CHILD OUTCOMES IN THE CONTEXT OF PARENTAL CHRONIC PAIN: EXAMINING SOCIAL TRANSMISSION PATHWAYS

by

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To my own parents, Joy and Kirk Higgins.
Thank you for everything

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ABSTRACT

Parents play a crucial role in children's pain experiences. Theory and empirical studies have highlighted the association between parental chronic pain and children's pain and mental health. The current dissertation aimed to: synthesize the results of existing literature on outcomes in offspring of parents with chronic pain (Paper 1); examine theorized social transmission pathways in predicting child pain and internalizing symptoms in children of parents with chronic pain (Paper 2); and examine the role of a novel construct, child catastrophizing about parental chronic pain, in predicting child outcomes (Paper 3). Paper 1 describes a systematic review of 59 studies of outcomes in offspring of parents with chronic pain. Meta-analyses indicated that offspring of parents with chronic pain had poorer outcomes than other offspring in several areas (e.g., increased pain, internalizing outcomes, and adverse birth outcomes; poorer social competence and family outcomes), while meta-ethnography results identified both negative and positive (e.g., development of compassion) impacts on offspring. In Paper 2, 72 dyads comprised of adults with chronic pain and their children (ages 8-15 years) completed questionnaires (regarding pain, pain catastrophizing, child internalizing symptoms) and children completed the cold pressor task (CPT) in the presence of their parent. Verbalizations during the CPT were coded and pain outcomes (observational, child-reported, parent-reported) were recorded. Support was found for parental modeling in predicting child internalizing symptoms but not the presence of child recurrent pain, and parental reinforcement of child pain behavior predicted increased child CPT pain. Paper 3 presents a dyadic analysis of children's catastrophizing about their parent's chronic pain and its role in predicting child pain and internalizing outcomes in the same sample of dyads. Children's increased catastrophizing about their parent's pain predicted increased child- and parent-reported child internalizing symptoms and CPT pain intensity over and above the influence of parent and child catastrophizing about their own pain. The findings of the dissertation suggest that parents' own management of their chronic pain and reinforcement of child pain behavior, as well as children's catastrophizing about their own and their parent's chronic pain, could be potential targets for intervention and prevention programs in this vulnerable population.

LIST OF ABBREVIATIONS AND SYMBOLS USED

APIM	Actor Partner Interdependence Model
B	Unstandardized estimate of indirect effect
BASC-2	Behavior Assessment System for Children-2
°C	Degrees Celsius
CAMPIS-R	Child-Adult Medical Procedure Interaction Scale-Revised
CI	Confidence interval
CNCP	Chronic non-cancer pain
cm	Centimeter
CPT	Cold pressor task
EFA	Exploratory factor analysis
FPS-R	Faces Pain Scale-Revised
GRADE	Grading of Recommendations Assessment, Development and Evaluation
I^2	I-squared value of heterogeneity
n	Sample size
M	Mean
MCAR	Missing Completely at Random
MeSH	Medical Subject Headings
NICU	Neonatal Intensive Care Unit
NR	Not reported
NRS	Numerical rating scale
OR	Odds ratio
p	P-value for testing significance
PCS	Pain Catastrophizing Scale
PCS-C	Pain Catastrophizing Scale for Children
PCS-P	Pain Catastrophizing Scale for Parents
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-Analyses
PROMIS	Patient-Reported Outcomes Measurement Information System
r	Pearson's correlation coefficient
R^2	Proportion of explained variance
RMSEA	Root Mean Square Error of Approximation
SE	Standard error
SMD	Standardized mean difference
SD	Standard deviation
TLI/NNFI	Tucker-Lewis Index/Non-Normed Fit Index
Z	Z statistic from meta-analysis
α	Cronbach's alpha (internal consistency)
β	Standardized estimate of indirect effect
κ	Cohen's kappa (inter-rater reliability)
$\Delta \chi^2$	Change in chi-squared value

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CHAPTER 1: INTRODUCTION

1.1 Overview of Chronic Pain

Pain is defined as “an unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage” (Task Force on Taxonomy of the International Association for the Study of Pain, 2002, p. 210). While acute pain resolves within approximately the normal healing time for a given injury, chronic pain is defined as “persistent or recurrent pain lasting longer than three months” (Treede et al., 2015, p. 1004), or longer than six months for the purposes of research (Task Force on Taxonomy of the International Association for the Study of Pain, 2002). Chronic pain is considered to be a biopsychosocial phenomenon; that is, it is impacted by biological (e.g., genetics, tissue damage, nociception), psychological (e.g., beliefs about pain, affective responses to pain such as depressive and anxiety symptoms), and social (e.g., others’ responses to an individual’s pain, communication about pain) factors (Turk & Monarch, 2002). Chronic pain is experienced by approximately 20% of adults and children (King et al., 2011; Schopflocher, Taenzer, & Jovey, 2011; Stanford, Chambers, Biesanz, & Chen, 2008; Vos et al., 2015) and represents a large burden to individuals and society (Gaskin & Richard, 2012; Vos et al., 2015). It is estimated that approximately $60B is spent on chronic pain each year in Canada (Canadian Pain Society, 2014). Although access to pain management is considered a human right (Cousins & Lynch, 2011), pain continues to be under-recognized and under-treated in adults and children (Burke, Mathias, & Denson, 2018; Friedrichsdorf et al., 2015; Hadi, Alldred, Briggs, Marczewski, & Closs, 2017; Johnston, Barrington, Taddio, Carbajal, & Filion, 2011; Lynch et al., 2008; Seers, Derry, Seers, & Moore, 2018).

Adults' and children's experiences with pain can be understood on a variety of dimensions, within the domains of pain, physical functioning, and emotional functioning (Turk et al., 2003). Pain can vary in intensity, quality, frequency, and duration, and reliable and valid measures of these variables exist for both adults and children (Dworkin et al., 2005; McGrath et al., 2008). In addition to the pain itself, individuals may cope with their pain in different ways, such as using approach or problem-focused strategies (e.g., seeking information, social supports, problem-solving strategies), emotion-focused strategies (e.g., worrying about pain), or avoidance-oriented strategies (e.g., reducing activity to avoid pain) (Blount et al., 2008; Reid, Gilbert, & McGrath, 1998). The extent to which pain interferes with a person's mental, physical, and social activities is described as pain interference (Amtmann et al., 2010). The impact of a disease or health condition on a person's daily functioning is known as functional disability (Walker & Greene, 1991). Difficulties with psychological health can also accompany pain, such as depressive and anxiety symptoms (Asmundson & Katz, 2009; de Heer et al., 2018; Turk et al., 2003).

1.2 Families and Pain

Much research has established the important role that families play in children's pain experiences (Birnie, Boerner, & Chambers, 2013; K. S. Higgins, Chambers, Birnie, & Boerner, In Press). Parents in particular are often responsible for identifying children's pain and making decisions about seeking pain assessment and treatment. Theories of parental and family-level influences on pediatric chronic pain (Palermo & Chambers, 2005; Palermo, Valrie, & Karlson, 2014) describe children's pain and functional disability as being influenced by individual child factors (e.g., physiology, emotional responses, pain behaviours), the child's developmental trajectory (including physiological, psychological, social, and emotional processes associated with development), individual parent factors (e.g., parents' own emotions, cognitions about pain, behaviours in response to child pain, health status) and family level factors (e.g., family environment and functioning). Each of these factors are believed to have bidirectional influences on one another; for example, child developmental stage (e.g., preschooler versus adolescent) may influence a parent's response to their child's chronic pain, and parental responses to child pain may influence the child's development of autonomy (Palermo et al., 2014).

Given theory and empirical evidence supporting several parent- and child-level factors as predictive of children's pain, these factors are likely important to consider in parents with chronic pain and their children. Two areas that have received much attention in the broader literature on children's pain are parents' and children's pain catastrophizing, and parents' responses to children's pain. The following sections (1.3 and 1.4) provide overviews of each of these key areas, before reviewing the literature on the aggregation of pain in families (Section 1.5) and a theoretical model that provides hypothesized mechanisms through which parental chronic pain may influence children's outcomes (Section 1.6).

1.3 Pain Catastrophizing in Parents and Children

As described above, pain experiences can be impacted by biological, social, and psychological factors, including cognitions about pain. One of the most robust predictors of poor pain-related outcomes in adults and children with and without chronic pain is pain catastrophizing, the tendency to view presently occurring or anticipated pain in an exaggerated, negative way (Quartana, Campbell, & Edwards, 2009; Sullivan et al., 2001). It encompasses three constructs: rumination about pain (excessive focus on pain, inability to inhibit pain-related thoughts when anticipating or experiencing pain), magnification (the tendency to amplify the threat value of pain), and helplessness (the tendency to feel helpless with regards to pain, lack of belief in ability to cope with pain) (Quartana et al., 2009; Sullivan, 2012) and has been found to be conceptually distinct from anxiety, depression, negative affectivity, and neuroticism (Goubert, Crombez, & Van Damme, 2004; Malfliet et al., 2017; Tran et al., 2015; Vervoort, Goubert, Eccleston, Bijttebier, & Crombez, 2006). It is most commonly measured using the Pain Catastrophizing Scale (Sullivan, Bishop, & Pivik, 1995) in adults and the Pain Catastrophizing Scale for Children (Crombez et al., 2003) in children, which are both self-report questionnaires assessing one's tendency to catastrophize about one's own pain. Pain catastrophizing has most often been conceptualized as a trait variable representing an underlying tendency (Quartana et al., 2009), although research on state pain catastrophizing (i.e. pain catastrophizing in the present moment, specific to a particular situation) has more recently been conducted (Durand et al., 2017). There are several hypothesized mechanisms through which pain catastrophizing might impact pain-related outcomes, including influencing pain appraisals and behavioural responses to pain, representing attention biases to pain stimuli and associated negative affect, soliciting interpersonal support that positively reinforces pain behaviours, altering central nervous system mechanisms in pain processing, impacting physiological responses to pain (e.g., activity in the hypothalamic-pituitary-adrenal axis), or resulting in increased activity in brain areas related to affective pain processing (Quartana et al., 2009).

Higher levels of trait pain catastrophizing (with regards to one's own experienced or anticipated pain) have been associated with a number of adverse pain-related outcomes in adults and children with chronic pain, including greater pain intensity, disability, and internalizing and depressive symptoms (Buenaver, Edwards, Smith, Gramling, & Haythornthwaite, 2008; Craner, Sperry, Koball, Morrison, & Gilliam, 2017; Fisher, Heathcote, Eccleston, Simons, & Palermo, 2017; Miller, Meints, & Hirsh, 2018; Sullivan et al., 2001; Wertli et al., 2014). A recent meta-analytic review found that across a total of 38 unique samples of children and adolescents with chronic pain, pain catastrophizing demonstrated relationships of moderate effect sizes with pain intensity and physical functioning, and of large effect sizes with anxiety, depression, and quality of life (Miller et al., 2018). None of the potential moderators tested (including child sex, age, and pain characteristics, measure of catastrophizing used, and study quality) were found to significantly moderate relationships between catastrophizing and any outcomes (Miller et al., 2018). Changes in pain catastrophizing have been found to mediate the relationship between pain interventions and reduced pain intensity and disability in adults (Junghans-Rutelonis et al., 2018; Smeets, Vlaeyen, Kester, & Knottnerus, 2006; Turner, Holtzman, & Mancl, 2007). In longitudinal studies, decreases in pain catastrophizing early in multidisciplinary chronic pain treatment were found to predict subsequent decreases in pain intensity in adults (Burns, Glenn, Bruehl, Harden, & Lofland, 2003; Racine et al., 2016). Experimental research has established that manipulations of state pain catastrophizing in adults with and without chronic pain predict changes in pain intensity and unpleasantness (Kjogx et al., 2016). Higher levels of trait pain catastrophizing have also been found to predict poorer pain-related outcomes in adults and children experiencing acute clinical (Roh et al., 2014; Vervoort, Goubert, et al., 2011) or laboratory pain (Birnie, Chambers, Chorney, Fernandez, & McGrath, 2016; Kjogx et al., 2016) and postsurgical pain (Birnie, Chorney, El-Hawary, & PORSCHE Study Group, 2017; Sobol-Kwapinska, Bąbel, Plotek, & Stelcer, 2016).

In addition to the intrapersonal influences of pain catastrophizing (i.e. the influence of one's own pain catastrophizing on one's own outcomes), interpersonal impacts in parents and children have also been studied. Much research has examined the association between parental catastrophizing about child pain and children's outcomes. This construct describes parents' tendencies to view their children's pain in an exaggerated, negative way and includes the components of magnification, rumination, and helplessness (Goubert, Eccleston, Vervoort, Jordan, & Crombez, 2006). Mothers generally report higher levels of catastrophizing about their child's pain compared to fathers (Goubert, Vervoort, Sullivan, Verhoeven, & Crombez, 2008), particularly rumination about child pain (Hechler et al., 2011). In the context of acute pain, higher levels of parental catastrophizing about child pain have been associated with higher child procedural fear (Vervoort, Goubert, et al., 2011) and parent ratings of child pain intensity (Birnie et al., 2016; Vervoort, Goubert, et al., 2011). Higher levels of state catastrophizing about child pain by parents of generally healthy schoolchildren were associated with greater parent-child congruence in their reports of children's pain intensity during a laboratory pain task (Goubert, Vervoort, Cano, & Crombez, 2009). Parental trait catastrophizing about child pain was identified as a presurgical predictor of children's chronic postsurgical pain in a recent systematic review and meta-analysis (Rabbitts, Fisher, Rosenbloom, & Palermo, 2017).

In children with chronic pain, higher levels of parental catastrophizing about child pain have been associated with children's clinically elevated depressive symptoms (Goldstein-Leever, Cohen, Dampier, & Sil, 2018), poorer functional disability and school attendance (Goubert et al., 2006; Sinclair, Meredith, Strong, & Feeney, 2016), lower readiness for change regarding pain treatment (Guite et al., 2014), and greater parent-reported child verbal and nonverbal expressions of pain (Lynch-Jordan, Kashikar-Zuck, Szabova, & Goldschneider, 2013). In a sample of children with treatment refractory chronic pain participating in an intensive day hospital program, decreases in child functional disability over the course of the program were associated with decreases in parent catastrophizing about their child's pain (Sieberg et al., 2017). The relationship between parental catastrophizing about child pain and children's functioning was found to be mediated by children's own pain catastrophizing in samples of adolescents with and without chronic pain (Vowles, Cohen, McCracken, & Eccleston, 2010; Wilson, Moss, Palermo, & Fales, 2014). Greater parent catastrophizing about child pain was also associated with negative outcomes for parents, such as increased parental depression, anxiety, and parenting stress associated with parenting a child with chronic illness (Goubert et al., 2006), and greater self-oriented distress when imagining their child in pain (Goubert et al., 2008).

A limited number of studies have examined the role of parents' catastrophizing about their own pain in predicting children's pain-related outcomes. These studies have found that parents' higher levels of catastrophizing about their own pain are associated with children having greater pain intensity one year after major surgery (Pagé, Campbell, Isaac, Stinson, & Katz, 2013) and with children's greater tendency to focus their attention away from pictures of individuals in pain during a cognitive task (Vervoort, Trost, & Van Ryckeghem, 2013). Additionally, the relationship between children's attentional avoidance of pain and their pain tolerance during a laboratory pain task was moderated by parents' rumination about their own pain; for children whose parents reported high levels of rumination, pain tolerance was significantly lower when they attentionally avoided pain as compared to when they attended to it, while this pattern was not observed in children of parents reporting low levels of rumination (Vervoort et al., 2013).

A robust literature supports the association between greater pain catastrophizing (about one's own or one's child's pain) and poorer pain-related outcomes in children and parents as described above. While the relationship between parent catastrophizing about child pain and child outcomes has been frequently studied, child catastrophizing about a parent's chronic pain has not been examined. This novel construct may confer risk to children through similar mechanisms as does child catastrophizing about their own pain, but may be increased in children of parents with chronic pain due to their increased exposure to their parent's pain experiences. The current dissertation addresses this gap in the literature by examining greater child catastrophizing about their parent's chronic pain as a predictor of poorer child pain-related outcomes.

1.4 Parental Responses to Child Pain

Research has examined the impact of parent-child interactions during children's pain experiences on children's pain and coping. In particular, studies have examined the impact of parent verbalizations on child pain and distress during children's medical procedures (e.g., needle procedures). Blount and colleagues (Blount et al., 1997) developed a detailed observational coding system to classify parent and child verbalizations during procedures (Child-Adult Medical Procedure Interaction Scale – Revised; CAMPIS-R). They classified the verbalizations of parents (and other adults present during medical procedures) into Coping Promoting (e.g., humor, talk not focused on the procedure, commands for the child to use coping strategies), Distress Promoting (e.g., reassurance, criticism, apologies), and Neutral (e.g., talk to other adults in the room, discussing the child's general status, managing the child's behaviour) statements. A large body of research shows that parents' use of Distress Promoting behaviours such as reassurance is associated with increased child pain and distress, in both clinical and experimental studies (Blount et al., 1997; Campbell, DiLorenzo, Atkinson, & Riddell, 2017; Chambers, Craig, & Bennett, 2002; Manimala, Blount, & Cohen, 2000; Spagrud et al., 2008). A study employing sequential analysis techniques found that parental reassurance did not predict the start of children's distress behaviours, but maintained existing child distress after surgery (Martin, Chorney, Cohen, & Kain, 2013). A recent systematic review found that combinations of Coping Promoting behaviours by parents are the most consistent predictors of children using adaptive coping responses during needle procedures (Campbell et al., 2017).

Studies have adapted the CAMPIS-R coding system to examine parent verbalizations that focus attention on, and thus positively reinforce, child pain (i.e. Attending Talk) versus those that direct attention away from child pain (i.e. Non-Attending Talk) (Moon, Chambers, & McGrath, 2011; Walker et al., 2006). Parent Attending Talk was associated with increased pain and distress in children (with and without chronic pain) experiencing acute procedural or laboratory pain (Campbell et al., 2017; Evans et al., 2016; Moon et al., 2011; Spagrud et al., 2008; Walker et al., 2006;

Williams, Blount, & Walker, 2011). It has also been found to predict children’s decreased use of adaptive strategies to cope with pain (Blount, Bunke, Cohen, & Forbes, 2001; Spagrud et al., 2008) and children’s increased attending to their pain (Walker et al., 2006). Similar results have been found in studies of parents’ responses to their children’s chronic pain (Pielech, Wallace, Fitzgerald, & Hoffart, 2018; Simons, Claar, & Logan, 2008; Sinclair et al., 2016; Walker, Claar, & Garber, 2002).

Parent and child pain catastrophizing also influence parent responses to child acute and chronic pain. In children with chronic pain, increased parent catastrophizing about child pain predicted increased child functional disability indirectly through parents’ use of more protective (e.g., giving special attention to child, reducing their responsibilities) and avoidant (e.g., canceling activities when the child is in pain) responses to child chronic pain (Simons, Smith, Kaczynski, & Basch, 2015). Parent catastrophizing about child pain was also found to moderate the relationship between parent attending talk and child pain behaviour, with a positive relationship between these variables occurring only in mothers who reported low levels of catastrophizing about their child’s pain (Langer et al., 2017). In parents of children with and without chronic pain, those who reported high levels of catastrophizing about their child’s pain reported greater distress and desire to stop their child’s laboratory pain task (Caes, Vervoort, Eccleston, Vandenhende, & Goubert, 2011). Higher levels of catastrophizing about their child’s pain was associated with increased use of solicitous responses to child pain in both mothers and fathers, and with increased use of distracting responses in fathers only (Hechler et al., 2011). Parent catastrophizing about their own pain was found to predict increased use of protective responses to their child's functional abdominal pain (Langer, Romano, Levy, Walker, & Whitehead, 2009).

Regarding the role of children's catastrophizing about their own pain in predicting parent responses to child pain, one study found that among parents of generally healthy children, higher levels of pain catastrophizing in children were associated with greater use of discouraging responses (e.g., ignorance or criticism of child's pain) in mothers and greater use of solicitous responses (e.g., reinforcing responses to pain such as spending more time with the child, reducing their responsibilities when in pain) in fathers (Vervoort, Huguet, Verhoeven, & Goubert, 2011). During a laboratory pain task completed by children with functional abdominal pain, parent verbalizations focused on child pain symptoms were associated with more child talk about pain symptoms, and this relationship was stronger for children who reported high levels of pain catastrophizing (Williams et al., 2011). In a sample of generally healthy children completing a laboratory pain task, children engaged in a greater proportion of utterances focused on their pain and the pain task when they and their parent both reported high levels of state catastrophizing about the child's pain during the task (Birnie et al., 2016).

Prior research in this area has clearly established parental attending responses to child pain as a predictor of poorer child pain-related outcomes, using both observational and self-report assessment of parent responses and child outcomes. Given the theoretical and empirical importance of parental attending responses to child pain in predicting poorer child outcomes, parental responses to child pain in the context of parental chronic pain are a possible means through which risk for chronic pain and related outcomes is transmitted from parents with chronic pain to their children. The current dissertation builds on previous work by applying rigorous methods (i.e. use of a reliable and valid observational coding system to characterize parent's and children's responses during the child's laboratory pain task) to examine the role of parental attending responses to child pain in predicting child pain-related outcomes in children of parents with chronic pain.

1.5 Aggregation of Pain in Families

Numerous previous studies have found that chronic pain tends to aggregate in families (Chaturvedi & Kanakalatha, 1988; Hoftun, Romundstad, & Rygg, 2013; Mikail & Von Baeyer, 1990; Ramchandani, Stein, Hotopf, & Wiles, 2006; Wilson et al., 2014). Several large, population-based studies have found a positive relationship between parental chronic pain and offspring chronic pain (M Aromaa, Rautava, Sillanpaa, Helenius, & Ojanlatva, 1999; Arruda, Guidetti, Galli, Albuquerque, & Bigal, 2010; Hoftun et al., 2013; O'Sullivan, Straker, Smith, Perry, & Kendall, 2008; Saunders, Korff, Leresche, & Mancl, 2007). Studies have found support for both genetic and environmental explanations of the aggregation of pain in families (Hoftun et al., 2013; Lee & Tracey, 2013; Ståhl, Mikkelsson, Salminen, Pulkkinen, & Rose, 2013). For example, in adolescents who lived primarily with their mothers, a significant relationship between maternal (but not paternal) pain and adolescent pain was observed, while in adolescents who lived primarily with their fathers, both maternal and paternal chronic pain were associated with adolescent chronic pain, with paternal pain having a stronger relationship (Hoftun et al., 2013). Relationships between parental and child pain may also be site- (Saunders et al., 2007) and sex-specific (Evans, Meldrum, Tsao, Fraynt, & Zeltzer, 2010). Rates of parental chronic pain were found to be higher in parents of children with acute or chronic musculoskeletal pain compared to parents of children without musculoskeletal pain (Clementi et al., 2018), and in parents of children with headaches compared to those without headaches (Galli et al., 2017). Studies of children with chronic pain (Anttila, Metsähonkala, Helenius, & Sillanpää, 2000; Beveridge, Neville, Wilson, & Noel, 2018) and adults with a childhood history of chronic pain (Sherman, Bruehl, Smith, & Walker, 2013) found that parental chronic pain was associated with poorer offspring pain-related outcomes. Although many studies have found evidence for an association between parental chronic pain and increased offspring pain, differences between children of parents with and without chronic pain in rates of pain problems were not found in all studies (G T Jones, Silman, & Macfarlane, 2004).

In addition to the aggregation of pain in families, the association between parental chronic pain and other offspring outcomes (e.g., broader health outcomes, mental health) has also been explored. Numerous studies have found higher rates of adverse birth outcomes (e.g., low birthweight, small for gestational age, preterm delivery) in newborns of mothers with chronic pain (H. M. Chen, Chen, Chen, & Lin, 2010; Nørgaard et al., 2010; Wallenius, Salvesen, Daltveit, & Skomsvoll, 2014). While some studies have found that parental chronic pain is associated with poorer offspring psychological health (Beveridge et al., 2018; Kaasbøll, Lydersen, & Indredavik, 2012), other studies have not identified differences between offspring of parents with and without chronic pain on these variables (K. B. Smith & Chambers, 2006). Given the mixed results of studies in this area, a comprehensive, mixed-methods systematic review and synthesis of the literature on the association between parental chronic pain and child pain, health, psychological, and family outcomes was conducted and is presented in Chapter 2.

1.6 Theoretical Basis

Stone and Wilson recently proposed a comprehensive theoretical model of the transmission of risk for chronic pain, disability, and poor psychological functioning from parents with chronic pain to their offspring (Stone & Wilson, 2016). This model proposes several mechanisms through which this transmission might occur: 1) genetics, 2) early neurobiological development, 3) pain-specific social learning, 4) general parenting and family health, and 5) exposure to stressful environment. The model suggests that these mechanisms predict vulnerability factors in children that may increase their risk of developing adverse pain-related outcomes. The hypothesized vulnerability factors in this model are 1) altered pain processing, 2) pain-related cognitions and affect, 3) pain coping behaviours, 4) physical health factors, and 5) difficulty regulating emotions (Stone & Wilson, 2016). The model also proposes several potential moderators of the relationship between parental chronic pain and offspring outcomes, including the presence of chronic pain in a second parent, characteristics of the parent's chronic pain (e.g., when it began, course of symptoms), and demographic characteristics of the child (e.g., sex, stage of development, race). Thus far, this model has been directly tested in one study of adolescents with functional abdominal pain and their parents, some of whom had chronic pain themselves (Stone, Bruehl, Smith, Garber, & Walker, 2018). In a test of pain-specific social learning mechanisms, support was found for a parental modeling mechanism; that is, the relationship between the presence of parental chronic pain and adolescent pain severity and functional disability was explained by adolescents observing parent pain behaviours and developing high threat views of pain (Stone et al., 2018). However, the other pain-specific social learning mechanism tested, parental reinforcement, was not supported; that is, the relationship between parental chronic pain and adolescent pain severity and functional disability was not mediated by parents' solicitous responses to adolescent pain (as measured by parent responses to self-report questionnaires) and adolescent pain threat (Stone et al., 2018). This study suggests that adolescents' observations of parents' pain behaviour play an important role in the experience of pain and disability for adolescents with chronic pain themselves.

Several knowledge gaps exist in this area requiring further research to support or refute the mechanisms described by Stone and Wilson (2016). While initial support has been found for the parental modeling mechanism in predicting increased pain and functional disability in children with chronic pain themselves (Stone et al., 2018), it is unclear whether these results indicate that parental modeling plays a role in the development of children's chronic pain, or the maintenance of pain in children who have already developed chronic pain themselves and for whom pain modeling may be more salient. Additionally, clarification regarding whether the parental modeling mechanism predicts poorer child psychological outcomes, in addition to pain outcomes, is needed. Previous work finding no support for the parental reinforcement of child pain mechanism measured parents' self-reported responses to child pain (Stone et al., 2018), which may be subject to retrospective or social desirability biases. The present dissertation builds on existing work by testing the parental modeling and reinforcement mechanisms hypothesized by Stone and Wilson (2016) using observational methodology in parents with chronic pain and their children.

1.7 Methodological Considerations

The current dissertation used several rigorous methods to examine the relationship between parental pain and offspring outcomes: systematic review and synthesis methods, reliable and valid measures of child pain, and an experimental pain paradigm. This section provides a summary of key methods used, including background information and rationale for their use.

