q1), marital status (Q6), education (Q7), and annual household income (Q7). Children self-reported on their gender (Q9) and racial/ethnic identity while their age (Q9) was calculated from their birth date.

Statistical Analyses

Analyses were conducted using SPSS (version 29; IBM Statistics, Armonk, NY, USA). Descriptive statistics characterized the sample, independent samples t-tests or chi-square tests compared mothers with (versus without) chronic pain and children on sociodemographic and study variables, and correlation analyses examined bivariate associations between the study variables. Continuous data are presented as mean \pm standard deviation (SD) and count data are presented as number (percentage). Binary logistic regressions were conducted to examine moderators of the association between maternal chronic pain and child chronic pain. Maternal chronic pain and the risk or protective/promotive factor were entered simultaneously as predictors in the first step to examine the main effects (risk or *promotive* effects for all children in the sample) of these variables on child chronic pain and then the interaction term of these variables (maternal chronic pain x the risk or protective/promotive factor) was entered in the next step to examine their moderating effect (risk or *protective* effect for children of mothers with versus without chronic pain). Separate logistic regression models were conducted for each risk or protective/promotive factor. Significant interactions were visualized and probed using the analysis of simple slopes through the PROCESS Macro for SPSS (Version 4.2; A. F. Hayes,

2022). For the analysis of simple slopes, the 16th, 50th, and 84th percentile of the distribution of the moderator variable was used to define low, moderate, and high levels of the moderator, as recommended by Hayes (2022). For the logistic regression results, odds ratios (OR) and 95% confidence intervals (CI) are reported. For the simple slope analyses, the regression or beta coefficient (b) is reported. Statistical assumptions of regression models were explored through examination of model residuals.

Unadjusted analyses were first conducted with study variables and then adjusted for sociodemographic variables that were selected *a priori* based on their association with parent and/or child chronic pain (maternal age, racial/ethnic identity, education, marital status, household income; child age and gender; Beveridge et al., 2018; Chambers et al., 2024; Wilson & Fales, 2015). Categorical covariates were dichotomized as follows: maternal racial/ethnic identity (0 = white; 1 = racialized), education (0 = at least some post-secondary; 1 = high school or less), marital status (0 = married or common-law; 1 = single, divorced/separated, or widowed), and household income (0 = \$80,000 or more; 1 = less than \$80,000) and child gender (0 = male; 1 = female or gender diverse). Maternal and child age were entered as continuous variables. Covariates were entered in the first step before the key study variables. Unadjusted and adjusted results are presented in tables and adjusted results are discussed in text.

The significance level was set at $p < 0.05$ and no corrections were applied as hypotheses were generated *a priori*, models were not rerun in different subsamples, and this was an exploratory (versus confirmatory) study (Althouse, 2016; Bender & Lange, 2001; Perneger, 1998). No *a priori* sample size calculations were conducted as this study used a convenience sample from a larger cohort. Missing data varied across timepoints, with < 2% of data missing from Q8, 16-17% missing from CV2, and 26-27% missing from CV3. Missing data on CV2 and

CV3 were not significantly associated with maternal or child chronic pain status ($ps > 0.05$). See Table 4.2 for the number of participants that completed each study variable.

Analyses were conducted with complete cases to allow for the use of the PROCESS Macro. Two sets of sensitivity analyses for the primary analyses were also conducted, first with bootstrapping applied to account for any violations to the assumptions of normality and homoscedasticity and then with multiple imputation to account for missing data (Field, 2018). More detailed methods and results of these sensitivity analyses are presented as supplementary material (Appendix C) and briefly described and compared to the results of the complete case analyses in the text.

Results

Descriptive Statistics

In the current sample, 9.8% ($n = 111$) of mothers and 15.6% ($n = 176$) of children reported chronic pain. Children whose mothers had chronic pain were significantly more likely to report chronic pain themselves than children whose mothers did not endorse chronic pain (26.1% vs. 14.5%; $\chi^2 = 10.81, p = 0.001$). Descriptive statistics of study variables grouped by maternal chronic pain status are reported in Table 4.2. Mothers with chronic pain reported significantly greater anxiety and depressive symptoms and significantly lower social support, optimism, and active coping than mothers without chronic pain. Children of parents with versus without chronic pain reported significantly lower peer belonging and community engagement. Mothers with versus without chronic pain did not significantly differ on ineffective or positive parenting, and their children did not significantly differ on optimism or connections with adults. Correlations between the study variables are displayed in Table 4.3.

Moderator Analyses

Maternal Factors

Results of the logistic regression models examining maternal factors moderating the association between maternal and child chronic pain are presented in Table 4.4. For main effects, maternal chronic pain was a significant predictor of child chronic pain across all models, when controlling for any of the maternal risk or protective/promotive factors and sociodemographic covariates (odds ratios [ORs] ranged from 1.76 to 2.09). Maternal depressive symptoms were the only maternal factor that predicted significantly increased risk of child chronic pain, while controlling for maternal chronic pain and covariates (OR = 1.03 [1.00, 1.05], $p = 0.019$).

In terms of moderation effects, the interaction term between maternal chronic pain and maternal anxiety symptoms was significant, indicating that the association between maternal and child chronic pain varied as a function of maternal anxiety symptoms. Simple slope analysis revealed that the odds of child chronic pain was significantly increased when mothers with chronic pain had higher anxiety symptoms ($b = 0.90$, $p = 0.002$), but not when mothers with chronic pain had moderate ($b = 0.11$, $p = 0.765$) or lower ($b = -0.36$, $p = 0.499$) anxiety symptoms. The level of maternal anxiety symptoms had no statistically significant effect on the odds of child chronic pain when mothers did not have chronic pain. See Figure 4.2a. The interaction term with maternal ineffective parenting was also significant, indicating that the association between mother and child chronic pain varied as a function of maternal ineffective parenting. Simple slope analysis revealed that the odds of child chronic pain was significantly increased when mothers with chronic pain reported high levels of ineffective parenting ($b = 1.23$, $p < 0.001$), but not when mothers with chronic pain had moderate ($b = 0.53$, $p = 0.069$) or lower ($b = 0.11$, $p = 0.795$) levels of ineffective parenting. See Figure 4.2b.

Child Factors

Results of the logistic regression models examining child factors moderating the association between maternal and child chronic pain are presented in Table 4.5. In terms of main effects, maternal chronic pain remained a significant predictor of child chronic pain across all models including when controlling for any of the child protective/promotive factors and sociodemographic variables (ORs ranged from 1.99 to 2.22). Several child factors were also significant predictors of reduced risk for child chronic pain while controlling for maternal chronic pain and covariates. Specifically, greater child optimism (OR = 0.91 [0.85, 0.97], $p = 0.004$), greater child connections with adults at home (OR = 0.87 [0.77, 0.98], $p = 0.023$), and greater child connections with adults at school (OR = 0.89 [0.81, 0.99], $p = 0.025$) had promotive effects on child chronic pain. None of the interaction terms between maternal chronic pain and child protective factors were significant.

Sensitivity Analyses

Results of the first set of sensitivity analyses for the primary analyses, which applied bootstrapping to address any violations to model assumptions, were comparable to the described results (see Supplementary Tables 4.1 and 4.2 in Appendix C). Results of the second set of sensitivity analyses, which used multiple imputation to address missing data, were also largely comparable to the described results that used complete cases (see Supplementary Tables 4.3 and 4.4 in Appendix C). For these analyses, imputed cases ranged from 11 to 311 for the unadjusted analyses and from 158 to 412 for the adjusted analyses. The statistical significance of several results differed across analyses, mostly when statistical significance was marginal or trending. For example, in the multiple imputation analyses, the interaction terms with maternal anxiety symptoms ($p = 0.077$) and maternal ineffective parenting ($p = 0.066$) were no longer statistically significant while the main effects of maternal optimism (OR = 0.96 [0.93, 1.00], $p = 0.039$) and

maternal active coping (OR = 0.93 [0.87, 1.00], $p = 0.040$) were statistically significant. Similarly, the main effects of child connections with adults at home (OR = 0.90 [0.80, 1.01], $p = 0.080$) and at school (OR = 0.95 [0.86, 1.04], $p = 0.241$) were no longer statistically significant. However, all effect sizes remained closely comparable and well within 95% confidence intervals.

Discussion

The aim of the current multi-wave, multi-informant study was to identify individual and interpersonal factors across childhood and adolescence that moderated the association between maternal and child chronic pain, either increasing or decreasing the risk of transmission among children of parents with (versus without) chronic pain. Hypotheses were partially supported. First, children were at significantly increased risk for chronic pain in early adolescence (age 13) when their mother reported chronic pain earlier in their childhood (from ages 5 to 8), consistent with a large body of research on the intergenerational transmission of chronic pain (K. S. Higgins et al., 2015). Second, maternal anxiety symptoms and ineffective parenting moderated the association between maternal and child chronic pain. Specifically, children had significantly increased odds of reporting chronic pain in early adolescence when their mothers had both chronic pain and higher (versus lower) anxiety symptoms or ineffective parenting practices earlier in their childhood. No other maternal or child factors significantly moderated the association between maternal and child chronic pain; however, several factors were significant predictors of child chronic pain in the overall sample (i.e., children of parents with and without chronic pain). Specifically, maternal depressive symptoms in earlier childhood (age 8) increased the risk of child chronic pain while child optimism and child connections with adults at home and at school in early adolescence (age 12) decreased the risk of chronic pain at age 13 for all children. Effect sizes were small in magnitude, which is consistent with previous research on

intergenerational associations (Beveridge et al., 2024; Goodman et al., 2011; K. S. Higgins et al., 2015; Racine et al., 2023) and suggests that other factors not examined in this study also contribute to risk-resiliency for pediatric chronic pain; however, these associations may still be clinically meaningful.

This study contributes to an emerging literature on resilience and pediatric chronic pain. To date, research has focused on identifying factors that promote adaptive outcomes for children already living with chronic pain and accessing tertiary pain treatment. Our results extend this research by identifying factors that may predict reduced risk for developing chronic pain among children at-risk for chronic pain due to their parent's chronic pain as well as children at lower-risk of chronic pain within a community-based sample. As highlighted by resilience researchers, the inclusion of both higher- and lower- risk groups allows for the differentiation of factors that may be particularly important for disrupting the intergenerational transmission of chronic pain (protective factors) and factors that may help to prevent chronic pain in the general population (promotive factors; Masten et al., 2021).

Interestingly, none of the investigated factors were found to have a significant *protective* effect, buffering the transmission of chronic pain from mothers to children. Rather, several child factors, specifically optimism and connections with adults, had a *promotive* effect, reducing the risk of chronic pain for all children. Maternal optimism and active coping may also act as promotive factors, as they bordered on statistical significance between complete case and multiple imputation analyses. These variables have been posited as potential resilience factors within pediatric chronic pain (Cousins, Kalapurakkel, et al., 2015; Stone & Wilson, 2016), and thus warrant further study.

Optimism is associated with better adaptation to pain in clinical samples of children and adults, with several studies showing this association is explained through decreased use of maladaptive coping strategies (i.e., catastrophic thinking about pain; Cousins, Cohen, et al., 2015; Goodin et al., 2013; Hood et al., 2012). Our findings suggest that optimism may also protect against the development of chronic pain, supporting conceptual models that position optimism as a resilience factor for chronic pain and adding to the evidence that positive pain-related outcomes can occur not just when there are lower levels of risk factors but also when there are greater levels of protective or promotive factors (Cousins, Kalapurakkel, et al., 2015; Goubert & Trompetter, 2017; Sturgeon & Zautra, 2010). Indeed, risk and resilience factors have been shown to act separately, underscoring the importance of dual-factor models that include both risk and resilience factors (Beeckman, Simons, et al., 2019; B. W. Smith & Zautra, 2008).

The promotive effect of child connections with adults at home and school in our study adds to a robust literature on the positive impact of social support for children's physical and mental health. For example, research on 'positive childhood experiences' consistently shows that children with more supportive and close relationships with adults inside or outside of the home have reduced risk of physical and mental health disorders, including chronic pain, even when also exposed to adverse childhood experiences (Bethell et al., 2019; Guo et al., 2022; Hinojosa & Hinojosa, 2024; Narayan et al., 2018; Pugh et al., 2023; Qu et al., 2022). Contrary to this literature, in which high-quality peer relationships and greater engagement with the community (e.g., connections with community members, participating in community traditions) is associated with better child outcomes (Cousins, Kalapurakkel, et al., 2015; Guo et al., 2022; Pugh et al., 2023; Qu et al., 2022; Ross et al., 2018), child connections with peers or adults in the community and engagement in community activities did not predict reduced risk of child chronic pain. These

non-significant findings may reflect the impact of the COVID-19 pandemic on forming connections with peers and engaging with the community. They may also reflect the relative importance of factors at more proximal (e.g., family, school) versus distal (e.g., community) levels of the child's ecology. The type of community engagement may also matter for chronic pain. For example, activities that involve more physical activity may be confounded by both maternal and child chronic pain. Further investigation of the role of these factors within pediatric chronic pain is needed.

Contrary to the protective factors, two risk factors – maternal anxiety symptoms and ineffective parenting – moderated the relationship between maternal and child chronic pain. Mothers with chronic pain and greater anxiety symptoms may be more sensitive or reactive to pain in their child and engage in behaviours that draw attention and concern to their child's pain, which could in turn impact the child's response to pain. This aligns with the interpersonal fear avoidance model, a guiding framework for understanding how child and parent factors (e.g., pain catastrophizing, pain-related fears, avoidance) may lead to children's pain-related disability (Goubert & Simons, 2013; Neville et al., 2021; Simons et al., 2015). A recent study demonstrated that parent physical and mental health significantly related to child pain-related disability through interpersonal fear avoidance mechanisms (i.e., parent and child pain catastrophizing, parent protective responses), lending further support to the important role that both parent chronic pain and mental health play in the onset and maintenance of pediatric chronic pain (Beveridge et al., 2024; Birnie et al., 2020; Poppert Cordts et al., 2019). Our findings that maternal chronic pain, anxiety symptoms, and depressive symptoms contributed to children's risk for chronic pain in different ways (main effect or moderated effect) add to a growing literature demonstrating the need to incorporate parent physical and emotional

functioning into models of pediatric chronic pain to disentangle their complex interactions with children's pain (Coenders et al., 2014; Helgeland et al., 2010; Hoftun et al., 2013; Incledon et al., 2016; Poppert Cordts et al., 2019; Ramchandani et al., 2006).

While pain-specific parenting practices (e.g., protective responses to child pain) have been well-studied within pediatric pain, general parenting practices including hostile and inconsistent parenting have received minimal attention. Our results found that, even though mothers with and without chronic pain reported similar parenting practices, children were more likely to report chronic pain when their mother endorsed both chronic pain and more ineffective parenting. Similar results have been found in studies on the intergenerational transmission of depression, wherein lower protective and controlling parenting predicted better outcomes for children of parents with depression but not children of parents without depression (Brennan et al., 2003; Lewandowski et al., 2014; Pargas et al., 2010). Thus, children at higher-risk for poor outcomes due to their parent's health appear to be particularly sensitive to maladaptive parenting practices. Interestingly, positive parenting did not act as a protective or promotive factor, which suggests that reducing hostile parenting may be more important for improving child outcomes than increasing positive parent interactions.

Clinical Implications

An understanding of factors that not only contribute to the continuation of, but also to the lack thereof, chronic pain across generations is necessary to inform interventions to prevent the development and maintenance of chronic pain (Branje et al., 2020). Findings from the current study suggest that targeting elevated anxiety symptoms and ineffective parenting among mothers with chronic pain may improve the effectiveness of primary prevention efforts aimed at reducing the transmission of pain from mothers to children whereas targeting elevated depressive

symptoms among all mothers may help to prevent chronic pain among children in general. In addition, interventions that help children develop close and supportive relationships with adults in their home or school and teach strategies for learned optimism may mitigate children's general risk for chronic pain. As suggested by calls for the evidence on childhood adversity to be applied to clinical practice, these targets may be best addressed through pediatric primary care, for example through screening for poor parent mental and physical health, providing education to parents on self-care and parenting, referring to community-based services and helping families overcome barriers to engage in these services, and offering group-based supports to parents and children (Traub & Boynton-Jarrett, 2017).

Strengths and Limitations

The current study had several notable strengths including the longitudinal design, multi-informant data, and community sample including mothers and children with and without chronic pain. However, several limitations are worthy of note. First, the measure of maternal chronic pain asked about the presence, but not frequency, severity, or impact, of chronic pain, all of which can vary across individuals (Elliott et al., 2014; Goubert et al., 2004; Häuser et al., 2014; Wilson et al., 2020). In fact, a recent framework emphasized the need to include impact (low vs. high) along with duration (acute vs. chronic) in studies of pain development, maintenance, and resolution (Eccleston et al., 2023). This is also important for studies of risk-resilience to chronic pain as resilience can be defined as 'better than expected' outcomes based on the risk exposure (Collishaw et al., 2016). Thus, a child who develops low-impact chronic pain, but has a parent with high-impact chronic pain, could be demonstrating resilience. In addition, mothers had to report chronic pain across two timepoints (child ages 5 and 8) to be coded as having chronic pain to ensure that children had sufficient exposure to maternal chronic pain. Perhaps due to this more

conservative approach, the prevalence of maternal chronic pain in the current study (10%) was lower than previous studies estimating the point prevalence of chronic pain in adults (20-40%; Dahlhamer et al., 2018; Schopflocher et al., 2011; Yong et al., 2022). Future research in this area should include more comprehensive assessments of parent chronic pain and carefully consider how ‘resilience’ is defined.

The measurement of child chronic pain also captured the presence, but not new onset, of chronic pain in children at a specific developmental period (age 13). While the incidence of chronic pain increases in early adolescence (Perquin et al., 2000; Stanford et al., 2008), children who did not report chronic pain in the current study may go on to develop chronic pain while children who did report chronic pain may experience a resolution of their pain. Moreover, the chronic pain reported by children at age 13 may have developed at an earlier age, prior to the measurement of maternal chronic pain and/or the risk and protective factors. Prospective research that follows children throughout their childhood and into later adolescence and young adulthood is needed to understand how the processes of risk-resilience may shift across different developmental periods to predict the onset and/or resolution of chronic pain.

The current sample also focused on maternal chronic pain. The inclusion of both parents in intergenerational research has been emphasized to allow for a comprehensive examination of patterns (e.g., same-sex vs. mixed-sex parent-child dyads), predictors (co-parent moderators), and processes (genes vs. environment; Branje et al., 2020). Indeed, children are more likely to develop chronic pain, particularly multisite and/or disabling chronic pain, when both parents have chronic pain as compared to just one parent (Assadi et al., 2015; Hoftun et al., 2013; Lier et al., 2016; Zadro et al., 2020). However, several studies have found that maternal chronic pain is a stronger predictor of child chronic pain than paternal chronic pain (Assadi et al., 2015;

Buonavolontà et al., 2010; Evans & Keenan, 2007; Lier et al., 2016; Ramchandani et al., 2006; Zadro et al., 2020). Thus, the focus of this study on mothers is an appropriate first step; however, future research should include both parents.

Only a handful of potential moderators were examined at various timepoints between child ages 8 and 12. Due to the design of the larger study, the child protective/promotive factors were examined closer in time to the outcome of child chronic pain than many of the parent risk and protective/promotive factors, which may have contributed to the strength of the associations between these parent and child factors and child chronic pain. Future research should measure parent and child variables at similar timepoints throughout childhood and adolescence to confirm these associations. Other psychosocial factors, such as exposure to other stressful or adverse events and lifestyle behaviours (e.g., diet, physical activity, screen time, sleep), biological factors (e.g., genetics, heart rate variability), and community factors (e.g., neighbourhood safety and support) that have been shown to relate to the presence of child chronic pain (Coenders et al., 2014; Helgeland et al., 2010; Incledon et al., 2016; Laird et al., 2015; Lier et al., 2016; Meredith et al., 2008; Pugh et al., 2023; Tremblay & Sullivan, 2010; Vandeleur et al., 2024; Zadro et al., 2020) and/or moderate the association between maternal depression and child outcomes (Collishaw et al., 2016; Goodman, 2020), should be investigated as potential moderators of the intergenerational transmission of chronic pain. Pain-specific factors including adaptive (e.g., pain acceptance, pain-related self-efficacy) and non-adaptive (e.g., pain catastrophizing, avoidance) coping strategies likely also play a role and should be investigated in future studies (Beeckman, Hughes, et al., 2019; Beeckman, Simons, et al., 2019; Cousins, Kalapurakkel, et al., 2015; Feinstein et al., 2018; Lee et al., 2020). Future research with greater diversity is also needed to better understand how the transmission of pain may differ based on sociodemographic

characteristics. While we controlled for relevant sociodemographic variables, a more nuanced investigation of their role could not be conducted due to the homogeneity of the current sample (i.e., predominately white, married, university-educated, higher income).

Lastly, the sample size varied across timepoints in the current study, with the lowest rates for the COVID-19 timepoints, as these were add-on surveys. Sensitivity analyses were conducted with multiple imputation to account for this missing data; however, this attrition may have still introduced bias. Due to the novelty of this research, potential moderators were examined in separate analyses and adjustments for multiple comparisons were not made based on recommendations for when adjustments should not be applied (Althouse, 2016; Bender & Lange, 2001; Perneger, 1998). Nonetheless, the likelihood of type I errors may have been inflated and replication of the current findings is needed. For example, the lower bound of the confidence interval for the effect between maternal depressive symptoms and child chronic pain borders on statistical significance, suggesting that this association may not be meaningful. Further research that confirms the current findings is needed. As additional risk and protective factors are identified in empirical studies, more complex statistical models can be built that examine interactions and bidirectional associations between factors at various levels of the child's ecology, in line with a developmental systems approach (Masten, 2016).

Conclusion

The current study examined risk and protective factors across childhood that predicted either increased or decreased risk for chronic pain in early adolescence among children of mothers with versus without chronic pain. Overall, children had significantly increased risk for chronic pain when their mothers reported chronic pain, greater depressive symptoms, greater anxiety symptoms *and* chronic pain, or more ineffective parenting practices *and* chronic pain.

Children had a significantly reduced risk for chronic pain when they endorsed higher levels of optimism and more connections with adults at home and school. These initial findings, which merit replication in other samples, highlight factors that could be targeted in prevention efforts. Further research that assesses additional factors at the various levels of the child's ecology in more diverse samples is needed for a comprehensive understanding of the multidimensional and biopsychosocial nature of the intergenerational continuation (and discontinuation) of pain.

Table 4.1*Sociodemographic Characteristics of the Sample*

	Total sample (<i>N</i> = 1128)	Mothers with chronic pain (<i>n</i> = 111)	Mothers without chronic pain (<i>n</i> = 1017)
	M ± SD or n (%)		
Parent characteristics			
Age, years	36.34 (4.30)	36.62 (4.05)	36.31 (4.33)
Racial/ethnic identity			
Asian	156 (13.8)	12 (10.8)	144 (14.2)
Black	13 (1.2)	1 (0.9)	12 (1.2)
First Nations, Metis, or Inuit	7 (0.6)	2 (1.8)	5 (0.5)
Latin American	17 (1.5)	2 (1.8)	15 (1.5)
Mixed or other	35 (3.1)	1 (0.9)	34 (3.3)
White	897 (79.5)	92 (82.9)	805 (79.2)
Missing	3 (0.3)	1 (0.9)	2 (0.2)
Marital status			
Married or common-law	1065 (94.4)	106 (95.5)	959 (94.3)
Single, separated, divorced, or widowed	63 (5.6)	5 (4.5)	58 (5.7)
Annual household income, CAD			
< \$80,000	193 (17.1)	22 (19.8)	171 (16.8)
≥ \$80,000	930 (82.4)	88 (79.3)	842 (82.8)
Missing	5 (0.4)	1 (0.9)	4 (0.4)
Education level			
High school or less	56 (5.0)	6 (5.4)	50 (4.9)

At least some post-secondary	778 (69.0)	76 (68.5)	702 (69.0)
At least some graduate school	192 (17.0)	21 (18.9)	171 (16.8)
Missing	102 (9.0)	8 (7.2)	94 (9.2)
Child characteristics			
Age, years	12.83 (0.78)	12.73 (0.83)	12.87 (0.77)
Gender			
Female	481 (42.6)	53 (47.7)	428 (42.1)
Male	572 (50.7)	51 (45.9)	521 (51.2)
Gender diverse	37 (3.3)	4 (3.6)	33 (3.2)
Other or prefer not to answer	32 (2.8)	2 (1.8)	30 (3.0)
Missing	6 (0.5)	1 (0.9)	5 (0.5)
Racial/ethnic identity			
Asian	125 (11.1)	9 (8.1)	116 (
Black	17 (1.5)	1 (0.9)	16 (1.6)
Indigenous	17 (1.5)	7 (6.3)	10 (1.0)
Latin American	13 (1.2)	2 (1.8)	11 (1.1)
Middle Eastern	18 (1.6)	0 (0.0)	18 (1.8)
White	860 (76.2)	83 (74.8)	777 (76.4)
Other	74 (6.6)	8 (7.2)	66 (6.5)
Missing	4 (0.4)	1 (0.9)	3 (0.3)
Presence of chronic pain	176 (15.6)	29 (26.1)	147 (14.5)
Pain frequency			
Not at all	212 (18.8)	14 (12.6)	198 (19.5)
Once per month	279 (24.7)	26 (23.4)	253 (24.9)
2 to 3 times per month	390 (34.6)	40 (36.0)	350 (34.4)

Almost everyday	201 (17.8)	25 (22.5)	176 (17.3)
Everyday	46 (4.1)	6 (5.4)	40 (3.9)
Pain locations			
Head	375 (33.2)	50 (45.0)	325 (32.0)
Muscles and joints	437 (38.7)	49 (44.1)	388 (38.2)
Stomach	307 (27.2)	38 (34.2)	269 (26.5)
Chest	123 (10.9)	16 (14.4)	107 (10.5)
Leg(s)	374 (33.2)	34 (30.6)	340 (33.4)
Other	119 (10.5)	14 (12.6)	105 (10.3)
Two or more locations	489 (43.4)	61 (55.0)	428 (42.1)

Note. More than one pain location could be selected.

Table 4.2*Descriptive Statistics of the Study Variables Grouped by Maternal Chronic Pain Status*

	Mothers with chronic pain	Mothers without chronic pain	Difference test	Complete cases
Variable	M (SD)	M (SD)		N
Maternal anxiety symptoms	12.29 (4.24)	9.84 (3.41)	<i>t</i> = 6.95 , <i>p</i> < 0.001	1110
Maternal depressive symptoms	14.93 (9.78)	8.27 (8.09)	<i>t</i> = 8.05 , <i>p</i> < 0.001	1125
Maternal ineffective parenting	10.05 (3.89)	9.83 (4.00)	<i>t</i> = 0.53, <i>p</i> = 0.594	1112
Maternal positive parenting	13.80 (2.69)	13.48 (2.71)	<i>t</i> = 1.20, <i>p</i> = 0.233	1122
Maternal social support	19.32 (4.55)	20.80 (3.96)	<i>t</i> = -3.70 , <i>p</i> < 0.001	1113
Maternal optimism	14.55 (5.17)	17.27 (4.81)	<i>t</i> = -5.02 , <i>p</i> < 0.001	938
Maternal active coping	14.37 (2.60)	15.20 (2.67)	<i>t</i> = -2.81 , <i>p</i> = 0.005	950
Child optimism	12.03 (2.62)	12.27 (2.96)	<i>t</i> = -0.70, <i>p</i> = 0.485	832
Child connections with adults at home	10.78 (1.66)	10.90 (1.55)	<i>t</i> = -0.69, <i>p</i> = 0.490	834
Child connections with adults at school	9.87 (2.15)	9.95 (2.09)	<i>t</i> = -0.30, <i>p</i> = 0.768	831
Child connections with adults in community	8.65 (3.05)	8.41 (3.06)	<i>t</i> = 0.66, <i>p</i> = 0.509	834
Child peer belonging	9.35 (2.29)	9.90 (2.15)	<i>t</i> = -2.16 , <i>p</i> = 0.031	838
Child community engagement	1.96 (1.25)	2.33 (1.13)	<i>t</i> = -2.72 , <i>p</i> = 0.007	823

Table 4.3*Bivariate Correlations Between Study Variables*

Variable	2	3	4	5	6	7	8	9	10	11	12	13	14
1. Maternal anxiety symptoms	0.61**	0.26**	-0.17**	-0.39**	-0.36**	-0.23**	-0.06	-0.11**	-0.06	-0.03	-0.11**	-0.07*	0.05
2. Maternal depressive symptoms	-	0.29**	-0.13**	-0.49**	-0.43**	-0.26**	-0.10**	-0.12**	-0.09*	-0.06	-0.13**	-0.08*	0.08**
3. Maternal ineffective parenting		-	-0.32**	-0.18**	-0.15**	-0.21**	-0.11**	-0.17**	-0.01	-0.07*	-0.05	0.01	-0.01
4. Maternal positive parenting			-	0.14**	0.16**	0.22**	0.09**	0.11**	0.07*	0.07*	0.08*	0.03	-0.02
5. Maternal social support				-	0.31**	0.17**	0.02	0.09**	0.14**	0.13**	0.13**	0.14**	-0.02
6. Maternal optimism					-	0.52**	0.06	0.08*	0.11**	0.15**	0.14**	0.12**	-0.07*
7. Maternal active coping						-	0.07*	0.10**	0.07*	0.12**	0.09*	0.06	-0.07*
8. Child optimism							-	0.47**	0.41**	0.31**	0.46**	0.09**	-0.11**
9. Child connections with adults at home								-	0.47**	0.36**	0.39**	0.07*	-0.08*

10. Child connections with adults at school	-	0.41**	0.39**	0.12**	-0.06
11. Child connections with adults in community	-	0.27**	0.19**		-0.05
12. Child peer belonging			-	0.13**	-0.07*
13. Child community engagement				-	-0.02
14. Child chronic pain					-

Note. * $p < 0.05$, ** $p < 0.01$

Table 4.4

Logistic Regression Analyses Examining Maternal Moderators of the Association Between Maternal and Child Chronic Pain

	Unadjusted				Adjusted			
	B (SE)	Wald	<i>p</i>	OR (95% CI)	B (SE)	Wald	<i>p</i>	OR (95% CI)
Step 1								
Maternal chronic pain	0.66 (0.24)	7.41	.006	1.94 (1.20, 3.13)	0.64 (0.27)	5.74	.017	1.90 (1.12, 3.20)
Maternal anxiety symptoms	0.03 (0.02)	1.17	.279	1.03 (0.98, 1.07)	0.04 (0.03)	2.92	.088	1.05 (0.99, 1.10)
Step 2								
Interaction	0.12 (0.06)	4.26	.039	1.13 (1.01, 1.27)	0.16 (0.07)	5.37	.020	1.17 (1.03, 1.34)
Step 1								
Maternal chronic pain	0.65 (0.24)	7.14	.008	1.92 (1.19, 3.09)	0.56 (0.27)	4.36	.037	1.76 (1.04, 2.98)
Maternal depressive symptoms	0.02 (0.01)	3.62	.057	1.02 (1.00, 1.04)	0.03 (0.01)	5.51	.019	1.03 (1.00, 1.05)
Step 2								
Interaction	0.03 (0.02)	1.05	.306	1.03 (0.98, 1.08)	0.03 (0.03)	1.41	.236	1.03 (0.98, 1.09)
Step 1								
Maternal chronic pain	0.76 (0.24)	10.40	.001	2.13 (1.35, 3.38)	0.74 (0.26)	8.04	.005	2.09 (1.26, 3.46)
Maternal ineffective parenting	-0.01 (0.02)	0.29	.593	0.99 (0.95, 1.03)	-0.003 (0.02)	0.01	.910	1.00 (0.95, 1.04)
Step 2								

Interaction	0.12 (0.06)	3.96	.047	1.13 (1.00, 1.26)	0.14 (0.07)	4.52	.034	1.15 (1.01, 1.31)
Step 1								
Maternal chronic pain	0.76 (0.24)	10.60	.001	2.15 (1.36, 3.40)	0.74 (0.26)	8.11	.004	2.09 (1.26, 3.47)
Maternal positive parenting	-0.02 (0.03)	0.57	.449	0.98 (0.92, 1.04)	-0.01 (0.03)	0.13	.716	0.99 (0.93, 1.06)
Step 2								
Interaction	-0.04 (0.09)	0.24	.622	0.96 (0.81, 1.14)	-0.06 (0.10)	0.39	.531	0.94 (0.78, 1.14)
Step 1								
Maternal chronic pain	0.76 (0.24)	10.23	.001	2.13 (1.34, 3.39)	0.72 (0.26)	7.72	.005	2.06 (1.24, 3.44)
Maternal social support	-0.01 (0.02)	0.07	.799	1.00 (0.96, 1.04)	-0.01 (0.02)	0.07	.786	0.99 (0.95, 1.04)
Step 2								
Interaction	-0.04 (0.05)	0.49	.485	0.97 (0.87, 1.07)	-0.02 (0.06)	0.15	.698	0.98 (0.87, 1.10)
Step 1								
Maternal chronic pain	0.66 (0.27)	5.88	.015	1.93 (1.13, 3.28)	0.67 (0.30)	5.16	.023	1.96 (1.10, 3.49)
Maternal optimism	-0.03 (0.02)	2.60	.107	0.97 (0.94, 1.01)	-0.03 (0.02)	2.82	.093	0.97 (0.93, 1.01)
Step 2								
Interaction	0.06 (0.05)	1.34	.248	1.06 (0.96, 1.18)	0.08 (0.06)	2.08	.149	1.08 (0.97, 1.21)
Step 1								
Maternal chronic pain	0.66 (0.27)	6.12	.013	1.93 (1.15, 3.26)	0.70 (0.29)	5.93	.015	2.02 (1.15, 3.56)

Maternal active coping	-0.06 (0.03)	3.54	.060	0.94 (0.88, 1.00)	-0.06 (0.04)	3.00	.083	0.94 (0.87, 1.01)
Step 2								
Interaction	0.08 (0.10)	0.62	.431	1.09 (0.89, 1.33)	0.14 (0.11)	1.50	.221	1.15 (0.92, 1.43)

Note. Adjusted analyses include maternal age, maternal racial/ethnic identity, maternal education, maternal marital status, annual household income, child age, and child gender as covariates in the first step of model building. Reported statistics for maternal chronic pain and the risk or protective/promotive factor are main effects, from the step prior to including the interaction term.

Abbreviations. CI, confidence interval; OR, odds ratio

Table 4.5

Logistic Regression Analyses Examining Child Moderators of the Association Between Maternal and Child Chronic Pain

	Unadjusted				Adjusted			
	B (SE)	Wald	<i>p</i>	OR (95% CI)	B (SE)	Wald	<i>p</i>	OR (95% CI)
Step 1								
Maternal chronic pain	0.80 (0.28)	8.00	.005	2.23 (1.28, 3.88)	0.80 (0.31)	6.55	.010	2.22 (1.21, 4.10)
Child optimism	-0.10 (0.03)	9.74	.002	0.91 (0.85, 0.96)	-0.10 (0.04)	8.38	.004	0.91 (0.85, 0.97)
Step 2								
Interaction	0.05 (0.10)	0.23	.631	1.05 (0.86, 1.29)	-0.11 (0.12)	0.82	.366	0.90 (0.71, 1.13)
Step 1								
Maternal chronic pain	0.75 (0.28)	7.04	.008	2.11 (1.22, 3.66)	0.76 (0.31)	5.95	.015	2.14 (1.16, 3.94)
Child connections with adults at home	-0.13 (0.06)	5.53	.019	0.88 (0.78, 0.98)	-0.14 (0.06)	5.19	.023	0.87 (0.77, 0.98)
Step 2								
Interaction	-0.08 (0.16)	0.27	.606	0.92 (0.68, 1.26)	-0.19 (0.18)	1.07	.301	0.83 (0.58, 1.18)
Step 1								
Maternal chronic pain	0.70 (0.29)	6.02	.014	2.01 (1.51, 3.52)	0.69 (0.32)	4.68	.031	1.99 (1.07, 3.71)

Child connections with adults at school	-0.08 (0.05)	3.11	.078	0.92 (0.85, 1.01)	-0.11 (0.05)	5.01	.025	0.89 (0.81, 0.99)
Step 2								
Interaction	-0.14 (0.13)	1.14	.286	0.87 (0.67, 1.12)	-0.23 (0.15)	2.33	.127	0.80 (0.60, 1.07)
Step 1								
Maternal chronic pain	0.79 (0.28)	7.83	.005	2.20 (1.27, 3.81)	0.77 (0.31)	6.08	.014	2.15 (1.17, 3.96)
Child connections with adults in community	-0.05 (0.03)	2.73	.099	0.95 (0.89, 1.01)	-0.05 (0.04)	1.70	.192	0.96 (0.89, 1.02)
Step 2								
Interaction	-0.05 (0.09)	0.27	.601	0.95 (0.79, 1.14)	-0.04 (0.10)	0.15	.700	0.96 (0.79, 1.17)
Step 1								
Maternal chronic pain	0.73 (0.28)	6.71	.010	2.07 (1.19, 3.60)	0.75 (0.31)	5.73	.017	2.11 (1.15, 3.88)
Child peer belonging	-0.08 (0.04)	3.33	.068	0.92 (0.85, 1.01)	-0.09 (0.05)	2.99	.084	0.92 (0.84, 1.01)
Step 2								
Interaction	-0.17 (0.13)	1.72	.190	0.85 (0.66, 1.09)	-0.26 (0.15)	3.13	.077	0.77 (0.58, 1.03)
Step 1								
Maternal chronic pain	0.73 (0.29)	6.38	.012	2.07 (1.18, 3.63)	0.72 (0.32)	5.13	.023	2.06 (1.10, 3.85)
Child community engagement	-0.02 (0.09)	0.06	.812	0.98 (0.83, 1.16)	0.02 (0.10)	0.02	.879	1.02 (0.84, 1.22)

Step 2

Interaction	-0.07 (0.24)	0.09	.760	0.93 (0.59, 1.48)	-0.13 (0.25)	0.25	.615	0.88 (0.54, 1.44)
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Note. Adjusted analyses include maternal age, maternal racial/ethnic identity, maternal education, maternal marital status, annual household income, child age, and child gender as covariates in the first step of model building. Reported statistics for maternal chronic pain and the child protective/promotive factor are main effects, from the step prior to including the interaction term.