1.7.1 Systematic Review and Synthesis Methods

Systematic review methodology aims to answer specific research questions by identifying and synthesizing all relevant evidence on a particular topic, based on a priori eligibility criteria (J. P. T. Higgins & Green, 2011). This comprehensive methodology involves setting clear objectives and eligibility criteria, conducting a systematic search to identify all relevant research, systematically describing the included studies, and synthesizing the results (J. P. T. Higgins & Green, 2011; Uman, 2011). A systematic review was conducted and described in Chapter 2 in order to identify and synthesize existing information on pain, health, psychological, and family outcomes in offspring of parents with chronic pain to determine whether parental chronic pain was associated with difficulties in each type of outcome. This was necessary because previous studies found mixed results and examined a wide variety of samples (e.g., pain types, offspring ages) and outcomes, making overall conclusions unclear. Meta-analysis was used to synthesize the results of quantitative studies identified in the systematic search. This method statistically combines results from multiple studies to provide a summary statistic and/or assess dispersion of results across studies (Borenstein, Hedges, Higgins, & Rothstein, 2009; J. P. T. Higgins & Green, 2011; Uman, 2011). Systematic review and meta-analysis methodology is more explicit and transparent than narrative reviews, which do not necessarily incorporate systematic searches and may synthesize study results using descriptive summaries or by counting the number of studies finding a particular result (Littell, Corcoran, & Pillai, 2008).

In order to synthesize results of studies of various methodologies, so as not to miss important information, qualitative research was included in the systematic review and synthesis conducted in Chapter 2. Qualitative synthesis is recognized as helpful in adding depth to traditional systematic reviews by incorporating the perspectives of people with lived experience of the topic being studied (Atkins et al., 2008). Synthesis of the results of qualitative studies identified in the systematic search was conducted using meta-ethnography, a well-established method for qualitative synthesis (Britten & Pope, 2012; Noblit & Hare, 1988; Paterson, 2012). This inductive, interpretive method involves translating the findings of included studies into one another in order to develop key concepts or themes across studies, explaining contradictions in results between studies, and providing an overall interpretation and synthesis of the topic studied (Paterson, 2012). Meta-ethnography is completed in seven steps: 1) Getting started (i.e. determining research questions), 2) Determining the scope of the synthesis, 3) Reading the included studies, 4) Determining how the studies are related to one another, 5) Translating the studies into one another, 6) Synthesizing the translations, and 7) Expressing the synthesis (i.e. writing up the results; Atkins et al., 2008; Noblit & Hare, 1988). Meta-ethnography was chosen as the qualitative synthesis method for Chapter 2 given its focus on developing higher order interpretations of the results of primary research, its interpretive basis which is consistent with most qualitative research methodologies, and its suitability for synthesizing homogeneous primary studies (Atkins et al., 2008; Britten, Campbell, & Pope, 2002; Paterson, 2012).

1.7.2 Measurement of Pediatric Pain

Pain is a subjective experience, which makes it difficult to assess in another person (Task Force on Taxonomy of the International Association for the Study of Pain, 2002). While research has generally identified parents as more accurate assessors of their children's pain than physicians and nurses (Brudvik, Moutte, Baste, & Morken, 2017; Hla et al., 2014), parents tend to underestimate their children's pain (Boerner, Chambers, Craig, Pillai, & Parker, 2013; Brudvik et al., 2017; Chambers, Reid, Craig, McGrath, & Finley, 1998). Multiple factors may influence parents' assessment of their children's pain, including both the parent's and child's levels of pain catastrophizing (Birnie et al., 2016; Goubert et al., 2009) and the parent's affect regulation (Loopstra, Strodl, & Herd, 2015). Research suggests that child self-report of pain intensity is a primary source of pain assessment information that should be obtained whenever possible, and should be used in combination with other relevant factors (e.g., context, developmental stage, psychological factors) to make pain management decisions for individual children (Schiavenato & Craig, 2010; Twycross, Voepel-Lewis, Vincent, Franck, & Von Baeyer, 2015; von Baeyer, 2009). The current dissertation has incorporated multiple sources of child pain assessment, in line with this research, including self- and parent-report of child pain intensity and behavioural indicators of child pain (i.e. pain tolerance during the cold pressor task).

Much research has examined appropriate means for assessing pain in children, who may have difficulty reliably reporting their pain depending on their developmental level (Cohen et al., 2008). Numerous self-report measures of pain intensity for children have been developed and psychometrically evaluated (Stinson, Kavanagh, Yamada, Gill, & Stevens, 2006; Tomlinson, von Baeyer, Stinson, & Sung, 2010). The Faces Pain Scale-Revised (FPS-R; Hicks, von Baeyer, Spafford, van Korlaar, & Goodenough, 2001) is one such measure that has strong psychometrics and is considered well-established for use in children ages 4-16 years (Cohen et al., 2008). It consists of six line drawings of gender neutral faces depicting different amounts of pain expression, anchored from “no pain” (scored 0 points) to “very much pain” (scored 10 points; Hicks et al., 2001). Importantly, this scale is not anchored with a smiling face, which in other faces scales for pain has been found to confound pain and affect (Chambers, Giesbrecht, Craig, Bennett, & Huntsman, 1999). In the current dissertation, parents reported on their children’s pain intensity during the cold pressor task using the FPS-R. This measure was chosen in order to allow for a common metric between both informants’ ratings of the child’s pain intensity, consistent with previous research (Birnie et al., 2016; Boerner et al., 2013; Spagrud et al., 2008). Evidence of the validity of this measure in adults has also been found (Ferreira-Valente, Pais-Ribeiro, & Jensen, 2011).

1.7.3 Use of Experimental Pain Paradigm in Children

Experimental pain paradigms are used to study topics that would not be feasible to study clinically and to allow for additional rigor which would not be possible in clinical settings (Birnie, Caes, Wilson, Williams, & Chambers, 2014). The most common experimental pain paradigm used in children is the cold pressor task (CPT; Birnie et al., 2014). This task involves the child submerging their hand in a bath of cold water, typically 10°C for 3-4 minutes, with the option to remove their hand whenever they choose (Birnie, Petter, Boerner, Noel, & Chambers, 2012). The task is considered to be ethically acceptable given that the potential risks of pain induction are balanced by children's control over the task, slow onset of pain, low rates of adverse events, and findings suggesting that parents and children have positive experiences with the task (Birnie, Noel, Chambers, Von Baeyer, & Fernandez, 2011; von Baeyer, Piira, Chambers, Trapanotto, & Zeltzer, 2005). The CPT is believed to be most representative of children's real-world experiences of acute pain, rather than chronic pain (von Baeyer et al., 2005), although some aspects of the task may differ from clinical acute pain (e.g., lower child anxiety levels before beginning the task; Birnie et al., 2014). It is appropriate for studying aspects of children's pain experiences that require several minutes to observe, such as interactions between parents and children during child pain. In one study where children completed the CPT in the presence of a parent, children and parents rated each other's behaviour during the task as being "a little different" than how they normally respond to the child's pain (Birnie et al., 2016). Studies of parent-child interactions during child pain have found similar results in studies of children's procedural pain (e.g., needle procedures) and children's pain during the CPT (Campbell et al., 2017; Evans et al., 2016; Moon et al., 2011; Spagrud et al., 2008; Walker et al., 2006; Williams et al., 2011), supporting the ecological validity of the CPT.

1.8 Introduction to Dissertation Papers

The present dissertation aimed to synthesize existing literature on outcomes (pain, health, psychological, family) in offspring of parents with chronic pain, and to examine theorized pathways through which parental pain might impact children. These goals were accomplished by conducting a systematic review and synthesis (using meta-analysis and meta-ethnography methodologies) and by conducting a laboratory-based study with a sample of parents with chronic pain and their children. The results of these studies are presented in three separate papers (Chapters 2-4). Chapter 5 presents discussion of the overall results of the dissertation, integration with previous work, theoretical and clinical implications, and study strengths and limitations.

The first goal was accomplished by conducting a comprehensive, mixed methods review and synthesis of the literature on offspring of parents with chronic pain (Chapter 2). This review included studies of a variety of designs (population-based, clinical, and qualitative studies) in order to examine the full range of empirical research available on this topic. A systematic search was conducted to identify papers published in English that examined the impact of parental chronic pain on offspring of any age. Data from studies with appropriate quantitative information provided were pooled based on offspring outcome (pain, health, psychological, or family outcomes) and study types, and this information was meta-analyzed. Additionally, information from identified qualitative studies was synthesized using meta-ethnography. Studies examining the relationship between parental chronic pain and offspring outcomes had previously found mixed results across a variety of samples and outcomes (e.g., Hoftun et al., 2013; Kaasbøll, Lydersen, & Indredavik, 2012; Smith & Chambers, 2006). The review and synthesis were conducted in order to clarify the relationship between parental chronic pain and several categories of offspring outcomes while including a wide range of study types, which may provide different information and perspectives on this topic.

The goal of examining theorized pathways through which parental chronic pain might impact offspring was addressed by conducting a laboratory-based study. Parents with chronic pain lasting for at least six months were recruited from a local adult chronic pain management clinic and using community methods. Seventy-two eligible parents and their children (ages 8-15 years) attended one laboratory visit in which they answered questionnaires about pain and mental health, and the child participated in the CPT in the presence of their parent. Parents and children were videotaped while the child completed the CPT, and verbalizations made were coded using an established observational coding system (Blount et al., 1997; Moon et al., 2011). Parents and children rated the child's pain intensity, and children's pain tolerance was recorded. The study was designed to incorporate both self-report and observational methodologies, as well as multiple informants (child and parent) for the measurement of a variety of variables in order to improve upon past work in this area. The results of two distinct sets of research questions examined in this study are presented in Chapters 3 and 4.

Chapter 3 describes the examination of potential social transmission mechanisms through which parental chronic pain may impact children as hypothesized in Stone and Wilson's model of intergenerational transmission of risk (Stone & Wilson, 2016). It was expected that parental chronic pain would predict children's pain and mental health outcomes through two such mechanisms; 1) parental pain modeling, in which parents' higher levels of pain interference would predict children's higher levels of pain catastrophizing, which would in turn predict the presence of child recurrent pain and greater internalizing symptoms (as reported on by both children and parents), and 2) parental pain reinforcement, in which parents' greater proportion of pain-attending verbalizations during the cold pressor task would predict children's pain outcomes (greater intensity and lower tolerance) through the mediator of children's own higher levels of pain-attending verbalizations. Support has been found for the parental modeling mechanism, but not the parental reinforcement mechanism, in children with functional abdominal pain and their parents (Stone et al., 2018); however, it is possible that these mechanisms may differ in children who do not necessarily have chronic pain themselves. For example, parental pain modeling may be more salient to children who already have their own chronic pain.

In Chapter 4, the novel construct of children's catastrophizing about their parent's pain is examined. Catastrophizing about one's own pain is a robust predictor of pain and mental health outcomes in children and adults with and without chronic pain (Birnie et al., 2016; Craner et al., 2017; Crombez et al., 2003; Sullivan, Tripp, & Santor, 2000), and parents' catastrophizing about their children's pain is predictive of negative outcomes for both parents and children (Goubert et al., 2006, 2008; Wilson et al., 2014). However, previous research has not examined children's catastrophizing about their parent's chronic pain as a potential risk factor for pain and mental health outcomes in children of parents with chronic pain. This construct may confer risk through similar mechanisms as catastrophizing about one's own pain, but may be particularly salient in children of parents with chronic pain because of their increased exposure to parent pain. It was hypothesized that 1) parents' and children's catastrophizing about their own pain would predict their catastrophizing about one another's pain, and 2) children's greater levels of catastrophizing about their parents' pain would predict greater child pain (over the past three months and in the context of the cold pressor task) and internalizing symptoms, beyond the influence of catastrophizing about one's own pain. These hypotheses were tested using the Actor-Partner Interdependence Model (Kenny, Kashy, & Cook, 2006), a form of dyadic analysis which allowed for taking into account the relationships between parents’ and children’s catastrophizing about their own and each other’s pain and each person’s outcomes.

CHAPTER 2: OFFSPRING OF PARENTS WITH CHRONIC PAIN: A SYSTEMATIC REVIEW AND META-ANALYSIS OF PAIN, HEALTH, PSYCHOLOGICAL, AND FAMILY OUTCOMES

The manuscript prepared for this study is presented below. Kristen Higgins, under the supervision of Dr. Christine Chambers, was responsible for developing the research questions, search strategy (in consultation with library services), and data extraction procedures. She completed the systematic search, screening, and data extraction processes. She was the lead on the data analysis and wrote the current manuscript. Co-authors reviewed the manuscript and their feedback was incorporated prior to submission. The manuscript was peer-reviewed and Ms. Higgins led the relevant revisions. The manuscript was published in PAIN. The full reference for this manuscript is:

Higgins, K. S., Birnie, K. A., Chambers, C. T., Wilson, A. C., Caes, L., Clark, A. J., Lynch, M., Stinson, J., & Campbell-Yeo, M. (2015). Offspring of parents with chronic pain: A systematic review and meta-analysis of pain, health, psychological, and family outcomes. *PAIN*, *156*, 2256-2266. doi: 10.1097/j.pain.0000000000000293

2.1 Abstract

Offspring of parents with chronic pain may be at risk for poorer outcomes than offspring of healthy parents. The objective of this research was to provide a comprehensive mixed-methods, systematic synthesis of all available research on outcomes in offspring of parents with chronic pain. A systematic search was conducted for published articles in English examining pain, health, psychological, or family outcomes in offspring of parents with chronic pain. Fifty-nine eligible articles were identified (31 population-based, 25 clinical, 3 qualitative), including offspring from birth to adulthood and parents with varying chronic pain diagnoses (e.g., mixed pain samples, arthritis). Meta-analysis was used to synthesize the results from population-based and clinical studies, while meta-ethnography was used to synthesize the results of qualitative studies. Increased pain complaints were found in offspring of mothers and of fathers with chronic pain, and when both parents had chronic pain. Newborns of mothers with chronic pain were more likely to have adverse birth outcomes, including low birthweight, preterm delivery, caesarean section, intensive care admission, and mortality. Offspring of parents with chronic pain had greater externalizing and internalizing problems and poorer social competence and family outcomes. No significant differences were found on teacher-reported externalizing problems. The meta-ethnography identified six key concepts (developing independence, developing compassion, learning about health and coping, missing out, emotional health, and struggles communicating with parents). Across study designs, offspring of parents with chronic pain had poorer outcomes than other offspring, although the meta-ethnography noted some constructive impact of having a parent with chronic pain.

Keywords: chronic pain, parents, offspring, children, systematic review, meta-analysis, meta-ethnography

2.2 Introduction

Chronic pain tends to aggregate in families (Hoftun et al., 2013; Saunders et al., 2007), and vulnerability to chronic pain is thought to be the result of complex interactions between both environment and genetics (Hoftun et al., 2013; Kato, Sullivan, Evengård, & Pedersen, 2006; Lee & Tracey, 2013; Ståhl et al., 2013; Young, Lariviere, & Belfer, 2012). It is known that parental mental and physical health conditions generally put offspring at risk for poorer outcomes. For example, children of mothers with depression are more likely to experience depression and other mental health problems compared to children of mothers without depression (Cho, Kim, Lim, Lee, & Shin, 2015; Goodman & Gotlib, 1999; Matijasevich et al., 2014; Sanger, Iles, Andrew, & Ramchandani, 2014), and children of parents with cancer have higher levels of anxiety, depression, and behaviour problems than children of parents without cancer (Siegel et al., 1992). Fewer studies have been conducted to examine the effects of parental chronic pain on their offspring.

Previous studies that have examined offspring of parents with chronic pain have studied the effects of parental chronic pain on the offspring's own pain complaints (Hoftun et al., 2013), other aspects of their physical health (Lin, Chen, Lin, & Chen, 2010; Logan, Riley, & Barker, 2008), their psychological health (Kaasbøll et al., 2012), and variables related to family relationships (K. B. Smith & Chambers, 2006). While several studies have found that offspring of parents with chronic pain have poorer pain (Hoftun et al., 2013), health (Lin et al., 2010), and psychological (Kaasbøll et al., 2012) outcomes than other offspring, other studies have found no between-group differences on pain complaints (G T Jones et al., 2004), and psychological and family outcomes (K. B.

Smith & Chambers, 2006). Additionally, few studies have explored possible differences in the effects of maternal versus paternal chronic pain on offspring, although at least one study has found such differences (Hoftun et al., 2013), and other research has explored differences in mothers' and fathers' interactions with children during child pain (Goubert et al., 2008; Moon et al., 2011). These conflicting results highlight the need for a comprehensive review of the literature including studies conducted in various disciplines looking at a variety of outcomes in offspring of parents with chronic pain.

One prior narrative review described 18 studies of children of parents with chronic pain (Umberger, 2014). The review concluded that children and adolescents whose parents have chronic pain are at risk for more pain complaints (particularly those similar to their parents), and more internalizing and externalizing symptoms than children and adolescents of parents without chronic pain. However, the previous review (Umberger, 2014) contained several critical limitations, including an overly narrow search strategy, lack of meta-analysis, and lack of inclusion of population-based and qualitative studies. It is important to conduct a mixed methods review of the literature on offspring of parents with chronic pain in order to better understand the conflicting results found in the literature. In particular, qualitative research may offer a different perspective on this topic, as participants may address topics that researchers had not previously considered. Better understanding the effects of parental chronic pain on offspring may identify opportunities to provide interventions to improve outcomes in offspring of parents with chronic pain.

The aim of the present study was to improve on previous work by conducting a rigorous mixed-methods systematic review, employing both meta-analysis and qualitative synthesis of the full range of empirical research (population-based, clinical, and qualitative studies) examining the relationship between parental chronic pain and pain, health, psychological, and family outcomes in their offspring.

2.3 Method

2.3.1 Search Strategy

The electronic databases searched included PubMed, PsycINFO, CINAHL, and EMBASE. PubMed was chosen over Web of Science as it has been shown to be an optimal biomedical database (Falagas, Pitsouni, Malietzis, & Pappas, 2008). Databases were searched from inception to August 2014. The search strategy included combinations of terms for parent (e.g., parent, mother, father, caregiver, family), pain (e.g., pain, arthritis, fibromyalgia, headache, neuropathy), child (e.g., infant, child, adolescent, young adult), and association (e.g., risk, influence, predictor, association), and was developed in consultation with a librarian specializing in systematic searches. The particular pain conditions included in the search strategy were decided upon through consensus with expert adult and pediatric pain researchers and clinicians. Medical Subject Headings (MeSH) terms and other key terms were included where possible; text terms were searched in all databases for entries including these terms in the title or abstract. See Appendix A for the complete search strategy for each database. Additional hand searching of previous relevant research was also conducted.

2.3.2 Eligibility Criteria

Eligible studies were empirical studies (population-based, clinical, or qualitative) examining the relationship between parental pain and offspring pain, health, psychological, or family outcomes. Studies had to include measures of offspring outcomes (e.g., not just measures of the parent's parenting abilities). Studies including offspring of any age from infancy to adulthood were eligible. Only published articles written in English were included, given that research suggests that this does not significantly affect meta-analysis results (Moher et al., 2000). Included studies had to have parent(s) as the identified pain patient(s) in their samples (rather than offspring).

2.3.3 Study Selection and Data Extraction

Study selection and data extraction were completed following accepted standards for systematic review procedures (J. P. T. Higgins & Green, 2011; Kisely et al., 2015; Uman, 2011). The titles of all of the citations generated in the systematic search were reviewed for relevance by the first author (KH). For titles that were identified as relevant, abstracts were retrieved and reviewed to determine whether they met the review inclusion criteria. For any studies where there was uncertainty about exclusion, input was sought from a co-author (KB). Full articles identified for inclusion were retrieved and coded using data extraction sheets designed for the present review. Data extraction sheets were developed by the first author (KH) with consultation from co-authors. Characteristics of the parent and offspring samples and outcome measures were recorded, and data was extracted for all outcomes measured in each study by the first author (KH). A subset of approximately 20% of identified studies was coded by an additional coder and disagreements were solved with consensus. The outcomes measured in the population and clinical studies were grouped into four core domains (pain, health, psychological, family) and other outcomes to assist in the analysis of the wide variety of outcomes measured across studies. For continuous variables, means and standard deviations were extracted from each study; for dichotomous variables, odds ratios with 95% confidence intervals or frequencies and group sizes were recorded.

2.3.4 Meta-Analysis

Meta-analyses were completed following accepted procedures for using this method (J. P. T. Higgins & Green, 2011; Lipsey & Wilson, 2001). Outcome data extracted from population and clinical studies were grouped by domain (pain, health, psychological, family, other). Within each core domain, similar constructs were grouped together for meta-analysis. At least two studies measuring the same outcome were required in order to conduct a meta-analysis on the outcome. Studies were only combined in meta-analyses if their measurement of the outcome was similar enough that the combining of data in a meta-analysis would result in an interpretable construct (J. P. T. Higgins & Green, 2011). Psychological outcomes were grouped into internalizing problems (including scales measuring general internalizing symptoms and symptoms of depression, anxiety, and obsessive-compulsive disorders) and externalizing problems (including scales measuring general externalizing problems, child behaviour problems, delinquency and hyperactivity). Given the lack of agreement between measures completed by different informants in previous studies (Achenbach, McConaughy, & Howell, 1987; Rescorla et al., 2013), outcomes were additionally grouped based on the source reporting on the measure (parent-, child-, and teacher-reported outcomes). Analyses were conducted using RevMan 5.3 software. Random effects analyses were used for all analyses, as this statistic is appropriate for meta-analyses in which the true effect size is thought to differ across studies (J. P. T. Higgins & Green, 2011). This is the case in the present review given the variety of parental pain conditions studied and the variety of measures used for each outcome. In order to perform a meta-analysis for a particular outcome, at least two studies providing appropriate data on the outcome were required. Heterogeneity amongst studies was measured using the I^2 statistic, and was interpreted using the following recommended guidelines: 0%-40% indicates that heterogeneity might not be important, 30%-60% may represent moderate heterogeneity, 50%-90% may represent substantial heterogeneity, 75%-100% represents considerable heterogeneity (J. P. T. Higgins & Green, 2011).

Continuous outcomes were analyzed using standardized mean difference. This is considered an appropriate statistical test for meta-analyses in which outcomes are measured on different scales across studies and when comparing mean scores across groups which are not randomly assigned (J. P. T. Higgins & Green, 2011; Lipsey & Wilson, 2001), which is the case in the present review. Data were entered in the analysis such that higher scores for each variable were indicative of greater levels of problems. Consequently, a positive standardized mean difference with both limits of the confidence interval being positive indicates that children in the parental chronic pain group had higher scores (poorer functioning) for the outcome compared to children of control parents. When the same outcome was rated by both the offspring's mother and father, data were pooled using the following formulas: mean = [(mean1*N1)+(mean2*N2)]/(N1+N2) and pooled SD = square root of [($SD1^2$(N1-1)+$SD2^2$(N2-1))/(N1+N2-2)].

For dichotomous data, odds ratios were calculated using random effects generic inverse variance analyses. This type of analysis weights study effect estimates using the inverse variance of the effect estimate, which allows effect sizes to be combined across studies in which frequencies and group sizes were not reported (J. P. T. Higgins & Green, 2011). Given that many studies reported odds ratios and 95% confidence intervals, but did not provide the frequency data necessary to calculate them, this analysis was considered most appropriate. In studies where odds ratios were not reported, frequency data were used to calculate odds ratios in RevMan to be included in the generic inverse variance analyses. Subgroup analyses were conducted using a chi-square test in RevMan 5.3 software.

2.3.5 Qualitative Synthesis

Qualitative studies identified in the systematic search were synthesized using meta-ethnographic techniques. Meta-ethnography is a well-accepted form of qualitative synthesis that involves translating the results of primary studies into one another, in order for overarching themes or concepts to be generated across studies (Britten et al., 2002; Noblit & Hare, 1988; Paterson, 2012). This process involves seven steps: 1) Getting started (determining research questions); 2) Deciding what is relevant to the initial interest (determining the scope of the synthesis); 3) Reading the studies; 4) Determining how the studies are related; 5) Translating the studies into one another; 6) Synthesizing translations; and 7) Expressing the synthesis (Noblit & Hare, 1988). These steps were followed for the qualitative studies identified in the search. Each study was read three times and the key concepts identified in each study were recorded independently by two coders, with disagreements resolved through consensus. Key concepts were reviewed across studies to determine those that commonly occurred across studies. Upon identification of these concepts, a table was created describing each concept in each primary study (“translating” studies into one another). Translations were synthesized by reading each study and concept definition and describing the relationship between the studies based on the concepts. The synthesis is expressed in the results section of the present review. These methods are consistent with suggested methods for using meta-ethnographic techniques (Atkins et al., 2008; Britten et al., 2002; Britten & Pope, 2012).

2.4 Results

2.4.1 Search Results

The screening process is illustrated in a flow diagram in following the PRISMA model (Moher, Liberati, Tetzlaff, & Altman, 2009) in Figure 2.8.1. The systematic search identified 16,450 unique citations after removal of duplicates. Titles of all identified articles were screened for relevance to the topic, resulting in 117 articles for possible inclusion. Abstracts of the 117 articles were reviewed for meeting the eligibility criteria. From this, 59 articles were identified for inclusion. Abstracts were excluded for several reasons, including not published articles (e.g., conference abstracts; *n* = 19), not examining parental pain (*n* = 12), focusing on offspring with chronic pain rather than parents with chronic pain (*n* = 9), not measuring offspring outcomes (*n* = 8), not empirical studies (e.g., topical reviews; *n* = 5), not written in English (*n* = 4). Additionally, one study was excluded because it included case data from only one offspring of a parent with chronic pain. The 59 studies identified for inclusion fell into three categories. Two categories utilized quantitative methodology: population-based studies (studies in which outcomes were measured quantitatively and a population-based sample was used; that is, participants were not specifically recruited to be part of a parental pain group or control group) and clinical studies (studies in which outcomes were measured quantitatively and a parental pain group was specifically recruited; a control group may or may not have been recruited). The third category of identified studies was qualitative studies (studies conducting qualitative analyses; e.g., grounded theory analysis). Thirty-one population-based studies, 25 clinical studies, and three qualitative studies were included in the review. Table 2.9.1 shows the studies included in the systematic review, along with the parental pain samples used and types of outcomes measured.

Population-based studies. Thirty population studies reported on a total of 4,626,806 offspring (sample size not reported in (Holmes & Deb, 2003)). Fourteen (45.16%) examined the relationship between maternal chronic pain and offspring outcomes, while the remaining 17 studies (54.84%) examined pain in either parent in relation to offspring outcomes. Population studies examined mixed forms of parental chronic pain (*n* = 13, 41.94%) and parental arthritis (*n* = 6, 19.35%), headaches (*n* = 6, 19.35%), migraines (*n* = 4, 12.90%), and back pain (*n* = 2, 6.45%). The offspring examined varied in age across studies, including newborns (defined as ages 1-30 days; *n* = 10, 32.26%), infants (1-23 months; *n* = 3, 9.68%), preschoolers (ages 2-5 years; *n* = 4, 12.90%), children (ages 6-12 years; *n* = 10, 32.36%), adolescents (ages 13-18 years; *n* = 13, 41.94%), and adult offspring (> 18 years; *n* = 5, 16.13%).