Abbreviations. CI, confidence interval; OR, odds ratio

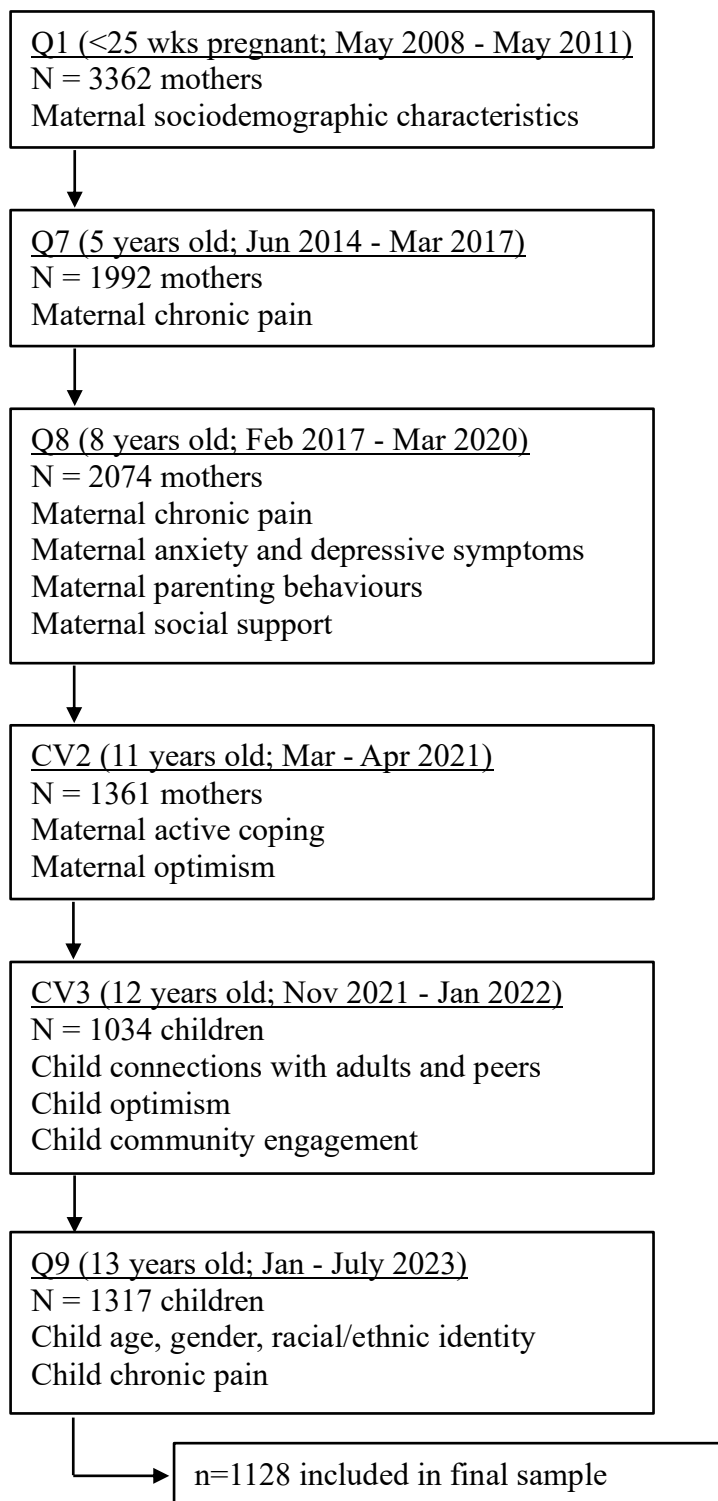
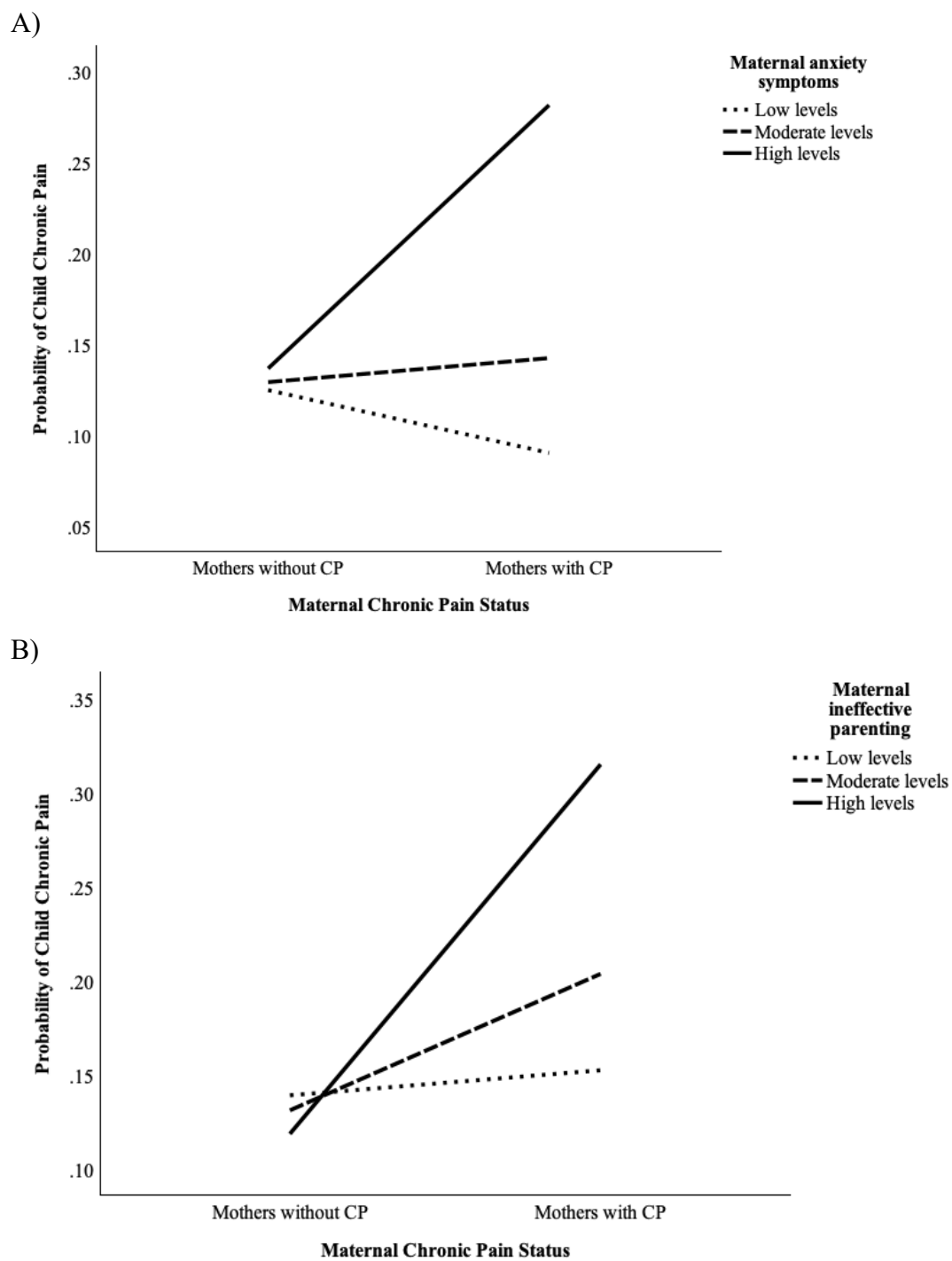
Figure 4.1*Flow Diagram of Data Collection for Current Research*

Figure 4.2

Interaction Effects of (A) Maternal Anxiety Symptoms and (B) Maternal Ineffective Parenting with Maternal Chronic Pain when Predicting Child Chronic Pain



CHAPTER FIVE: General Discussion and Conclusion

General Discussion

Parent factors have been recognized to play an important role in pediatric chronic pain for several decades (Palermo & Eccleston, 2009). While the majority of this research has focused on parent responses to child pain (e.g., protective behaviours, catastrophic thinking), recent findings have drawn increased attention to parent functioning, specifically parents' own chronic pain and mental health problems (e.g., anxiety and depressive symptoms). Indeed, accumulating evidence from both community-based and clinical research studies suggests that poor physical and mental health in parents can contribute to the development, maintenance, and severity of chronic pain in children, and interfere with children's response to psychological interventions for their chronic pain (Brown et al., 2022; Donnelly et al., 2020; K. S. Higgins et al., 2015; Law et al., 2017; Poppert Cordts et al., 2019). However, many gaps exist in our understanding of the associations between parent functioning and child chronic pain including mixed findings on the role of parent mental health and its interaction with their physical health and limited focus on general (versus pain-specific) factors as well as resilience (versus risk) factors that may moderate the association between parent functioning and child chronic pain. The broad aim of this dissertation was to address these gaps using a multi-method approach that considered both risk and resilience factors at various levels of the child's ecology in clinical and community samples.

Summary of Findings

The first study (Chapter 2) systematically reviewed and synthesized the existing literature examining the association between parent mental health and children's chronic pain and related functioning to generate estimates of the general magnitude of these associations. Meta-analytic findings from the 49 included studies demonstrated that mental health problems were common among parents of children with chronic pain, with pooled prevalence estimates of 28.8% for

anxiety, 20.0% for depression, and 32.4% for general distress. Poorer mental health in parents, including higher (versus lower or no) symptoms of anxiety, depression, and general distress, was significantly associated with the presence of chronic pain in children and the severity of their chronic pain-related functioning (i.e., greater pain intensity, lower physical functioning, higher anxiety and depression symptoms). Effect sizes were generally small (odds ratios for presence of chronic pain ranged from 1.74-1.91 and correlations for functioning ranged from 0.10-0.25) and there was substantial heterogeneity for many of the effect sizes. Nonetheless, effect sizes were comparable in magnitude to other meta-analyses examining the associations between parent and child chronic pain, and parent responses to child pain and children's pain-related functioning (Donnelly et al., 2020; Harrison et al., 2020; K. S. Higgins et al., 2015).

The second study (Chapter 3) investigated daily variability among parents with and without chronic pain, and its impact on their child's daily chronic pain-related functioning, using daily diary data from a clinical sample of 76 youth with chronic pain and one of their parents, of which 50% reported their own chronic pain. Multilevel modeling revealed that parents with chronic pain were significantly more variable in their parenting stress across the week, but not in their anxiety, mood, or protective responses, than parents without chronic pain. Moreover, parent daily variability was not significantly related to youth's daily chronic pain, and parent chronic pain status was not a significant predictor or moderator of the associations between parent daily variability and youth daily pain. Instead, parents' baseline depressive symptoms and mean levels of anxiety, protective responses, and parenting stress across the week significantly predicted youth's daily pain interference. These results suggested that parent distress, including general distress (anxiety and depressive symptoms), distress related to their child's pain, and distress related to parenting, may be more important for youth's daily chronic pain-related functioning

than parent chronic pain. Of note, parents with chronic pain reported higher levels of anxiety and depressive symptoms, protective responses, and parenting stress than parents without chronic pain; thus, these factors may need to be addressed among parents with chronic pain more often than parents without chronic pain.

The third study (Chapter 4) aimed to identify individual and interpersonal factors in childhood that either increased or decreased the risk for chronic pain in early adolescence among children of mothers with chronic pain as compared to children of mothers without chronic pain. This study used data from a community sample of 1128 mother-child dyads who were recruited for a larger longitudinal cohort study. In logistic regression analyses, two risk factors – maternal anxiety symptoms and ineffective parenting practices – moderated the association between maternal and child chronic pain, such that children had significantly increased risk for chronic pain in early adolescence when their mothers reported both chronic pain and greater anxiety symptoms or ineffective parenting in middle childhood (age 8 years). No protective factors were found to moderate or buffer the transmission of chronic pain from mothers to children; however, higher levels of child optimism and more child connections with adults in early adolescence decreased the risk of chronic pain for all children (regardless of maternal chronic pain status). Of note, maternal chronic pain was a significant predictor of child chronic pain across all analyses.

Research Implications

The findings from this dissertation add to a growing literature on the importance of parent functioning (i.e., their own physical and mental health) within pediatric chronic pain. First, meta-analytic findings from the first study (Chapter 2) confirmed that parent mental health is an important parent factor to consider within pediatric chronic pain given its prevalence and similar-sized associations with children's chronic pain as other parent factors (i.e., protective behaviours,

pain catastrophizing, chronic pain status) that have historically received greater attention in the pediatric pain literature than parent mental health. The second and third studies also support this conclusion with the findings that parent anxiety and depressive symptoms were significantly associated with the daily pain-related functioning of a clinical sample of youth with chronic pain (Chapter 3) as well as the presence of chronic pain in a community-based sample of children (Chapter 4). Indeed, in the second study, children's daily pain interference was significantly predicted by parent mental health but *not* parent chronic pain. However, in the third study, parent chronic pain was more strongly associated with the presence of child chronic pain than parent mental health. These mixed findings are consistent with existing literature wherein the stronger predictor of child chronic pain (parent chronic pain versus parent mental health) differs between studies, likely due to differences in study methodology and samples. While the findings from the three studies in this dissertation cannot be directly compared due to differences in the samples and study designs, the findings suggest that parent chronic pain may play a more important role in the onset or development of pediatric chronic pain while parent mental health may play a more important role in the maintenance or severity of pediatric chronic pain. Critically, findings from the third study indicated that parent chronic pain and mental health can interact to increase the risk for child chronic pain. Thus, it is likely that chronic pain and mental health interact with one another in complex ways that differ across parents and parent-child dyads depending on other individual and interpersonal factors within the child's ecology. Given the comorbidity between chronic pain and mental health problems, it may not be essential to tease out the effects of each for children's chronic pain. Rather, it may be more pertinent for future research to assess both the physical *and* mental health of parents when investigating how parent functioning impacts children's adaptation to pain, similar to Birnie et al. (2020) and Stone et al. (2019) who used the

PROMIS measure to assess various domains of parent health including their pain, physical function, anxiety and depression, fatigue, sleep, and social functioning.

This dissertation also extended the existing research on parent factors within pediatric chronic pain by exploring general (i.e., not pain-specific) parent and child factors. Previous research examining factors mediating or moderating the association between parent factors and child chronic pain has focused on pain-specific coping strategies – primarily maladaptive (e.g., protective responses to child pain, catastrophic thinking about pain) but more recently adaptive (e.g., instructions to engage in activities, pain acceptance) strategies – that parents may model or reinforce in their children that in turn impacts children’s pain adaptation (Beeckman, Simons, et al., 2019; Fussner et al., 2018; Lee et al., 2020; Sieberg et al., 2011; Stone et al., 2018). This dissertation found that general parent factors, specifically greater parenting stress and ineffective parenting practices, were significantly associated with poor chronic pain-related outcomes for children including greater daily pain interference in a clinical sample of youth with chronic pain and presence of chronic pain in a community sample of children. These general parent factors were also related and/or interacted with parent chronic pain. These findings contribute to the few studies that have examined general parent factors in the context of parent functioning and children’s chronic pain. For example, in a community-based study by Darlington et al. (2012), greater parenting stress mediated the association between maternal anxiety symptoms and the presence of child chronic pain. In a case-control study by Evans et al. (2006), maladaptive parenting practices, specifically over-reactivity, mediated the association between parent chronic pain and children’s physical health. Taken altogether, general (i.e., non-pain-specific) parent factors also appear to contribute to the association between parent functioning and child chronic pain and thus warrant further investigation. This dissertation did not directly examine

interactions between parent mental health and general parent factors, and thus further research is needed to understand how these variables may interact to contribute to children's pain-related outcomes.

The third study of this dissertation also demonstrated that several general child factors, specifically greater optimism and connections with adults, significantly *reduced* the risk of child chronic pain in a community sample. These findings are consistent with the few studies that have examined general risk and resilience factors in clinical samples of youth with chronic pain. Specifically, these studies have shown that greater child optimism and social support are associated with more positive adaptation to chronic pain including lower disability and greater quality of life (Cousins, Cohen, et al., 2015; Ross et al., 2018; Walker et al., 2008). While other studies investigating resilience within pediatric chronic pain have identified general child and parent factors, such as self-esteem and psychological flexibility, that predict more positive child pain adaptation, the majority of studies have focused on pain-specific factors including child pain self-efficacy and child and parent pain acceptance (Beeckman, Hughes, et al., 2019; Beeckman, Simons, et al., 2019; Feinstein et al., 2018; Kalapurakkal et al., 2015; Lee et al., 2020; Mikkelsen et al., 2021). The current findings extend previous research by examining general parent and child factors that decreased the risk for developing chronic pain in a community-based sample of children (versus decreased risk for poor pain-related functioning in clinical samples of youth with chronic pain referred to tertiary services). Interestingly, no parent factors were found that significantly decreased the risk for child chronic pain and no protective factors (parent or child) interacted with parent chronic pain to significantly reduce the transmission of chronic pain from mothers to children. Instead, three child factors – optimism and connections with adults at home and at school – decreased the risk of chronic pain for all children (regardless of maternal chronic

pain status). Further research on resilience *to* pediatric chronic pain in community samples is needed to identify additional factors, both general and pain-specific, that may help to prevent the development of chronic pain in children.

Theoretical Implications

The findings of this dissertation support the use of a broader developmental systems approach to conceptualizing the role of parent factors – specifically parent physical and mental health – in pediatric chronic pain. While previous research has found support for the role of social learning mechanisms (i.e., parent modeling and/or reinforcement of maladaptive or adaptive responses to pain) in the association between parent functioning and child chronic pain and related disability (Birnie et al., 2020; K. S. Higgins et al., 2019; Stone et al., 2018), this is likely just one pathway through which parent factors may impact children’s adaptation to pain. Indeed, as previously suggested by conceptual models including Palermo & Chambers’ (2005) integrative model on parent and family factors in pediatric pain, Stone & Wilson’s (2016) integrative model on the intergenerational transmission of risk for chronic pain, and Cousins et al.’s (2015) ecological model on risk and resilience within pediatric chronic pain, many other individual and interpersonal factors and processes across the child’s ecology likely contribute to the association between parent functioning and child chronic pain.

The current findings suggest that poor functioning in parents (i.e., their own pain and related disability, greater anxiety and depressive symptoms) may increase children’s risk for poor pain-related outcomes through a more stressful home environment. The second and third studies found that children had an increased risk for poor chronic pain-related outcomes when their parents reported more parenting stress, such as feeling frustrated, unhappy, tense, or worried in response to parenting, and more ineffective parenting practices, which encompasses inconsistent

discipline and hostile parenting responses. While these parenting responses may model poor coping strategies to the child, and thus impact children through social learning mechanisms, these parenting responses likely also contribute to a more stressful home environment. Exposure to a stressful environment at home can impact children's physical and mental health through physiological (e.g., disruptions in the stress response system), psychological (e.g., lack of support, deficits in ability to effectively manage emotions), and behavioural (e.g., poor health behaviours) mechanisms that interact with genetic vulnerabilities (Repetti et al., 2002). Across the second and third studies of this dissertation, parents with chronic pain reported greater anxiety and depressive symptoms as well as lower social support, optimism, and active coping strategies. Parent chronic pain status also related or interacted with parenting stress and ineffective parenting to increase children's risk for poor pain-related outcomes. In these ways, poor functioning in parents may contribute to a stressful home environment, which may in turn interact with a child's vulnerabilities and increase their risk for poor adaptation to chronic pain.

The third study's finding that child factors, including optimism and connections with adults at home and school, reduce the risk for child chronic pain also lend support to the stressful home environment pathway, as these factors have been suggested in related literatures to buffer the negative impact of stress on physical and emotional health through both biological (e.g., greater immune health) and psychological (e.g., use of adaptive coping strategies, sense of belonging) pathways (Holt-Lunstad, 2018; Rasmussen et al., 2009). These findings are also consistent with the broader resilience literature, which has found a number of individual and interpersonal factors, including optimism and connections with capable adults, are associated with children's capacity to successfully adapt to a wide variety of adverse or stressful situations (e.g., maltreatment, poverty, war; Masten, 2015). These factors are posited to promote resilience

through several adaptive systems, including the attachment system, central nervous system, and mastery motivation and related reward systems, which evolved through biological and cultural means to promote human survival and positive development (Masten, 2015). Thus, similar to other potentially adverse or stressful childhood experiences, children whose parents have poor physical and/or mental health may experience increased stress, which could have negative effects on their development, but which can also be buffered by protective factors to foster resilience.

Taken altogether, findings from this dissertation suggest that a broader developmental systems approach that includes multiple pathways at various system levels is needed when conceptualizing the role of parent functioning within pediatric chronic pain. This dissertation did not directly examine specific mechanisms of the transmission (or buffering) of risk. Thus, further research is needed to better understand the ‘stressful home environment’ pathway.