Clinical studies. Twenty-three clinical studies reported on a total of 1,610 offspring of parents with chronic pain (sample size not reported in (Raphael, Dohrenwend, & Marbach, 1990; R. Smith, 1998)). Eighteen (72.00%) included control groups, with a total of 1,305 offspring of parents without chronic pain being included. Seven (28.00%) clinical studies examined offspring of mothers with chronic pain while 17 (68.00%) examined offspring of either parent with chronic pain and one (4.00%)

examined offspring of fathers with chronic pain. Similarly to the population studies, clinical studies (*n* = 25) included mixed parent pain samples (*n* = 9, 36.00%), as well as parent samples with arthritis (*n* = 5, 20.00%), fibromyalgia (*n* = 3, 12.00%), headaches (*n* = 2, 8.00%), irritable bowel syndrome (*n* = 2, 8.00%), migraines (*n* = 1, 4.00%), inflammatory bowel disease (*n* = 1, 4.00%), back pain (*n* = 1, 4.00%), and temporomandibular pain (*n* = 1, 4.00%). These studies also included offspring of a range of ages, including newborns (*n* = 1, 4.00%), infants (*n* = 3, 12.00%), preschoolers (*n* = 5, 20.00%), children (*n* = 16, 64.00%), adolescents (*n* = 14, 56.00%), and adult offspring (*n* = 7, 28.00%). One study did not report on the age of included offspring (Raphael et al., 1990).

Qualitative studies. Three qualitative articles describing two participant samples were identified in the systematic search. One study (Evans & de Souza, 2008) interviewed 21 children (ages 6-12) of mothers with various chronic pain diagnoses regarding their understanding of their mothers' pain and health, their feelings about their mothers' pain, and their own health. The remaining two studies described the same sample of participants (Umberger et al., 2013, 2014). This sample comprised 30 young adults (18-21 years) with either a mother or father with a variety of chronic pain disorders. Participants were interviewed and asked to reflect on their experiences as an adolescent growing up with a parent with chronic pain, including the strategies they used to manage these experiences.

2.4.2 Meta-Analysis

Pain outcomes.

Population-based studies. Sixteen population studies (M Aromaa et al., 1999; Arruda et al., 2010; Assadi, Zerafati, Velazquez-rodriquez, Contreras, & Leone, 2013; Borge & Nordhagen, 2000; Deubner, 1977; Hasvold & Johnsen, 1996; Hoftun et al., 2013; G T Jones et al., 2004; Koutantji, Pearce, & Oakley, 1998; Kovacs et al., 2003; O'Sullivan et al., 2008; Ramchandani et al., 2006; Saunders et al., 2007; Thomas, Roy, & Cook, 1992; Wilson et al., 2014; Wolff et al., 2009) examined offspring pain outcomes, although not all provided data sufficient for inclusion in the meta-analysis. Meta-analysis of six studies (Borge & Nordhagen, 2000; Deubner, 1977; Hasvold & Johnsen, 1996; Hoftun et al., 2013; G T Jones et al., 2004; Saunders et al., 2007) (N = 9965 offspring) providing appropriate data for meta-analysis found that offspring of mothers with chronic pain were more likely to report pain complaints compared to offspring of control mothers (OR = 1.59, 95% CI [1.37, 1.85], Z = 6.00, $p < 0.00001$, I^2 = 44%). The same pattern was revealed in offspring-reported pain complaints in five studies (Borge & Nordhagen, 2000; Deubner, 1977; Hasvold & Johnsen, 1996; Hoftun et al., 2013; G T Jones et al., 2004) (N = 6453) of offspring of fathers with chronic pain (OR = 1.30, 95% CI [1.08, 1.57], Z = 2.77, p = 0.006, I^2 = 30%) and in four studies (Borge & Nordhagen, 2000; Deubner, 1977; Hoftun et al., 2013; G T Jones et al., 2004) (N = 2380) of offspring with both parents having chronic pain (OR = 1.61, 95% CI [1.29, 2.01], Z = 4.22, $p < .0001$, I^2 = 0%). Additionally, a similar pattern was found in five studies (Deubner, 1977; Hasvold & Johnsen, 1996; Hoftun et al., 2013; G T Jones et al., 2004; O'Sullivan et al., 2008) (N = 7034) that examined offspring with any one parent having chronic pain (regardless of sex) compared to control offspring (OR = 1.59, 95% CI [1.32, 1.92], Z = 4.90, $p <$ 0.00001, I^2 = 57%). A test of subgroup differences revealed no significant differences in odds of pain complaints between offspring of mothers versus fathers with chronic pain ($X^2 = 2.60$, $p = 0.11$, $I^2 = 61.5\%$).

The remaining population studies measuring offspring pain outcomes could not be combined for meta-analysis. One study examining mother-reported offspring pain complaints found that offspring of mothers with chronic daily headaches were more likely to have chronic daily headaches compared to offspring of mothers with no lifetime history of headaches (Arruda et al., 2010). The remaining studies used dissimilar parental pain or control definitions and could not be combined with the other studies (Assadi et al., 2013; Koutantji et al., 1998; Ramchandani et al., 2006), did not report associations between parental pain and offspring pain despite measuring these variables (M Aromaa et al., 1999; Kovacs et al., 2003; Wolff et al., 2009), or reported data in a format that could not be combined with the other meta-analyzed studies (e.g., regression coefficients that could not be combined with the OR data in the generic inverse variance analysis)(Thomas et al., 1992; Wilson et al., 2014).

Clinical studies. Thirteen clinical studies reported on various offspring pain outcomes that could not be appropriately grouped for meta-analysis. These studies found significant between-group differences in the outcomes they measured, including offspring-reported number of pain sites (Evans & Keenan, 2007), teacher-reported presence of offspring pain complaints (Evans, Keenan, & Shipton, 2007), parent-reported frequency of stomachaches or abdominal pain (Jamison & Walker, 1992), number of tender points determined using dolorimetry assessment (Dan Buskila & Neumann, 1997), fibromyalgia diagnosis (Saito et al., 2010), self-reported presence of headaches (Messinger, Spierings, Vincent, & Lebbink, 1991), mean frequency of headaches (Mikail

& Von Baeyer, 1990), and pain-related responses to hypothetical scenarios (Rickard, 1988). Two studies (Aguas et al., 2011; Hudson, Arnold, Keck, Auchenbach, & Pope, 2004) reported on pain outcome analyses in groups of relatives including offspring, but did not report the offspring results separately.

The remaining three clinical studies examined relationships between parental pain and offspring pain outcomes but did not include control groups of unrelated healthy parents and their offspring. One study (D. Buskila, Neumann, Hazanov, & Carmi, 1996) found a prevalence rate of fibromyalgia of 28% in offspring of parents with fibromyalgia (reported as higher than the prevalence in the general population), while another (Kalantar, Locke III, Zinsmeister, Beighley, & Talley, 2003) found prevalence rates of irritable bowel syndrome of 12%-20% in offspring of parents with irritable bowel syndrome (reported as comparable with the general population prevalence). A third study (Chaturvedi & Kanakalatha, 1988) found that 16.67% of their sample of offspring of parents with chronic pain had pain complaints themselves.

Health outcomes.

Population-based studies. Eight population studies (Bánhidy, Ács, Horváth-Puhó, & Czeizel, 2007; Blair & Nelson, 2011; H. M. Chen et al., 2010; J. S. Chen, Ford, Roberts, Simpson, & March, 2013; Lin et al., 2010; Nørgaard et al., 2010; Wallenius et al., 2011, 2014) examined birth outcomes in newborns of women with chronic pain conditions and provided appropriate data for meta-analysis. Results of these meta-analyses are shown in Table 2.9.2. Significant group differences were found for low birthweight, being small for gestational age, preterm delivery, caesarian section, perinatal mortality or stillbirth, and admission to neonatal intensive care unit (NICU), with newborns of mothers with chronic pain being more likely to experience these adverse birth outcomes. Significant group differences were not found for Apgar score below seven at five minutes after birth, congenital abnormalities, and instrument assisted delivery.

Four population studies examined other health outcomes that could not be appropriately combined for meta-analysis. Two studies found that children of parents with chronic pain had increased self-reported medication use for headache (Andersen, Holstein, Berntsson, & Hansen, 2012) and increased odds of emergency department visits and hospitalizations (Logan et al., 2008) compared to control children. Two other studies found no significant group differences when measuring author-defined newborn risk categories (Minna Aromaa, Rautava, Helenius, & Sillanpää, 1996) and parent-reported use of over-the-counter analgesics by children (Jensen et al., 2014).

Clinical studies. Four clinical studies examined measures of offspring general health (Evans & Keenan, 2007; Evans et al., 2007; Evans, Shipton, & Keenan, 2006) and were combined within meta-analyses. Significant group differences were found for offspring-reported (*SMD* = 0.79, 95% CI [0.38, 1.19], $Z = 3.80$, $p = 0.0001$, $I^2 = 44\%$; $N = 241$) (Evans & Keenan, 2007; Evans et al., 2007, 2006) and parent-reported (*SMD* = 0.51, 95% CI [0.18, 0.84], Z = 3.05, $p = 0.002$, $I^2 = 0\%$; $N = 153$) (Dura & Beck, 1988; Evans & Keenan, 2007; Evans et al., 2007) general health scales, with offspring of parents with chronic pain having poorer scores on these measures. Two clinical studies (Mikail & Von Baeyer, 1990; Turner Cobb, Steptoe, Perry, & Axford, 1998) ($N = 132$) examined parent-reported questionnaire measures of somatization, and meta-analysis revealed no significant group differences (*SMD* = 0.30, 95% CI [-0.10, 0.69], $Z = 1.46$, *p*

= 0.15, $I^2 = 18\%$). Other clinical studies found that parental pain groups had offspring with lower birthweight (Bowden, Barrett, Fallow, & Silman, 2001), increased medication use for gastrointestinal symptoms (Jamison & Walker, 1992), and more illnesses and injuries (Raphael et al., 1990) compared to control groups, but found no differences regarding daily physical activity or sleep habits (Turner Cobb et al., 1998).

Psychological outcomes.

Population-based studies. Three population studies examined offspring psychological outcomes but could not be appropriately combined for meta-analysis. One study (Kaasbøll et al., 2012) found that adolescents with both parents having chronic pain had increased risk of experiencing anxiety and depressive symptoms than adolescents of control parents, while this was not seen in children with only one parent with chronic pain. They also found an increased risk of conduct problems in girls whose mothers had chronic pain. Another study (Arruda & Bigal, 2012) found that maternal headache status predicted clinical-range scores on parent-reported general behaviour measures in children without migraines, but not in children with migraines. A third study (Holmes & Deb, 2003) did not report on offspring psychological outcomes for parental pain groups separately from other parental condition groups (e.g., cancer, mental health diagnoses).

Clinical studies. Fourteen clinical studies reported on offspring psychological outcomes. The combined results of seven studies (Chun, Turner, & Romano, 1993; Dura & Beck, 1988; Evans & Keenan, 2007; Evans et al., 2007, 2006; Mikail & Von Baeyer, 1990; K. B. Smith & Chambers, 2006) ($N = 502$) revealed that offspring of parents with chronic pain scored higher on parent-rated measures of externalizing problems compared to control offspring ($SMD = 0.58$, 95% CI [0.31, 0.85], $Z = 4.23$, $p < 0.0001$, $I^2 = 47\%$).

However, two studies (Chun et al., 1993; Evans et al., 2007) ($N = 106$) measuring teacher-reported child externalizing problems revealed no difference between groups on this variable ($SMD = 0.63$, 95% CI [-0.12, 1.37], $Z = 1.64$, $p = 0.10$, $I^2 = 71\%$). Meta-analysis of five studies (Evans & Keenan, 2007; Evans et al., 2007, 2006; Mikail & Von Baeyer, 1990; K. B. Smith & Chambers, 2006) ($N = 428$) found increased levels of parent-reported internalizing problems in offspring of parents with chronic pain ($SMD = 0.60$, 95% CI [0.39, 0.81], $Z = 5.62$, $p < 0.00001$, $I^2 = 7\%$). Five studies (Dura & Beck, 1988; Evans & Keenan, 2007; Evans et al., 2007, 2006; Barton J Hirsch, Moos, & Reischl, 1985) ($N = 469$) found the same pattern of results for offspring-reported internalizing problems ($SMD = 1.13$, 95% CI [0.85, 1.41], $Z = 7.83$, $p < 0.00001$, $I^2 = 40\%$). Offspring-reported internalizing problems included measures of anxiety (Dura & Beck, 1988; Evans & Keenan, 2007; Evans et al., 2007; Barton J Hirsch et al., 1985), depression (Dura & Beck, 1988; Evans et al., 2007, 2006; Barton J Hirsch et al., 1985), and obsessive compulsive disorder symptoms (Barton J Hirsch et al., 1985).

Six studies (Chun et al., 1993; Dura & Beck, 1988; Evans et al., 2007, 2006; Mikail & Von Baeyer, 1990; Turner Cobb et al., 1998) ($N = 413$) examined parent-reported offspring social competence and meta-analysis revealed a significant difference between groups on this variable, with offspring of parents with chronic pain having poorer social abilities ($SMD = 0.47$, 95% CI [0.27, 0.67], $Z = 4.62$, $p = 0.00001$, $I^2 = 0\%$). Two studies (Chun et al., 1993; Evans et al., 2007) ($N = 106$) examining teacher-reported child social competence found a similar pattern of results ($SMD = 0.76$, 95% CI [0.23, 1.28], $Z = 2.83$, $p = 0.005$, $I^2 = 41\%$). One study reported that adolescents of parents with chronic pain had lower self-esteem than control adolescents (Barton J Hirsch et al., 1985), while others measured offspring self-concept (Roy, Thomas, Mogilevsky, & Cook, 1994) and internalizing problems (Chaturvedi & Kanakalatha, 1988; Zelkowitz, Looper, Mustafa, Purden, & Baron, 2013) without control groups.

Family outcomes.

Population-based studies. No population studies examined family outcomes.

Clinical studies. Three clinical studies (Evans et al., 2006; Mikail & Von Baeyer, 1990; K. B. Smith & Chambers, 2006) (N = 232) measured parent-reported family functioning and meta-analysis indicated that family functioning was poorer in families with parental chronic pain compared to healthy control families (SMD = 0.47, 95% CI [0.20, 0.74], Z = 3.46, p = 0.0005, I^2 = 0%). One study did not provide sufficient data for meta-analysis (Dura & Beck, 1988).

Other Outcomes.

Population-based studies. No population studies examined offspring outcomes other than the pain, health, psychological, and family outcomes described above.

Clinical studies. Eight clinical studies examined offspring outcomes not falling into any of the other categories (pain, health, psychological, family). Two studies examined offspring days absent from school during one school year (Chun et al., 1993; Dura & Beck, 1988) (N = 45) and meta-analysis revealed no significant differences between groups (SMD = 0.36, 95% CI [-0.42, 1.14], Z = 0.90, p = 0.37, I^2 = 34%). The outcomes measured in the remaining clinical studies were not sufficiently similar to be combined in meta-analyses. These studies examined physiological (Turner Cobb et al., 1998), school related (e.g., school activity participation) (Barton J Hirsch et al., 1985; Jamison & Walker, 1992; Rickard, 1988), and peer-relationship variables (B J Hirsch & Reischl, 1985). An additional study examined children's behaviours in response to their parent's pain (e.g., keeping distance, rubbing parent's neck) (R. Smith, 1998). Two of these studies did not include control groups (Jamison & Walker, 1992; R. Smith, 1998); the remaining studies did not find group differences on these variables.

2.4.3 Qualitative Synthesis

The three identified qualitative studies were synthesized using meta-ethnographic techniques. One study (Evans & de Souza, 2008) described a sample of 21 children ages 6-12 of mothers with various chronic pain diagnoses, while the remaining two studies (Umberger et al., 2013, 2014) described one sample of 30 young adults (ages 18-21) who reflected on their adolescence with either a mother or father having various chronic pain diagnoses. A thorough literature review revealed no recommendations for handling overlapping samples in meta-ethnography; thus, both samples were included given that they presented analyses of separate data collected from the group of participants. A summary of the key study characteristics and results of the meta-ethnography is provided in Appendix B. Six key concepts regarding the effects of parental chronic pain were identified across studies. The six concepts were: (1) development of children's independence at an early age; (2) development of compassion or empathy for others; (3) learning about health and coping; (4) missing out on normal activities or aspects of childhood or adolescence; (5) effects on participants' emotional health; (6) struggles with communication.

All three studies discussed the *development of children's independence at an early age*, and this was viewed both positively and negatively by participants. Many mothers and children in Evans and de Souza (Evans & de Souza, 2008) reported that children were independent and caring, and while mothers typically viewed this as positive, some children did not enjoy being left alone or unsupervised. Participants in Umberger et al. (Umberger et al., 2014) described hardships relating to developing independence at a young age, and one of the “life lessons” that emerged from the study was “growing up too fast is not a good thing”. Some participants in Umberger et al. (Umberger et al., 2013) used independence from their parent as a strategy for coping with their parent’s pain. Participants also discussed the difficulty of having to take care of their parents physically and emotionally.

The second key concept that emerged across studies was the *development of compassion or empathy for others*. Children in Evans and de Souza (Evans & de Souza, 2008) were reported to be caring and helpful with household tasks, and most children reported enjoying being able to help their mothers. In Umberger et al. (Umberger et al., 2013), some young adults reported being able to empathize with their parents as adolescents and consider the complexity of their pain problems. They also reported learning the life lesson that “it is important to look beneath the surface” and being able to empathize with others (Umberger et al., 2014).

The third key concept that emerged across studies was *learning about health and coping*. Children learned about health, illness, their own bodies, and strategies for coping with stress because of their mother’s pain (Evans & de Souza, 2008). Many young adults in Umberger et al. (Umberger et al., 2014) reported the positive experience of having learned “life lessons” as adolescents because of growing up with a parent with chronic pain. Learning about health and coping did not emerge as a key concept from Umberger et al. (Umberger et al., 2013).

The fourth key concept was *missing out* on normal activities or aspects of childhood and adolescence because of living with a parent with chronic pain. Children reported missing out on social activities because of their mother’s pain, and some expressed anger because of this (Evans & de Souza, 2008). Some young adults reported intense feelings of grief as adolescents regarding the loss of their normal childhood and questioned whether their parents loved them (Umberger et al., 2013), while some learned to accept missing out on certain aspects of life because of parental chronic pain (Umberger et al., 2014).

The fifth key concept that emerged was *effects on participants’ emotional health*. Participants in all samples indicated that they had suffered emotionally at times due to the experience of living with parental chronic pain. Children were described as “clingy” and showing anxiety and sadness related to parental pain (Evans & de Souza, 2008). Young adults reported experiencing negative emotions as adolescents including sorrow, anger, frustration, and fear, both because of the difficulties of having a parent with chronic pain and the worry that their parent would not be around for important future events (Umberger et al., 2013). Some young adults also described worrying during their adolescence about whether their parent’s pain was life threatening and whether they would experience chronic pain themselves in the future, as well as experiencing guilt when they questioned whether their parents were truly in as much pain as they described (Umberger et al., 2014).

The final key concept that emerged was *struggles with communication*. Across studies, children tended to hide their true thoughts and feelings from their parent with pain and avoided communicating their needs. In Evans & de Souza (Evans & de Souza, 2008), several children reported anxiety about their mother's pain, of which their mother was not aware. Some young adults reported distancing themselves from their parents both physically and emotionally in their adolescence, and did not discuss their feelings regarding their parent's pain with anyone (Umberger et al., 2013). However, those who were able to share their feelings with their parents as adolescents experienced an increased closeness in the relationship (Umberger et al., 2013). Some participants explained that they avoided initiating conversations with their parent about pain for fear that it would cause their parent additional pain and suffering (Umberger et al., 2014). Few participants across studies described receiving clear information about their parent's pain.

One main difference in results emerged between the three qualitative studies; namely, the effects of parental chronic pain on offspring pain experiences. Evans and de Souza (Evans & de Souza, 2008) reported that one of the most profound effects of maternal chronic pain was on the children's physical health, as many children reported experiencing pain complaints and discussed the positive aspects of having pain (e.g., receiving special attention from parents). This concept was not described in either of the other two included qualitative studies examining the young adult sample. While some participants in Umberger et al. (Umberger et al., 2014) reported worrying about whether or not they would experience chronic pain problems in the future, they did not discuss effects of their parents' pain on their own pain or other aspects of physical health.

2.5 Discussion

This comprehensive and rigorous mixed-methods systematic review and syntheses indicates that overall, offspring of parents with chronic pain have poorer outcomes in the areas of pain, health, psychological, and family functioning as compared to offspring of parents without pain. Thirty-one population-based studies, 25 clinical studies, and three qualitative studies were included, representing parents with a variety of pain conditions and offspring from newborns to adulthood. Population studies indicated that offspring of parents with chronic pain were more likely to have pain complaints than other offspring, including when either one or both parents had chronic pain. No differences were found for maternal versus paternal chronic pain. They also indicated that newborns of mothers with chronic pain had increased rates of many, but not all, adverse birth outcomes, including low birthweight, small for gestational age, preterm birth, caesarian section, perinatal mortality, and admission to neonatal intensive care.

Clinical studies found that offspring of parents with chronic pain had poorer scores on general health measures, psychological outcomes, and family functioning than other offspring, including higher levels of parent-reported externalizing and internalizing problems and parent- and teacher-reported social competence. Differences were not found for teacher-reported externalizing problems, parent-reported somatization, or days absent from school.

Synthesis of the included qualitative studies also revealed negative effects of parental chronic pain on offspring, including a sense of missing out on a normal childhood or adolescence, negative effects on emotional health, and struggles with parent-child communication. However, the qualitative synthesis also uniquely identified positive outcomes associated with having a parent with chronic pain, including developing independence, developing compassion or empathy for others, and learning about health and coping. This may have revealed positive outcomes associated with having a parent with chronic pain because qualitative studies were designed in such a way that participants could discuss any of their experiences related to parental chronic pain, whether positive or negative, while the quantitative studies (population and clinical) focused on outcomes that were hypothesized to be poorer in children of parents with chronic pain.

This review synthesized findings regarding the relationship between parental pain and offspring outcomes. A relationship between maternal chronic pain and several adverse birth outcomes was found, and the meta-analysis of outcomes based on the informant assessing the outcome (e.g., parent, offspring, teacher) highlighted important findings. Offspring of parents with chronic pain were found to have greater parent-reported externalizing problems, but not teacher-reported externalizing problems. This suggests that future studies should take into account different informants when studying the effects of parental chronic pain on offspring. This review also synthesized results that are generally consistent with the previous narrative review completed in this area (Umberger, 2014); offspring of parents with chronic pain were more likely to have pain complaints and internalizing and externalizing problems compared to other offspring.

While it has been established that offspring of parents with chronic pain have poorer outcomes than offspring of healthy parents, it is unclear whether this finding is directly related to parental pain per se, or to other characteristics commonly associated with chronic pain, such as comorbid mental health issues, medication use, and/or pain-related disability. For example, parental depression is associated with poorer outcomes in offspring (Cho et al., 2015; Goodman & Gotlib, 1999; Matijasevich et al., 2014; Sanger et al., 2014) and the prevalence of major depressive disorder is higher among adults with chronic pain than in the general population (McWilliams, Cox, & Enns, 2003). Two studies included in our review (B J Hirsch & Reischl, 1985; Barton J Hirsch et al., 1985) controlled for parental mental health by including a comparison group of parents with depression in addition to chronic pain. These studies reported poorer outcomes for offspring of parents in both clinical groups compared to healthy control groups, but were inconsistent in observed differences between clinical groups. Two other studies found mixed results regarding correlations between parental mental health and child outcomes (Mikail & Von Baeyer, 1990; Raphael et al., 1990). Similarly, medication use in mothers could have influenced birth outcomes in newborns, although this was not controlled for in any of the included population studies. Moreover, studies included in our review focused primarily on the impact of the presence of parental chronic pain on offspring outcomes. However, individuals with chronic pain can have highly variable levels of associated pain-related disability and interference (Severeijns, Vlaeyen, van den Hout, & Weber, 2001; Vlaeyen, Kole-Snijders, Rotteveel, Ruesink, & Heuts, 1995) and therefore the impact of level of impairment as a result of pain on offspring outcomes will be an important focus for future work. Only two studies in our review included parental pain-related disability in their analyses of offspring outcomes (K. B. Smith & Chambers, 2006; Turner Cobb et al., 1998). Attempts are needed to disentangle the unique contribution of parental chronic pain relative to other associated parental factors in influencing outcomes in offspring.

The present systematic review, meta-analysis, and qualitative synthesis has many strengths. Firstly, a broad search strategy was used, which was successful in identifying a large number of relevant articles including a variety of study designs and samples (population, clinical, and qualitative studies). The inclusion of population studies is important because these studies are more representative of the general population of families dealing with chronic pain than matched control group studies. Completing a synthesis of qualitative studies allowed for the incorporation of the rich data gained in qualitative research in the review and for the discussion about both positive and negative self-perceived effects of having a parent with chronic pain. Secondly, this review is unique in its inclusion of both meta-analysis and meta-ethnographic techniques. Completing formal syntheses allowed for conclusions to be drawn from combinations of studies that would not have been possible in a narrative synthesis. In particular, the meta-analysis allows for estimates of the effect sizes of the studied relationships to be calculated. The use of a mixed methods approach to knowledge synthesis is innovative in the field of pain research and provides a richer basis for understanding the state of the research in this topic area.

A limitation of this work is that an evaluation of the quality of the primary studies included in the review was not possible given that existing quality assessment tools could not be applied across the different types of studies (e.g., Grading of Recommendations Assessment, Development, and Evaluation (GRADE) criteria (Balshem et al., 2011; Huguet et al., 2013)). Furthermore, application of the GRADE criteria require that prognostic studies must use longitudinal designs, and only two of 31 population studies (M Aromaa et al., 1999; Ramchandani et al., 2006) and two of 25 clinical studies (Raphael et al., 1990; Turner Cobb et al., 1998) employed longitudinal designs. The meta-ethnographic synthesis of qualitative studies may have been limited by the small number of included studies. Furthermore, several of the quantitative meta-analyses included small numbers of studies.

This review addresses an important knowledge synthesis gap and identifies areas in need of further exploration in the study of offspring of parents with chronic pain. Future studies should use prospective, longitudinal designs to examine parental pain as a prognostic factor in the development of chronic pain in offspring and to further examine the processes by which parental pain may transfer to offspring. While prior studies have examined other prognostic factors in pain development (Walker, Sherman, Bruehl, Garber, & Smith, 2012), few studies have used longitudinal designs to examine the role of parental chronic pain in offspring outcomes. Additionally, future studies should include multiple measures of outcomes and multiple informants, as relying only on parent reports of offspring pain may be biased by the parents' own pain experiences. Another gap in this area is the role of particular aspects of parental pain such as specific pain parameters (e.g., pain frequency, onset, and duration), pain-related disability, and parent mental health in the relationship between parental pain and offspring outcomes. No studies have examined the timing of onset of parent pain with respect to offspring age or developmental stage. While no differences in outcomes of offspring of mothers versus fathers with chronic pain were found in this review, few included studies provided data separated by parent sex for meta-analysis. Future studies should examine possible differences in this area, given that at least one well-powered study has found such a difference (Hoftun et al., 2013).