Clinical Implications

Consistent with the focus on parent responses to child pain in the empirical literature, parent interventions for the management of pediatric chronic pain have focused on reducing parent responses that may reinforce maladaptive child pain behaviours, in particular protective responses. Two randomized controlled trials (RCTs) examining the efficacy of family-based cognitive-behavioural therapies for the management of pediatric chronic pain found that the interventions significantly decreased parents’ protective responses at follow-up (Palermo et al., 2016). However, a secondary analysis of the data from one of these RCTs revealed that observed decreases in parent protective responses were not related to decreases in children’s self-reported disability (Law et al., 2017). These results suggest that reducing parent protective responses may not be the most effective treatment target for improving children’s pain and related disability.

The current findings suggest that parent chronic pain and mental health may be important treatment targets. Several recent studies have investigated psychological interventions that more directly address parent distress in the context of pediatric chronic pain including problem-solving skills therapy (PSST) and acceptance-and-commitment therapy (ACT). PSST aimed to reduce parent distress by developing a positive problem-solving orientation (greater optimism and self-efficacy) and teaching rational problem-solving strategies (Palermo et al., 2016) to help parents feel less stress related to their child's chronic pain. The main aim of ACT has been to increase psychological flexibility among parents so they are better able to remain focused on the present, accept distress associated with their child's pain, and align their behaviour with specific values (Benjamin et al., 2020). Both of these interventions have demonstrated effectiveness in reducing parent distress and improving children's pain-related outcomes, even when the child was not directly involved in the therapeutic process. Another recent pilot study investigated the feasibility and acceptability of an ACT-based intervention for parents who have chronic pain themselves as well as a child with chronic pain (Pavlova et al., 2024). This intervention focused on parents' own chronic pain, mental health, and parenting; however, parent feedback at the end of the trial indicated that parents would have liked more content on parenting and the intergenerational transmission of chronic pain.

These previous findings suggest that parent interventions for pediatric chronic pain should acknowledge and address parent functioning alongside parent responses to child pain. Many of the strategies described above, from PSST and ACT, can be used by the parent to help manage their child's pain as well as their own mental or physical health difficulties. In this way, parent interventions that target their own functioning do not have to be solely focused on the parent's health, which is likely not feasible within the pediatric health care system, but could

focus on strategies that parents could use for themselves as well as their child. Empirical research has shown that parent distress can interfere with children's response to psychological treatments, such that children had less improvement in disability at 12 months post-treatment when their parent reported greater distress at pre-treatment (Law et al., 2017). The authors of this study suggested that parents may have been less able to engage with the treatment, which focused on modifying their responses to their child's pain, when they had high levels of distress. In this way, interventions that do *not* acknowledge and address parent functioning, and continue to only focus on modifying parent responses to child pain, may be less effective overall.

Findings from this dissertation suggest that parent functioning may also be an important target for efforts to *prevent* the onset of pediatric chronic pain. In the first study, poor mental health in parents was significantly related to the presence of child chronic pain and, in the third study, maternal chronic pain in childhood significantly predicted the presence of child chronic pain in early adolescence. Thus, interventions that aim to improve the mental and physical health of parents in the general population may have downstream positive effects on the mental and physical health of children. These interventions could occur through public health campaigns and/or primary or community-based care. For example, as suggested by efforts to reduce the effects of childhood adversity, primary care practitioners could screen for poor mental and physical health in parents, provide parents with education on self-care and parenting, help parents access community-based services, and offer group-based supports to parents and children (Traub & Boynton-Jarrett, 2017). While further research is needed to evaluate the effectiveness of specific interventions targeting parent distress in the prevention and treatment of pediatric chronic pain, findings from this dissertation, as well as previous research, are clear that the

mental and physical health of parents – not just their parenting responses – needs to be acknowledged and addressed to improve the well-being of families.

Strengths and Limitations

This dissertation had several notable strengths including the inclusion of both clinical and community samples, the use of a diverse set of methodologies to examine both micro (daily diary data) and macro (meta-analysis of a large literature; nine-year longitudinal cohort) processes, and the investigation of pain-specific and general factors at various levels of the child's ecology.

There were also several limitations, predominately due to the fact that the second and third studies used data from pre-existing studies and thus the methods were not tailored to the specific aims of the dissertation studies. First, in both studies, only one parent was included and thus interactions with the second parent, most often the father, could not be investigated. This limitation is consistent with the broader pediatric psychology literature, wherein fathers are often not included (Phares et al., 2005). While maternal factors, including their chronic pain and mental health symptoms, have been shown to be more strongly related to children's chronic pain than paternal factors (Evans & Keenan, 2007; Hoftun et al., 2013; Ramchandani et al., 2006), the inclusion of the second parent is critical for a more comprehensive understanding of how parent functioning – in both parents – may interact and contribute to children's adaptation to pain. The samples were also mostly white and higher socioeconomic status. While representative of the populations in the settings of the studies (i.e., families treated in tertiary pain clinics in North America and the general population of Calgary, Alberta, Canada), the studies were not inclusive to other sociodemographic groups and thus the findings are likely not generalizable, particularly to groups that may have less access to resources and more exposure to stressors, such as racism and poverty, which could impact child and parent adaptation to chronic pain. Pediatric chronic

pain research tends to recruit from tertiary pain clinics and enroll the subset of the population that has the social capital to access tertiary care (i.e., white, educated, middle to upper class). As such, knowledge of the pain experiences of racialized and minoritized groups is currently limited. Future research that adopts anti-racism practices to increase inclusivity is needed to extend current knowledge beyond white, middle-class families (Letzen et al., 2022).

The examination of parent mental health in this dissertation was largely limited to anxiety and depressive symptoms. While the first study planned to include post-traumatic stress disorder, there were too few distinct studies in the existing literature for a meta-analysis. A handful of studies within pediatric chronic pain have examined mental health disorders other than anxiety and depression in parents, including post-traumatic stress disorder and substance use disorders, with mixed findings on their association with children's chronic pain (Beveridge et al., 2018; Campo et al., 2007; Ibeziako et al., 2021; Noel et al., 2016). Further research on these and other mental health disorders that may also contribute to a more stressful home environment (e.g., psychotic disorders) and impact children's pain adaptation is needed.

The examination of parent chronic pain was also limited as it was measured with single items that assessed the presence but not impact of chronic pain. As such, there was likely variability among parents with chronic pain that was not captured. A recent study that examined the pain characteristics of 400 mothers with chronic pain found differences in their pain severity, with relatively even distributions across four *a priori* classifications. Specifically, about 25% of the mothers were grade I (low intensity, low disability), 28% were grade II (high intensity, low disability), 21% were grade III (high disability, moderately limiting), and 26% were grade IV (high disability, severely limiting; Wilson et al., 2020). Due to this variety of presentations, there have been recent calls for research to consider not only the duration (acute versus chronic), but

also the impact (low versus high), of pain (Eccleston et al., 2023). However, as noted, recent findings suggest other dimensions of parent functioning should also be included when assessing the impact of parent chronic pain on child chronic pain. A study by Stone et al. (2019) examined the same data as the above-mentioned study by Wilson et al. (2020) but used a different approach (i.e., latent profile analysis) to identify profiles of mothers with chronic pain based on their patterns of pain and related physical, emotional, and role functioning. Interestingly, Wilson et al.'s (2020) *a priori* maternal chronic pain groups, which were based just on pain severity and disability, were not significantly related to child functioning whereas Stone et al.'s (2019) statistically-derived maternal chronic pain groups were related to child functioning, such that children reported more mental health symptoms when their mothers had more severe pain, related disability, and poorer psychosocial functioning. These findings suggest that parent chronic pain and mental health symptoms should both be included in future studies aiming to investigate the role of parent functioning in pediatric chronic pain.

This dissertation focused on individual and interpersonal factors in children and parents and was thus limited in its investigation of how broader social and cultural factors across the child's ecology may interact with parent functioning and contribute to children's adaptation to chronic pain. The importance of the broader social context is being increasingly recognized in the chronic pain literature, with recent calls for research to examine the social determinants and consequences of pain across multiple levels (e.g., interpersonal, group, societal; Kapos et al., 2024). While several measures in the third study aimed to investigate the child's social context, including connections with adults and peers outside of the home and engagement in community activities, other group and societal factors were not examined such as affiliation with a cultural or religious group, access to health resources, and neighbourhood characteristics such as safety

and support (Cousins, Kalapurakkel, et al., 2015). Future research examining risk and resilience factors in the intergenerational transmission of chronic pain should include these broader social factors. Due to the novelty of the second and third studies in this dissertation, statistical analyses were relatively simple (i.e., linear and logistic regression) and examined variables in separate analyses. As research continues to progress in this area, more advanced statistical analyses that incorporate multiple variables into models are needed to add nuance to the current findings. For example, structural equation modeling would allow for an examination of the interrelationships between parent functioning variables (i.e., physical and mental health), mediating or moderating variables (i.e., risk and protective factors), and children's adaptation to chronic pain. These models could examine developmental cascades of the transmission of risk (or resilience) from poor parent functioning to child chronic pain or the interactions between ecological systems that contribute to children's adaptation to pain.

Conclusion

Parents have been shown to play a critically important role in the onset, maintenance, severity, and treatment of children's chronic pain. This dissertation aimed to extend the existing literature by examining lesser studied factors including parents' own functioning, specifically their physical (i.e., chronic pain) and mental (i.e., anxiety and depressive symptoms) health, general parent and child factors (i.e., not specific to pain such as general parenting stress), and potential parent and child resilience factors (e.g., optimism, social support) using a multi-method approach. Findings of this dissertation demonstrate that parent chronic pain and mental health is significantly related to the presence and severity of chronic pain in clinical and community samples of children and across micro (i.e., weekly) and macro (i.e., across years) levels. Moreover, several general factors were found to contribute to this association, either increasing

or decreasing the risk of poor child adaptation to pain, suggesting that parent functioning may relate to children's chronic pain through general pathways such as a stressful home environment. Overall, results suggest that parent functioning should be addressed in interventions aimed at preventing or treating pediatric chronic pain. However, further research is needed to more comprehensively understand the pathways linking parent functioning and children's chronic pain and the factors that may interact to increase or decrease the risk of poor functioning in families.

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Appendices

Appendix A: Letters of Permission

Letter of Permission

I hereby provide permission to use the following co-authored work as part of Jaimie Beveridge's doctoral thesis:

Beveridge, J. K., Noel, M., Soltani, S., Neville, A., Orr, S. L., Madigan, S., & Birnie, K. A. (2024). The association between parent mental health and pediatric chronic pain: A systematic review and meta-analysis. *PAIN*, *165*(5), 997-1012. doi: 10.1097/j.pain.0000000000003125.

I am of the understanding that the purpose of this request is to include the material in Jaimie Beveridge's thesis, and that the thesis will be added to the institutional repository at the University of Calgary and the Library and Archives Canada.

University of Calgary Theses Repository – The Vault: <http://theses.ucalgary.ca/Library>

Archives Canada: <http://collectionsCanada.gc.ca/obj/s4/f2/frm-nl59-2-e.pdf>

Melanie Noel, PhD

Sabine Soltani, PhD

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Sheri Madigan, PhD

Kathryn Birnie, PhD

Letter of Permission

I hereby provide permission to use the following co-authored work as part of Jaimie Beveridge's doctoral thesis:

Beveridge, J. K., Walker, A., Orr, S. L., Wilson, A. C., Birnie, K. A., & Noel, M. (2024). Parent anxiety, depression, protective responses, and parenting stress in the context of parent and child chronic pain: A daily diary study of parent variability. *The Journal of Pain*, 25(8), 104512. doi: 10.1016/j.jpain.2024.03.008.

I am of the understanding that the purpose of this request is to include the material in Jaimie Beveridge's thesis, and that the thesis will be added to the institutional repository at the University of Calgary and the Library and Archives Canada.

University of Calgary Theses Repository – The Vault: <http://theses.ucalgary.ca/Library>

Archives Canada: <http://collectionscanada.gc.ca/obj/s4/f2/frm-nl59-2-e.pdf>

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Serena Orr, MD, MSc

Anna Wilson, PhD

Kathryn Birnie, PhD

Melanie Noel, PhD

Letter of Permission

I hereby provide permission to use the following co-authored work as part of Jaimie Beveridge's doctoral thesis:

Beveridge, J. K., Noel, M., McArthur, B.A., Madigan, S., Orr, S. L., McDonald, S., Tough, S., & Birnie, K. A. (In Preparation). Identifying risk and protective factors in the transmission of chronic pain from mothers to children: A multi-method longitudinal cohort study.

I am of the understanding that the purpose of this request is to include the material in Jaimie Beveridge's thesis, and that the thesis will be added to the institutional repository at the University of Calgary and the Library and Archives Canada.

University of Calgary Theses Repository – The Vault: <http://theses.ucalgary.ca/Library>

Archives Canada: <http://collectionsCanada.gc.ca/obj/s4/f2/frm-nl59-2-e.pdf>

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Appendix B: Supplementary Material for Chapter 2

Supplementary Table 2.1

Definitions of Included Constructs

Term	Definition of variables included in current study
<i>Related to parent mental health</i>	
General distress	Combined measures of generic or global mental health symptoms or disorders, including measures of overall health with a subscale for mental health, measures of specific mental health domains with either a total score for all symptoms endorsed or a total % of participants with a diagnosis of a psychiatric disorder, and measures of non-specific mental health domains (e.g., psychiatric disorder, psychological distress)
Clinically-elevated	At or above clinical cut-offs for moderate to severe mental health symptoms
<i>Related to child chronic pain and related functioning</i>	
Chronic primary pain	Pain persisting or recurring for > 3 months that has not been identified as being secondary to an underlying disease process
Presence of chronic pain	Included self-report of pain persisting or recurring for ≥ 3 months, physician or researcher diagnosis of chronic pain condition, or child seeking treatment for persistent or recurrent pain
Pain characteristics	Comprised various pain characteristics such as duration, intensity (using Visual Analogue Scale or Numerical Rating Scale), unpleasantness
Physical functioning	Combined measures of functional disability, functional impairment, pain-related disability, pain interference, and physical functioning
Psychological functioning	Comprised measures of anxiety, depression, posttraumatic stress disorder, and general mental health symptoms
Role functioning	Comprised various child roles such as school attendance, social functioning, psychosocial functioning, family functioning
Quality of life	Comprised measures of health-related quality of life (e.g., Pediatric Quality of Life Inventory; PedsQL)
Sleep	No studies were included that measured features of sleep
Pain-related experiences	Comprised various experiences such as pain catastrophizing, pain coping, pain-specific anxiety
Other	Comprised measures of development, somatic symptoms, anxiety sensitivity, and grouping of participants based on pain characteristics and functioning
<i>Related to moderator variables</i>	
Clinical study	Sample seeking treatment for the child's pain in a primary or tertiary care setting; a control group may or may not have been recruited
Community study	Sample not seeking treatment for the child's pain; recruited from settings in the community including schools and medical registries or as part of a population-based study that recruited representative samples of participants
RAP	Recurrent abdominal pain, typically defined by having at least 3-4 episodes of abdominal pain, occurring over ≥ 3 months, that cannot be fully explained by another medical condition
Headache	Included headache and migraine
Fibromyalgia	Juvenile primary fibromyalgia syndrome as defined by Yunus & Masi criteria
Mixed chronic pain conditions	Included clinical studies with a sample of children and adolescents with a variety of chronic pain conditions (e.g., headache, RAP, and musculoskeletal pain) and community studies that asked about the presence of any chronic or recurring pain
Self-report measure	Measure completed by respondent about their own mental health
Interview measure	Structured or semi-structured measure administered by interviewer
Current symptoms	Included measures that assessed current mental health symptoms
History of symptoms	Included measures that asked about current symptoms at a previous timepoint in prospective studies and measures that asked about the parent's history of mental health in retrospective studies

Supplementary Table 2.2

Search Strategy

<i>Databases: CINAHL, Embase Classic+ Embase, Medline, PsycINFO, Web of Science</i>	
#	Searches
1	exp child/ or adolescent/ or exp pediatrics/ or child, abandoned/ or exp child, exceptional/
2	(offspring* or pediatric* or paediatric* or child* or preschool* or pre-school* or kindergarten* or kindergarden* or elementary school* or nursery school* or (day care* not adult*) or schoolchild* or toddler* or boy or boys or girl* or middle school* or pubescen* or juvenile* or teen* or youth* or high school* or adolesc* or pre-pubesc* or prepubesc* or yound adult* or young person* or young people*).mp. or (child* or adolesc* or pediat* or paediat*).jn.
3	1 or 2
4	exp parents/ or caregivers/ or legal guardians/
5	(parent* or mother* or maternal* or father* or paternal* or family or families or stepfamil* or caregiv* or care-giver* or guardian* or carer or carers or care giver*).tw,kf.
6	4 or 5
7	mental health/
8	"mental health".tw,kf.
9	mental disorders/
10	(behaviour disorder* or behavior disorder* or mental illness* or mentally ill or neuropsychiatric or psychic or psychologic* or insanity or psychiatric or psychopathology).tw,kf.
11	((mental or mentally) adj1 (disorder* or diagnos* or condition* or care* or factor* or state* or status* or help* or service* or disturbance* or insufficien* or symptom* or defect* or abnormal* or confusion*).tw,kf.
12	exp anxiety disorders/ or catastrophization/
13	(anxiet* or anxious* or agoraphobi* or astheni* or asthaeni* or psychastheni* or psychasthaeni* or effort syndrome* or hyperkinetic heart syndrome* or neurocirculatory dystonia* or neurogenic heart or cardioneuros* or cardiophobia or neurosis or neuroses or neurotic or neuraxia or neurasthaeni* or neurastheni* or psychoneurosis or psychoneuroses or psychoneurotic or obsessive compulsive disorder* or obsessive-compulsive disorder* or anankastic personalit* or panic or phobia or phobias or phobic or acrophobia or claustrophobi* or catastrophisation or catastrophising* or neophobi* or koro or phonophobia or sonophobia or cothymia or depress* or hysteria or hystery or parapathia or neuroticism or mental fatigue or compulsi* or obsession* or obsessive* or ocd or recur* thought* or hoarding or distress* or avoidance or grief or horror or death* or nightmare* or night mare* or emotion*).tw,kf.
14	exp mood disorders/ or depression/
15	(depress or dysphori* or dysthymi* or melanchol* or affective or cyclothymic or blunted affect* or flat affect* or schizoaffective or pseudodementia).tw,kf.
16	(mood adj1 (disorder* or change* or disturbance* or fluctuation*).tw,kf.
17	exp "bipolar and related disorders"/
18	(bipolar or mania or manias or manic or hypomanic or hypomania*).tw,kf.
19	psychophysiologic disorders/
20	(psycho organic syndrome* or psychoorganic syndrome* or psychophysiologic disorder* or psychophysiological disorder* or psychosoautonomic syndrome* or psychosomat*).tw,kf.
21	"trauma and stressor related disorders"/
22	(adjustment disorder* or anniversary reaction* or reactive disorder* or transient situational disturbance*).tw,kf.
23	(stress or stressed or stressing or stressor* or trauma* or psychotrauma* or ptsd or posttrauma* or combat or post-trauma*).tw,kf.
24	or/7-23
25	exp Pain/
26	(pain or pains).tw,kf.
27	("abnormal feeling of chest" or allodynia or allodynias or alvealgia or alveolalgias or aphagia or aphasias or arthralgia or arthralgias or back ache or back aches or backache or backaches or backpain or backpains or breast tenderness or burning sensation or burning sensations or cephalalgia or cephalalgias or cephalgia or cephalgias or cephalodynia or cephalodynias or cervicalgia or cervicalgias or cervicodynia or cervicodynias or cheiragra or chest discomfort or chiragra or chiragras or colic or cruralgia or cruralgias or cystalgia or cystalgias or dorsalgia or dorsalgias or dry socket or dry sockets or dysmenorrhoeal or dysmenorrhoeas or dysmenorrhoea or dysmenorrhoeas or dyspareunia or dyspareunias or dysuria or dysurias or earache or earaches or ear ache or ear aches or headache or headaches or head ache or head aches or hemicranias or hyperalgesia or hyperalgesias or hyperalgia or hyperalgias or hyperpathia or hyperpathias or hypoalgesia or hypoalgesias or lumbago or lymphadenopath* or mastalgia or mastalgias or mastodynia or mastodynias or menstrual cramp or menstrual cramps or metatarsalgia or metatarsalgias or muscle soreness or myalgia or myalgias or myodynia or myodynias or neckache or neckaches or neck ache or neck aches or neuralgia or neuralgias or neuralgy or

	neurodynia or neurodynias or odontalgia or odontalgias or odyndophagia or odyndophagias or orchialgia or orchialgias or otalgia or otalgias or painful breathing or painful defecation or painful defecations or painful erection or painful erections or painful micturition or painful micturitions or painful respiration or painful respirations or painful scrotum or painful testis or paroxysmal nerve pain or paroxysmal nerve pains or pelipathia vegetativa or pelvic syndrome or pelvis syndromes or pharyngalgia or pharyngalgias or piriformis muscle syndrome or piriformis syndrome or piriformis syndromes or polyarthralgia or polyarthralgias or precordialgias or prostatalgia or prostatalgias or prostatodynia or prostatodynias or psychalgia or psychalgias or rachialgia or rachialgias or sciatica or sciaticas or slit ventricle syndrome or slit ventricle syndromes or sore throat or sore throats or stomatodynia or stomatodynias or stomatopyrosis or stranguria or strangurias or strangury or thoracic discomfort or thorax discomfort or throat ache or tooth ache or throat aches or tooth ache or tooth aches or toothache or toothaches or vulvodinia or vulvodynias).tw,kf.
28	or/25-27
29	(chronic* or recur or recurs or recurr* or persist* or continue* or continual).tw,kf.
30	28 and 29
31	chronic pain/
32	30 or 31
33	3 and 6 and 24 and 32
34	limit 33 to dt=20190620-20210826
34	limit 33 to dt=20210819-20221117

Note. Example of search conducted in Medline. Lines 34 display criteria added to update searches on August 26, 2021 and November 16, 2022, respectively. This search strategy was developed in collaboration with a librarian who has experience in systematic reviews, using a comprehensive list of search terms that was created by reviewing terms used in published systematic reviews and empirical studies and consulting with collaborators with expertise in pediatric chronic pain and adult mental health.