Thus far, population-based and clinical studies have established that in general, offspring of parents with chronic pain tend to have poorer outcomes in terms of pain, health, psychological, and family functioning as compared to offspring of parents without pain. However, as revealed in the qualitative synthesis, there are at least some perceived positive offspring outcomes of having a parent with chronic pain. Future research should move beyond simply comparing offspring with and without parental chronic pain, instead, focusing on possible predictors of *which* offspring of parents with chronic pain are at greatest risk for poorer pain, health, psychological, and family outcomes. Employing a risk-resilience framework (Sturgeon & Zautra, 2010) to understanding possible offspring, parent, dyadic, and family variables involved in predicting poorer and, conversely, better offspring outcomes will be important. A greater focus on the mechanisms involved in the relationship between parental chronic pain and offspring outcomes is also needed (i.e., *why* and *how* do these differences occur). Researchers should also consider the strengths of qualitative methodology when designing future studies, and consider whether this methodology may be appropriate for answering their particular research questions. While additional research is needed, the results of this review highlight the impact of parental chronic pain on offspring outcomes and the need for clinicians to incorporate this factor in routine chronic pain assessment and management.

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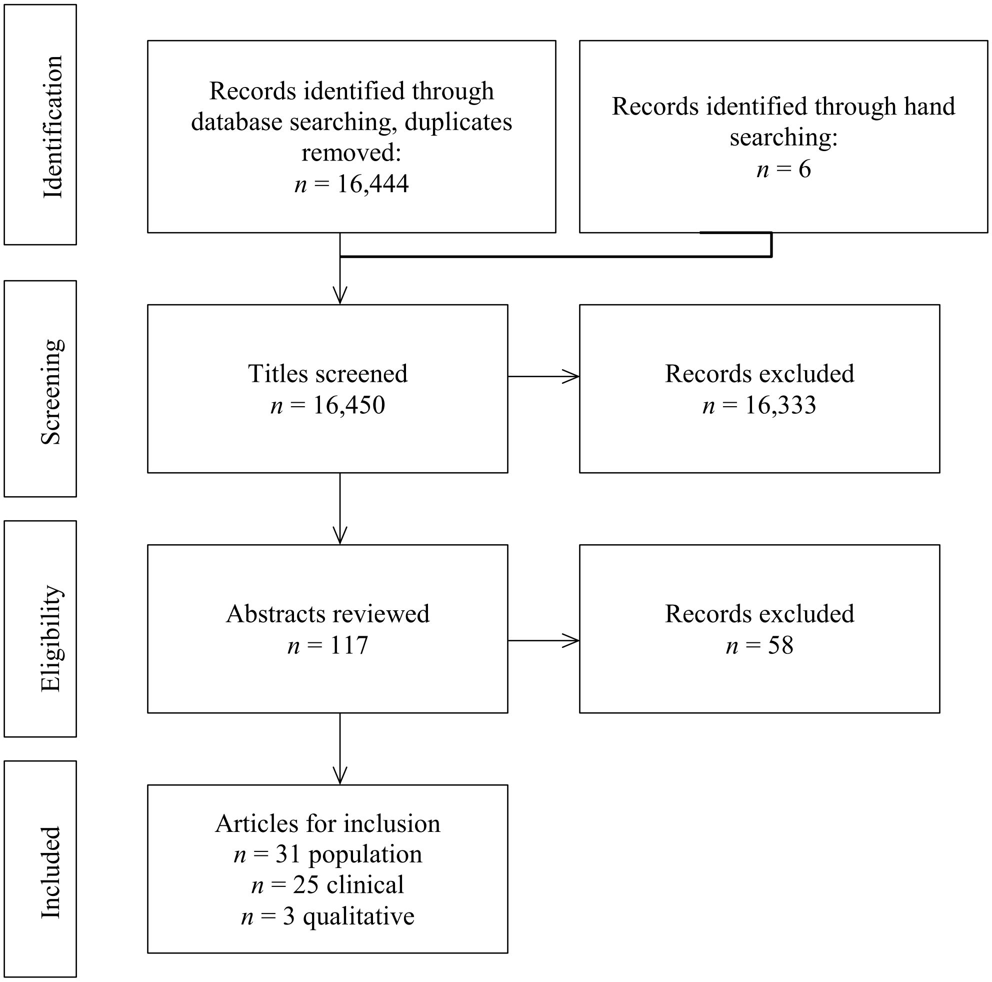
2.8 Figures

Figure 2.8.1 PRISMA flow diagram of identification and screening process for systematic review



2.9 Tables

Table 2.9.1 Characteristics of Studies Included in the Systematic Review

					Offspring Outcomes Measured				
First Author & Year	Parent with Pain	Type of Parental Pain	Offspring Age Range	Number of Offspring	Pain	Health	Psych	Family	Other
Population									
Andersen 2012	Either	Headaches	2-17 years	8256		x			
Aromaa 1996	Mother	Headaches	Newborn	1443		x			
Aromaa 1999	Either	Headaches	6 years	908	x				
*Arruda 2010	Mother	Headaches	5-12 years	1994	x				
Arruda 2012	Mother	Headaches	5-12 years	1856			x		
Assadi 2013	Either	Headaches	14-18 years	272	x				
*Banhidy 2007	Mother	Migraine	Newborn	38151		x			
*Blair 2011	Mother	Migraine	Newborn	660		x			
*Borge 2000	Either	Mixed	13-15 years	229	x				
*Chen 2010	Mother	Migraine	Newborn	29466		x			

*Chen 2013	Mother	Arthritis	Newborn	941574		x	
*Deubner 1977	Either	Migraine	10-20 years	232	x		
*Hasvold 1996	Either	Mixed	20-70 years	1939	x		
*Hoftun 2013	Either	Mixed	13-18 years	5370	x		
Holmes 2003	Either	Arthritis	> 18 years	NR			x
Jensen 2014	Mother	Mixed	6-11 years	131		x	
*Jones 2004	Either	Mixed	12-15 years	1293	x		
Kaasboll 2012	Either	Mixed	13-18 years	3227			x
Koutantji 1998	Either	Mixed	18-51 years	180	x		
Kovacs 2003	Either	Back pain	13-15 years	7361	x		
*Lin 2010	Mother	Arthritis	Newborn	11472		x	
Logan 2008	Either	Mixed	0-17 years	258313		x	
*Norgaard 2010	Mother	Arthritis	Newborn	871579		x	
*O'Sullivan 2008	Either	Back pain	14 years	1608	x		

Ramchandani 2006	Either	Mixed	6 years	8272	x			
*Saunders 2007	Mother	Mixed	11-17 years	2466	x			
Thomas 1992	Either	Mixed	> 18 years	141	x			
*Wallenius 2011	Mother	Arthritis	Newborn	1800827		x		
*Wallenius 2014	Mother	Arthritis	Newborn	627138		x		
Wilson 2014	Either	Mixed	11-14 years	173	x			
Wolff 2009	Mother	Mixed	14 months	275	x			
Clinical								
Aguas 2011	Either	IBD	> 18 years	$n_p = 74$	x			
Bowden 2001	Mother	Arthritis	0-8 months	$n_p = 133$, $n_c = 103$		x		
Buskila 1996	Mother	FM	5-46 years	$n_p = 58$	x			
Buskila 1997	Mother	FM	> 8 years	$n_p = 67$, $n_c = 310$	x			
Chaturvedi 1988	Either	Mixed	10-15 years	$n_p = 36$	x		x	
*Chun 1993	Either	Mixed	6-16 years	$n_p = 35$, $n_c = 29$			x	x

*Dura 1988	Mother	Mixed	7-13 years	$n_p = 7$, $n_c = 14$		x	x	x	x
*Evans 2007	Either	Mixed	6-12 years	$n_p = 24$, $n_c = 12$	x	x	x		
*Evans 2007	Mother	Mixed	6-12 years	$n_p = 55$, $n_c = 48$	x	x	x		
*Evans 2006	Mother	Mixed	6-12 years	$n_p = 55$, $n_c = 48$		x	x	x	
*Hirsch 1985	Either	Arthritis	12-18 years	$n_p = 16$, $n_c = 16$			x		x
Hirsch 1985	Either	Arthritis	12-18 years	$n_p = 16$, $n_c = 16$					x
Hudson 2004	Either	FM	> 18 years	$n_p = 109$, $n_c = 53$	x		x		
Jamison 1992	Either	Mixed	6-18 years	$n_p = 42$, $n_c = 55$	x	x			x
Kalantar 2003	Either	IBS	> 18 years	$n_p = 97$	x				
Messinger 1991	Either	Headaches	> 18 years	$n_p = 247$, $n_c = 279$	x				
*Mikail 1990	Either	Mixed	9-17 years	$n_p = 24$, $n_c = 30$	x	x	x	x	
Raphael 1990	Mother	TMPDS	NR	$n_p = NR$, $n_c = NR$		x			
Rickard 1988	Father	Back pain	8-12 years	$n_p = 21$,	x		x		x

				n_c = *21*					
Roy 1994	Either	Mixed	5-18 years	n_p = 31			x		
Saito 2010	Either	IBS	18-70 years	n_p = 357, n_c = 195	x				
Smith 1998	Either	Migraine	0-17 years	NR					x
*Smith 2006	Either	Headaches	8-15 years	n_p = 52, n_c = 23			x	x	
*Turner Cobb 1998	Either	Arthritis	4-16 years	n_p = 25, n_c = 53		x	x		x
Zelkowitz 2013	Either	Arthritis	1.5-18 years	n_p = 29			x		
Qualitative									
*Evans 2008	Mother	Mixed	6-12 years	n_p = 21					
*Umberger 2013 *Umberger 2014	Either	Mixed	18-21 years	n_p = 30					

Note. * indicates that the study was included in the meta-analyses or meta-ethnography. IBD = inflammatory bowel disease; FM = fibromyalgia; IBS = irritable bowel syndrome; NR = not reported; n_p = number of offspring in parental pain group; n_c = number of offspring in control group; TMPDS = temporomandibular pain and dysfunction syndrome.

Table 2.9.2 Results of Meta-Analyses of Newborn Birth Outcomes

Outcome	Studies	Number of Participants	*OR*	95% CI	I^2
Low birthweight	5	672408	1.35	[1.13, 1.60]	59%
Small for gestational age	6	2447410	1.24	[1.08, 1.42]	63%
Preterm delivery	8	2486221	1.59	[1.28, 1.97]	83%
C-section	6	2447410	1.51	[1.25, 1.83]	90%
Apgar score below 7 at 5 minutes	3	2071095	1.16	[0.82, 1.62]	0%
Perinatal mortality/stillbirth	3	1464898	2.03	[1.29, 3.20]	0%
NICU admission	3	1534893	1.69	[1.01, 2.81]	75%
Congenital abnormalities	3	1464898	1.22	[0.91, 1.64]	18%
Instrument assisted delivery	3	1534893	1.55	[0.82, 2.92]	88%

Note. *OR* = odds ratio; CI = confidence interval

2.10 Supplemental Materials

Supplementary Materials - Appendix A

Search Strategy by Database

Table A1. Search Terms: PubMed

Parental Terms	**Pain Terms**	**Child Terms**	**Relationship Terms**
Title/abstract:	Title/abstract:	Title/abstract:	Title/abstract:
Parent* Mother* Maternal Father* Paternal Family Families Caregiver* Care-giver*	Pain* Fibromyalgia Irritable bowel syndrome Arthrit* Osteoarthrit* Headache* Migraine* Neuralgi* Neuropath* Complex regional pain syndrome	Child* Adolescen* Teen* Youth* Infant* Young adult*	Impact* Influenc* Predict* Factor* Association* Risk* Relation* Correlat* Aggregat*
MeSH Terms:	MeSH Terms:	MeSH Terms:	MeSH Terms:
Parents (Exp)	Pain (Not Exp) Chronic Pain (Exp) Fibromyalgia (Not Exp) Irritable Bowel Syndrome (Not Exp) Arthritis (Not Exp) Osteoarthritis (Not Exp) Headache (Not Exp) Migraine (Not Exp) Neuralgia (Not Exp) Peripheral Nervous System Diseases (Not Exp) Complex Regional Pain Syndromes (Not Exp)	Child (Exp) Child, Preschool (Exp) Adolescent (Exp) Infant (Exp) Young adult (Exp)	Risk (Not Exp) Association (Not Exp)

Note. Exp = exploding

Table A2. Search Terms: CINAHL

Parental Terms	**Pain Terms**	**Child Terms**	**Relationship Terms**
Title/abstract:	Title/abstract:	Title/abstract:	Title/abstract:
Parent* Mother* Maternal Father* Paternal Family Families Caregiver* Care-giver*	Pain* Fibromyalgia Irritable bowel syndrome Arthrit* Osteoarthrit* Headache* Migraine* Neuralgi* Neuropath* Complex regional pain syndrome	Child* Adolescen* Teen* Youth* Infant* Young adult*	Impact* Influenc* Predict* Factor* Association* Risk* Relation* Correlat* Aggregat*
CINAHL Headings:	CINAHL Headings:	CINAHL Headings:	CINAHL Headings:
Parents (Exp)	Pain (Not Exp) Chronic Pain (Exp) Fibromyalgia (Not Exp) Irritable Bowel Syndrome (Not Exp) Arthritis (Not Exp) Osteoarthritis (Not Exp) Headache (Not Exp) Migraine (Not Exp) Neuralgia (Not Exp) Peripheral Nervous System Diseases (Not Exp) Complex Regional Pain Syndromes (Not Exp)	Child (Exp) Child, Preschool (Not Exp) Adolescence (Exp) Infant (Exp) Young adult (Not Exp)	Predictive Research (Not Exp) Correlational Studies (Not Exp) Risk Assessment (Not Exp)

Table A3. Search Terms: EMBASE

Parental Terms	**Pain Terms**	**Child Terms**	**Relationship Terms**
Title/abstract:	Title/abstract:	Title/abstract:	Title/abstract:
Parent* Mother* Maternal Father* Paternal Family Families Caregiver* Care-giver Care-givers	Pain* Fibromyalgia Irritable bowel syndrome Arthrit* Osteoarthrit* Headache* Migraine* Neuralgi* Neuropath* Complex regional pain syndrome	Child* Adolescen* Teen* Youth* Infant* Young adult Young adults	Impact* Influenc* Predict* Factor* Association* Risk* Relation* Correlat* Aggregat*
Emtree Terms:	Emtree Terms:	Emtree Terms:	Emtree Terms:
Parent (Exp)	Pain (Not Exp) Chronic Pain (Not Exp) Fibromyalgia (Not Exp) Irritable Colon (Not Exp) Arthritis (Not Exp) Osteoarthritis (Not Exp) Headache (Not Exp) Migraine (Not Exp) Neuralgia (Not Exp) Peripheral Neuropathy (Not Exp) Complex Regional Pain Syndrome (Not Exp)	Child (Exp) Preschool Child (Not Exp) Adolescence (Exp) Infant (Exp) Young adult (Not Exp)	Predictive Research (Not Exp) Correlational Studies (Not Exp) Risk Assessment (Not Exp)

Table A4. Search Terms: PsycINFO

Parental Terms	**Pain Terms**	**Child Terms**	**Relationship Terms**
Title/abstract:	Title/abstract:	Title/abstract:	Title/abstract:
Parent* Mother* Maternal Father* Paternal Family Families Caregiver* Care-giver*	Pain* Fibromyalgia Irritable bowel syndrome Arthrit* Osteoarthrit* Headache* Migraine* Neuralgi* Neuropath* Complex regional pain syndrome	Child* Adolescen* Teen* Youth* Infant* Young adult*	Impact* Influenc* Predict* Factor* Association* Risk* Relation* Correlat* Aggregat*
Subject Terms:	Subject Terms:	Subject Terms:	Subject Terms:
Parents (Exp)	Pain (Not Exp) Chronic Pain (Not Exp) Fibromyalgia (Not Exp) Irritable Bowel Syndrome (Not Exp) Arthritis (Not Exp) Headache (Not Exp) Migraine Headache (Not Exp) Neuralgia (Not Exp) Peripheral Neuropathy (Not Exp) Complex Regional Pain Syndrome (Type I) (Not Exp)	Offspring (Exp)	Risk Assessment (Not Exp) Risk Factors (Not Exp) Prediction (Not Exp) Statistical Correlation (Not Exp)

Supplementary Materials - Appendix B

Summary of Meta-Ethnography Results for the Three Included Qualitative Studies

	Evans & de Souza, 2008	Umberger et al., 2013	Umberger et al., 2014
Sample	• 21 children and their 16 mothers with chronic pain • 7 girls, 14 boys, ages 6-12 (*M* = 8 years, 9 months)	• 30 young adults ages who identified as having lived during their adolescence with a parent suffering with chronic non-cancer pain (CNCP) • 21 females, 9 males, ages 18-21	
Data Collection	• Interviews conducted in participants' homes; mothers and children interviewed separately • "Themes explored in interviews with children included: mother's health and pain, how children knew about their mother's health, children's feelings about their mother's pain, and the child's health." (p. 492)	• Interviews conducted at university or other accessible public places • "Participants were interviewed once and encouraged to tell their stories about what it was like growing up with a parent who suffered with CNCP and their day-to-day management strategies." (pg. 346, Umberger et al., 2013)	
Setting	• Small city in New Zealand	• Ohio, USA	
Types of parental pain	• Lupus (*n* = 1), arthritis (*n* = 1), polycystic ovary syndrome (*n* = 1), migraine (*n* = 2), repetitive strain injury (*n* = 2), spinal pain (*n* = 9)	• 18 had mothers with CNCP, 12 had fathers with CNCP • Arthritis (*n* = 12), fibromyalgia (*n* = 6), migraine headache (*n* = 5), herniated disc (*n* = 4), degenerative joint disease (*n* = 3), neuropathy (*n* = 3), fractured vertebrae (n = 3), carpel tunnel (*n* = 2), Crohn's disease (*n* = 2), chronic regional pain syndrome (*n* = 1), multiple sclerosis (n = 1), paraneoplastic syndrome (*n* = 1)	
Study perspective	• Developmental gains-and-loss perspective	• Grounded theory	

	- Strengths perspective		
Research questions	"a) understand the challenges that mothers with chronic pain face, as well as their children; b) examine how mothers and children overcome these challenges; and c) discuss maternal chronic pain as a two-way reciprocal process, with mothers and children impacting on the well-being of each other." (pg. 491)	"a) how do adolescents manage living with a parent suffering with CNCP? And b) what environmental factors help or hinder this process?" (pg. 345, Umberger et al., 2013)	
Data analysis	- Qualitative conceptual framework approach - Categories determined before the interviews, but also included topics participants discussed outside of those categories	- Grounded theory - Constant comparative methods - 3 levels of coding, 6 investigators involved in coding	
Themes			
Developing independence	Children developed increased independence and cared for themselves and their mothers.	Some adolescents described having to endure hardships related to developing independence (e.g., having to fulfill a parental role before being ready to do so). Some also used their independence as means of coping with their parent's pain.	Participants described "life lessons", including "growing up too fast is not such a good thing". They described the hardships of having to be independent from an early age.
Developing compassion or empathy for	Mothers and children reported that children were very caring and helped with household tasks. Most	Some adolescents were able to empathize with their parents, including trying to understand the	Participants described learning the "life lesson" that "it is important to look beneath the surface". They

others	children reported enjoying being able to help their mother.	motives behind their parents' behaviours and the complexity of their parents' pain problems.	became able to empathize with others, especially those experiencing adversity.
Learning about health and coping	Children learned more about health, illness, their own bodies, and how to handle stress because of having a mother with chronic pain.	Not described in this analysis	"Many participants described themselves as being "better off" because of having learned some sort of "life lesson" while growing up with a parent with chronic pain." (pg. 6)
Missing out	Activities were often missed because of the mother's chronic pain. Some children expressed anger because of the experiences they had to miss out on because of their mother's chronic pain.	Some participants described experiencing strong negative emotions related to the loss of their childhood and authentic parents.	Participants learned the life lesson that "sometimes you have to accept that 'it's just the way it is'" and came to accept missing out on certain things.
Effects on emotional health	Children became "clingy" and experienced anxiety and sadness related to their mother's pain. In several cases, the extent of the child's anxiety regarding their mother's pain was not known to their mother.	Participants experienced deep, negative emotions regarding their experiences with parental chronic pain. Others worried about whether their parent would be present for important events in the future.	Some participants experienced anxiety and worry related to not knowing whether their parent's pain problems were life-threatening. Others worried whether they would experience chronic pain themselves.

Struggles with communication	As described above, in some cases mothers were unaware of the worries their children had regarding their mother's pain.	Some adolescents distanced themselves emotionally and physically from their parents, not sharing their feelings about their parent's pain. Some were able to seek emotional support from other family members or friends, while others were not.	Few participants described receiving clear information about their parent's pain. Many avoided initiating conversations about pain because they believed that it would cause their parent more pain and distress. They described trying to construct meaning about their parent's pain on their own.

CHAPTER 3: TESTING THE INTERGENERATIONAL MODEL OF TRANSMISSION OF RISK FROM PARENTS WITH CHRONIC PAIN TO THEIR CHILDREN: AN EMPIRICAL INVESTIGATION OF SOCIAL TRANSMISSION PATHWAYS

The manuscript based on this study is presented below. Kristen Higgins, under the supervision of Dr. Christine Chambers, developed the research questions, methodology and data analysis approaches, and obtained ethical approval and funding. Ms. Higgins developed data collection and coding procedures, contributed substantially to completing data collection and coding, and oversaw staff and volunteers who contributed to these activities. She conducted the data analyses and wrote the manuscript. Co-authors reviewed the manuscript and provided feedback, which Ms. Higgins incorporated before submitting the manuscript to *PAIN* on February 22, 2019. The reference for this manuscript is:

Higgins, K. S., Chambers, C. T., Rosen, N. O., Sherry, S., Mohammadi, S., Lynch, M., Campbell-Yeo, M., & Clark, A. J. (Under Review). Testing the Intergenerational Model of Transmission of Risk from Parents with Chronic Pain to their Children: An Empirical Investigation of Social Transmission Pathways. *PAIN*

3.1 Abstract

Children of parents with chronic pain have higher rates of pain and internalizing symptoms (e.g., anxiety, depressive symptoms) than children of parents without chronic pain. Parental modeling of pain behaviour and reinforcement of child pain have been hypothesized to underlie these relationships. These mechanisms were tested in a sample of 72 parents with chronic pain and their children (ages 8 to 15 years). Standardized measures were completed by parents (pain characteristics, pain interference, child internalizing symptoms) and children (pain catastrophizing, pain over the previous three months, and internalizing symptoms). In a laboratory session, children completed the cold pressor task (CPT) in the presence of their parent, and parent-child verbalizations were coded. Significant indirect effects of parental pain interference on child self-reported ($B = 0.12$, 95% CI: 0.01, 0.29) and parent-reported ($B = 0.16$, 95% CI: 0.03, 0.40) internalizing symptoms through child pain catastrophizing were found (parental modeling mechanism). Significant indirect effects were found between parent pain-attending verbalizations and child self-reported ($B = 2.58$, 95% CI: 1.03, 5.31) and parent-reported ($B = 2.18$, 95% CI: 0.93, 4.27) CPT pain intensity and tolerance ($B = -1.02$, 95% CI: -1.92, -0.42) through child pain-attending verbalizations (parental reinforcement mechanism). While further understanding of the temporal relationships between these variables is needed, the current study identifies potential targets (e.g., parent pain interference, child pain catastrophizing, parent reinforcement of child pain) for prevention and intervention of pain and internalizing symptoms in children of parents with chronic pain.

Keywords: Chronic pain, Parents, Children, Social learning, Modeling, Reinforcement

3.2 Introduction

Parents play a critical role in children's pain experiences (Palermo & Chambers, 2005; Palermo et al., 2014). Offspring of parents with chronic pain have higher rates of pain and more internalizing symptoms (i.e., anxiety, depressive symptoms) than offspring of parents without chronic pain (K. S. Higgins et al., 2015). Improved understanding of factors underlying these relationships is needed to support this vulnerable population (K. S. Higgins et al., 2015; Stone & Wilson, 2016). Stone and Wilson's (Stone & Wilson, 2016) model of intergenerational transmission of risk posits that parental chronic pain impacts children's pain and psychological functioning through several mechanisms (e.g., pain-specific social learning) impacting child vulnerabilities (e.g., pain-related cognitions). Pain-specific social learning is hypothesized to occur through parental pain modeling and/or reinforcement of child pain, as in other chronic conditions (Goodman & Gotlib, 1999; Levy et al., 2004). In adolescents with abdominal pain, parental chronic pain predicted adolescent pain severity and disability through parental modeling, not reinforcement (Stone et al., 2018). Modeling may be more salient in children with chronic pain, and predict the maintenance (versus development) of child chronic pain; clarification is crucial for developing effective interventions.

Parental pain modeling may occur through parents' pain interference (extent to which pain interferes with mental, physical, and social activities; Amtmann et al., 2010), which is highly correlated with adolescent- and self-reports of parent pain behaviour (Stone & Walker, 2017). Greater parental pain interference is associated with greater child pain (Evans & Keenan, 2007; Jamison & Walker, 1992; Wilson & Fales, 2015) and parent-reported internalizing (Evans et al., 2006). Observing parental pain interference may impact children's outcomes through its effect on pain-related cognitions (Stone & Wilson, 2016). Increased pain catastrophizing is a robust predictor of poorer pain-related (Fisher et al., 2017; Pielech et al., 2014) and internalizing outcomes (Eccleston, Crombez, Scotford, Clinch, & Connell, 2004; Fisher et al., 2017; Pielech et al., 2014) in children with chronic pain and generally healthy children (Birnie et al., 2016, 2017; Durand et al., 2017; Vervoort, Goubert, et al., 2011). Greater child pain catastrophizing was associated with greater parental pain (Wilson et al., 2014) and mediated the parent-child pain relationship in children with rheumatic disease (Schanberg et al., 2001). Observing greater parental pain interference may increase children's trait catastrophizing about their pain, resulting in increased child pain and internalizing (Stone & Wilson, 2016).

Parent reinforcement of child pain is another proposed social learning mechanism for the transmission of risk in families with chronic pain (Stone & Wilson, 2016). Parental responses directing increased attention to (i.e. positively reinforcing) child pain predict poorer pain-related outcomes in children experiencing chronic (Pielech et al., 2018; Simons et al., 2008; Sinclair et al., 2016; Walker et al., 2002) and acute pain (Campbell et al., 2017; Evans et al., 2016; Moon et al., 2011; Spagrud et al., 2008; Walker et al., 2006; Williams et al., 2011). Parent attending to child pain also predicted children's decreased use of adaptive pain coping strategies (Blount et al., 2001; Spagrud et al., 2008) and increased pain attending (Walker et al., 2006), in turn predicting increased child pain and distress (Blount et al., 1997). Preliminary evidence indicates that parents with chronic pain report more pain-reinforcing responses than other parents (Wilson & Fales, 2015). Increased parent pain-attending behaviours may increase child attending to pain, ultimately resulting in greater child pain in this population.

This study examined pain-specific social learning mechanisms (parental modeling and reinforcement) in the intergenerational transmission of pain and internalizing symptoms (Stone & Wilson, 2016). It builds on previous research by examining these mechanisms in adults with chronic pain and their children, incorporating multi-informant outcome assessment, and directly observing parents and children during child pain. It was hypothesized that 1) child pain catastrophizing would mediate the relationship between parental pain interference and the presence of child chronic pain and internalizing symptoms (parental modeling), and 2) during an experimental pain task, parents' pain-attending verbalizations would predict child pain outcomes through child pain-attending verbalizations (parental reinforcement).