Supplementary Table 2.3

Studies that Met Inclusion Criteria but were not Included in Meta-analyses

Study	Study characteristics				Child characteristics					Parent characteristics			
	Country	Setting	Data type ^a	N ^b	Age, M	% female	% White or nationality of study country	Chronic pain condition	Pain variables measured	Age, M	Included parent, % mothers	Mental health variables measured	Timing, measure type
Clinical samples with cohort/cross-sectional design													
Benjamin, 2019 ^c	USA	Outpatient pain day program	C	160	15.81	62	94	Mixed	Phys ^f , Psyc ^f	49.26	Either, 50	DEP, DIS	Cur, SR
Beveridge, 2018 ^c	Canada	Outpatient pain clinics	C	204	13.43	67	80	Mixed	Phys ^f , Psyc ^f , QoL ^f	NR	Either, 91	PTSD	Cur, SR
Beveridge, 2021 ^c	Canada	Outpatient pain clinics	C	170	14.32	71.2	80	Mixed	Prev	45.03	Either, 90.6	PTSD	Cur, SR
Connelly, 2012 ^c	USA	Outpatient pain clinic	C	87	13.5	75	85	Mixed	QoL ^f	42.0	Either, 85	DIS	Cur, SR
Helgeland, 2011 ^c	Norway	Outpatient pediatric clinics	L	94	11.1	62	95	RAP	Phys ^f	NR	Only mothers	DIS	His, SR
Kalomiris, 2022 ^d	USA	Outpatient GI clinics	C	79	11.71	59.49	89.9	RAP	Char ^f , Phys ^f , Psyc ^f	NR	Either, 87.3	ANX	Cur, SR
Kaminsky, 2006 ^c	Canada	Outpatient GI and community clinics	C	50	11.00	82	96	RAP	Psyc ^f	NR	Only mothers	DIS	Cur, SR
Khu, 2019 ^d	Canada	Outpatient pain clinics	C	135	13.1	64.4	91.1	Mixed	Char ^f , Phys ^f	NR	Either, 89.6	ANX, DEP, PTSD	Cur, SR
Neville, 2018 ^d	Canada	Outpatient pain clinics	C	102	13.5	72.5	78.2	Mixed	Char ^f , Phys ^f , Exp ^f , Psyc ^f	NR	Either, 93.1	PTSD	Cur, SR
Robins, 2005 ^c	USA	Outpatient GI and primary care clinics	C	62	11.2	57	79	RAP	Presence ^h	NR	Either, 96.8	DIS	Cur, SR
Stone, 2020 ^c	USA	Outpatient GI clinic	C	278	14.62	66.2	86	RAP	Other ^f	NR	Either, 95.3	ANX, DEP	Cur, SR
Clinical samples with case-control design													
Coenders, 2014 ^e	Australia	-	C	-	-	-	-	Mixed	Presence ^f	-	NR	ANX, DEP	Cur & His, SR
Cases	-	Outpatient pain clinic	-	45	14.1	66.7	NR	-	-	NR	-	-	-
Controls	-	Hospital service program and secondary schools	-	56	15.5	62.5	NR	-	-	NR	-	-	-
Noel, 2016 ^c	USA	-	C	-	-	-	-	Mixed	Presence ^h	NR	Either	PTSD	Cur, SR
Cases	-	Outpatient pain clinics	-	95	15.0	72.2	88.2	-	Char ^f , Phys ^f	-	90.7	-	-

									Psyc ^f , QoL ^f				
Controls	-	Community ads, participant research database	-	100	14.3	55.4	75.0	-	-	-	93.1	-	-
Ramchandani, 2011 ^c	UK	-	C	-	-	-	-	RAP	Presence ^h	-	Only mothers	ANX, DEP, DIS	Cur, SR
Cases	-	Primary care clinics	-	30	7.62	68.7	NR	-	-	NR	-	-	-
Controls	-	Registry of primary care clinics	-	32	7.75	71.9	NR	-	-	NR	-	-	-
Walker, 1993 ^c	USA	-	C	-	-	-	-	RAP	Presence ^h	-	Both	DIS	Cur, SR
Cases	-	Outpatient pediatric clinics	-	88	10.19	61.4	92	-	-	NR	-	-	-
Controls	-	Same clinics or emergency room	-	56	11.45	50.0	93	-	-	NR	-	-	-
Community samples with cohort/cross-sectional design													
Fryer, 2017 ^c	UK	Representative sample of UK children	L	8463	NR	49.4	92.1	Mixed	Presence ^g	NR	99.2	DIS	His, SR
Perquin, 2003 ^e	Netherlan ds	Representative community sample and primary care registry	L	109	10.0	62.5	97.3	Mixed	Presence ^{f/g}	NR	NR	DIS	His, SR
Ramchandani, 2005 ^d	UK	Community, and routine prenatal visits	L	9312	NR	47.1	96.2	RAP	Presence ^g	NR	Both	ANX, DEP	His, SR

Abbreviations. ANX, anxiety; C, cross-sectional; Char, pain characteristics; Cur, current symptoms assessed; DEP, depression; Exp, pain-related experiences; Fibro, fibromyalgia; GI, gastroenterology or gastrointestinal; DIS, general distress; His, history of symptoms/diagnosis assessed; IN, diagnostic interview; L, longitudinal; NR, not reported; Phys, physical functioning; Presence, presence of chronic pain; Prev, examined prevalence of parent mental health problems; Psyc, psychological functioning; PTSD, posttraumatic stress disorder; QoL, quality of life; RAP, recurrent abdominal pain; Role, role functioning; SR, self-report measure

Note. ^a Based on data extracted for meta-analysis (e.g., only cross-sectional data extracted from longitudinal data); ^b Ns vary slightly between variables/analyses; ^c Excluded from meta-analysis because did not report data that could be combined with other studies; ^d Excluded from meta-analysis because sample overlapped with other studies and did not have any distinct data that could be combined with other studies; ^e Excluded from meta-analysis because did not report statistics in the article; ^f Child self-report; ^g Parent-report; ^h Clinician/researcher diagnosed

Supplementary Table 2.4

Quality Assessment Ratings for Observational Cohort and Cross-Sectional Studies Included in Systematic Review

Study	Research question or objective clearly stated?	Study pop. specified and defined?	Participation rate \geq 50%?	Subjects recruited from similar pop.? Uniform eligibility criteria?	Sample size justified?	Exposure measured prior to outcome?	Sufficient time frame between exposure and outcome?	Different levels of exposure as related to outcome?	Exposure measures clearly defined, valid, reliable, uniformly applied?	Exposure assessed more than once?	Outcome measures clearly defined, valid, reliable, uniformly applied?	Outcome assessors blind to exposure status?	Loss to follow-up \leq 20%?	Analyses adjusted for potential confounding variables?
Benjamin, 2019	Yes	Yes	No	Yes	Yes	No	No	Yes	Yes	No	Yes	NA	NA	Yes
Benjamin, 2020	Yes	Yes	CD	Yes	Yes	No	No	Yes	Yes	Yes	CD	NA	No	No
Beveridge, 2018	Yes	No	Yes	Yes	No	No	No	Yes	Yes	No	Yes	NA	NA	No
Beveridge, 2021	Yes	Yes	Yes	Yes	No	No	No	Yes	Yes	No	Yes	NA	Yes	No
Beveridge, 2022	Yes	Yes	Yes	Yes	No	Yes	CD	Yes	Yes	No	Yes	NA	Yes	Yes
Birmie, 2020	Yes	Yes	Yes	Yes	No	No	No	Yes	Yes	No	Yes	NA	NA	Yes
Brown, 2021	Yes	Yes	No	Yes	No	No	No	Yes	Yes	No	Yes	NA	NA	Yes
Brown, 2022	Yes	Yes	No	Yes	No	No	No	Yes	Yes	No	Yes	NA	NA	Yes
Cohen, 2010	Yes	No	CD	CD	No	No	No	Yes	Yes	No	Yes	NA	NA	No
Connelly, 2012	Yes	No	Yes	Yes	No	No	No	Yes	Yes	No	Yes	NA	NA	Yes
Darlington, 2012	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes	No	Yes	NA	Yes	Yes
Dutta, 2021	Yes	Yes	Yes	Yes	No	No	No	Yes	Yes	No	Yes	NA	NA	Yes
Eccleston, 2004	Yes	No	Yes	Yes	No	No	No	Yes	Yes	No	Yes	NA	NA	Yes
Evans, 2010	Yes	No	CD	Yes	No	No	No	Yes	Yes	No	Yes	NA	NA	No
Feldman, 2010	Yes	Yes	Yes	Yes	No	No	No	No	Yes	No	No	NA	NA	Yes
Fryer, 2017	Yes	Yes	Yes	Yes	No	Yes	Yes	No	Yes	No	No	NA	No	Yes
Galli, 2009	Yes	No	CD	Yes	No	No	No	No	CD	No	Yes	CD	NA	No
Goubert, 2006	Yes	No	Yes	CD	No	No	No	Yes	Yes	No	Yes	NA	NA	Yes
Hammond, 2019	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes	Yes	Yes	NA	No	Yes
Helgeland, 2010	Yes	Yes	Yes	Yes	No	Yes	Yes	No	Yes	Yes	Yes	NA	No	Yes
Helgeland, 2011	Yes	Yes	Yes	Yes	No	Yes	CD	Yes	Yes	No	Yes	NA	Yes	Yes
Hinze, 2023	Yes	Yes	Yes	Yes	No	No	No	No	Yes	No	No	NA	No	Yes
Ibeziako, 2021	Yes	Yes	Yes	Yes	No	No	No	No	CD	No	Yes	NA	NA	Yes
Inceland, 2016	Yes	Yes	Yes	Yes	No	Yes	Yes	No	Yes	No	No	NA	Yes	Yes
Jamison, 1992	Yes	No	CD	Yes	No	No	No	Yes	Yes	No	No	NA	NA	No
Kalomiris, 2021	Yes	Yes	Yes	Yes	Yes	No	No	Yes	Yes	Yes	Yes	NA	Yes	No
Kaminsky, 2006	Yes	No	No	Yes	No	No	No	Yes	No	No	No	NA	NA	Yes
Khu, 2019	Yes	No	Yes	Yes	No	No	No	Yes	Yes	No	Yes	NA	NA	Yes

Kolaitis, 2022	Yes	Yes	CD	Yes	No	Yes	Yes	Yes	Yes	No	No	NA	No	Yes
Law, 2019	Yes	Yes	Yes	Yes	Yes	No	No	Yes	Yes	Yes	Yes	NA	Yes	No
Logan, 2005	Yes	No	CD	Yes	No	No	No	Yes	Yes	No	Yes	NA	NA	Yes
Lommel, 2011	Yes	No	CD	Yes	No	No	No	Yes	Yes	No	Yes	NA	NA	No
Mikkelsen, 2021	Yes	Yes	No	Yes	No	No	No	Yes	Yes	No	Yes	NA	NA	No
Moore, 2020	Yes	No	Yes	Yes	No	Yes	Yes	Yes	Yes	No	No	NA	NA	Yes
Mortimer, 1992	Yes	No	Yes	Yes	No	No	No	No	Yes	No	Yes	CD	NA	No
Neville, 2018	Yes	No	Yes	Yes	Yes	No	No	Yes	Yes	No	Yes	NA	NA	Yes
Perquin, 2003	Yes	Yes	No	Yes	No	Yes	Yes	Yes	Yes	Yes	No	NA	No	No
Poppert Cordts, 2019	Yes	No	CD	Yes	No	No	No	Yes	Yes	No	Yes	NA	NA	No
Ramchandani, 2005	Yes	Yes	Yes	Yes	No	Yes	CD	No	Yes	No	No	NA	No	Yes
Ramchandani, 2006	Yes	Yes	Yes	Yes	No	Yes	Yes	No	Yes	No	No	NA	No	Yes
Ramchandani, 2007	Yes	Yes	Yes	Yes	No	Yes	Yes	No	Yes	Yes	No	NA	No	Yes
Robins, 2005	Yes	Yes	Yes	Yes	Yes	No	No	Yes	Yes	No	CD	NA	NA	No
Schneider, 2019	Yes	Yes	Yes	Yes	No	No	No	No	CD	No	CD	NA	NA	No
Sieberg, 2011	Yes	Yes	Yes	Yes	No	No	No	Yes	Yes	No	Yes	NA	NA	Yes
Soltani, 2022	Yes	Yes	Yes	Yes	No	No	No	Yes	Yes	Yes	Yes	NA	Yes	No
Stone, 2020	Yes	Yes	Yes	Yes	Yes	No	No	Yes	Yes	No	Yes	NA	NA	Yes
Tran, 2021	Yes	Yes	CD	Yes	No	No	No	Yes	Yes	No	Yes	NA	NA	No
Vetter, 2013	Yes	Yes	Yes	Yes	No	No	No	Yes	Yes	No	Yes	NA	NA	Yes
Wendland, 2010	Yes	No	CD	Yes	No	No	No	Yes	Yes	No	Yes	NA	NA	No
Wiwe Lipsker, 2016	Yes	No	CD	Yes	No	No	No	Yes	Yes	No	Yes	NA	NA	No
Total 'Yes'	50/50	32/50	32/50	48/50	7/50	13/50	10/50	38/50	46/50	8/50	35/50	0/50	8/50	30/50
Total 'No'	0/50	18/50	6/50	0/50	43/50	37/50	37/50	12/50	1/50	42/50	12/50	0/50	10/50	20/50
Total 'CD'	0/50	0/50	12/50	2/50	0/50	0/50	3/50	0/50	3/50	0/50	3/50	2/50	0/50	0/50
Total 'NA'	0/50	0/50	0/50	0/50	0/50	0/50	0/50	0/50	0/50	0/50	0/50	48/50	32/50	0/50

Abbreviations. CD, could not determine; NA, not applicable

Note. Study quality was assessed using the National Institutes of Health Quality Assessment Tool for Observational Cohort and Cross-Sectional Studies. See website for full description of criteria: <https://www.nhlbi.nih.gov/health-topics/study-quality-assessment-tools>

Supplementary Table 2.5

Quality Assessment Ratings for Case-Control Studies Included in Systematic Review

Study	Research question or objective clearly stated?	Study pop. specified and defined?	Sample size justified?	Controls recruited from similar populations as cases?	Processes to identify cases and controls valid, reliable, uniformly applied?	Cases clearly defined and differentiated from controls?	Cases and/or controls randomly selected?	Use of concurrent controls?	Exposure occurred prior to condition or event that defined case?	Measures of exposure defined, valid, reliable, uniformly applied?	Exposure assessors blinded to case or control status?	Analyses adjusted for potential confounding variables?
Buonavolonta, 2010	Yes	Yes	No	Yes	Yes	Yes	No	CD	No	Yes	NA	No
Campo, 2007	Yes	No	No	Yes	Yes	Yes	No	CD	No	Yes	Yes	Yes
Coenders, 2014	Yes	Yes	Yes	Yes	Yes	Yes	CD	CD	No	Yes	NA	CD
Czyzewski, 2007	Yes	No	No	Yes	Yes	Yes	NA	CD	No	Yes	NA	No
Garber, 1990	Yes	No	No	Yes	Yes	Yes	CD	CD	No	Yes	NA	No
Hodges 1985a	Yes	No	No	CD	Yes	Yes	No	CD	No	Yes	NA	Yes
Hodges, 1985b	Yes	No	No	CD	Yes	Yes	No	CD	No	Yes	NA	Yes
Kashikar-Zuck, 2008	Yes	No	Yes	Yes	Yes	Yes	No	CD	No	Yes	NA	Yes
Kaufman, 1997	Yes	No	No	Yes	Yes	Yes	CD	CD	No	Yes	NA	Yes
Liakopoulou-Kairis, 2002	Yes	No	No	CD	Yes	Yes	No	CD	No	Yes	NA	No
Moayedi, 2015	Yes	No	No	CD	Yes	Yes	CD	CD	No	Yes	NA	No
Noel, 2016	Yes	No	No	Yes	Yes	Yes	CD	CD	No	Yes	NA	Yes
Ramchandani, 2011	Yes	No	No	Yes	Yes	Yes	No	CD	No	Yes	NA	No
Reid, 1997	Yes	No	Yes	Yes	Yes	Yes	CD	CD	No	Yes	NA	No
Robinson, 1990	Yes	No	No	Yes	Yes	Yes	No	CD	Yes	CD	NA	No
Walker, 1989	Yes	No	No	Yes	Yes	Yes	No	CD	No	Yes	NA	Yes
Walker, 1993	Yes	No	No	Yes	Yes	Yes	No	CD	No	Yes	NA	Yes
Total 'Yes'	17/17	2/17	3/17	13/17	17/17	17/17	0/17	0/17	1/17	16/17	1/17	8/17
Total 'No'	0/17	15/17	14/17	0/17	0/17	0/17	10/17	0/17	16/17	0/17	0/17	8/17
Total 'CD'	0/17	0/17	0/17	4/17	0/17	0/17	6/17	17/17	0/17	1/17	0/17	1/17
Total 'NA'	0/17	0/17	0/17	0/17	0/17	0/17	1/17	0/17	0/17	0/17	16/17	0/17

Abbreviations. CD, could not determine; NA, not applicable

Note. Study quality was assessed using the National Institutes of Health Quality Assessment Tool for Case-Control Studies. See website for full description of criteria: <https://www.nhlbi.nih.gov/health-topics/study-quality-assessment-tools>

Supplementary Table 2.6

Results of Analyses Examining Moderators of the Prevalence of Mental Health Problems Among Parents of Children with Chronic Pain

<i>Parent Anxiety</i>					
Categorical moderators	k	Prevalence	95% CI	<i>Q</i>	<i>p</i>-value
Type of child chronic pain				.06	.811
Mixed	7	28.9	17.3, 44.1		
RAP	4	31.8	16.1, 53.1		
Timing of parent MH				.10	.753
Current	9	27.6	17.2, 41.1		
History	3	31.4	14.5, 55.4		
Continuous moderators	k	<i>B</i>	95% CI	<i>z</i>-value	<i>p</i>-value
Child age (mean)	12	0.02	-0.17, 0.21	0.22	.822
Child sex/gender (% female)	12	-0.03	-0.08, 0.02	-1.14	.253
Child race/ethnicity (% White)	7	0.03	-0.03, 0.08	1.05	.295
<i>Parent Depression</i>					
Categorical moderators	k	Prevalence	95% CI	<i>Q</i>	<i>p</i>-value
Type of sample				0.33	.564
Clinical	14	20.9	15.5, 27.7		
Community	5	17.8	10.8, 27.9		
Type of child chronic pain				0.25	.884
Mixed	8	21.7	14.9, 30.4		
RAP	5	18.9	10.8, 30.9		
Headache	5	19.2	11.6, 30.1		
Timing of parent MH				0.00	.983
Current	13	19.9	14.5, 26.8		
History	6	20.0	12.8, 29.9		
Continuous moderators	k	<i>B</i>	95% CI	<i>z</i>-value	<i>p</i>-value
Child age (mean)	18	0.09	-0.03, 0.22	1.48	.138
Child sex/gender (% female)	18	0.01	-0.02, 0.04	0.71	.480
Child race/ethnicity (% White)	10	0.03	-0.01, 0.07	1.29	.196
<i>Parent General Distress</i>					
Continuous moderators	k	<i>B</i>	95% CI	<i>z</i>-value	<i>p</i>-value
Child age (mean)	8	-0.35	-0.68, -0.03	-2.15	.032
Child sex/gender (% female)	8	-0.02	-0.06, 0.03	-0.78	.438
Child race/ethnicity (% White)	5	< -0.01	-0.04, 0.04	-0.01	.992

Abbreviations. CI, confidence interval; k, studies; MH, mental health; RAP, recurrent abdominal pain.