3.3 Method

The data described in the current manuscript were collected as part of a larger study examining two separate research questions which are described in two papers. The current paper tests a theoretical model of the mechanisms through which parental chronic pain impacts child pain and mental health outcomes. The other paper examines the measurement of a novel construct, child catastrophizing about parental chronic pain, and the intra- and interpersonal effects of parent and child catastrophizing about one another's pain (Higgins et al., in preparation). The study procedures were approved by the IWK Health Centre Research Ethics Board with reciprocal approval from the Nova Scotia Health Authority Research Ethics Board.

3.3.1 Participants

Seventy-two parent-child dyads participated in the current study. Parents with chronic pain were recruited through a local adult pain management clinic (*n* = 50) and using community recruitment methods (e.g., posters placed in community locations, social media advertisements; *n* = 22). Parents were considered eligible for the current study if they: 1) had experienced pain for at least six months (consistent with the International Association for the Study of Pain recommendations for research on chronic pain; (Task Force on Taxonomy of the International Association for the Study of Pain, 2002); 2) had bodily pain over the previous four weeks rated as moderate, severe, or very severe on a verbal rating scale (from the Short Form-36 Health Survey; (Ware, Jr. & Sherbourne, 1992)), and 3) had an eligible child between the ages of 8 years 0 months and 15 years 11 months. Exclusion criteria for parents included cognitive impairments that would impact their ability to participate in study activities (e.g., dementia), not living with the eligible child at least 50% of the time, being unable to read, write, and speak English well enough to participate in study activities, or having uncorrected hearing or vision impairments.

Children were eligible to participate if they were the child of an eligible parent (one child per parent) and were between the ages of 8 years, 0 months and 15 years, 11 months. Exclusion criteria for children included having developmental delays that would impact their ability to participate in study activities, inability to read, write, and speak English well enough to participate in study activities, having uncorrected hearing or vision impairments, or having contraindications to participating in the cold pressor task (e.g., blood or circulation disorders, current injury or history of frostbite to the non-dominant hand or arm; von Baeyer et al., 2005).

3.3.2 Experimental Pain Task

Children completed the cold pressor task (CPT), a safe and ethically acceptable method for inducing brief, mild-moderate pain in the laboratory (Birnie et al., 2011). Children were asked to place their non-dominant hand in a bath of cold water (10 ± 0.2°C) up to their wrist. They were asked to keep their hand in the water for as long as they could, but were informed that they could remove their hand at any point if it became too uncomfortable or painful. Children were able to keep their hand in the water for a maximum of four minutes, but were not informed of this limit beforehand. These parameters are consistent with published safety guidelines for this task (von Baeyer et al., 2005). During the CPT, parents sat across from their child and watched, and dyads were asked to speak to one another as they normally would elsewhere. Parents' and children's interactions during the CPT were videotaped throughout the duration of time that the child's hand was in the water.

3.3.3 Measures

Demographics. Parents completed an author-developed questionnaire assessing demographic characteristics of themselves and their child including age, sex, racial/ethnic identity, level of education, and marital status. Parents were also asked whether any other members of the child's immediate or extended family had chronic pain.

Pain characteristics. Parents and children each reported on their own pain over the past three months using a questionnaire modified from previous research (Petter, Chambers, McGrath, & Dick, 2013) and based on established guidelines for pain assessment in research with children and adults (McGrath et al., 2008; Turk et al., 2003). Parents completed the written questionnaire while children completed the questionnaire verbally with a research assistant. The research assistant began by establishing the three month timeline (e.g., by asking children about the activities they had engaged in over the past three months) and asking children about any pain they may have experienced during this time period. They were asked to indicate which of these pains had occurred most often over the past three months, and answered questions about the location, duration, frequency, and usual intensity of this pain (using an 11-point numeric rating scale). Children were classified as having chronic pain if they had experienced pain (other than muscle soreness due to physical activity) at least once per week for at least the past three months. Parents completed the same questions in written form, and this information was used to characterize the sample.

Parent pain interference. Parents completed the PROMIS Pain Interference Short Form 8a measuring the extent to which pain limits or interferes with an individuals' functioning (e.g., physical and social activities) and enjoyment of life (Amtmann et al., 2010). This measure contains eight items (e.g., "How much did pain interfere with your ability to participate in social activities?"), each responded to on a 5-point scale from "Not at all" to "Very much". Total scores can range from 8-40, and higher scores indicate greater interference with functioning. Evidence of the measure's internal consistency and validity (including construct, discriminant, and ecological validity) have been found in general samples and individuals with a variety of chronic health conditions including chronic pain (Amtmann et al., 2010; Cella et al., 2010; K. F. Cook et al., 2016). The internal consistency of this measure in the current study was $\alpha = .95$.

Child pain catastrophizing. Children completed the Pain Catastrophizing Scale for Children (Crombez et al., 2003) to assess trait tendency to engage in catastrophic thinking about their own pain (e.g., “When I am in pain, I worry all the time about whether the pain will end”, “When I am in pain, I can’t keep it out of my mind”). Evidence of internal consistency and validity (including factorial, construct, and predictive validity) has been found for this measure in community samples and in samples of children with chronic pain ages 8 to 16 (Crombez et al., 2003; Parkerson et al., 2013; Pielech et al., 2014). It is considered well-established in terms of its psychometric properties in youth with chronic pain (Fisher et al., 2017). In the current study, the internal consistency of this measure was α = .92.

Child internalizing symptoms. Child and parent reports of child internalizing symptoms were collected using the Internalizing Problems composite scale of the Behavior Assessment System for Children (Reynolds & Kamphaus, 2004), a widely used system of multidimensional clinical measures of children’s overall behaviour and emotional functioning. The Internalizing Problems composite scale assesses the child’s inwardly directed distress (i.e. symptoms of anxiety and depressive disorders). T scores (*M* = 50, *SD* = 10) for this scale were generated using the BASC-2 ASSIST computer scoring program. *T* scores of 70 or above on this scale represent clinically significant levels of internalizing symptoms (Reynolds & Kamphaus, 2004). Children completed the Self-Report of Personality Child version (for ages 8-11 years; 139 items) or Adolescent version (for ages 12-15 years; 176 items), comprised of items answered on true/false or 4-point response scales (ranging from “never” to “almost always”) assessing a wide range of behaviors and emotions. Parents completed the Parent Rating Scale Child version (160 items) or Adolescent versions (150 items); all items on the parent forms were answered using the 4-point response scale (“never” to “almost always”). Evidence for the reliability (internal consistency and test-retest reliability) and validity (including factorial, concurrent, and discriminant validity) of both forms has been found (Reynolds & Kamphaus, 2004).

Child experimental pain outcomes. After completion of the CPT, children and parents were independently asked to rate the child's average pain intensity during the task using the Faces Pain Scale-Revised, a well-validated scale considered appropriate for assessing acute pain intensity in children ages 4-16 years (Cohen et al., 2008; Hicks et al., 2001). Evidence of strong test-retest reliability and concurrent validity has been found (Stinson et al., 2006). This scale has also been used in studies to provide parent reports of child experimental and procedural pain intensity (Birnie et al., 2016; Boerner et al., 2013; Spagrud et al., 2008). The scale includes six faces ranging from "no pain" to "very much pain", resulting in a score ranging from 0-10.

Children's pain tolerance during the CPT was measured as the time in seconds that the child kept their hand in the cold water, up to a maximum of four minutes (240 seconds). Given the typically bimodal distribution of this variable, it was dichotomized, with children classified as having reached the ceiling tolerance time or not.

Observed parent and child verbalizations during experimental pain task. Parents and children's verbalizations during the CPT were transcribed verbatim and coded utterance by utterance using a version of the Child and Adult Medical Procedure Interaction Scale Revised (CAMPIS-R; Blount et al., 1997) modified for use in the context of the CPT (Moon et al., 2011). The CAMPIS-R is a well-established observational coding system for parent-child interactions during painful procedures, and the modified CAMPIS-R is considered to be approaching well-established (Bai, Swanson, & Santacroce, 2018). Given that several parents and children discussed the parent's chronic pain during the cold pressor task, a subcode for these verbalizations was added to the coding system for the current study. Parent verbalizations were coded into three categories following previous studies (Birnie et al., 2016; Moon et al., 2011; Walker et al., 2006). The primary focus for the current study was the attending talk category (i.e. verbalizations drawing attention to child cold and pain symptoms), which included four subcodes: 1) symptom-focused talk and commands to child, 2) sympathy to child, 3) procedure-related praise to child, and 4) procedure time talk and commands to child. Other coded categories were non-attending talk (i.e. verbalizations aimed at drawing attention away from child cold and pain symptoms; comprised of subcodes 1) non-symptom-focused talk and commands to child, and 2) humor to child) and other talk (i.e. represented verbalizations not fitting into the above categories; comprised of subcodes 1) other procedure talk and commands to child, 2) criticism to child, 3) parent talk about own pain symptoms, and 4) other talk to child such as sentences cut off before meaning could be ascertained and parents asking for clarification about child statements). Children's verbalizations during the CPT were grouped into two categories. The focus for the current study was on the child symptom complaints category, which was composed of four subcodes: 1) cold/pain symptom talk to parent, 2) anxiety talk to parent, 3) procedure time talk to parent, and 4) resistance talk to parent. The other coded category was child other talk, which included five subcodes: 1) child non-symptom focused talk to parent, 2) other procedure talk to parent, 3) child coping talk to parent, 4) child talk about parent pain symptoms to parent, and 5) other talk to parent. Proportion scores were calculated for each category (number of utterances made in each category divided by total number of utterances made by the participant) in order to account for between-participant differences in the number of utterances made during the CPT, consistent with previous studies (Birnie et al., 2016; Moon et al., 2011). These proportion scores were used in all analyses of coded variables.

Coding of all participant transcripts was completed by a primary coder who was trained on the coding system by the first author. The first author was trained on the coding system and demonstrated inter-rater reliability with coded training videos developed for previous studies. The first author coded a randomly selected subset of 20% of the transcripts coded by the primary coder to determine inter-rater reliability. Excellent inter-rater reliability was established for both parent (subcode level: 90.10% agreement, κ=0.85, SE=0.02; code grouping level: 90.60% agreement, κ =0.85, SE=0.03) and child utterances (subcode level: 86.60% agreement, κ =0.81, SE=0.03, code grouping level: 93.20% agreement, κ =0.80, SE=0.04) (Banerjee, Capozzoli, Mcsweeney, & Sinha, 1999).

3.3.4 Procedure

Parents and children attended one study visit, which began with completing informed consent and assent procedures. Children and parents completed the questionnaire and interview measures as described above, and children completed the CPT in the presence of their parent. The order of these tasks was counterbalanced across parent-child pairs (CPT first: *n* = 36 pairs; 50.00%). While completing the questionnaires, children were accompanied by a research assistant. Parents completed the questionnaires independently in a separate room, but could approach the research assistant at any time to ask questions. The study visit took approximately 90 minutes to complete, and parents and children were compensated for their time and travel expenses.

3.3.5 Data Analysis

Missing data on study variables was minimal (1.00%) and found to be missing completely at random (Little's MCAR test $\chi 2$ (3326) = 0.00, $p = 1.00$). For participants missing 10% or less of a particular questionnaire, missing data was handled using expectation maximization imputation ($n = 3$ children, $n = 2$ parents) (Enders, 2001; Little, Jorgensen, Lang, & Whitney Moore, 2014; Tabachnick & Fidell, 2007). In mediation models with continuous outcomes, full information maximum likelihood was used (Enders, 2001; Little et al., 2014). Correlations were used to examine relationships between study variables and potential covariates (parent and child age, parent and child sex, parent chronic pain duration, first task completed during study visit) to be controlled for in mediation analyses. Where correlation coefficients were $r \geq 0.30$, a covariate was included in analyses involving that study variable (Frigon & Laurencelle, 1993).

Mediation models were tested with path analysis in R using lavaan (Rosseel, 2012). Two theoretical models based on Stone and Wilson's theory (Stone & Wilson, 2016) were tested with child-reported, parent-reported, and observed outcomes. The parental modeling mechanism was tested by examining the indirect effects of parental pain interference on child outcomes (child chronic pain status, child self-reported internalizing symptoms, parent-reported child internalizing symptoms) through the mediator of child pain catastrophizing. The parental reinforcement mechanism was tested within the context of the CPT; the indirect effects of parental attending talk during the task on child outcomes (child self-reported average pain intensity during CPT, parent-reported child average pain intensity during CPT, observed child CPT tolerance) through child symptom complaints were examined. Separate models were run for each outcome.

Mediation models were tested using MLR estimation for models with continuous outcomes and using WLSMV for models with dichotomous outcomes; standardized and unstandardized path coefficients are reported. In each model, indirect effects were tested using bias-corrected bootstrapped (5,000 samples) 95% confidence intervals (MacKinnon, Lockwood, & Williams, 2004; Preacher & Hayes, 2008). If the 95% confidence interval for the indirect effect did not include 0, the indirect effect was considered significant (Preacher & Hayes, 2008). Based on simulation study results, the current study was estimated to be adequately powered (power of 0.80 or higher) to detect indirect effects using bias-corrected bootstrapped confidence intervals in cases when both paths of the indirect effect were medium in effect size (Fritz & MacKinnon, 2010).

3.4 Results

3.4.1 Demographics

Parent participants had a mean age of 42.91 years (*SD* = 6.49). Most parent participants were mothers (*n* = 57; 79.17%), and all parents reported being the participating child's biological parent. Parents identified most often as white (*n* = 64; 88.89%), followed by bi- or multi-racial (*n* = 4, 5.55%) and "other" racial/ethnic identities (*n* = 4, 5.55%). Regarding highest level of education completed, parents most commonly reported completing trade school or community college (*n* = 27; 37.50%). Others reported having some university education (at least one year; *n* = 16, 22.22%), having completed an undergraduate degree (*n* = 10; 13.89%) or high school education (*n* = 10; 13.89%), or completing graduate school or professional training (*n* = 7; 9.72%) or part of high school (*n* = 2, 2.78%). Most parents reported being married (*n* = 47; 65.28%) or in a common-law relationship (*n* = 10; 13.89%). The majority of parents reported that the participating child lived with them full time (*n* = 62; 86.11%) and that they typically spoke English together at home (*n* = 71; 98.61%). Twenty-two parents (30.56%) reported that another member of their or their child's extended family also has chronic pain.

Child participants had a mean age of 12.12 years (*SD* = 2.45) and were approximately evenly split between girls (*n* = 39; 54.17%) and boys (*n* = 33; 45.83%). Parent reports identified children most often as white (*n* = 57; 79.17%), followed by bi- or multi-racial (*n* = 9, 12.50%) and "other" racial/ethnic identities (*n* = 6; 8.33%).

3.4.2 Descriptive Statistics

Parent pain characteristics are in Table 3.9.1. Back pain was the most common type of pain among parents. On average, parents reported the usual pain intensity for their most common pain over the past three months to be nearly 7 out of 10 (moderate pain (Boonstra et al., 2016)). Descriptive statistics and correlations among study measures included in the mediation models are provided in Table 3.9.2. Approximately one third of child participants had chronic pain based on self-report responses. Six children (8.33%) had Internalizing Problems composite scores that fell within the clinically significant range based on child report; based on parent report, 13 children (19.12%) scored in this range.

3.4.3 Potential Covariates

Correlations where $r \geq 0.30$ were identified between the child reaching ceiling CPT tolerance and child age ($r = 0.36$, $p = .002$) and parent age ($r = 0.38$, $p = .001$). Mediation analyses were run controlling for these covariates; however, given that the pattern and significance of results was the same as the results without covariates included, only the most parsimonious model is presented. No other correlations between potential covariates and study variables where $r \geq .30$ were found.

3.4.4 Testing Parental Modeling Mechanism

Figure 3.8.1(a) shows the path analyses testing the parent modeling mechanism with the outcomes child chronic pain status, child self-reported internalizing, parent-reported child internalizing, and child CPT tolerance. Tests of the indirect effects are in Table 3.9.3. Parent pain interference had significant indirect effects on child self-reported internalizing and parent-reported child internalizing through child pain catastrophizing, supporting the hypothesized parental modeling mechanism. The indirect effect of parent pain interference on child chronic pain status through child pain catastrophizing was not significant.

3.4.5 Testing Parental Reinforcement Mechanism

Figure 3.8.1(b) shows the path analyses testing the parent reinforcement mechanism in the context of the CPT with the outcomes child self-reported average pain intensity, parent-reported average pain intensity, and child reaching ceiling CPT tolerance. Parent attending talk had a significant indirect effect on each of these outcomes through child symptom complaints (Table 3.9.3), supporting the hypothesized parental reinforcement mechanism.

3.5 Discussion

Results of the current study partially supported the hypothesized pain-specific social learning mechanisms underlying the intergenerational transmission of risk from parents with chronic pain to their children (Stone & Wilson, 2016). The parental pain modeling mechanism, represented by parental pain interference predicting child outcomes through child pain catastrophizing, was supported for child internalizing symptoms, but not for the presence of child chronic pain. These results are consistent with studies examining relationships between parental pain and disability and child internalizing (Evans et al., 2006; Kaasbøll, Lydersen, Ranøyen, Nilsen, & Indredavik, 2018). However, they differ from studies finding support for the parental pain modeling mechanism in adolescents with functional abdominal pain (predicting adolescents' pain severity and disability; Stone et al., 2018) and children of mothers with irritable bowel syndrome (predicting child-reported frequency of stomachaches and bother of gastrointestinal symptoms; Levy et al., 2004). Parental pain modeling may have greater influence on the maintenance rather than development of child chronic pain, or on the impact of child pain (e.g., children's disability) rather than on its presence. These effects may only be evident in children who have chronic pain themselves; future studies could examine child chronic pain status as a moderator in this relationship. Other factors beyond pain interference may contribute to the pain modeling mechanism; for example, parent-child communication about parental pain (Umberger et al., 2013) could impact children's interpretations of parents' pain behaviours. Children may vary in their awareness of parental pain behaviour, perhaps due to differences in empathy (Goubert et al., 2005).

The current results supported the parental reinforcement mechanism of transmission, with increased proportion of parental attending responses to child pain during the CPT predicting increased child pain intensity and decreased tolerance through the mediator of child pain attending. This is consistent with previous studies of children's acute (Campbell et al., 2017; Evans et al., 2016; Moon et al., 2011; Spagrud et al., 2008; Williams et al., 2011) and chronic pain (Pielech et al., 2018; Simons et al., 2008; Sinclair et al., 2016; Walker et al., 2002). Increased parent attending to child pain may contribute to the development of child chronic pain through its influence on parent responses to child pain expression. For example, parents who could more easily shift their attention to pain-related stimuli showed more pain control behaviours in response to increased child facial cues of pain (Vervoort, Karos, Johnson, Sütterlin, & Van Ryckeghem, 2019), while those with reduced attention-shifting responded similarly regardless of child facial cues.

The results of the current paper were inconsistent with a previous study that did not find support for the parental reinforcement mechanism in adolescents with functional abdominal pain and their parents (Stone et al., 2018). Several methodological differences may explain the divergent results. Firstly, the current study used a psychometrically sound observational coding system to identify parent and child pain-attending verbalizations during a laboratory pain task, while the other used a parent self-report measure of solicitous responding to child chronic pain. Social desirability or retrospective reporting bias may impact parents' reporting on their own solicitous responses. Secondly, parents and children may have acted differently during the novel experience of the CPT than in day-to-day child pain, although results of CPT studies often mimic those found in studies of acute and day-to-day pain (Birnie et al., 2016; Chambers et al., 2002; Evans et al., 2016) and parents and children report similar responding to one another during experimental and other pain contexts (Birnie et al., 2016; Walker et al., 2006). Finally, parents in the current study were a clinical sample of adults with chronic pain with fairly high levels of pain intensity and interference (Boonstra et al., 2016; PROMIS Pain

Interference Scoring Manual, 2017) and higher self-reported average pain intensity than in the previous study (Stone et al., 2018). Parents with more severe chronic pain may engage in increased pain-reinforcing responses; in the current and previous studies of parents with chronic pain (Wilson & Fales, 2015), those with greater pain interference engaged in increased child pain reinforcing responses, as did parents with increased depressive symptoms (Fussner, Schild, Lewandowski Holley, & Wilson, 2018).

Overall, the current results suggest parental pain modeling predicts the development of child (self- and parent-reported) internalizing symptoms in children of parents with chronic pain, and that parental attending to child pain may influence children’s pain experiences through increases in child pain attending. These results present the first examination of parental pain modeling as a mechanism underlying the relationship between parental chronic pain and child internalizing symptoms. While much previous research has focused on intergenerational transmission of pain, children of parents with chronic pain are at risk for increased internalizing symptoms compared to other children (K. S. Higgins et al., 2015). Improved understanding of the mechanisms underlying these relationships is essential for the development of effective prevention and intervention programs for this population (K. S. Higgins et al., 2015; Stone & Wilson, 2016).

The study has several strengths, including providing empirical tests of a theoretical model of the intergenerational risk transmission in a clinical sample of adults with chronic pain and their children. Previous research has often focused on clinical samples of children with chronic pain and their parents (Schanberg et al., 2001; Stone et al., 2018; Walker et al., 2006). Assessment of relevant variables utilized multiple methods (psychometrically sound questionnaires and observational coding system) and informants (child, parent, trained coders), while much previous research has relied exclusively on questionnaires (Levy et al., 2004; Stone et al., 2018) or on parent-reported child variables (Chun et al., 1993; Jamison & Walker, 1992; Wilson & Fales, 2015). This approach is particularly important given concerns that parental chronic pain may bias parents' reports of their children's pain (Graungaard et al., 2016).

The current findings must be interpreted in the context of study limitations. The cross-sectional design limits the interpretation of the directions of relationships between variables, although the analyses were conducted based on existing relevant theory (Stone & Wilson, 2016). The transmission of risk from parents with chronic pain to their children is theorized to occur through multiple mechanisms, and only social learning mechanisms were examined in this study; other mechanisms are likely involved in these relationships (e.g., genetics). This study also used parental pain interference to represent parents' modeling of overt pain behaviour to their children. While this construct correlates highly with adolescent- and self-reports of parental pain behaviour (Stone & Walker, 2017), parental pain behaviour was not directly assessed. Incorporating observational assessment of parental pain behaviour into future studies of these mechanisms could help disentangle these relationships. While the use of a child pain interview likely assisted with child understanding of the measure, social desirability may have influenced children's responses. The current sample of parents was largely mothers (79.17%) and identified as white (88.89%); thus the generalization of these results to fathers and parents of other races is unknown.

Several important clinical implications can be drawn from the present results. Consistent with family theories of pain and health (Bavelas & Segal, 1982; Palermo & Chambers, 2005; Rolland, 1999; Stone & Wilson, 2016), parents' own health and coping is associated with children's wellbeing. While the temporal directions of these relationships still need to be established, the findings suggest that parental pain interference may impact children's own pain catastrophizing and internalizing symptoms. Ensuring that parents with chronic pain can access appropriate pain management services could positively impact children's mental health and pain coping strategies. These results highlight the importance of access to appropriate treatment for individuals with chronic pain; not only is it a human right (Cousins & Lynch, 2011) and important for their own health and wellbeing (Turk, Wilson, & Cahana, 2011), it may also benefit to their family members. The present findings identify child pain catastrophizing and parental responses to child pain as potential targets for prevention and intervention in children of parents with chronic pain. Programs that help children identify their cognitions about pain and teach alternative strategies to interpret and respond to pain could be effective in preventing or treating internalizing symptoms in children of parents with chronic pain (Umberger, Risko, & Covington, 2015). Educating parents with chronic pain about helpful ways to interact with their children during pain may assist in preventing the intergenerational transmission of chronic pain. Such interventions may be particularly important to this population given parents' worry about their children developing problems with pain (Wilson & Fales, 2015), and could be incorporated into adult pain management programs along with support to connect parents with additional child resources as needed.

Additional research is needed to clarify the mechanisms underlying the vulnerability of children of parents with chronic pain to poorer pain and psychological outcomes. Longitudinal, prospective studies are needed to determine the temporal relationships between parental pain modeling, parental reinforcement of child pain, and child outcomes. Consideration of other variables which may impact child health such as parental mental health (Cho et al., 2015; Goodman & Gotlib, 1999; Neville, Soltani, Pavlova, & Noel, 2018), neighborhood characteristics (Schild, Reed, Hingston, Dennis, & Wilson, 2016), and exposure to adverse childhood events (Gareth T. Jones, Power, & Macfarlane, 2009; Varinen, Kosunen, Mattila, Koskela, & Sumanen, 2017) will also be important to disentangle the influences of these factors on child health in the context of parental chronic pain. Identification of children most at risk for poor outcomes (e.g., based on sex, racial/ethnic background, socioeconomic status) may assist in appropriately targeting interventions. While studies have focused on the important task of identifying potential risk factors in families with chronic pain, research on protective factors is also needed (K. S. Higgins et al., 2015; Stone & Wilson, 2016). Several such factors have been hypothesized (e.g., pain acceptance (Stone & Wilson, 2016)), and should be examined in future research. Research aimed at developing prevention and intervention programs for this vulnerable population should be a priority. Potential targets for such programs have been identified (e.g., parental pain interference and reinforcement of child pain, child pain catastrophizing; parent-child communication about parental pain (Umberger et al., 2013, 2015)), and qualitative research has described child- and parent-perceived needs of children living with parental chronic pain (Umberger et al., 2015). Intervention programs should be co-created with families with parental chronic pain so that such programs are not only evidence-based, but also relevant and feasible for intended users. Coordinated efforts from researchers, health professionals, and families are needed to build on our current understanding of pain-specific social learning and better support children of parents with chronic pain.