Note. Analyses included subgroup meta-analyses for categorical moderators and univariable meta-regression for continuous moderators. Categorical moderator variables were not examined when there were <10 studies in the analysis or <3 studies for a specific variable. For example, too few studies examined fibromyalgia for it to be included in analyses examining the moderator variable of ‘type of child chronic pain’ in the parent anxiety and parent depression analyses and there were too few studies overall in the parent general distress analyses to examine categorical moderators.

Supplementary Table 2.7

Summary Statistics and Sensitivity Analysis for Each Study Included in Meta-Analyses

Examining the Prevalence of Mental Health Problems Among Parents of Children with Chronic Pain

Study	Summary Statistics for Each Study			Leave-one-out Sensitivity Analysis for Each Study	
	Estimate	95% CI	Weight	Estimate	95% CI
<i>Parent Anxiety</i>					
Beveridge, 2022	38.50	31.88-45.57	9.30	27.79	18.82-38.99
Birmie, 2020	8.90	6.59-11.91	9.22	32.12	23.69-41.91
Buonavolonta, 2010	20.40	13.70-29.27	8.75	29.64	20.44-40.86
Campo, 2007	50.00	37.20-62.80	8.59	27.08	18.68-37.51
Cohen, 2010	55.90	46.16-65.20	9.03	26.59	18.62-36.45
Eccleston, 2004	62.00	50.58-72.23	8.79	26.21	18.42-35.85
Hodges, 1985a	39.00	23.49-57.11	7.77	27.97	19.29-38.70
Ibeziako, 2021	24.90	19.55-31.14	9.25	29.07	19.66-40.71
Ramchandani, 2006	23.50	20.90-26.31	9.56	29.04	18.79-41.98
Reid, 1997	3.12	0.19-35.03	2.11	29.91	21.13-40.46
Sieberg, 2011	7.00	3.92-12.20	8.25	32.04	22.93-42.76
Wiwe Lipsker, 2016	35.00	29.48-40.96	9.38	28.04	18.76-39.67
Total	28.78	20.29-39.08	-	-	-
<i>Parent Depression</i>					
Benjamin, 2020	31.30	26.03-37.10	6.29	19.39	15.02-24.67
Beveridge, 2022	16.10	11.55-22.00	5.95	20.27	15.67-25.82
Birmie, 2020	6.70	4.72-9.42	5.99	21.51	17.18-26.57
Buonavolonta, 2010	3.90	1.47-9.91	3.80	21.22	16.69-26.59
Campo, 2007	48.20	35.52-61.11	5.48	18.90	14.81-23.81
Cohen, 2010	31.40	23.15-41.02	5.84	19.43	15.05-24.72
Eccleston, 2004	40.00	29.58-51.41	5.70	19.13	14.91-24.21
Hammond, 2019a	20.20	15.73-25.55	6.18	19.96	15.33-25.57
Hammond, 2019b	6.30	2.53-14.82	3.95	20.92	16.38-26.33
Hodges, 1985b	25.00	11.88-45.18	4.11	19.83	15.40-25.15
Ibeziako, 2021	20.70	15.78-26.67	6.11	19.94	15.34-25.51
Law, 2019a	16.60	11.56-23.26	5.84	20.23	15.64-25.76
Law, 2019b	30.50	21.52-41.25	5.67	19.49	15.09-24.79
Mortimer, 1992	25.70	22.36-29.35	6.44	19.59	14.92-25.29
Ramchandani, 2006	11.40	9.53-13.59	6.41	20.89	16.53-26.04
Reid, 1997	3.12	0.19-35.03	0.96	20.35	15.93-25.63
Sieberg, 2011	21.00	15.33-28.07	5.95	19.93	15.37-25.45
Tran, 2021	27.00	9.81-55.72	3.13	19.82	15.43-25.10
Wiwe Lipsker, 2016	21.00	16.50-26.34	6.20	19.91	15.28-25.52
Total	20.03	15.67-25.23	-	-	-
<i>Parent General Distress</i>					
Birmie, 2020	26.10	22.24-30.37	13.34	33.46	21.84-47.49
Campo, 2007	57.10	43.95-69.32	11.80	29.55	20.15-41.07
Galli, 2009	58.10	51.11-64.78	13.08	29.00	21.42-37.97
Helgeland, 2010	24.40	14.05-38.92	10.83	33.51	22.90-46.10
Ibeziako, 2021	39.60	33.17-46.42	13.10	31.42	20.45-44.94

Inclendon, 2016	16.50	11.96-22.32	12.67		35.34	24.95-47.34
Schneider, 2019	19.00	13.22-26.53	12.37		34.70	23.96-47.26
Sieberg, 2011	30.00	23.35-37.61	12.83		32.80	21.68-46.25
Total	32.44	22.72-43.97	-		-	-

Abbreviation. CI, confidence interval.

Supplementary Table 2.8

Results of Analyses Examining Moderators of the Associations Between Parent Mental Health and the Presence of Child Chronic Pain

<i>Parent Anxiety</i>					
Categorical moderators	k	OR	95% CI	Q	p-value
Type of sample				1.46	.226
Clinical	10	2.21***	1.58, 3.08		
Community	4	1.65**	1.17, 2.31		
Type of child chronic pain				1.33	.249
Mixed	3	1.52	0.97, 2.39		
RAP	9	2.11***	1.53, 2.92		
Timing of parent MH				0.06	.809
Current	9	1.99***	1.38, 2.86		
History	5	1.87***	1.33, 2.62		
Continuous moderators	k	B	95% CI	z-value	p-value
Child age (mean)	12	0.04	-0.04, 0.12	1.06	.289
Child sex/gender (% female)	13	0.005	-0.02, 0.03	0.44	.660
Child race/ethnicity (% White)	8	-0.01	-0.04, 0.01	-0.95	.342
<i>Parent Depression</i>					
Categorical moderators	k	OR	95% CI	Q	p-value
Type of sample				21.53	< .001
Clinical	10	3.29***	2.40, 4.51		
Community	8	1.41***	1.18, 1.67		
Type of child chronic pain				8.33	.016
Mixed	4	1.50	0.96, 2.33		
RAP	9	2.80***	1.93, 4.04		
Headache	3	1.26	0.79, 2.01		
Timing of parent MH				10.77	.001
Current	8	3.28***	2.19, 4.92		
History	10	1.53***	1.24, 1.89		
Continuous moderators	k	B	95% CI	z-value	p-value
Child age (mean)	15	< -0.01	-0.10, 0.09	-0.11	.916
Child sex/gender (% female)	16	0.01	-0.01, 0.04	1.02	.308
Child race/ethnicity (% White)	7	-0.01	-0.04, 0.02	-0.75	.453
<i>Parent General Distress</i>					
Categorical moderators	k	OR	95% CI	Q	p-value
Type of sample				0.01	.922
Clinical	3	1.70*	1.08, 2.68		
Community	8	1.74***	1.44, 2.11		
Type of child chronic pain				3.83	.050
Mixed	6	1.56***	1.24, 1.96		
RAP	3	2.62***	1.64, 4.19		
Timing of parent MH				1.34	.248
Current	7	1.60***	1.26, 2.03		
History	4	2.01***	1.48, 2.74		
Continuous moderators	k	B	95% CI	z-value	p-value
Child age (mean)	10	0.01	-0.10, 0.13	0.21	.831

Child sex/gender (% female)	11	0.005	-0.01, 0.02	0.46	.644
Child race/ethnicity (% White)	6	0.002	-0.01, 0.01	0.52	.601

Abbreviations. CI, confidence interval; k, studies; MH, mental health; RAP, recurrent abdominal pain.

Note. Analyses included subgroup meta-analyses for categorical moderators and univariable meta-regression for continuous moderators. Categorical moderator variables were not examined when there were <10 studies in the analysis or <3 studies for a specific variable (e.g., too few studies examined fibromyalgia for it to be included in analyses examining the moderator variable of 'type of child chronic pain'). *** $p < .001$, ** $p < .01$, * $p < .05$

Supplementary Table 2.9

Summary Statistics and Sensitivity Analysis for Each Study Included in Meta-Analyses

Examining the Associations Between Parent Mental Health and the Presence of Child Chronic

Pain

Study	Summary Statistics for Each Study			Leave-one-out Sensitivity Analysis for Each Study	
	Estimate	95% CI	Weight	Estimate	95% CI
<i>Parent Anxiety</i>					
Buonavolonta, 2010	1.26	0.56-2.82	6.00	1.97	1.54-2.52
Campo, 2007	4.78	2.16-10.59	6.12	1.78	1.44-2.20
Czyzewski, 2007	2.12	1.05-4.28	7.22	1.90	1.48-2.45
Darlington, 2012	1.71	1.14-2.56	12.78	1.96	1.49-2.57
Garber, 1990	6.38	1.51-26.96	2.35	1.85	1.47-2.32
Helgeland, 2010	1.90	0.91-3.96	6.84	1.92	1.49-2.47
Kashikar-Zuck, 2008	2.86	1.35-6.06	6.60	1.86	1.45-2.37
Kaufman, 1997	0.52	0.17-1.56	3.72	1.97	1.59-2.45
Kolaitis, 2022	1.31	0.81-2.12	10.99	2.00	1.56-2.58
Liakopoulou-Kairis, 2002	1.56	0.83-2.93	8.29	1.95	1.51-2.52
Ramchandani, 2006	1.74	1.48-2.05	18.74	1.98	1.47-2.67
Reid, 1997	0.31	0.01-8.24	0.50	1.93	1.52-2.44
Tran, 2021	5.24	1.75-15.67	3.75	1.83	1.46-2.29
Walker, 1989	2.48	1.12-5.50	6.10	1.88	1.47-2.41
Total	1.91	1.51-2.41	-	-	-
<i>Parent Depression</i>					
Buonavolonta, 2010	2.66	0.29-24.91	0.97	1.89	1.50-2.39
Campo, 2007	4.89	2.17-10.98	4.81	1.78	1.43-2.22
Darlington, 2012	1.20	0.82-1.76	8.98	1.99	1.56-2.55
Garber, 1990	4.54	1.10-18.69	2.16	1.86	1.47-2.34
Hammond, 2019a	1.32	0.89-1.96	8.86	1.98	1.54-2.54
Hammond, 2019b	0.85	0.44-1.65	6.02	1.99	1.57-2.50
Helgeland, 2010	1.90	0.99-3.65	6.08	1.91	1.50-2.43
Hodges, 1985b	9.31	3.51-24.67	3.80	1.73	1.41-2.12
Kashikar-Zuck, 2008	3.43	1.61-7.33	5.20	1.83	1.45-2.30
Kolaitis, 2022	1.01	0.61-1.67	7.64	1.99	1.57-2.53
Moore, 2020	2.45	1.17-5.16	5.31	1.87	1.48-2.37
Mortimer, 1992	1.51	1.22-1.88	10.83	2.00	1.52-2.61
Liakopoulou-Kairis, 2002	2.15	1.14-4.06	6.26	1.89	1.48-2.40
Ramchandani, 2006	1.56	1.25-1.94	10.83	1.99	1.52-2.61
Reid, 1997	0.17	0.01-3.97	0.52	1.91	1.52-2.40
Robinson, 1990	2.39	1.07-5.36	4.84	1.88	1.48-2.38
Tran, 2021	4.25	0.97-18.71	2.00	1.86	1.48-2.35
Walker, 1989	2.80	1.26-6.23	4.88	1.86	1.47-2.35
Total	1.90	1.51-2.38	-	-	-
<i>Parent General Distress</i>					
Brown, 2022	1.39	0.99-1.94	13.51	1.80	1.50-2.15
Campo, 2007	3.65	1.75-7.63	4.40	1.69	1.45-1.96
Feldman, 2010	1.74	1.34-2.25	17.35	1.74	1.42-2.14
Helgeland, 2010	3.20	1.68-6.11	5.46	1.68	1.44-1.97

Hinze, 2023	1.87	1.67-2.10	26.11		1.71	1.38-2.13
Incleidon, 2016	1.39	0.88-2.21	9.08		1.78	1.49-2.12
Jamison, 1992	3.82	1.19-12.24	1.93		1.71	1.45-2.02
Liakopoulou-Kairis, 2002	1.09	0.58-2.05	5.73		1.78	1.51-2.10
Lommel, 2011	2.17	0.82-5.71	2.70		1.73	1.45-2.06
Mikkelsen, 2021	1.41	0.91-2.17	9.91		1.78	1.49-2.13
Moayedi, 2015	1.37	0.62-3.03	3.83		1.75	1.47-2.09
Total	1.74	1.47-2.05	-		-	-

Abbreviations. CI, confidence interval.

Supplementary Table 2.10

Results of Analyses Examining Moderators of the Associations Between Parent Mental Health and the Functioning of Children with Chronic Pain

<i>Parent Anxiety and Child Pain Intensity</i>					
Continuous moderators	k	B	95% CI	z-value	p-value
Child age (mean)	8	0.02	-0.08, 0.13	0.43	.666
Child sex/gender (% female)	8	0.003	-0.01, 0.01	0.54	.586
Child race/ethnicity (% White)	5	-0.01	-0.02, 0.01	-1.21	.227
<i>Parent Anxiety and Child Depression Symptoms</i>					
Continuous moderators	k	B	95% CI	z-value	p-value
Child age (mean)	8	0.05	-0.05, 0.15	0.94	.347
Child sex/gender (% female)	8	0.002	-0.01, 0.01	0.54	.589
Child race/ethnicity (% White)	6	0.004	-0.01, 0.02	0.61	.542
<i>Parent Depression and Child Depression Symptoms</i>					
Continuous moderators	k	B	95% CI	z-value	p-value
Child age (mean)	8	0.06	-0.05, 0.16	1.01	.313
Child sex/gender (% female)	8	0.004	-0.004, 0.01	0.92	.359
Child race/ethnicity (% White)	6	-0.002	-0.02, 0.01	-0.33	.741

Abbreviations. CI, confidence interval; k, studies.

Note. Analyses included univariable meta-regression for continuous moderators. Categorical moderator variables were not examined as there were <10 studies in each analysis.

Supplementary Table 2.11

Summary Statistics and Sensitivity Analysis for Each Study Included in Meta-Analyses

Examining the Associations Between Parent Mental Health and the Functioning of Children with Chronic Pain

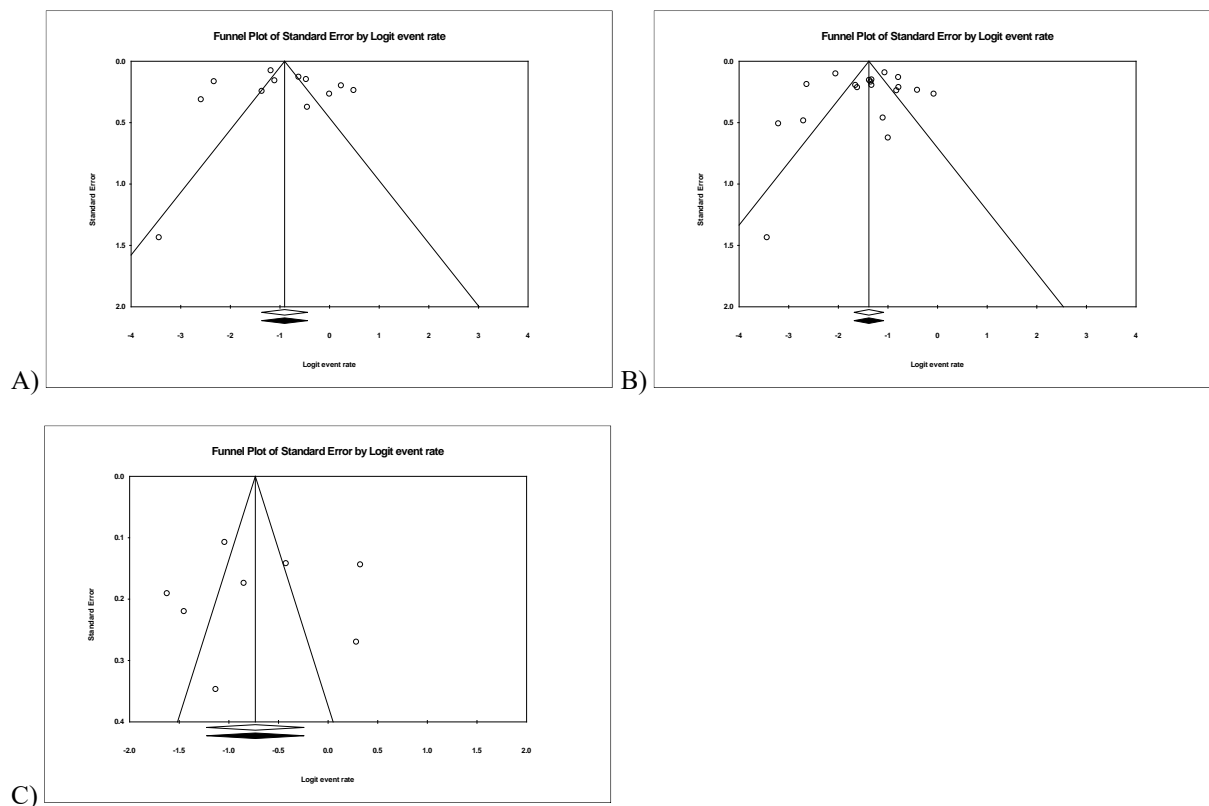
Study	Summary Statistics for Each Study			Leave-one-out Sensitivity Analysis for Each Study	
	Estimate	95% CI	Weight	Estimate	95% CI
<i>Parent Anxiety and Child Pain Intensity</i>					
Beveridge, 2022	-0.06	-0.21-0.09	15.74	0.13	0.05-0.21
Dutta, 2021	0.01	-0.19-0.22	11.61	0.12	0.02-0.22
Eccleston, 2004	0.22	-0.0-0.43	10.09	0.09	-0.01-0.19
Evans, 2010	0.30	0.02-0.53	7.49	0.09	0.00-0.18
Goubert, 2006	0.16	-0.03-0.34	12.63	0.10	-0.01-0.20
Poppert Cordts, 2019	0.24	0.08-0.39	14.92	0.08	-0.01-0.16
Sieberg, 2011	0.02	-0.14-0.18	15.46	0.12	0.02-0.22
Vetter, 2013	0.05	-0.15-0.25	12.06	0.11	0.01-0.21
Total	0.10	0.01-0.19	-	-	-
<i>Parent Anxiety and Child Physical Functioning</i>					
Beveridge, 2022	0.10	-0.06-0.25	12.92	0.18	0.12-0.25
Brown, 2021	0.09	-0.06-0.23	14.48	0.19	0.12-0.25
Cohen, 2010	0.35	0.17-0.51	8.72	0.15	0.10-0.21
Dutta, 2021	0.16	-0.04-0.36	8.00	0.17	0.11-0.24
Eccleston, 2004	0.01	-0.22-0.24	6.53	0.18	0.12-0.24
Evans, 2010	0.28	0.00-0.52	4.39	0.17	0.10-0.23
Goubert, 2006	0.20	0.01-0.38	9.11	0.17	0.10-0.24
Poppert Cordts, 2019	0.28	0.12-0.42	12.01	0.16	0.10-0.22
Reid, 1997	0.07	-0.30-0.42	2.58	0.18	0.11-0.24
Sieberg, 2011	0.12	-0.04-0.27	12.78	0.18	0.11-0.25
Vetter, 2013	0.23	0.03-0.41	8.48	0.17	0.10-0.23
Total	0.17	0.11-0.23	-	-	-
<i>Parent Anxiety and Child Anxiety Symptoms</i>					
Brown, 2021	0.18	0.04-0.32	19.74	0.23	0.16-0.29
Cohen, 2010	0.34	0.16-0.50	10.92	0.20	0.13-0.27
Dutta, 2021	0.23	0.03-0.41	9.92	0.22	0.15-0.28
Eccleston, 2004	0.10	-0.13-0.32	7.94	0.23	0.16-0.29
Poppert Cordts, 2019	0.26	0.10-0.41	15.77	0.21	0.14-0.28
Ramchandani, 2007	0.27	0.07-0.45	9.92	0.21	0.15-0.28
Soltani, 2022	0.14	-0.03-0.30	15.22	0.23	0.16-0.30
Vetter, 2013	0.23	0.03-0.41	10.58	0.22	0.15-0.28
Total	0.22	0.15-0.28	-	-	-
<i>Parent Anxiety and Child Depression Symptoms</i>					
Brown, 2021	0.25	0.11-0.38	15.26	0.19	0.07-0.30
Cohen, 2010	0.45	0.28-0.59	12.11	0.17	0.09-0.24
Dutta, 2021	0.04	-0.17-0.24	11.57	0.22	0.12-0.32
Eccleston, 2004	-0.04	-0.26-0.19	10.32	0.23	0.13-0.31
Evans, 2010	0.27	0.05-0.47	10.40	0.19	0.08-0.30
Poppert Cordts, 2019	0.23	0.07-0.38	14.11	0.19	0.08-0.30
Soltani, 2022	0.15	-0.01-0.30	14.29	0.21	0.09-0.32

Vetter, 2013	0.17	-0.03-0.36	11.94		0.20	0.09-0.31
Total	0.20	0.10-0.29	-		-	-
<i>Parent Depression and Child Pain Intensity</i>						
Beveridge, 2022	0.03	-0.12-0.18	18.48		0.14	0.07-0.21
Dutta, 2021	0.11	-0.10-0.30	10.39		0.12	0.05-0.19
Eccleston, 2004	0.05	-0.18-0.27	8.31		0.13	0.06-0.19
Evans, 2010	0.30	0.02-0.53	5.43		0.11	0.04-0.18
Goubert, 2006	0.22	0.03-0.39	12.01		0.11	0.04-0.18
Poppert Cordts, 2019	0.19	0.03-0.34	16.51		0.11	0.03-0.18
Sieberg, 2011	0.08	-0.08-0.23	17.78		0.13	0.06-0.20
Vetter, 2013	0.09	-0.11-0.28	11.09		0.12	0.05-0.19
Total	0.12	0.05-0.18	-		-	-
<i>Parent Depression and Child Physical Functioning</i>						
Beveridge, 2022	0.20	0.05-0.34	13.11		0.21	0.14-0.27
Brown, 2021	0.15	0.00-0.29	14.84		0.21	0.15-0.28
Cohen, 2010	0.22	0.03-0.40	8.62		0.20	0.14-0.27
Dutta, 2021	0.30	0.11-0.48	7.88		0.20	0.14-0.25
Eccleston, 2004	-0.01	-0.24-0.22	6.38		0.22	0.16-0.27
Evans, 2010	0.34	0.07-0.56	4.23		0.20	0.14-0.26
Goubert, 2006	0.29	0.11-0.45	9.03		0.20	0.13-0.26
Poppert Cordts, 2019	0.27	0.11-0.41	12.12		0.20	0.13-0.26
Reid, 1997	0.12	-0.25-0.46	2.46		0.21	0.15-0.27
Sieberg, 2011	0.09	-0.07-0.24	12.96		0.22	0.16-0.28
Vetter, 2013	0.29	0.10-0.46	8.37		0.20	0.14-0.26
Total	0.20	0.15-0.26	-		-	-
<i>Parent Depression and Child Anxiety Symptoms</i>						
Brown, 2021	0.17	0.03-0.31	19.35		0.24	0.15-0.33
Cohen, 2010	0.30	0.11-0.47	12.89		0.22	0.13-0.30
Dutta, 2021	0.14	-0.06-0.34	12.00		0.24	0.15-0.32
Eccleston, 2004	0.09	-0.14-0.31	10.07		0.24	0.16-0.32
Poppert Cordts, 2019	0.26	0.10-0.41	16.74		0.22	0.13-0.31
Soltani, 2022	0.18	0.02-0.34	16.34		0.24	0.14-0.32
Vetter, 2013	0.42	0.24-0.57	12.60		0.20	0.13-0.27
Total	0.23	0.15-0.30	-		-	-
<i>Parent Depression and Child Depression Symptoms</i>						
Brown, 2021	0.27	0.13-0.40	15.06		0.24	0.12-0.36
Cohen, 2010	0.38	0.20-0.53	12.15		0.23	0.12-0.33
Dutta, 2021	-0.04	-0.24-0.16	11.64		0.28	0.21-0.36
Eccleston, 2004	0.10	-0.13-0.32	10.45		0.26	0.16-0.36
Evans, 2010	0.35	0.14-0.53	10.52		0.23	0.12-0.34
Poppert Cordts, 2019	0.37	0.22-0.50	14.01		0.23	0.12-0.33
Soltani, 2022	0.16	0.00-0.31	14.18		0.26	0.15-0.37
Vetter, 2013	0.33	0.14-0.50	11.98		0.24	0.12-0.34
Total	0.25	0.15-0.34	-		-	-
<i>Parent General Distress and Child Physical Functioning</i>						
Birmie, 2020	0.11	0.02-0.20	41.98		0.12	0.02-0.23
Brown, 2021	0.08	-0.01-0.17	42.55		0.13	0.05-0.21
Logan, 2005	0.29	0.06-0.49	6.32		0.10	0.03-0.16
Wendland, 2010	0.11	-0.09-0.30	9.15		0.11	0.04-0.19
Total	0.11	0.05	-		-	-

Abbreviations. CI, confidence interval.

Supplementary Figure 2.1

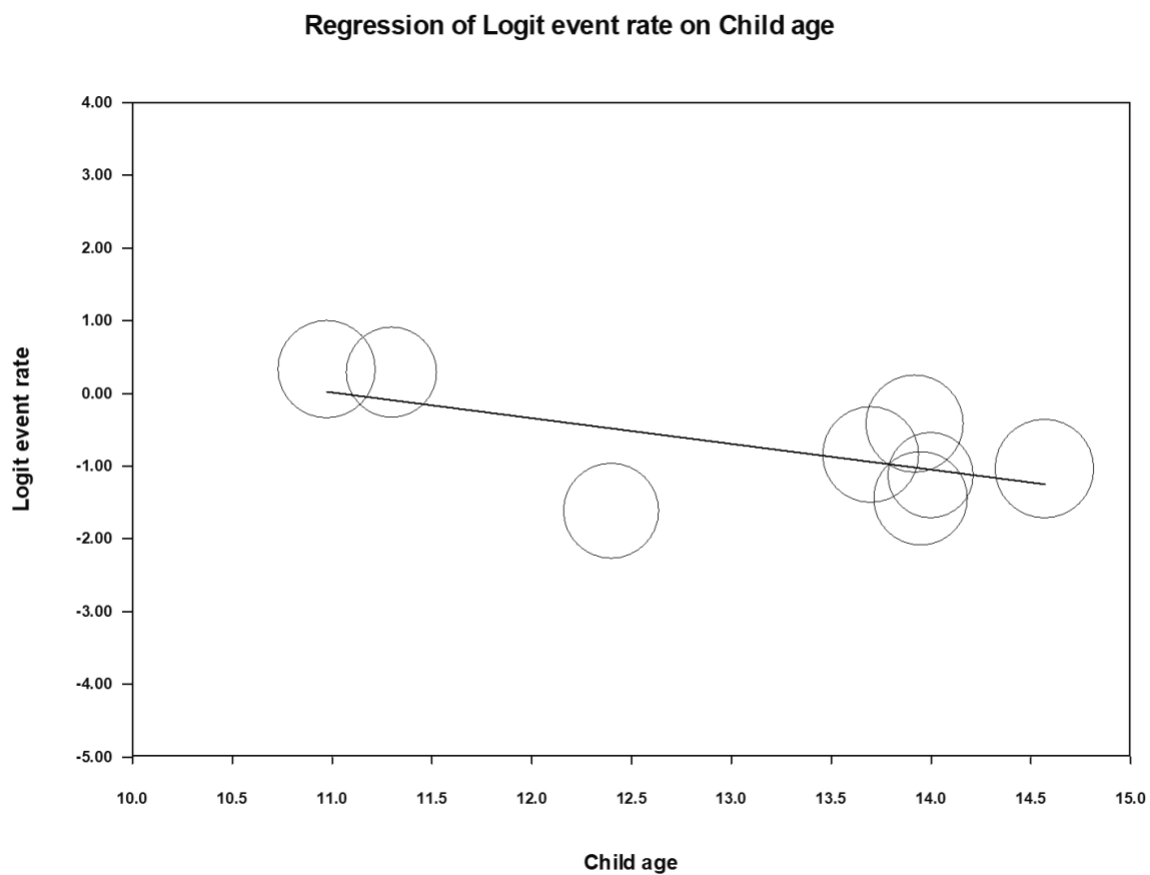
Funnel Plots of Studies Examining the Prevalence of Mental Health Problems Among Parents of Children with Chronic Pain



Note. Funnel plots of studies examining the prevalence of (A) parent anxiety problems (Egger test $p=.740$); (B) parent depression problems (Egger test $p=.872$); (C) parent general distress problems (Egger test $p=.912$).

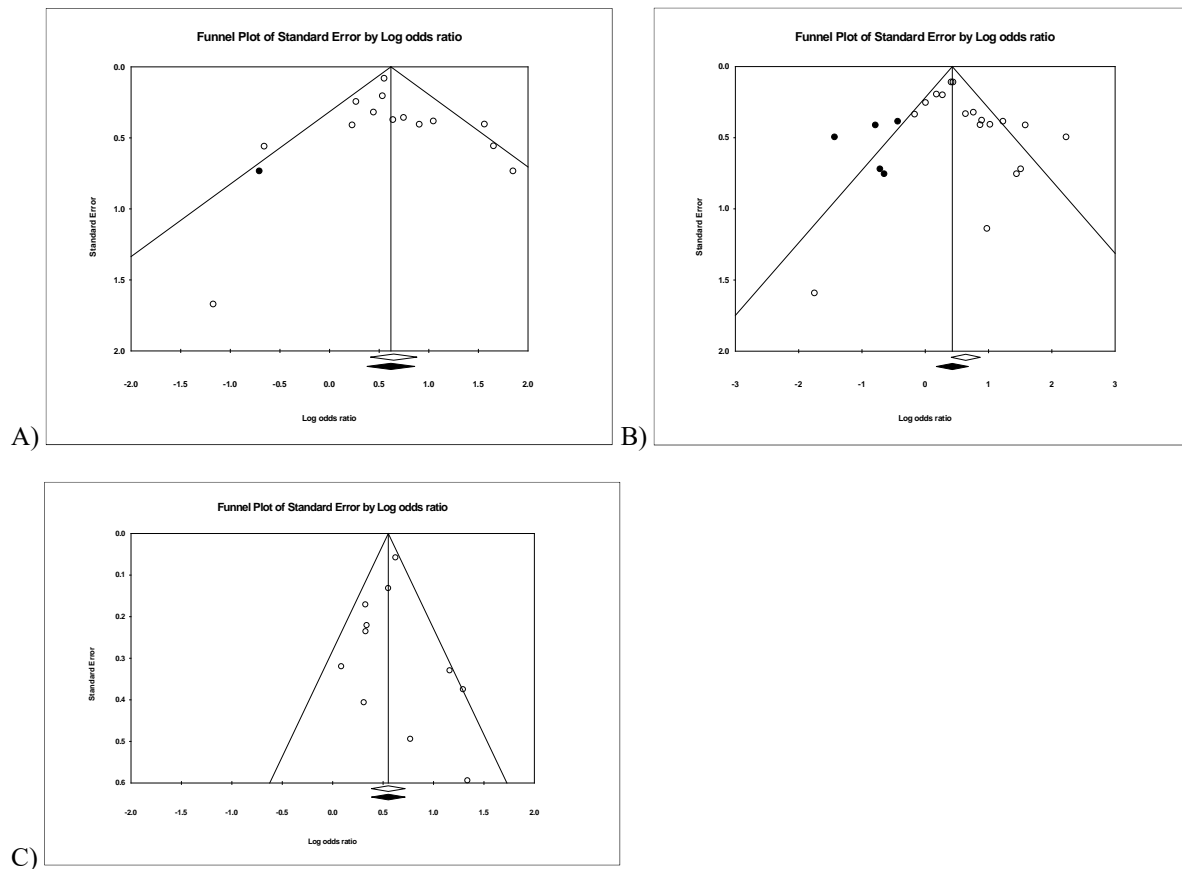
Supplementary Figure 2.2

Meta-Regression Scatterplot Examining the Moderator of Child Age on the Prevalence of General Distress Problems in Parents of Children with Chronic Pain



Supplementary Figure 2.3

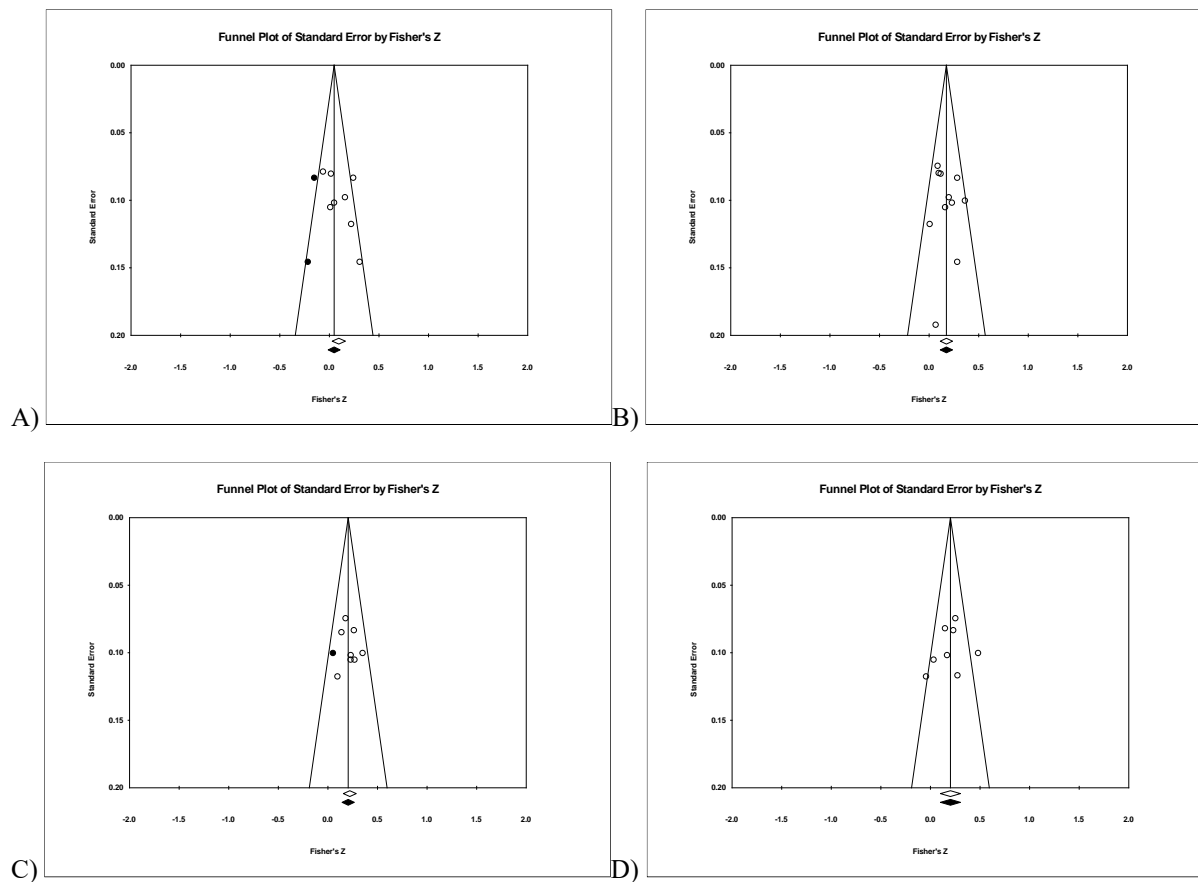
Funnel Plots of Studies Examining the Associations Between Parent Mental Health and the Presence of Child Chronic Pain



Note. Funnel plots of studies examining associations between (A) parent anxiety and the presence of child chronic pain (Egger test $p=.496$); (B) parent depression and the presence of child chronic pain (Egger test $p=.061$); (C) parent general distress and the presence of child chronic pain (Egger test $p=.998$).