3.6 References

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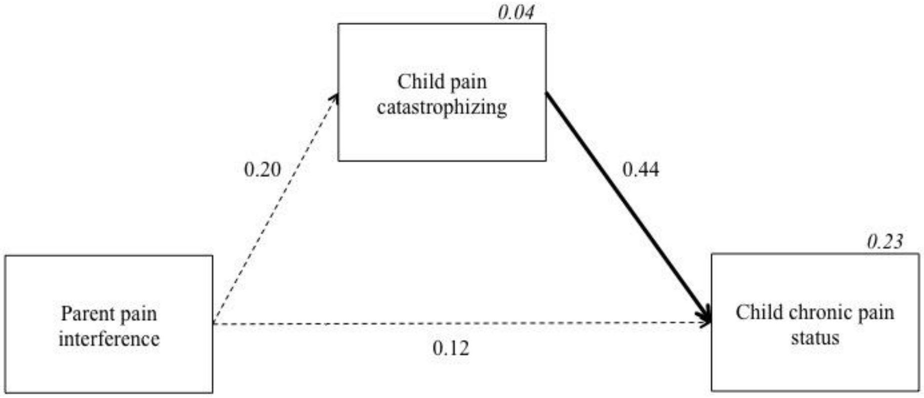
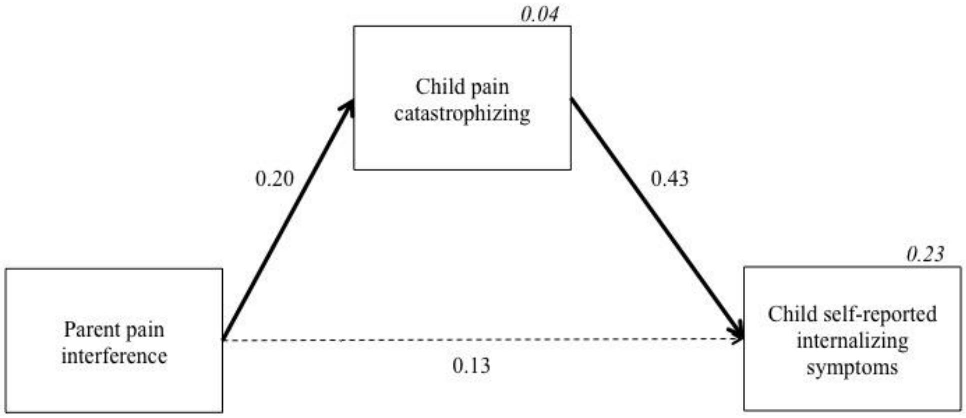
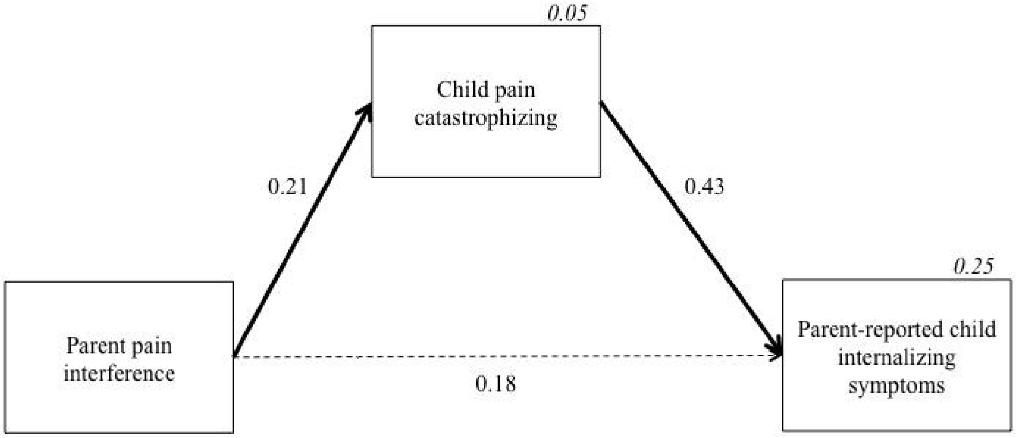
3.8 Figures

Figure 3.8.1 Path diagrams for tested mechanisms of transmission of risk from parents with chronic pain to their children

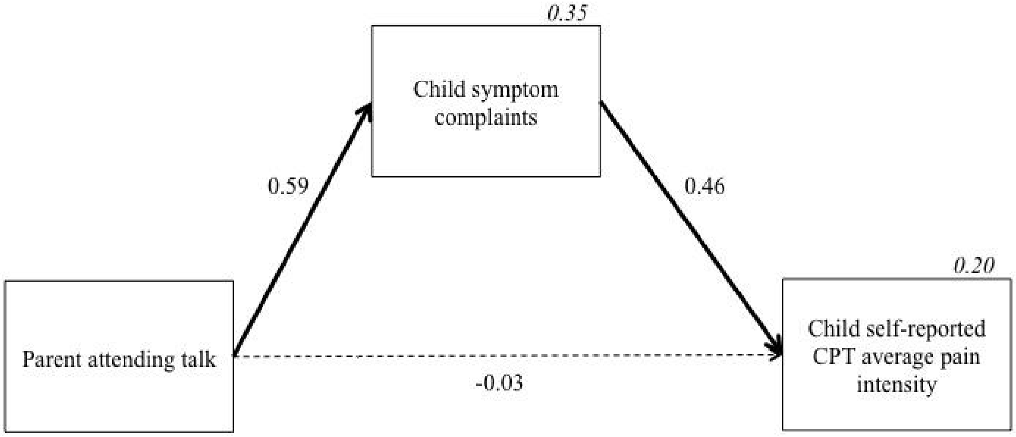
a) Parental modeling mechanism

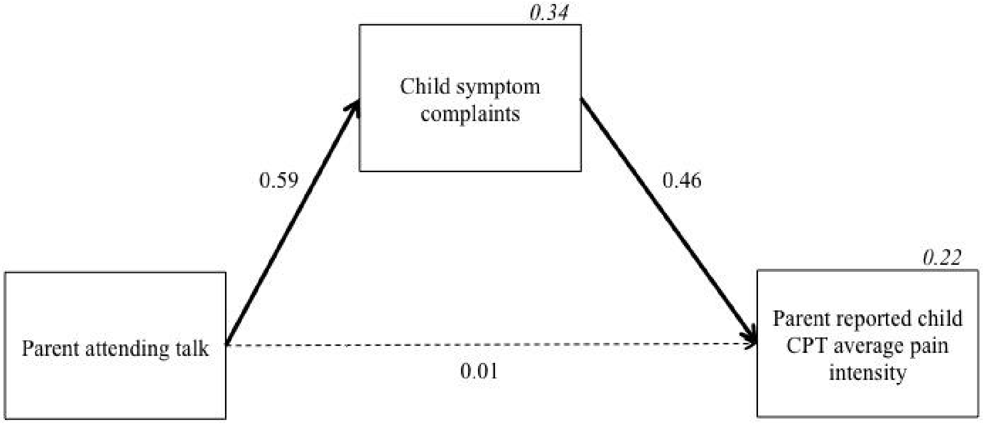


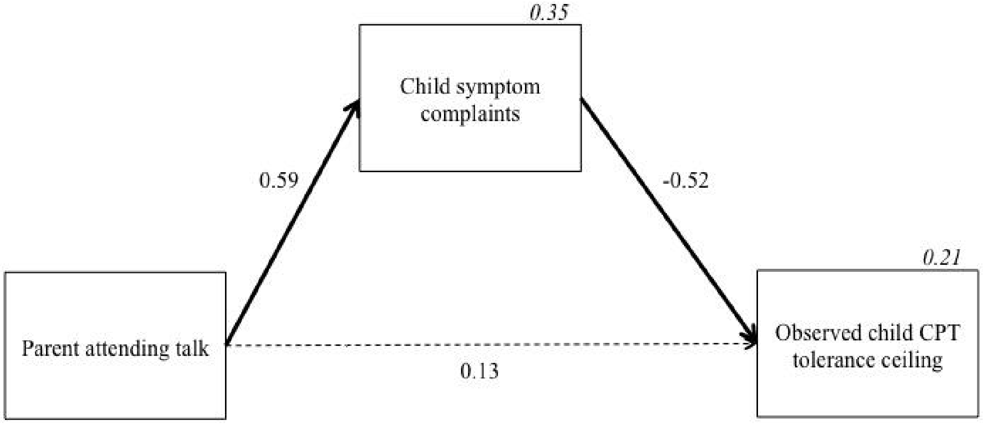




b) Parent reinforcement mechanism







Solid black lines represent significant paths ($p < 0.05$). Dashed lines represent non-significant paths ($p \geq 0.05$). Statistics presented are standardized path coefficients for each path. Italicized values above the upper right corner of mediator and outcome variables represent the proportion of variance explained for that variable (R^2).

3.9 Tables

Table 3.9.1 Parent Pain Characteristics

	Mean (*SD*) or *n* (%)	Observed range
Duration of chronic pain	10.92 years (9.46)	0.42-37.60 years
Most common pain over the past 3 months	Back pain: 21 (29.17%) Muscle pain: 14 (19.44%) Multisite pain: 11 (15.28%) Headaches: 7 (9.73%) Joint pain: 5 (6.94%) Nerve pain: 3 (4.17%) Abdominal pain: 2 (2.78%) Migraines: 2 (2.78%) Neck pain: 2 (2.78%) Other types (including bone, thoracic, tooth, pelvic pain): 5 (6.94%)	
Frequency of most common pain over the past 3 months	More than once per week: 68 (94.44%) About once per week: 2 (2.78%) Once or twice per month or less: 2 (2.78%)	
Usual intensity of most common pain over the past 3 months	6.94 (1.91)	3-10

Table 3.9.2 Descriptive Statistics and Correlations

	Mean (SD) or *n* (%)	Observed range	1	2	3	4	5	6	7	8	9	10
1. Parent pain interference	29.26 (8.15)	10-40	--	0.20	0.17	0.22	0.24	0.27^{*}	0.12	0.14	0.16	-0.02
2. Child pain catastrophizing	18.25 (11.46)	0-51		--	0.39^{**}	0.46^{***}	0.46^{***}	-0.16	-0.08	0.25^{*}	0.04	-0.16
3. Child chronic pain status	Yes: *n*=24 (33.33%) No: *n*=48 (66.67%)				--	0.31^{**}	0.31^{*}	-0.09	-0.01	0.13	0.00	-0.16
4. Child self-reported internalizing problems	51.06 (10.99)	36-79				--	0.58^{***}	-0.14	-0.01	0.29^{*}	-0.05	-0.23
5. Parent-reported child internalizing problems	57.74 (14.40)	36-104					--	-0.01	0.10	0.23	0.07	-0.21
6. Observed parent attending talk	0.48 (0.30)	0-1.00						--	0.59^{***}	0.24^{*}	0.28^{*}	-0.14
7. Observed child symptom complaints	0.52 (0.31)	0-1.00							--	0.44^{***}	0.47^{***}	-0.35^{**}
8. Child self-reported average CPT pain intensity	3.72 (2.87)	0-10								--	0.30^{*}	-0.40^{***}
9. Parent-reported child average CPT	3.58 (2.42)	0-10									--	-0.47^{***}

pain intensity												
10. Child CPT tolerance	Reached ceiling (240 sec): *n*=32 (44.44%) Did not reach ceiling: *n*=40 (55.56%)											--

* $p < 0.05$; ** $p < 0.01$; ***$p < 0.001$

Child- and parent-reported child internalizing symptoms are presented as T scores. Observed parent attending talk and observed child symptom complaints are presented as the proportion of total utterances the individual engaged in during the CPT. Children were classified as having chronic pain if their interview answers indicated they had experienced pain (other than muscle pain due to physical activity) at least once per week for at least three months. Given the bimodal distribution of child CPT tolerance, this variable was dichotomized; children were classified as either reaching the ceiling tolerance (240 seconds) or not.

Table 3.9.3 Tests of the Indirect Effect in Each Mediation Model

Indirect effect	Unstandardized estimate of indirect effect (B)	Standardized estimate of indirect effect (β)	Standard error	95% bias-corrected bootstrapped confidence interval (lower, upper)
Parent modeling mechanism				
Parent pain interference -> Child pain catastrophizing -> Child chronic pain status	0.01	0.09	0.01	(0.00, 0.03)
Parent pain interference -> Child pain catastrophizing -> Child self-reported internalizing problems	0.12	0.09	0.07	(0.01, 0.29)
Parent pain interference -> Child pain catastrophizing -> Parent-reported child internalizing problems	0.16	0.09	0.09	(0.03, 0.40)
Parent reinforcement mechanism				
Parent attending talk -> Child symptom complaints -> Child self-reported average CPT pain intensity	2.58	0.27	1.09	(1.03, 5.31)
Parent attending talk -> Child symptom complaints -> Parent-reported child average CPT pain intensity	2.18	0.27	0.83	(0.93, 4.27)
Parent attending talk -> Child symptom complaints -> Observed child CPT tolerance	-1.02	-0.30	0.38	(-1.92, -0.42)

CHAPTER 4: CHILD CATASTROPHIZING ABOUT PARENT CHRONIC PAIN: A POTENTIAL CHILD VULNERABILITY FACTOR

The manuscript based on this study is presented below. Kristen Higgins, under the supervision of Dr. Christine Chambers, developed the research questions, methodology and data analysis approaches, and obtained ethical approval and funding. Ms. Higgins developed data collection procedures, contributed substantially to completing data collection, and oversaw staff who contributed to these activities. She conducted the data analyses and wrote the manuscript. Co-authors reviewed the manuscript and provided feedback, which Ms. Higgins incorporated before submitting the manuscript to *Health Psychology* on March 12, 2019. The current reference for this manuscript is:

Higgins, K. S., Chambers, C. T., Rosen, N. O., Sherry, S., Mohammadi, S., Lynch, M., Campbell-Yeo, M., & Clark, A. J. (Submitted). Child Catastrophizing About Parent Chronic Pain: A Potential Child Vulnerability Factor. *Health Psychology.*

4.1 Abstract

Objective: Robust evidence suggests children's catastrophizing about their own pain is a risk factor for poor child pain-related outcomes. In children of parents with chronic pain, child catastrophizing about their parents' pain might be a unique predictor of child pain-related outcomes given their increased exposure to parental chronic pain and disability. The objective of this study was to examine associations between child and parent catastrophizing about their own and each other's pain and child and parent pain-related outcomes. **Methods:** Seventy-two parents with chronic pain and their children (ages 8-15) completed questionnaires assessing their trait catastrophizing about their own and each other's pain, their own pain, and the child's internalizing symptoms. Children completed the cold pressor task (CPT) in the presence of their parent. Parents and children rated children's worst pain intensity and their own anxiety during the task. Analyses were guided by the Actor-Partner Interdependence Model. **Results:** Greater child catastrophizing about parent pain was predicted by children's and parents' increased catastrophizing about their own pain. Child catastrophizing about parent pain predicted greater child- and parent-reported child internalizing symptoms and greater CPT pain intensity for the child, but not parent/child usual pain or CPT anxiety, over and above the influence of parent and child catastrophizing about their own pain.
Conclusions: Child catastrophizing about parent pain is a potential vulnerability factor associated with poor pain-related outcomes in children of parents with chronic pain that should be considered in future research and clinical settings.

Keywords: Catastrophizing, Chronic Pain, Parent-Child Relations, Child, Adolescent

4.2 Introduction

Chronic pain is a common health problem in children and adults (Johannes, Le, Zhou, Johnston, & Dworkin, 2010; King et al., 2011) that tends to aggregate in families (Hoftun et al., 2013). A large population-based study found that the prevalence of chronic pain was 43.2% in offspring with one parent with chronic pain and 50.9% when both parents had chronic pain (Hoftun et al., 2013). Children of parents with chronic pain have been identified as a population particularly vulnerable to problems with pain and mental health (e.g., internalizing symptoms; Higgins et al., 2015). Improved understanding of risk and protective factors associated with child outcomes is needed to support this vulnerable population.

A recently proposed theoretical model of the intergenerational transmission of risk for chronic pain posits that several mechanisms account for the transmission of risk from parents to children (e.g., genetics, early neurobiological development, social learning), resulting in children developing characteristics that make them more vulnerable to pain and psychological disorders (e.g., altered pain processing, pain-related cognitions; Stone & Wilson, 2016). Preliminary support for the social learning (Stone et al., 2018) and neurobiological development (Cservenka, Stein, Wilson, & Nagel, 2015) mechanisms has been found. Pain-related cognitions are one class of child vulnerability characteristics through which parental chronic pain might impact children (Stone & Wilson, 2016). Pain catastrophizing, the tendency to view (presently occurring or future) pain in an exaggerated, negative way (Sullivan, 1995) is a pain-related cognition which may act as a child vulnerability factor in the context of parental chronic pain (Stone & Wilson, 2016). The current study examines a novel form of pain catastrophizing, child catastrophizing about parent pain, and its associations with pain-related outcomes in children of parents with chronic pain.

Much research has examined the intrapersonal influences of pain catastrophizing. Robust evidence of the role of increased catastrophizing about one's own pain in predicting poorer pain and mental health outcomes is found in adults and children with and without chronic pain (Birnie et al., 2016; Buenaver et al., 2008; Craner et al., 2017; Fisher et al., 2017). Catastrophizing about one's own pain can also have interpersonal effects. Increased parent catastrophizing about their own pain is associated with greater child pain intensity one year after major surgery (Pagé et al., 2013) and greater child attentional avoidance of pain-related stimuli (Vervoort, Trost, & Van Ryckeghem, 2013). Increased child catastrophizing about child pain predict parents' reports of increased child pain intensity (Birnie et al., 2016). Catastrophizing about one's own pain predicts one's own pain-related outcomes beyond the influence of similar constructs such as neuroticism, negative affectivity, and anxiety (Goubert et al., 2004; Tran et al., 2015; Vervoort et al., 2006).

Intra- and interpersonal effects of catastrophizing about another person's pain have also been found. A large body of research has examined the impact of parental catastrophizing about child pain on child and parent outcomes in children with and without chronic pain. For example, increased parent catastrophizing about child pain has been associated with higher parent ratings of child pain (Birnie et al., 2016; Goubert et al., 2009), higher child pain catastrophizing, pain-related disability, and depressive symptoms (Goubert et al., 2006; Wilson et al., 2014), greater self-oriented distress in response to imagining their child in pain (Goubert et al., 2008), and greater parental depression, anxiety, and stress associated with parenting a child with a chronic illness (Goubert et al., 2006). In parents with and without chronic pain, parent catastrophizing about child pain has been associated with increased parental protective responses to child pain (Wilson & Fales, 2015). Increased number of parent chronic pain locations has been associated with increased parent and child catastrophizing about child pain (Wilson et al., 2014). Overall, higher levels of parent catastrophizing about child pain have been associated with poorer pain-related outcomes in children and psychological outcomes in parents.

While research has established the intra- and interpersonal effects of parent catastrophizing about child pain, child catastrophizing about parent pain has not previously been studied. Child catastrophizing about parent pain may confer risk to children through similar mechanisms as catastrophizing about their own pain, but may be increased in this population compared to children of parents without chronic pain given their increased exposure to parental pain and disability. This exposure may result in additional opportunities to develop negative pain-related cognitions through social learning mechanisms (Stone et al., 2018; Stone & Wilson, 2016), particularly if exposed to increased parent catastrophizing about their own pain. Thus, child catastrophizing about parent pain may be a factor through which children of parents with chronic pain become vulnerable to pain and psychological difficulties (Stone & Wilson, 2016).

The aims of the current study were to assess child catastrophizing about parent pain in a sample of children of parents with chronic pain, explore associations between child and parent catastrophizing about their own and one another’s pain, and examine the dyadic relationships between child and parent catastrophizing about one another’s pain and parent and child pain-related outcomes. Primary study hypotheses were that 1) parent's and children's catastrophizing about their own pain would predict their catastrophizing about each other's pain and 2) higher levels of child catastrophizing about parent pain would be associated with greater child pain (over the previous three months and during a laboratory task) and internalizing symptoms, beyond the influence of catastrophizing about one's own pain. The examined variables are interdependent and were tested within a dyadic model, allowing for testing of secondary hypotheses regarding associations between parent catastrophizing about child pain, parent and child catastrophizing about their own pain, and parent and child pain-related outcomes. It was hypothesized that higher levels of parent catastrophizing about child pain would be associated with greater child pain and greater parent anxiety during the child's laboratory pain task, and higher parent and child catastrophizing about their own pain would be associated with greater parent and child pain over the previous three months.

4.3 Methods

This study was conducted as part of a larger study of parents with chronic pain and their children examining two distinct research questions. The current paper describes the examination of child catastrophizing about parent pain as a potential vulnerability factor for child pain and psychological outcomes. The other paper (Higgins et al., in preparation) describes tests of specific social learning mechanisms of the intergenerational transmission of risk for chronic pain. All study procedures were approved by the IWK Health Centre Research Ethics Board with reciprocal approval from the Nova Scotia Health Authority Research Ethics Board.

4.3.1 Participants

Participants were 72 parent-child dyads in which the parent experienced chronic pain (for six months or more with at least moderate bodily pain over the past four weeks) and the child was 8 to 15 years old. Parents were recruited from an adult pain clinic (*n* = 50) or using community recruitment methods (*n* = 22; posters, social media). Parents and children were ineligible to participate if their ability to consent/assent to or participate in study tasks was affected by cognitive impairments or developmental disabilities, uncorrected hearing/vision impairments, English language difficulties, or if they did not live together at least 50% of the time. Children were also ineligible if they had contraindications for the cold pressor task (CPT; e.g., blood or circulation disorders, current injury or history of frostbite to non-dominant arm).

4.3.2 Measures

Child Measures.

Catastrophizing about Own Pain. Children completed the Pain Catastrophizing Scale for Children (PCS-C) (Crombez et al., 2003), containing 13 items answered on a scale from “Not at all” (0 points) to “Extremely” (4 points). Total scores can range from 0 to 52; higher scores indicate greater catastrophizing about one’s pain. Evidence suggests the measure is reliable and valid in children with and without chronic pain (Crombez et al., 2003; Fisher et al., 2017). Internal consistency in the current study was Cronbach’s α = .92.

Catastrophizing about Parent Pain. Children completed a modified version of the Pain Catastrophizing Scale – Parent Version (Goubert et al., 2006). The stem of all questions was modified from “When my child is in pain…” to “When my parent is in pain…”. The questionnaire includes 13 items answered on the same 5-point scale as the previous measure (total scores 0-52). While the original measure has been found to be reliable and valid in parents of children with and without chronic pain (Goubert et al., 2006), the current study presents the first known use of this questionnaire in children. Exploratory factor analysis was used to examine the structure of the modified measure in this sample (see Appendix A). Results indicated that the items loaded on three highly correlated factors. In line with prior research on the PCS and related measures (Fisher et al., 2017; Pagé et al., 2013), total scores were used in analyses. Internal consistency in the current study was Cronbach’s α = .94.

Relationship with Parent. Children were asked “How close would you say your relationship is with your parent?”, and “How important to you is your relationship with your parent?”. Both questions were answered on an 11-point numerical rating scale (NRS) from 0 (“not close/important at all”) to 10 (“very close/important”). These measures were developed for the current study to be used as potential covariates.

Pain in Previous Three Months. Children completed an interview with a research assistant that was developed for a previous research study (Petter et al., 2013) and followed guidelines for child pain assessment in research (McGrath et al., 2008). The interview began by discussing events over the past three months to establish the timeline (e.g., holidays, school events, etc.). Children were asked about pains they had experienced during that time and identified which type of pain had occurred most often. They reported on the frequency, duration, locations, and usual intensity (0-10 NRS) of their most common pain.

Internalizing Symptoms. Children completed the Behavior Assessment System for Children-2 (Reynolds & Kamphaus, 2004) Self-Report of Personality Child version (ages 8-11 years; 139 items) or Adolescent version (12-15 years; 176 items), with questions answered on true/false and 4-point response scales ("never" to "almost always"). Evidence of its reliability and validity have been found (Reynolds & Kamphaus, 2004). Children's *T* scores (*M* = 50, *SD* = 10) on the Internalizing Problems composite scale, measuring inwardly directed distress (i.e., symptoms of anxiety and depression), were used (Reynolds & Kamphaus, 2004).

Experimental Pain Task Outcomes. Children reported on their worst pain intensity during the CPT using the Faces Pain Scale-Revised (Hicks et al., 2001), an established, validated measure of pain intensity appropriate for children ages 4-16 years (Cohen et al., 2008). It includes six faces anchored from "No pain" (neutral face; 0 points) to "Very much pain" (10 points). Children rated how nervous or anxious they felt "on average, taken all together" during the CPT by marking a line on a visual analogue scale from "Not nervous or anxious at all" (0 cm) to "Most nervous or anxious" (10 cm).

Parent Measures.

Demographics. Parents completed an author-developed demographics questionnaire about themselves (age, sex, racial/ethnic identity, marital status, education level, household income, relationship to their child) and their child (age, sex, racial/ethnic identity). They rated the closeness and importance of their relationship with their participating child using two NRS from 0 (not close/important at all) to 10 (very close/important).

Chronic Pain Characteristics. Parents reported on the location(s), duration, frequency, and usual pain intensity (0-10 NRS) of their most common pain in the previous three months using a questionnaire version of the interview that children completed (Petter et al., 2013). They reported on their pain interference (the extent to which pain interfered with their mental, physical, and social activities) using the PROMIS Pain Interference Short Form 8a. This reliable and valid measure (Amtmann et al., 2010; K. F. Cook et al., 2016) includes eight items answered on a 5-point scale from "Not at all" (1 point) to "Very much" (5 points; total scores 8-40).

Catastrophizing about Own Pain. Parents completed the Pain Catastrophizing Scale (Sullivan, 1995; Sullivan et al., 1995), containing 13 items on a 5-point scale from "Not at all" (0 points) to "All the time" (4 points; total scores 0-52). The reliability and validity of this measure in adults with and without chronic pain has been established (e.g., Osman et al., 2000). Internal consistency in the current study was Cronbach's α = .92.

Catastrophizing about Child Pain. Parents completed the Pain Catastrophizing Scale – Parent Version (Goubert et al., 2006). This measure contains 13 items answered on a 5-point response scale from "Not at all" to "Extremely" (total scores ranging 0-52). It has been found to be reliable and valid in parents of children with and without chronic pain (Goubert et al., 2006). Internal consistency in the current sample was Cronbach's α = .92.

Child Internalizing Symptoms. Parents completed the Parent Rating Scale of the Behavior Assessment System for Children-2 (Reynolds & Kamphaus, 2004) Child version (for children 8-11 years; 160 items) or Adolescent version (12-15 years; 150 items). Items are answered on a 4-point scale from "never" to "almost always". Support for its reliability and validity have been found (Reynolds & Kamphaus, 2004). Parents' *T* scores on the Internalizing Problems composite scale (assessing children's inwardly directed distress symptoms) were used.

Child Experimental Pain Intensity. Parents reported on the child's worst pain intensity during the CPT using the Faces Pain Scale-Revised (Hicks et al., 2001) so that parents and children could report on this construct with the same measure, as in previous work (Birnie et al., 2016). Evidence supports its validity in adults (Ferreira-Valente et al., 2011).

Anxiety During Child Experimental Pain Task. Parents reported on their own level of anxiety "on average, taken all together" during the child's CPT using a 10-cm visual analogue scale ranging from "Not nervous or anxious at all" (0 cm) to "Most nervous or anxious" (10 cm).

4.3.4 Procedure

Children and parents attended one 90-minute laboratory visit. Informed consent and assent procedures were completed with parents and children respectively, including assessing capacity to consent/assent by asking questions about the study purpose and activities. Dyads completed the questionnaires and CPT in a counterbalanced order (questionnaires first: n = 36 (50.00%)). Children and parents were debriefed about the study and each provided with a $20 gift card honorarium. Parents also received compensation for travel expenses ($15 or $30 based on distance traveled), and children received a Junior Scientist Certificate.

Questionnaires. Parents and children completed measures in separate rooms. The research assistant briefly reviewed written instructions for parents before they completed the measures alone, and for children read aloud instructions and recorded interview answers.

Experimental Pain Task. Children completed the cold pressor task (CPT), a safe and ethical laboratory pain task which induces mild-moderate pain (Birnie et al., 2011), following safety guidelines for this task (von Baeyer et al., 2005). Children placed their non-dominant hand in a bath of cold water (10 ± 0.2 °C) up to the wrist and were instructed to keep their hand in as long as they could (maximum of four minutes; von Baeyer et al., 2005), but were informed they could remove it whenever they wanted. Parents sat across from children during the task and dyads were encouraged to speak to each other as they would elsewhere. During the CPT, the research assistant provided instructions via intercom from another room and observed the task via closed circuit television. Afterwards, the research assistant returned to the room and asked parents and children to rate the child's pain intensity and their own anxiety during the task independently.

4.3.5 Data Analysis

Overall missing data was minimal (1.00%). For individuals missing less than 10% of the items on a particular measure ($n = 3$ parents, $n = 3$ children), missing data was handled using Expectation Maximization imputation (Little et al., 2014). Bivariate correlations were conducted between potential covariates (child and parent age, sex, and ratings of relationship closeness and importance; first study task completed) and outcome variables; the only variable that met criteria for inclusion as a covariate (i.e. correlation coefficient ≥ 0.30; Frigon & Laurencelle, 1993) was parent rating of relationship closeness with the outcome variable child self-reported internalizing symptoms ($r = -.37$, $p < .01$).

Bivariate correlations examined relationships between child catastrophizing about parent pain, parent pain characteristics (chronic pain duration, usual pain intensity, pain interference) and child- and parent-reported relationship closeness and importance. Dyadic analyses were conducted using the Actor-Partner Interdependence Model (APIM; Kenny, Kashy, & Cook, 2006) based on path analyses in R lavaan syntax (Rosseel, 2012). APIM estimates actor effects (e.g., the association between child catastrophizing about parent pain and the child's own CPT pain intensity) and partner effects (e.g., the association between parent catastrophizing about child pain and the child's CPT pain intensity) while controlling for the other type of effect (Kenny et al., 2006). Dyads were treated as distinguishable, MLR estimation was used to address any non-normality, and full information maximum likelihood addressed missing data (Little et al., 2014). Based on a power analysis conducted using APIMPowerR (Ackerman & Kenny, 2016), 72 dyads exceeded the required sample size (i.e. 56 dyads) to detect average actor and partner effects of medium effect sizes (0.25) at 80% power. For the first hypothesis, an APIM analysis examined whether child and parent catastrophizing about their own pain predicted catastrophizing about one another's pain. For the second hypothesis, a series of APIM analyses examined whether child and parent catastrophizing about one another's pain predicted outcomes in everyday life (child and parent usual pain intensity of most common pain over the previous three months, child- and parent-reported child internalizing symptoms) and in the context of the CPT (child- and parent-reported child worst pain intensity, child and parent anxiety). Separate models were tested for each outcome variable. Child and parent catastrophizing about their own pain were included as covariates, as they are already known to impact pain-related outcomes. Unstandardized coefficients and standard errors are presented (Kenny et al., 2006).

4.4 Results

Demographics of the sample are in Table 4.8.1. Most parents were mothers, married, and identified as white. All parents were the participating child's biological parent. Children were approximately evenly split between girls and boys and were most often identified by parents as white. Parent and child pain characteristics are in Table 4.8.2. Parents had chronic pain for, on average, over 10 years, and reported moderate pain intensity (Boonstra et al., 2016). One third of the children had chronic pain (pain other than muscle soreness due to physical activity at least once per week for at least three months). Descriptive statistics for study measures are in Table 4.8.3.

4.4.1 Correlational Analyses

Child catastrophizing about parent pain was significantly correlated with parent pain interference ($r = .46, p < .001$), but not parents' chronic pain duration ($r = -.08, p > .05$) or usual pain intensity ($r = .07, p > .05$). Child catastrophizing about parent pain was not significantly associated with parent ($r = -.12, p > .05$) or child ($r = .17, p > .05$) ratings of closeness or parent ratings of relationship importance ($r = .02, p > .05$). Child catastrophizing about parent pain was positively correlated with child ratings of relationship importance ($r = .32, p < .01$).

Correlations among study variables are in Table 4.8.3. Child catastrophizing about parent pain was significantly positively correlated with children's usual pain intensity, their worst CPT pain intensity and anxiety, parent ratings of child worst CPT pain intensity, child- and parent-reported child internalizing symptoms, and child and parent catastrophizing about their own pain.

4.4.2 Hypothesis 1: Catastrophizing (Own Pain) Predicting Catastrophizing (Other's Pain)

It was hypothesized that parents' and children's catastrophizing about their own pain would predict catastrophizing about the other's pain. Significant actor effects were found; greater catastrophizing about one's own pain was predictive of greater catastrophizing about the other's pain for both parents and children (Figure 4.9.1). There was one significant partner effect; parents' higher catastrophizing about own pain predicted higher child catastrophizing about parent pain.

4.4.3 Hypothesis 2: Catastrophizing (Other's Pain) Predicting Study Outcomes

It was hypothesized that higher child catastrophizing about parent pain would be associated with greater child pain intensity and internalizing symptoms, beyond associations with parent and child catastrophizing about their own pain. Secondary hypotheses were that higher parent catastrophizing about child pain would predict greater child pain and internalizing symptoms and greater parent CPT anxiety, and that parents' and children's greater catastrophizing about their own pain would predict their own higher three-month pain intensity.

Results of the APIM analyses examining these hypotheses are in Figures 4.9.2a-c. There were no significant actor ($b_{child} = -.01$, $SE = 0.02$, $p > .05$; $b_{parent} = -.03$, $SE = .03$, $p > .05$) or partner effects on children ($b = .02$, $SE = .02$, $p > .05$) or parents' ($b = .00$, $SE = .02$, $p > .05$) own usual pain intensity of the most common pain over the previous three months. However, both individuals' greater catastrophizing about their own pain significantly predicted their own higher usual pain intensity ($b_{child} = .11$, $SE = .02$, $p < .001$; $b_{parent} = .05$, $SE = 0.02$, $p < 0.05$). In predicting self- and parent-reported child internalizing symptoms, there was a significant positive actor effect of child catastrophizing about parent pain on child internalizing symptoms (Figure 2a). There was also a significant positive partner effect of child catastrophizing about parent pain on parent-reported child internalizing symptoms. Greater child catastrophizing about own pain predicted higher self-reported ($b = .23$, $SE = .11$, $p < .05$) and parent-reported child internalizing symptoms ($b = .34$, $SE = .15$, $p < .05$). The covariate of higher parent relationship closeness predicted lower child self-reported internalizing symptoms ($b = -2.92$, $SE = .90$, $p = .001$).

There were significant actor and partner effects of children's catastrophizing about their parents' pain during the CPT, with higher levels predicting higher child- and parent-reported child worst pain intensity (Figure 4.9.2b). There was a significant negative partner effect of parental catastrophizing about child pain, with higher levels predicting lower child self-reported worst pain intensity. For CPT anxiety, there was a significant actor effect for parents, with greater catastrophizing about child pain predicting greater parent CPT anxiety (Figure 2c). Higher child catastrophizing about own pain predicted higher child CPT anxiety ($b = .09$, $SE = .04$, $p < .05$).

4.5 Discussion

Overall, our study suggests child catastrophizing about parent pain plays a unique role in predicting child pain-related outcomes. Consistent with primary study hypotheses, higher levels of child and parent catastrophizing about their own pain were associated with greater child catastrophizing about parent pain, and greater child catastrophizing about parent pain predicted greater child self- and parent-reported internalizing symptoms and CPT pain intensity, beyond associations with children's and parents' catastrophizing about their own pain. Regarding secondary hypotheses, greater parent catastrophizing about child pain predicted lower child self-reported CPT pain intensity. Child and parent catastrophizing about the child's pain predicted their own anxiety during the CPT. Only child and parent catastrophizing about their own pain predicted their own usual intensity of their most common pain in the previous three months.

These results extend the existing literature on the intra- and interpersonal effects of pain catastrophizing in parents and children. Higher levels of parent catastrophizing about their own pain were associated with greater child catastrophizing about parent pain. Greater parent catastrophizing about their own pain has been associated with negative child pain-related outcomes in other studies (Pagé et al., 2013; Vervoort et al., 2013). As theorized, greater parent catastrophizing about their own pain may influence the behaviours they use in expressing and coping with their own pain, communicating higher threat value of pain to their children (Stone & Wilson, 2016). This may result in children's increased catastrophizing about their parent's pain.

Children's catastrophizing about parent pain predicted child self- and parent-reported outcomes (internalizing symptoms and CPT pain intensity) beyond children's catastrophizing about their own pain. It is theorized that children learn from parents with chronic pain (e.g., through modeling or reinforcement) to appraise their own pain as threatening, and these cognitions make children vulnerable to poor pain-related outcomes (Stone & Wilson, 2016). Observing others in pain is associated with negative observer outcomes including self-oriented distress (e.g., internalizing symptoms; Goubert et al., 2005). This may be particularly evident in children, for whom it is adaptive to appraise situations impacting a parent's ability to meet the child's needs (e.g., chronic pain) as particularly threatening (Mikail, Henderson, & Tasca, 1994). This may result in unique impacts on children beyond cognitions about their own pain.

Consistent with the robust literature on the intrapersonal effects of catastrophizing about one's own pain (Buenaver et al., 2008; Craner et al., 2017; Durand et al., 2017; Fisher et al., 2017), greater parent and child catastrophizing about their own pain was associated with higher usual pain intensity over the previous three months. Child catastrophizing about their own pain was also associated with greater child anxiety during the CPT. Contrary to research in parents of children with and without chronic pain (Birnie et al., 2016; Goubert et al., 2009; Wilson et al., 2014), few interpersonal effects of parental catastrophizing about child pain on child outcomes were found. Only an interpersonal association in the opposite direction from what was expected was found in the context of the CPT, with increased parent catastrophizing about child pain predicting decreased child self-reported worst pain intensity. Different relationships may exist between parent catastrophizing about child pain and child pain outcomes in children of parents with chronic pain compared to other samples. Children have reported hiding their distress to avoid upsetting parents with chronic pain (Evans & de Souza, 2008; Umberger et al., 2015), and may similarly hide their pain, particularly when their parents have higher trait catastrophizing about the child's pain. Alternatively, with higher parent catastrophizing about child pain, children may feel confident that their pain will be recognized and managed, leading to decreased pain expression. Increased parent catastrophizing about child pain has been associated with greater parent-child congruence in rating children’s pain (Goubert et al., 2009).

This study makes an important contribution to the literature on children of parents with chronic pain and the knowledge base on pain catastrophizing. It identifies a new construct of interest and makes use of a modified version of a well-established measure (Goubert et al., 2006). The modified measure showed strong internal consistency and a three-factor structure, and correlations with expected variables (e.g., parent pain interference, child ratings of parent relationship importance) were observed suggesting preliminary evidence of concurrent validity. Strong study methodology was employed, including the use of reliable and valid measures, multi-informant outcome measurement, and use of experimental pain methodology to study children’s immediate (rather than retrospective) pain experiences. Dyadic data analysis techniques were used to appropriately account for non-independence in the parent-child data.

Our study must be interpreted in the context of its limitations. Given the cross-sectional design, directions of the observed relationships cannot be determined. Assessment of child catastrophizing about parent pain used a measure modified from one created for parents to report on their catastrophizing about child pain (Goubert et al., 2006), and further study is needed to determine its validity and factor structure. Only trait catastrophizing was examined, and parents’ and children’s trait and state catastrophizing may have differential effects (Durand et al., 2017). Though the child pain interview (Petter et al., 2013) likely assisted children in establishing the relevant timeline, social desirability may have influenced responses. The sample was homogeneous (mostly mothers and identifying as white) and the generalizability of results to other groups is unknown.

Further study of child catastrophizing about parent chronic pain is needed to better understand this potential vulnerability factor. Prospective, longitudinal studies are needed to clarify the temporal relationships between parent and child catastrophizing about their own and each others' pain and their effects on children's outcomes. Improved understanding of how children's trait catastrophizing develops in the context of parental chronic pain is needed. Child catastrophizing about parent pain is likely impacted by child development, dispositional empathy (Goubert et al., 2005), and parent-child attachment (Donnelly & Jaaniste, 2016). Incorporation of these variables in future work will assist in clarifying their impacts on child pain catastrophizing.

While the role of child catastrophizing about parent pain as a risk factor needs to be clarified, it may be an important target for prevention and intervention programs. Screening for high levels of catastrophizing about a parent's chronic pain could identify children in need of increased support. Interventions to decrease child catastrophizing about parent pain could be developed based on interventions for decreasing catastrophizing about one's own pain (Schütze et al., 2018) or decreasing anxiety in children of parents with other chronic illnesses (e.g., Scholten et al., 2013). Parents have expressed concern about the how their chronic pain impacts their children (Evans & de Souza, 2008; Umberger et al., 2015), and providing education and evidence-based strategies to mitigate these effects could empower parents in improving child outcomes. While further research is needed, child catastrophizing about parental chronic pain is a potential risk factor that could be addressed to improve outcomes in this vulnerable population.

4.6 References

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4.8 Tables

Table 4.8.1 Demographics

	Parent		***Child***	
	Mean (*SD*) or *n* (%)	**Range**	**Mean (*SD*) or *n* (%)**	**Range**
Age	42.91 years (6.49)	28.25-57.25	12.12 years (2.45)	8.01-15.96
Sex	Women: *n* = 57 (79.17%) Men: *n* = 15 (20.83%)		Girls: *n*=39 (54.17%) Boys: *n*=33 (45.83%)	
Race/ethnicity	White: *n* = 64 (88.89%) Bi- or multi-racial: *n* = 4 (5.55%) Other: *n* = 4 (5.55%)		White: *n*=57 (79.17%) Bi- or multi-racial: *n*=9 (12.50%) Other: *n*=6 (8.33%)	
Highest education completed	High school or partial high school: *n* = 12 (16.67%) Trade school or community college: *n* = 27 (37.50%) Some university education: *n* = 16 (22.22%) Undergraduate degree: *n* = 10 (13.89%) Graduate school/professional training: *n* = 7 (9.72%)			
Marital status	Married: *n* = 47 (65.28%) Common-law relationship: *n* = 10 (13.89%) Other committed relationship: *n* = 6 (8.33%) Not in committed relationship: *n* = 9 (12.50%)			
Total annual household income	Less than $10,000: *n*=3 (4.17%) $10,000-$25,000: *n*=7 (9.72%) $25,000-$50,000: *n*=16 (22.22%) $50,000-$75,000: *n*=17 (23.61%) $75,000-$100,000: *n*=9 (12.50%) $100,000-$125,000: *n*=4 (5.56%) $125,000-$150,000: *n*=3 (4.17%) More than $150,000: *n*=7 (11.11%) Prefer not to answer: *n*=6 (8.33%)			
Percentage of time child lived with participating parent	100% of the time: *n*=62 (86.11%) 90% of the time: *n*=4 (5.56%) 50-70% of the time: *n*=4 (5.56%) Not reported: *n*=2 (2.78%)			
Rating of relationship closeness	9.24 (1.20)	4.00-10.00	8.80 (2.04)	0.00-10.00
Rating of importance of relationship	9.96 (0.20)	9.00-10.00	9.39 (1.80)	0.00-10.00

Table 4.8.2 Parent and Child Pain Characteristics

	Mean (*SD*) or *n* (%)	**Range**
Parents		
Duration of having chronic pain	10.92 years (9.46)	0.42-37.60
Time since parent began receiving treatment for their chronic pain	7.94 years (7.89)	0.12-32.48
Usual pain intensity of most common pain over the past 3 months (0-10)	6.94 (1.91)	3.00-10.00
Frequency of most common pain over the past 3 months	More than once per week: *n*=68 (94.44%) About once per week: *n*=2 (2.78%) Once or twice per month or less: *n*=2 (2.78%)	
Pain interference	29.26 (8.15)	10.00-40.00
Children		
Usual pain intensity of most common pain over the past 3 months (0-10)	4.93 (2.08) _*Three children reported not having had any pain over the past 3 months_	1.00-10.00
Frequency of most common pain over the past 3 months	More than once per week: *n*=26 (36.11%) About once per week: *n*=12 (16.67%) Once or twice per month or less: *n*=31 (43.06%) No pain reported in last 3 months: *n*=3 (4.17%)	
Duration of most common pain experienced over the past 3 months	Just this month: *n*=8 (11.11%) Less than three months: *n*=12 (16.67%) Over three months: *n*=20 (27.78%) Over a year: *n*=28 (38.89%) Missing: *n*=1 (1.4%) No pain reported in past 3 months: *n*=3 (4.17%)	
Presence of child chronic pain (i.e. pain other than muscle soreness following physical activity, occurring at least weekly over the previous three months)	Yes: *n*=24 (33.33%) No: *n*=48 (66.67%)	

Table 4.8.3 Descriptive Statistics And Correlations For Study Measures

	Mean (*SD*)	Range	1	2	3	4	5	6	7	8	9	10	11	12
1. Child catastrophizing (parent pain)	28.48 (12.83)	0-52	--	0.58^{***}	0.19	0.24^{*}	0.34^{**}	0.07	0.51^{***}	0.51^{***}	0.31^{**}	0.27^{*}	0.27^{*}	0.17
2. Child catastrophizing (own pain)	18.25 (11.46)	0-51		--	0.07	0.11	0.56^{***}	0.08	0.46^{***}	0.46^{***}	0.26^{*}	0.13	0.42^{***}	0.11
3. Parent catastrophizing (child pain)	20.65 (10.27)	4-47			--	0.56^{***}	0.13	0.00	0.08	0.18	-0.16	0.21	0.10	0.51^{***}
4. Parent catastrophizing (own pain)	21.00 (11.81)	0-47				--	0.13	0.22	0.05	0.24^{*}	0.06	0.14	0.07	0.30^{*}
5. Child usual pain intensity	4.93 (2.08)	1.00-10.00					--	0.24^{*}	0.44^{***}	0.41^{**}	0.16	0.07	0.21	0.07
6. Parent usual pain intensity	6.94 (1.91)	3.00-10.00						--	0.04	0.00	0.23^{*}	0.21	0.11	0.02
7. Child self-reported Internalizing Problems T score	51.06 (10.99)	36-79							--	0.58^{***}	0.20	-0.01	0.27^{*}	0.07
8. Parent-reported child Internalizing Problems T score	57.74 (14.40)	36-104								--	0.14	0.10	0.41^{**}	0.11
9. Child self-reported worst CPT pain intensity	4.89 (2.55)	0-10									--	0.24^{*}	0.33^{**}	-0.06
10. Parent-reported child worst CPT pain intensity	4.81 (2.48)	0-10										--	0.21	0.21
11. Child CPT anxiety	2.78 (2.72)	0.0-10.0cm											--	0.06
12. Parent CPT anxiety	2.74 (2.72)	0.0-9.8cm												--

* $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$. CPT=cold pressor task

4.9 Figures

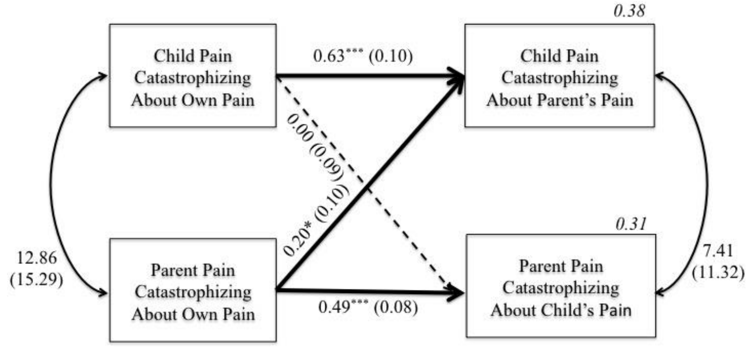

Figure 4.9.1 APIM Analysis of Child and Parent Catastrophizing About Their Own and The Other's Pain. Unstandardized coefficients and standard errors are presented. Bold lines indicate significant effects, dotted lines indicate non-significant effects. $^{*}p < 0.05$, $^{**}p < 0.01$, $^{***}p < 0.001$.

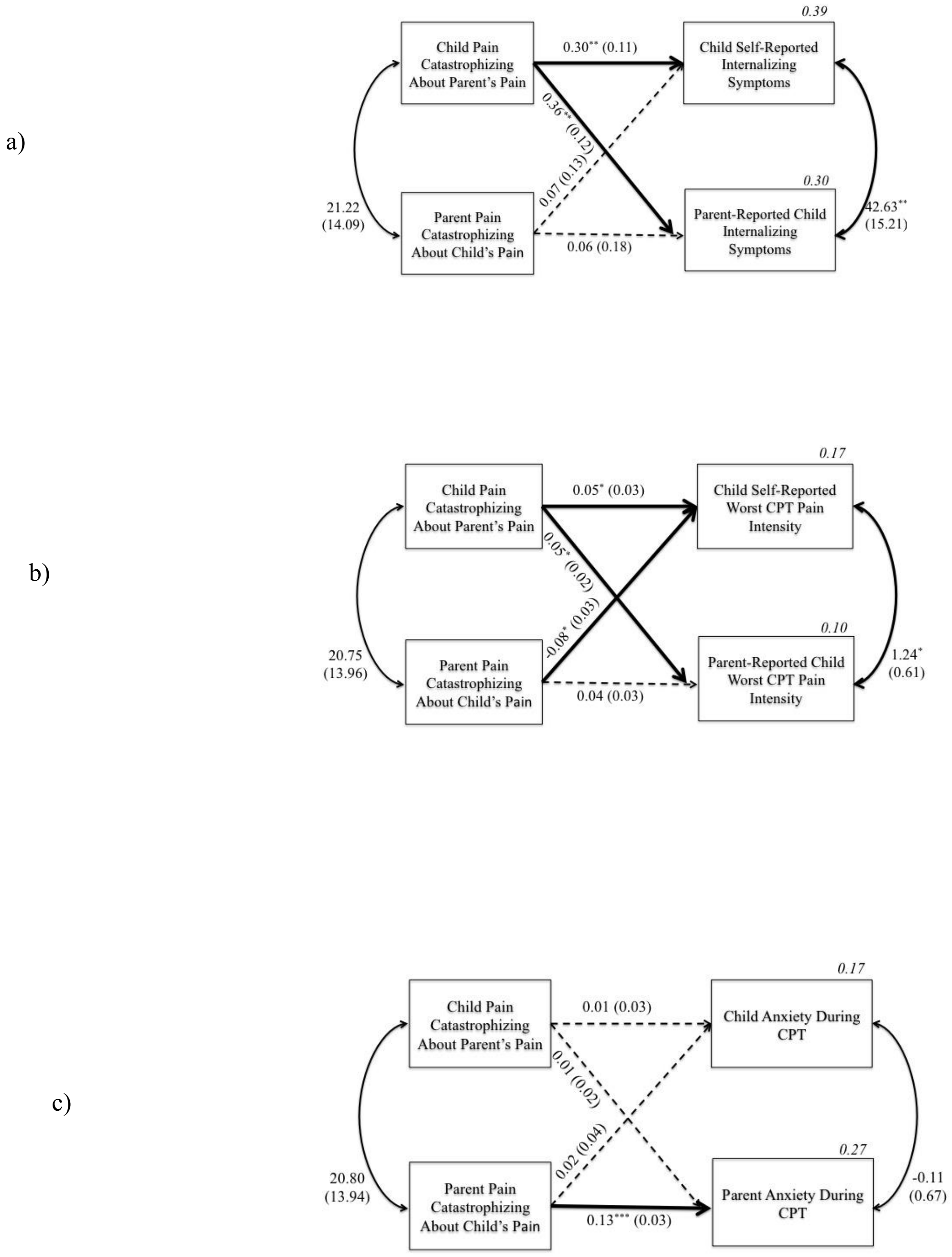

Figure 4.9.2 APIM Analyses of Child and Parent Catastrophizing About Each Other's Pain Predicting Study Outcomes. Unstandardized coefficients and standard errors are presented. Bold lines indicate significant effects, dotted lines indicate non-significant effects. $^{*}p < 0.05$, $^{**}p < 0.01$, $^{***}p < 0.001$

4.10 Supplementary Materials

Appendix A: Exploratory Factor Analysis

Given that the current study presents the first use of the modified Pain Catastrophizing Scale for Parents (PCS-P) in children to report on children's catastrophizing about their parent's pain, exploratory factor analysis (EFA) was used to examine the measure's structure. Best practices for EFA were followed (Costello & Osborne, 2005; Fabrigar & Wegener, 2012; Sakaluk & Short, 2017). Analyses were conducted in SPSS, with factors extracted using maximum likelihood estimation and promax rotation. The decision regarding the number of factors to retain was made using multiple procedures (Fabrigar & Wegener, 2012; Sakaluk & Short, 2017) including parallel analysis with eigenvalues from the reduced correlation matrix (O'Connor, 2000), descriptive model fit indices, nested model comparisons, and theoretical interpretability of resulting factor structures.

Parallel analysis suggested that a one-factor solution would be reasonable. A one-factor model did not adequately fit the data, *RMSEA* = 0.10, *TLI/NNFI* = 0.90 (Little, 2013). A two-factor model was examined, which was a significant improvement in fit compared to the one-factor model, $\Delta \chi^2$ (12) = 29.25, *p* = .004, but did not achieve adequate model fit, *RMSEA* = 0.09, *TLI/NNFI* = 0.92. Finally, a three-factor model was examined which demonstrated a significant improvement over the two-factor model, $\Delta \chi^2$ (11) = 24.94, *p* = .009, and acceptable model fit, *RMSEA* = 0.07, *TLI/NNFI* = 0.95. Factor loadings for each item are presented in Table A1.

The three-factor model produced a theoretically meaningful solution. Six items loaded on Factor 1. These items addressed high pain threat and passive coping with pain;

a similar factor has been identified in adults (referred to as "powerlessness"; Chibnall & Tait, 2005) and children reporting on catastrophizing about their own pain (Pielech et al., 2014). Four items loaded on Factor 2, addressing a lack of ability to cope with pain, similar to the helplessness factor identified in other measures based on the Pain Catastrophizing Scale (Crombez et al., 2003; Goubert et al., 2006; Sullivan et al., 1995). Three items loaded on Factor 3, addressing rumination about the parent's pain. The three factors were highly correlated with one another ($r_{1,2} = 0.80$, $r_{1,3} = 0.71$, $r_{2,3} = 0.73$) and were each internally consistent (Cronbach's $\alpha_1 = 0.89$, Cronbach's $\alpha_2 = 0.84$, Cronbach's $\alpha_3 = 0.87$).

Overall, the EFA results indicating three correlated factors support the use of the measure's total scores in the analyses throughout the current paper. These results should be interpreted with caution given the small sample size used (Costello & Osborne, 2005; Sakaluk & Short, 2017) and should be replicated in future studies.

Table A1. Factor loadings for each item of the modified PCS-P in the current sample.

When my parent is in pain…	Factor 1	Factor 2	Factor 3
1. I worry all the time about whether the pain will end.	**0.49**	0.28	0.03
2. I feel like I can't go on like this much longer.	0.02	**0.76**	0.04
3. It's terrible and I think it's never going to get better.	**0.83**	0.18	-0.21
4. It's awful and I feel that it overwhelms me.	0.00	**0.71**	0.23
5. I can't stand it anymore.	0.14	**0.80**	-0.13
6. I become afraid that the pain will get worse.	**0.89**	-0.06	0.17
7. I keep thinking of other painful events.	**0.37**	0.16	0.30
8. I want the pain to go away.	0.06	**0.42**	0.10
9. I can't keep it out of my mind.	-0.05	-0.04	**1.07**
10. I keep thinking about how much he/she is suffering.	0.29	0.16	**0.40**
11. I keep thinking about how much I want the pain to stop.	0.10	0.36	**0.42**
12. There is nothing I can do to stop the pain.	**0.53**	-0.01	0.08
13. I wonder whether something serious may happen.	**0.64**	0.02	0.09

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CHAPTER 5: GENERAL DISCUSSION

The goals of the current dissertation were to synthesize the existing literature on the impact of parental chronic pain on offspring outcomes and to test theorized pathways through which parental chronic pain might impact children's pain and mental health outcomes. The first goal was accomplished by conducting a comprehensive, mixed-methods systematic review of studies examining the impact of parental chronic pain on offspring outcomes in the areas of pain, health, psychological, and family outcomes (described in Chapter 2). Meta-analysis and meta-ethnography methods were used to synthesize results from the included studies. The second goal of the dissertation was addressed in Chapters 3 and 4. In Chapter 3, two social learning pathways (parental modeling of pain behaviour and parental reinforcement of child pain) that might underlie the relationships between parental chronic pain and child pain and internalizing outcomes as hypothesized by the intergenerational model of transmission of risk from parents with chronic pain to their children (Stone & Wilson, 2016) were tested. These relationships were examined based on questionnaire data from parents and children describing trait characteristics and pain experiences in their everyday life, as well as observational, self-report, and informant-report data collected while the child completed the cold pressor task (CPT) in the presence of their parent. Chapter 4 examined the novel construct of child catastrophizing about a parent's chronic pain and its relationship with children's pain and internalizing symptoms. Dyadic analysis using the Actor Partner Interdependence Model (W. Cook & Kenny, 2005) was used to examine these relationships, taking into account the non-independence of parent and child variables.