Supplementary Figure 2.4

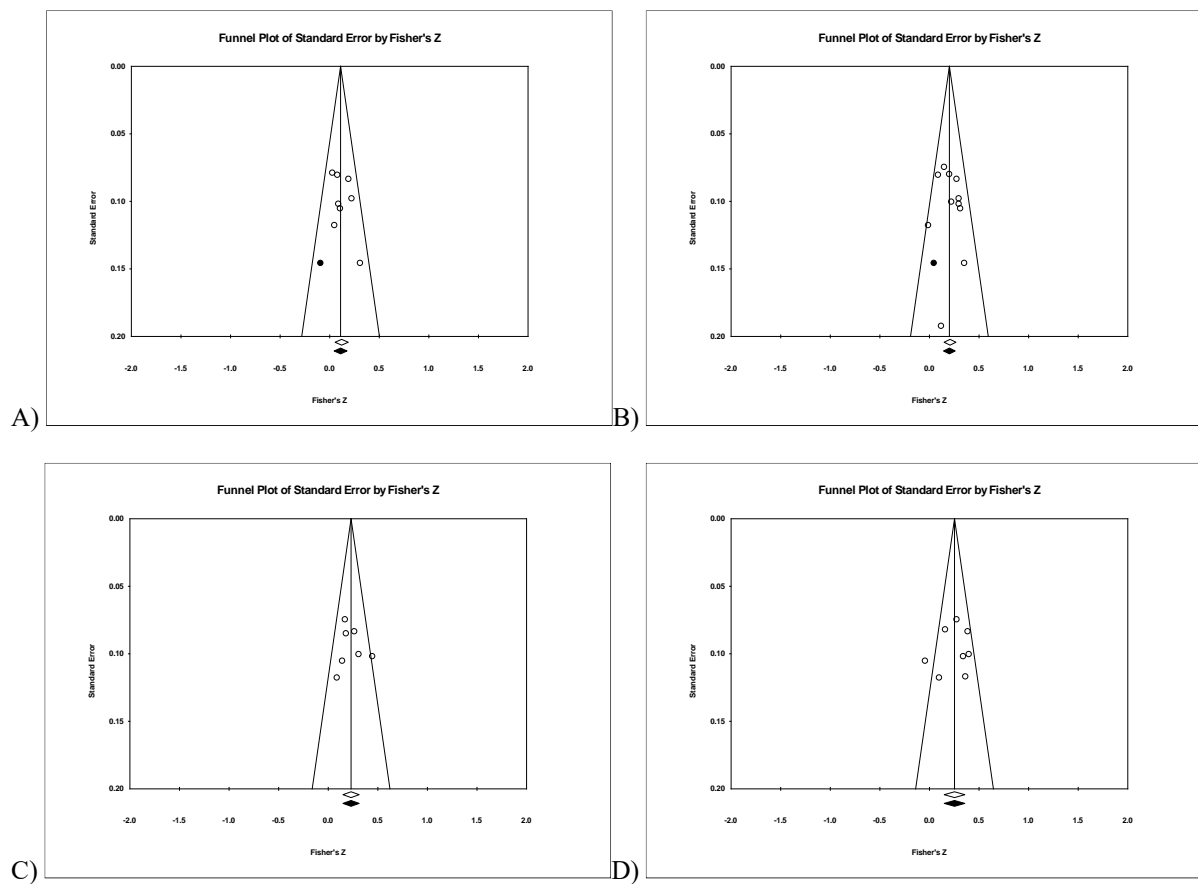
Funnel Plots of Studies Examining the Associations Between Parent Anxiety and the Functioning of Children with Chronic Pain



Note. Funnel plots of studies examining associations between parent anxiety and (A) child pain intensity (Egger test $p=.181$); (B) child physical functioning (Egger test $p=.748$); (C) child anxiety symptoms (Egger test $p=.745$); (D) child depression symptoms (Egger test $p=.562$)

Supplementary Figure 2.5

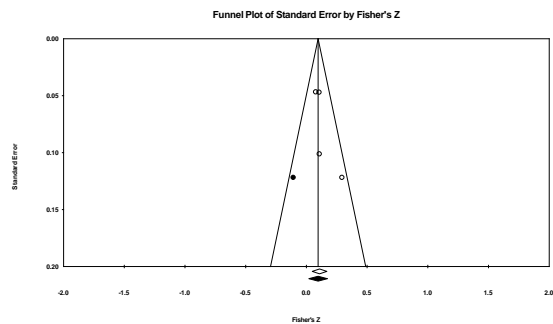
Funnel Plots of Studies Examining the Associations Between Parent Depression and the Functioning of Children with Chronic Pain



Note. Funnel plots of studies examining associations between parent depression and (A) child pain intensity (Egger test $p=.270$); (B) child physical functioning (Egger test $p=.705$); (C) child anxiety symptoms (Egger test $p=.856$); (D) child depression symptoms (Egger test $p=.676$)

Supplementary Figure 2.6

Funnel Plot of Studies Examining the Association Between Parent General Distress and Child Physical Functioning



Note. Funnel plot of studies examining the association between parent general distress and child physical functioning (Egger test $p=.228$)

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Appendix C: Supplementary Material for Chapter 4

Methods for bootstrapping

Bootstrapping analyses were conducted through the bootstrapping function in SPSS with 1000 samples and 95% bias-corrected and accelerated (BCa) confidence intervals (CIs).

Methods for multiple imputation

Multiple imputation data was generated through the Multiple Imputation procedure in SPSS. The fully conditional specification method was used, which is an iterative Markov Chain Monte Carlo (MCMC) method. The number of imputations was set at 25 while the number of iterations was set at 20. Predictive mean matching was used, with the 5 closest predictions. The total number of missing values for each variable that were imputed (times 25) are as follows: Maternal racial/ethnic identity (n=3), maternal education (n=102), maternal income (n=5), maternal age (n=12), maternal marital status (n=0), child age (n=0), child gender (n=30), maternal chronic pain (n=0), maternal depressive symptoms (n=3), maternal anxiety symptoms (n=18), maternal ineffective parenting (n=16), maternal positive parenting (n=6), maternal social support (n=15), maternal active coping (n=178), maternal optimism (n=190), child chronic pain (n=8), child connections with adults at home (n=294), child connections with adults at school (n=297), child connections with adults in community (n=294), child peer belonging (n=290), child optimism (n=296), child community engagement (n=305).

Supplementary Table 4.1

Results of Bootstrap Sensitivity Analyses for Logistic Regression Analyses with Maternal Moderators

Predictor variables	Unadjusted			Adjusted		
	B (SE)	<i>p</i>	BCa 95% CI	B (SE)	<i>p</i>	BCa 95% CI
Step 1						
Maternal chronic pain	0.66 (0.25)	.006	0.16, 1.15	0.64 (0.28)	.019	0.13, 1.11
Maternal anxiety symptoms	0.03 (0.02)	.260	-0.02, 0.06	0.04 (0.03)	.100	-0.01, 0.10
Step 2						
Interaction	0.12 (0.07)	.043	-0.004, 0.27	0.16 (0.07)	.011	0.04, 0.31
Step 1						
Maternal chronic pain	0.65 (0.24)	.005	0.19, 1.11	0.56 (0.29)	.041	-0.07, 1.06
Maternal depressive symptoms	0.02 (0.01)	.031	0.001, 0.04	0.03 (0.01)	.011	0.004, 0.05
Step 2						
Interaction	0.03 (0.02)	.265	-0.02, 0.07	0.03 (0.03)	.221	-0.02, 0.09
Step 1						
Maternal chronic pain	0.76 (0.25)	.003	0.23, 1.20	0.74 (0.28)	.009	0.13, 1.24
Maternal ineffective parenting	-0.01 (0.02)	.587	-0.05, 0.03	-0.003 (0.02)	.906	-0.05, 0.04
Step 2						
Interaction	0.12 (0.06)	.034	0.01, 0.24	0.14 (0.07)	.030	-0.01, 0.32
Step 1						
Maternal chronic pain	0.76 (0.24)	<.001	0.28, 1.14	0.74 (0.28)	.004	0.16, 1.21
Maternal positive parenting	-0.02 (0.03)	.434	-0.08, 0.04	-0.01 (0.03)	.709	-0.08, 0.05
Step 2						
Interaction	-0.04 (0.09)	.624	-0.24, 0.14	-0.06 (0.11)	.568	-0.27, 0.13
Step 1						
Maternal chronic pain	0.76 (0.25)	.003	0.24, 1.19	0.72 (0.27)	.009	0.11, 1.24
Maternal social support	-0.01 (0.02)	.800	-0.04, 0.04	-0.01 (0.02)	.790	-0.05, 0.05
Step 2						
Interaction	-0.04 (0.06)	.483	-0.14, 0.07	-0.02 (0.06)	.687	-0.14, 0.09
Step 1						
Maternal chronic pain	0.66 (0.27)	.017	0.14, 1.15	0.67 (0.31)	.022	0.01, 1.24
Maternal optimism	-0.03 (0.02)	.118	-0.07, 0.01	-0.03 (0.02)	.102	-0.08, 0.01
Step 2						
Interaction	0.06 (0.06)	.300	-0.05, 0.19	0.08 (0.07)	.206	-0.05, 0.26
Step 1						
Maternal chronic pain	0.66 (0.28)	.014	0.03, 1.18	0.70 (0.31)	.015	0.01, 1.34
Maternal active coping	-0.06 (0.04)	.066	-0.13, 0.01	-0.06 (0.04)	.095	-0.14, 0.03
Step 2						
Interaction	0.08 (0.10)	.389	-0.13, 0.28	0.14 (0.13)	.228	-0.12, 0.40

Note. Bootstrapping conducted with 1000 samples and bias-corrected and accelerated 95% confidence intervals (BCa 95% CI).

Supplementary Table 4.2

Results of Bootstrap Sensitivity Analyses for Logistic Regression Analyses with Child Moderators

Predictor variables	Unadjusted			Adjusted		
	B (SE)	<i>p</i>	BCa 95% CI	B (SE)	<i>p</i>	BCa 95% CI
Step 1						
Maternal chronic pain	0.80 (0.30)	.005	0.15, 1.33	0.80 (0.34)	.016	0.06, 1.42
Child optimism	-0.10 (0.03)	.005	-0.16, -0.03	-0.10 (0.04)	.003	-0.16, -0.04
Step 2						
Interaction	0.05 (0.12)	.642	-0.17, 0.33	-0.11 (0.14)	.380	-0.40, 0.16
Step 1						
Maternal chronic pain	0.75 (0.29)	.006	0.16, 1.24	0.76 (0.33)	.009	0.07, 1.35
Child connections with adults at home	-0.13 (0.06)	.015	-0.24, -0.01	-0.14 (0.07)	.021	-0.27, -0.02
Step 2						
Interaction	-0.08 (0.18)	.612	-0.44, 0.26	-0.19 (0.25)	.397	-0.67, 0.21
Step 1						
Maternal chronic pain	0.70 (0.30)	.016	0.07, 1.22	0.69 (0.34)	.028	-0.03, 1.32
Child connections with adults at school	-0.08 (0.05)	.089	-0.17, 0.02	-0.11 (0.05)	.028	-0.21, -0.01
Step 2						
Interaction	-0.14 (0.15)	.320	-0.45, 0.18	-0.23 (0.18)	.148	-0.57, 0.05
Step 1						
Maternal chronic pain	0.79 (0.30)	.007	0.13, 1.30	0.77 (0.35)	.021	0.04, 1.38
Child connections with adults in community	-0.05 (0.03)	.091	-0.11, 0.01	-0.05 (0.04)	.187	-0.12, 0.03
Step 2						
Interaction	-0.05 (0.11)	.625	-0.28, 0.16	-0.04 (0.12)	.721	-0.27, 0.14
Step 1						
Maternal chronic pain	0.73 (0.30)	.009	0.09, 1.28	0.75 (0.33)	.016	0.07, 1.34
Child peer belonging	-0.08 (0.05)	.079	-0.16, 0.02	-0.09 (0.06)	.120	-0.19, 0.04
Step 2						
Interaction	-0.17 (0.14)	.187	-0.47, 0.11	-0.26 (0.18)	.097	-0.62, 0.02
Step 1						
Maternal chronic pain	0.73 (0.31)	.016	0.10, 1.29	0.72 (0.34)	.023	0.06, 1.32
Child community engagement	-0.02 (0.09)	.805	-0.22, 0.16	0.02 (0.10)	.896	-0.18, 0.20
Step 2						
Interaction	-0.07 (0.26)	.767	-0.61, 0.44	-0.13 (0.29)	.629	-0.74, 0.43

Note. Bootstrapping conducted with 1000 samples and bias-corrected and accelerated 95% confidence intervals (BCa 95% CI).

Supplementary Table 4.3

Results of Multiple Imputation Sensitivity Analyses for Logistic Regression Analyses with Maternal Moderators

Predictor variables	Unadjusted			Adjusted			Imputed cases
	B (SE)	p	OR (95% CI)	B (SE)	p	OR (95% CI)	
Step 1							Unadjusted: n = 26
Maternal chronic pain	0.68 (0.24)	.005	1.97 (1.23, 3.16)	0.65 (0.25)	.009	1.92 (1.18, 3.11)	Adjusted: n = 171
Maternal anxiety symptoms	0.02 (0.02)	.334	1.02 (0.98, 1.07)	0.03 (0.02)	.227	1.03 (0.98, 1.08)	
Step 2							
Interaction	0.10 (0.06)	.078	1.11 (0.99, 1.25)	0.11 (0.06)	.077	1.11 (0.99, 1.26)	
Step 1							Unadjusted: n = 11
Maternal chronic pain	0.61 (0.24)	.012	1.84 (1.14, 2.97)	0.59 (0.25)	.019	1.80 (1.10, 2.93)	Adjusted: n = 158
Maternal depressive symptoms	0.02 (0.01)	.050	1.02 (1.00, 1.04)	0.02 (0.01)	.037	1.02 (1.00, 1.04)	
Step 2							
Interaction	0.02 (0.02)	.349	1.02 (0.98, 1.07)	0.03 (0.03)	.294	1.03 (0.98, 1.08)	
Step 1							Unadjusted: n = 24
Maternal chronic pain	0.74 (0.23)	.002	2.09 (1.32, 3.30)	0.72 (0.24)	.003	2.06 (1.29, 3.30)	Adjusted: n = 166
Maternal ineffective parenting	-0.01 (0.02)	.650	0.99 (0.95, 1.03)	-0.003 (0.02)	.896	1.00 (0.96, 1.04)	
Step 2							
Interaction	0.12 (0.06)	.047	1.12 (1.00, 1.26)	0.11 (0.06)	.066	1.12 (0.99, 1.26)	
Step 1							Unadjusted: n = 14
Maternal chronic pain	0.74 (0.23)	.002	2.10 (1.33, 3.33)	0.73 (0.24)	.003	2.07 (1.29, 3.31)	Adjusted: n = 159
Maternal positive parenting	-0.03 (0.03)	.417	0.98 (0.92, 1.04)	-0.02 (0.03)	.510	0.98 (0.92, 1.04)	
Step 2							
Interaction	-0.04 (0.09)	.629	0.96 (0.81, 1.14)	-0.02 (0.09)	.800	0.98 (0.82, 1.17)	
Step 1							Unadjusted: n = 22
Maternal chronic pain	0.72 (0.24)	.002	2.06 (1.30, 3.28)	0.72 (0.24)	.003	2.05 (1.27, 3.29)	Adjusted: n = 168
Maternal social support	-0.01 (0.02)	.743	0.99 (0.95, 1.03)	-0.004 (0.02)	.856	1.00 (0.96, 1.04)	
Step 2							
Interaction	-0.03 (0.05)	.554	0.97 (0.88, 1.07)	-0.03 (0.05)	.525	0.97 (0.87, 1.07)	
Step 1							Unadjusted: n = 196
Maternal chronic pain	0.64 (0.24)	.008	1.89 (1.18, 3.02)	0.61 (0.25)	.013	1.85 (1.14, 3.00)	Adjusted: n = 311
Maternal optimism	-0.04 (0.02)	.043	0.97 (0.93, 1.00)	-0.04 (0.02)	.039	0.96 (0.93, 1.00)	
Step 2							
Interaction	0.06 (0.05)	.225	1.06 (0.96, 1.18)	0.07 (0.05)	.178	1.08 (0.97, 1.19)	
Step 1							Unadjusted: n = 184
Maternal chronic pain	0.68 (0.24)	.004	1.98 (1.25, 3.15)	0.66 (0.24)	.006	1.94 (1.21, 3.12)	Adjusted: n = 300
Maternal active coping	-0.06 (0.03)	.062	0.94 (0.88, 1.00)	-0.07 (0.03)	.040	0.93 (0.87, 1.00)	
Step 2							
Interaction	0.06 (0.10)	.558	1.06 (0.87, 1.29)	0.08 (0.10)	.475	1.08 (0.88, 1.32)	

Abbreviations. CI, confidence interval; OR, odds ratio

Supplementary Table 4.4

Results of Multiple Imputation Sensitivity Analyses for Logistic Regression Analyses with Child Moderators

Predictor variables	Unadjusted			Adjusted			Imputed cases
	B (SE)	<i>p</i>	OR (95% CI)	B (SE)	<i>p</i>	OR (95% CI)	
Step 1							Unadjusted: n = 302
Maternal chronic pain	0.73 (0.24)	.002	2.07 (1.30, 3.29)	0.72 (0.24)	.003	2.05 (1.28, 3.28)	Adjusted: n = 403
Child optimism	-0.09 (0.03)	.004	0.91 (0.86, 0.97)	-0.07 (0.03)	.035	0.93 (0.87, 1.00)	
Step 2							
Interaction	0.04 (0.10)	.652	1.05 (0.86, 1.27)	0.02 (0.10)	.845	1.02 (0.84, 1.25)	
Step 1							Unadjusted: n = 300
Maternal chronic pain	0.72 (0.24)	.002	2.05 (1.29, 3.25)	0.71 (0.24)	.003	2.03 (1.27, 3.26)	Adjusted: n = 402
Child connections with adults at home	-0.13 (0.06)	.022	0.88 (0.78, 0.98)	-0.10 (0.06)	.080	0.90 (0.80, 1.01)	
Step 2							
Interaction	-0.05 (0.17)	.777	0.95 (0.69, 1.32)	-0.06 (0.17)	.730	0.94 (0.67, 1.33)	
Step 1							Unadjusted: n = 303
Maternal chronic pain	0.73 (0.24)	.002	2.08 (1.31, 3.29)	0.72 (0.24)	.003	2.05 (1.28, 3.29)	Adjusted: n = 406
Child connections with adults at school	-0.07 (0.05)	.137	0.94 (0.86, 1.02)	-0.06 (0.05)	.241	0.95 (0.86, 1.04)	
Step 2							
Interaction	-0.09 (0.12)	.471	0.92 (0.72, 1.16)	-0.11 (0.13)	.367	0.89 (0.70, 1.14)	
Step 1							Unadjusted: n = 300
Maternal chronic pain	0.75 (0.24)	.001	2.11 (1.33, 3.35)	0.73 (0.24)	.003	2.07 (1.29, 3.32)	Adjusted: n = 402
Child connections with adults in community	-0.04 (0.03)	.166	0.96 (0.90, 1.02)	-0.03 (0.03)	.308	0.97 (0.91, 1.03)	
Step 2							
Interaction	-0.02 (0.08)	.844	0.98 (0.84, 1.16)	-0.01 (0.09)	.909	0.99 (0.84, 1.17)	
Step 1							Unadjusted: n = 296
Maternal chronic pain	0.70 (0.24)	.003	2.01 (1.27, 3.19)	0.70 (0.24)	.004	2.02 (1.26, 3.25)	Adjusted: n = 400
Child peer belonging	-0.07 (0.04)	.113	0.94 (0.86, 1.02)	-0.03 (0.04)	.473	0.97 (0.89, 1.06)	
Step 2							
Interaction	-0.09 (0.12)	.467	0.92 (0.73, 1.16)	-0.10 (0.12)	.423	0.91 (0.71, 1.15)	
Step 1							Unadjusted: n = 311
Maternal chronic pain	0.73 (0.24)	.002	2.08 (1.31, 3.30)	0.73 (0.24)	.003	2.08 (1.29, 3.34)	Adjusted: n = 412
Child community engagement	-0.01 (0.08)	.931	0.99 (0.84, 1.17)	0.03 (0.09)	.701	1.04 (0.87, 1.23)	
Step 2							
Interaction	-0.03 (0.22)	.887	0.97 (0.62, 1.51)	-0.12 (0.23)	.606	0.89 (0.57, 1.39)	

Abbreviations. CI, confidence interval; OR, odds ratio