The current chapter discusses key findings across Chapters 2 to 4 and contextualizes these results within existing research. Subsequent sections provide discussion of theoretical and clinical implications of the results as well as strengths and limitations of the included studies. Directions for future research to expand current knowledge of relationships between parental pain and child pain and mental health outcomes are also provided.

5.1 Discussion of Key Findings And Integration With Existing Research

5.1.1 Parental Chronic Pain And Offspring Outcomes

As described in Chapter 2, meta-analyses of results from quantitative (population-based and clinical) studies indicated that parental chronic pain was associated with poorer offspring outcomes in each area as compared to offspring of parents without chronic pain, including increased rates of pain and adverse birth outcomes, greater internalizing and externalizing problems as reported by parents and offspring, poorer social competence, and poorer family functioning. Synthesis of results of qualitative studies using meta-ethnography indicated that parental chronic pain was associated with not only negative outcomes in offspring, such as missing out on aspects of normal childhood/adolescence, difficult emotions, and struggles communicating with parents, but also with positive offspring outcomes, including the development of compassion and learning about their own health and coping. Taken together, the results suggest that offspring of parents with chronic pain are a vulnerable population whose needs for intervention should be examined in future research, including ways to potentially build on constructive aspects of parental chronic pain. This comprehensive, mixed methods review and synthesis provides an important contribution to the literature in this area by clarifying the relationship between parental chronic pain and each category of offspring outcomes, as previous individual studies had found mixed results. The methodology utilized in Chapter 2 allowed for results to be synthesized across studies and overall conclusions to be generated using statistical and qualitative synthesis methods. A key strength of this study was the inclusion of a variety of study designs, which highlighted that positive outcomes associated with parental chronic pain may have been overlooked if only quantitative study designs had been included. The need to move beyond simply comparing groups of offspring with and without parental chronic pain exposure and to examine factors that may underlie the relationships between parental chronic pain and offspring outcomes was identified.

Given that the review and synthesis described in Chapter 2 were conducted several years ago, a potential limitation is that the results of the review may not reflect current research in this area. To address this, the systematic search completed for Chapter 2 was repeated on July 24, 2018. The same screening process was used, and 39 new relevant articles were identified (30 population-based, seven clinical, two qualitative). Several of the new population-based studies identified examined the same samples as studies included in the original review (Kaasbøll, Ranøyen, Nilsen, Lydersen, & Indredavik, 2015; Lier, Nilsen, Vasseljen, & Mork, 2015; Lier, Nilsen, & Mork, 2014; Zadro et al., 2018) or as one another (Rom et al., 2016, 2017, 2018). The newly identified studies generally support the same conclusions as in the original review, with population-based and clinical studies identifying relationships between parental pain and poorer offspring outcomes in the areas of pain (e.g., Buse et al., 2018; Kuo et al., 2015; Lier, Mork, Holtermann, & Nilsen, 2016; Lier, Nilsen, Vasseljen, & Mork, 2015; Lier, Nilsen,

& Mork, 2014; Wilson & Fales, 2015), health (e.g., Bröms et al., 2016; Carman, Accortt, Anthony, Iles, & Enger, 2017; Magnus et al., 2016), psychological (e.g., Instanes et al., 2017; Kaasbøll, Ranøyen, Nilsen, Lydersen, & Indredavik, 2015) and family outcomes (e.g., Bicking Kinsey, Baptiste-Roberts, Zhu, & Kjerulff, 2014; Kaasbøll et al., 2015). Few studies found evidence of no differences between offspring of parents with and without chronic pain (e.g., Pillemer, Riffin, Suitor, Peng, & Reid, 2017; Posfai, Banhidy, Urban, & Czeizel, 2015). The two qualitative studies identified included a secondary analysis of the same sample of young adult children of parents with chronic pain included in the original review (Umberger et al., 2015) and a study of adult children who were caregivers for their parents with osteoarthritis (Barker, Minns Lowe, & Toye, 2017). Both studies identified themes consistent with the results of the meta-ethnography in Chapter 2, including missing out on normal developmental activities (e.g., sense of loss of childhood, missing out on social activities and work and family responsibilities), negative impacts on emotional health (e.g., blaming self for parent's difficulties, distress associated with caregiving imbalances in the family), and development of compassion and empathy (e.g., positive feelings arising from their caregiving roles). Overall, the additional studies identified in the updated search use similar methodology to the originally included studies and are consistent with the results of the original review, and review methodology has not significantly changed since the publication. These factors suggest that an update to the conducted syntheses is not needed at this time based on decision-making frameworks for determining when to update systematic reviews (Garner et al., 2016) and meta-ethnographies (France, Wells, Lang, & Williams, 2016). Additionally, given the large sample sizes included in many of the meta-analyses and the critical sample size needed for stable correlation estimates (typically >250; Schönbrodt & Perugini, 2013), the results of many of the meta-analyses would be unlikely to change unless newly added studies showed inconsistent effects with existing research and very large sample sizes. However, given the rate of publication of new studies in this area, the possibility of an update should likely be reconsidered in the future (e.g., five years after publication of the review).

As discussed in Chapter 2, the specificity of the relationship between parental chronic pain and offspring outcomes needs to be clarified. Recent work has aimed to address this gap by studying other factors that may explain or contribute to this relationship, such as parental exposure to pain medications, parental pain interference, or comorbid parental mental health conditions. Recent research on parental medication exposure suggests that this variable does not fully explain the relationship between parental pain and offspring outcomes (Bröms et al., 2016; Carman et al., 2017; Magnus et al., 2016). The association between parental pain interference and child pain and mental health outcomes was examined in Chapter 3. The role of comorbid parental mental health conditions has begun to be examined; support for the role of parent psychological health (anxiety and depressive symptoms, parent catastrophizing about child) and chronic pain in predicting child pain, functioning, and psychological health in children with chronic pain was found (Poppert Cordts, Stone, Beveridge, Wilson, & Noel, 2019). Higher levels of parent depressive symptoms have also been associated with higher levels of parent catastrophizing about their child’s pain and protective responses to their child’s pain, beyond the influence of parental chronic pain status (Fussner et al., 2018). Chapter 2 identified important knowledge gaps in the literature on the relationship between parental chronic pain and offspring outcomes, and these areas continue to be explored in the broader literature. Chapter 3 built on this work by further clarifying the mechanisms through which the relationship between parental chronic pain and offspring outcomes might occur, and specifically by addressing the knowledge gap regarding the role of parental pain interference in predicting child outcomes.

5.1.2 Social learning mechanisms in the relationship between parental chronic pain and offspring outcomes.

Chapter 3 examined two pathways (parental modeling of pain behaviour and parental reinforcement of child pain) that might underlie the relationships between parental chronic pain and child pain and internalizing outcomes as hypothesized by the intergenerational model of transmission of risk from parents with chronic pain to their children (Stone & Wilson, 2016). As indicated by the results of the systematic review and synthesis in Chapter 2, little research thus far has focused on identifying factors underlying the relationship between parental chronic pain and children's pain and mental health outcomes. Chapter 3 makes an important contribution to the literature by directly examining theorized pathways in a sample of parents with chronic pain and their children (ages 8-15 years), incorporating multi-method (questionnaire, laboratory pain task) and multi-informant (child, parent, observational coding) assessment.

Results supported the parental modeling pathway for the outcome of children's internalizing symptoms, with significant indirect effects being found between greater parental pain interference and greater children's internalizing symptoms (as reported by both children and parents) through children's higher levels of trait catastrophizing about their own pain. However, this pathway was not supported for the outcome of the presence of children’s own recurrent pain. Support for the parental reinforcement pathway was found during the CPT, with significant indirect effects of parental attending talk (i.e. the proportion of their verbalizations during the task focused on children’s pain) on children’s pain outcomes (observed pain tolerance, pain intensity as reported by children and parents) through children’s symptom complaints (i.e. the proportion of their verbalizations focused on their own pain/cold symptoms) during the task. These results identify potential pathways through which child pain and internalizing symptoms may be impacted by parental pain, which could be further assessed in future studies to better understand the direction and temporal aspects of these relationships.

A novel finding of the current dissertation is support for the parental pain modeling pathway in children’s (self- and parent-reported) internalizing symptoms. As described in Chapter 2, an association between the presence of parental chronic pain and increased offspring internalizing symptoms has been established. Recently, a large population-based study of adolescents and their parents found support for this relationship only in adolescents for whom both parents had chronic pain, and found that adolescent social competence mediated this relationship in girls but not in boys (Kaasbøll et al., 2018). However, the relationship between parental pain interference, representing a possible proxy for parent pain behaviour, and child internalizing symptoms has been less frequently studied and mixed results have been identified. One study found no significant differences in child internalizing symptoms between children of parents with high versus low pain-related interference (K. B. Smith & Chambers, 2006). Another study found that lower maternal self-reported physical functioning due to pain (i.e. limitations in physical functioning due to pain demands) was associated with greater mother-reported child internalizing symptoms through the mediator of mothers' self-reported use of over-reactive parenting behaviours (Evans et al., 2006). This relationship was not seen for the outcome of child self-reported depressive symptoms; however, the authors evaluated the indirect effect using Sobel's test, a highly conservative method which is generally not recommended except in sample sizes above 500 (Hayes & Scharkow, 2013). A third study examined a structural equation model for predicting child pain, functioning, and psychological health (including child self-reported anxiety, depressive symptoms, and catastrophizing about their own pain) that included parent physical functioning (a latent factor represented by parent self-reported pain interference and physical functioning), parent psychological health (including anxiety, depressive symptoms, and catastrophizing about child pain), parent chronic pain features (e.g., chronic pain status, pain intensity) and parent behavioural responses to child pain (self-reported parent protectiveness and monitoring behaviour in response to child pain) (Poppert Cordts et al., 2019). The authors found that parent physical functioning predicted child outcomes in this sample of children with chronic pain (63.2% with headaches as primary pain complaint) and their parents; all other parental factors except behavioural responses to child pain also significantly predicted outcomes in this model. Building on the previous research in this area, Chapter 3 provided a unique contribution to the literature by specifically testing a hypothesized parental modeling pathway, representing the modeling of parent behaviour (with the proxy of parental pain interference) and the hypothesized child vulnerability factor of child catastrophizing about their own pain (Stone & Wilson, 2016).

The finding in Chapter 3 that the parental modeling mechanism was not supported for the outcome of child recurrent pain status can be contextualized amongst the mixed results that have been found in previous studies of this topic. Several such studies have examined samples of children with chronic pain and their parents (Poppert Cordts et al., 2019; Schanberg et al., 2001; Stone et al., 2018; Stone & Walker, 2017). Schanberg and colleagues (2001) found that greater self-reported parental pain interference was associated with greater child self-reported pain intensity. However, when examining child pain catastrophizing as a mediator in this relationship, different results were found. They found that a type of parent pain interference, i.e. changes in parents' ability to work due to pain, was not associated with increased child pain through the mediator of child pain catastrophizing, consistent with the results of the current dissertation. In studies of adolescents with recurrent abdominal pain and their parents (Stone et al., 2018; Stone & Walker, 2017), the parental modeling mechanism has been supported in predicting adolescent pain outcomes. As described above, Poppert Cordts and colleagues (2019) found support for parent physical functioning (including pain interference) predicting child psychological health; however, in this model child pain catastrophizing was included as an outcome rather than a mediator. These studies examined samples of children with chronic pain and their parents, while the current dissertation examined parents with chronic pain and their children. It is possible that parental pain modeling may be more salient in children who already have chronic pain themselves; that is, modeling may play a greater role in the maintenance of children's chronic pain or their coping with chronic pain, rather than with the development or presence of children's chronic pain.

Several additional factors may explain the mixed results observed across studies in the role of parental pain modeling in predicting children's pain. While self-reported parental pain interference has been found to be highly correlated with adolescent-reported parental pain behaviour (Stone & Walker, 2017) and has been used as part of a proxy measure of parental pain behaviour in previous research (Poppert Cordts et al., 2019), the current dissertation did not directly measure either pain behaviour engaged in by parents or children's observations of parents' pain behaviour. Self-reported parental pain interference may not adequately capture the amount of pain behaviour that a parent displays (e.g., due to self-report bias or retrospective reporting bias), particularly at extreme ends of the spectrum; for example, both parents with very low and very high pain interference might model less pain behaviour to their children, either because they rarely demonstrate pain behaviours or because their pain is so interfering that they are less available to interact with their children and display such behaviours. Parent-specific factors may also influence the amount and types of pain behaviours that parents model to children and children's interpretation of these behaviours. Parents' own coping with chronic pain (e.g., catastrophizing about own pain; acceptance of their chronic pain) may impact the messages children receive from observing their parent experiencing pain. Parent mental health likely impacts the display of pain behaviour as well. While adolescents' reports of parent pain behaviours are more strongly correlated with parent self-reported pain interference than with parent mental health (Stone & Walker, 2017), it is likely that parents' mental health impacts their functioning, and children may have difficulty differentiating between the effects of chronic pain and mental health on their parents' functioning.

Child-specific factors such as sex, developmental stage, and dispositional empathy may influence the effect of parental pain modeling on children's own pain.

Studies of parental pain modeling suggest that children may be more influenced by a same-sex parent with chronic pain (Evans et al., 2010) and that parental pain modeling may impact girls' pain experiences more than boys (Boerner, Chambers, McGrath, LoLordo, & Uher, 2017). Developmental stage may also impact children's awareness of parent pain behaviours and attribution of such behaviours to parents' chronic pain. The observation of another person's pain is thought to be a complex interaction influenced by the pain expression of the person in pain, inferences made by the observer (which may be impacted by characteristics of both the observer and the person in pain), and the observer's emotional and behavioural responses (Goubert et al., 2005; Hadjistavropoulos et al., 2011). A theoretical model of the impact of observing others' pain on observers themselves (Goubert et al., 2005) suggests that there are both bottom-up (e.g., verbal and facial cues of the person in pain, age of the person in pain, contextual information about the pain stimulus) and top-down (e.g., observer's own pain catastrophizing, learning experiences regarding pain) influences on an observer's understanding of another person's pain experiences. These factors may have impacts on the observer's affective responses (e.g., self-oriented distress, other-oriented sympathy), which in turn may influence their behavioural responses to the person in pain (Goubert et al., 2005). All of these factors are hypothesized to be impacted by contextual factors (e.g., the relationship between the observer and the person in pain, informational priming; Gagnon, Hadjistavropoulos, & MacNab, 2017; Goubert et al., 2005; Hampton, Hadjistavropoulos, & Gagnon, 2018). Research testing this model has shown that aspects of parents' self-reported dispositional empathy plays a role in predicting their responses to reading vignettes about their child being in pain; specifically, higher parent personal distress (tendency to experience distress/discomfort in response to others' distress) and lower empathic concern (tendency to experience sympathy and compassion for others in need) predicted increased self-oriented emotional responses (e.g., feeling worried, anxious, sad) in response to the pain vignettes about their child (Goubert et al., 2008). While these processes have not yet been examined in children of parents with chronic pain, children's dispositional empathy may also influence the impact of observing parent pain on their own experiences with pain and psychological wellbeing.

Aspects of the parent-child relationship likely impact the effects of parental pain modeling on children and were not assessed in the current dissertation. For example, attachment style is theorized to impact children's development and maintenance of chronic pain as well as their choice of strategies for coping with pain (Donnelly & Jaaniste, 2016). It is hypothesized to influence the appraisal of pain threat, of one's ability to cope with pain, and of other's abilities to provide effective support, and insecure attachment has been associated with negative pain-related outcomes in adults and children (Donnelly & Jaaniste, 2016; Laird, Preacher, & Walker, 2015; Meredith, Ownsworth, & Strong, 2008). Parents' chronic pain may negatively impact parent-child attachment (e.g., making parents less available or able to consistently meet children's needs). Insecure child attachment may influence the information that children learn from observing parental pain modeling (e.g., regarding how to cope with pain, the availability of others to provide pain coping support), and it has been shown to moderate the relationship between child catastrophizing and parent responses to child pain (Vervoort, Goubert, & Crombez, 2010). Parent-child communication about pain also likely impacts how parental pain modeling affects children. Qualitative research with offspring of parents with chronic pain has shown that when parents with chronic pain or their adolescents are “shrouded” (i.e. emotionally unavailable to one another, unable to openly communicate about the parent’s pain and its impacts on the adolescent and family), it is associated with negative impacts on the adolescent and the relationship, and “unshrouding” can be a key turning point in improving the relationship (Umberger et al., 2013). Further analysis from the same sample identified understanding chronic pain and communicating with parents as key challenges these adolescents faced in coping with chronic pain (Umberger et al., 2015). Reduced emotional closeness between parents and children, as well as poor child understanding of parental chronic pain, may change the effect that parents’ modeling of pain behaviours has on children. If children have a realistic understanding of how pain impacts their parent and there is emotional closeness in the relationship allowing for such issues to be discussed, children may be less negatively impacted by parental pain modeling (e.g., exposure to parents’ pain may be interpreted less negatively; children may be able to seek parental support to regulate associated distress).

As described in Chapter 3, the hypothesized parental reinforcement pathway was supported in the context of the CPT, with parent pain-attending verbalizations predicting child pain outcomes (child- and parent-reported child pain intensity, observed child pain tolerance) through the mediator of child pain-attending verbalizations. These results are consistent with many previous studies of parental responses to children’s acute (Campbell et al., 2017; Evans et al., 2016; Moon et al., 2011; Spagrud et al., 2008; Walker et al., 2006; Williams et al., 2011) and chronic pain (Pielech et al., 2018; Simons et al., 2008; Sinclair et al., 2016; Walker et al., 2002). Parental reinforcement by attending to child pain has been associated with increased child pain attending (Walker et al., 2006) and less use of adaptive coping strategies (Blount et al., 2001; Spagrud et al., 2008). However, two recent studies of parental chronic pain have not found support for parental reinforcement pathways in predicting child outcomes when examining multiple mechanisms (including both parental reinforcement and modeling) within the same models (Poppert Cordts et al., 2019; Stone et al., 2018). These studies examined children with chronic pain and their parents, whose self-reported average pain intensity was lower than that of parents in Chapter 2. It is possible that reinforcement of pain behaviour is less salient to children who have chronic pain themselves (e.g., higher overall rates of pain behaviour in these children may result in diluted effects of parental reinforcement of the behaviour) or that the relationship between attending to and reinforcing pain is more complex in children with chronic pain (e.g., non-attending or distraction responses from parents are less feasible when children experience frequent pain). Higher levels of parent pain interference were associated with greater parent reinforcement of child pain in Chapter 3 and in past research (Wilson & Fales, 2015), and thus sample differences in parental pain characteristics may explain differing study results. Additionally, different types of pain were examined in these studies as compared to Chapter 3 (everyday pain in children with chronic pain versus pain in the context of the CPT, a novel task for participants). Importantly, parent pain-reinforcing behaviours were measured in Chapter 3 using a well-established observational coding system in the context of a laboratory pain task, while the other studies relied on parent self-report of pain reinforcing behaviours, which may have been biased by retrospective reporting bias, social desirability concerns, or the measures' lack of capturing idiosyncratic, complex parent reinforcement behaviours which may occur in families' daily lives (Poppert Cordts et al., 2019).

Overall, the results of Chapter 3 are consistent with hypothesized social transmission pathways in predicting children's internalizing symptoms and pain during a laboratory task. However, the possibility that these results can be explained by other mechanisms (e.g., genetics, shared environmental stress) must be considered (Stone & Wilson, 2016). Existing research suggests that the presence of chronic pain is at least partly explained by genetics (Hocking et al., 2012; McIntosh et al., 2016; Veluchamy, Hébert, Meng, Palmer, & Smith, 2018) and there is some evidence that the child vulnerability factors examined (e.g., child catastrophizing about their own pain) may also be heritable (Trost et al., 2015). Future research should examine alternative potential mechanisms to explain the current findings. Additionally, the cross-sectional design of the study described in Chapter 3 does not allow for testing of temporal relationships between the variables examined. Children's experiences with chronic pain may influence parents' own coping with their chronic pain, or this relationship may be bidirectional.

5.1.3 Parents' and children's catastrophizing about their own and each other's pain.

Chapter 4 examined the novel construct of child catastrophizing about their parents' chronic pain and its relationship with children's pain and internalizing symptoms. Dyadic analysis using the Actor Partner Interdependence Model (W. Cook & Kenny, 2005) was used to examine these relationships beyond the associations with parent and child catastrophizing about their own pain. Results showed that higher levels of catastrophizing about their own pain in parents and children were associated with greater child catastrophizing about parent pain. Greater child catastrophizing about parent pain predicted higher levels of child internalizing symptoms and pain intensity during the cold pressor task (as reported by children and parents), beyond the influence of parent and child catastrophizing about their own pain. This pattern of results was not seen for the outcomes of children's usual pain intensity of their most commonly experienced pain over the previous three months or their anxiety experienced during the CPT. Results suggest that child catastrophizing about parent pain could be a vulnerability that makes children of parents with chronic pain more likely to develop difficulties with pain and internalizing symptoms. Further research is needed to understand how this variable might confer risk to children's wellbeing and its potential role as a target for prevention/intervention programs.

The findings of Chapter 4 are generally consistent with previous research on the robust intra- and interpersonal effects of pain catastrophizing. Higher levels of catastrophizing about one's own pain were associated with greater self-reported usual pain intensity in both parents and children, as in previous studies of adults and children with and without chronic pain (Birnie et al., 2016; Craner et al., 2017). Higher levels of parent catastrophizing about child pain were associated with greater parent anxiety during the child's CPT, and higher levels of child catastrophizing about their own pain were associated with greater child self-reported anxiety during the CPT, consistent with results of previous research (Hechler et al., 2011).

A divergent result of Chapter 4 in comparison to previous work was that greater parent catastrophizing about child pain was significantly associated with lower levels of child- and parent-reported child CPT pain intensity. Previous work has generally found that parent catastrophizing about child pain is associated with poorer child pain-related outcomes (Birnie et al., 2016; Rabbitts et al., 2017; Wilson et al., 2014) in studies of children with and without chronic pain and their parents (rather than studies of parents with chronic pain). It is possible that children of parents with chronic pain may attempt to alter their own pain expression to reduce parents' distress. For example, consistent with Goubert and colleagues' (2005) model of observation of others' pain, children of parents with chronic pain may experience increased other-oriented distress as a result of observing their parent in pain and aim to reduce upsetting their parent by downplaying their own pain expression. Children who observe their parents experiencing difficulty coping with pain may infer that their parents are unable to manage the child's distress, and may decrease their own pain expression through means such as using suppression or positive reappraisal of pain (Hampton, Hadjistavropoulos, Gagnon, Williams, & Clark, 2015). This may interact with child attachment style; for example, children with insecure attachment styles may view their parents as unable to effectively manage their distress and therefore decrease their own pain expression. If parents with pain have typically responded in the past with withdrawal or punishment to the child's pain expression, the child may learn to reduce their expressions to avoid these responses. Alternatively, when parents have greater levels of catastrophizing about child pain, children may be more confident in their parents' ability to accurately assess and manage their pain, resulting in lower child distress and pain expression. Higher levels of parent catastrophizing about child pain have been associated with better parent-child congruence in rating children's pain intensity (Goubert et al., 2009).

The present dissertation identified child catastrophizing about their own pain as a child vulnerability factor associated with higher levels of internalizing symptoms

(Chapter 3), and with greater self-reported usual pain intensity and parent-reported child internalizing symptoms (Chapter 4). Greater children's and parents' catastrophizing about their own pain were both associated with higher levels of child catastrophizing about parent pain. Greater child catastrophizing about parent pain was associated with poorer child outcomes (greater child- and parent-reported child internalizing symptoms and CPT pain intensity) beyond the influence of child catastrophizing about their own pain. This suggests that child catastrophizing about parent pain is an additional factor, beyond types of catastrophizing that have previously been studied, that is important to consider in the context of parental chronic pain. Previous theories (Stone and Wilson) have suggested that children learn from parents (e.g., through pain modeling and reinforcement of child pain behaviours) about how to appraise their own pain, which in turn impacts their pain, functioning, and psychological health. However, the current results suggest that children's catastrophizing about their parents' pain (i.e. a form of other-oriented distress) could be a separate vulnerability factor through which children's outcomes are influenced. The process of learning about pain and coping from parents may be altered in the context of parental chronic pain compared to parent-child dyads where the parent does not have chronic pain. From an evolutionary perspective, children rely on their parents for caregiving and emotional support, and it is adaptive to pay close attention to parental cues, particularly in situations of threat such as pain (Mikail et al., 1994). In the context of parental chronic pain, children may pay increased attention to parents' pain experiences and appraise their parent's pain as more threatening if it impacts the parents' ability to provide for the child's needs (e.g., emotional closeness). In this way, parents' pain experiences may uniquely impact children's outcomes through child catastrophizing about parent pain, rather than through the child developing increased catastrophizing about their own pain. Further research is needed to examine the complex intra- and interpersonal relationships between children's and parents' catastrophizing about their own and each others' pain and their pain and mental health outcomes, and the mechanisms that underlie these relationships.

5.2 Theoretical Implications

Theory underlying the intergenerational transmission of risk for chronic pain is in its early stages of empirical investigation (Stone & Wilson, 2016). The current dissertation examined social learning mechanisms in this relationship as hypothesized by Stone and Wilson's model (Stone & Wilson, 2016). This model posits that social learning mechanisms, including parental modeling of pain behaviour and parental reinforcement of child pain behaviour, are one type of mechanism through which risk for chronic pain, disability, and poor psychological functioning is transmitted from parents with chronic pain to their children. Preliminary support for the parental modeling mechanism has been found in samples of children with chronic pain and their parents (Poppert Cordts et al., 2019; Stone et al., 2018). Within Stone and Wilson's model, parents' catastrophizing about their own and their child's pain is hypothesized to transmit information (via modeling and reinforcement processes) to children, influencing children's catastrophizing about their own pain, which is thought to represent a child vulnerability to increased risk of poor outcomes (Stone & Wilson, 2016). The current dissertation makes an important contribution to theory in this area by providing support for social learning mechanisms in the relationship between parental chronic pain and child pain and internalizing symptoms in parents with chronic pain and their children, and by finding preliminary support for the differential roles of children’s catastrophizing about their own and their parent’s pain in predicting child outcomes. These results provide empirical support for hypothesized mechanisms and identify additional factors that may predict child vulnerability to pain and internalizing symptoms.

The findings of the current dissertation support the hypothesized parental pain behaviour modeling pathway (Stone & Wilson, 2016) in predicting children’s internalizing symptoms, but not the presence of children’s chronic pain, and support the parental reinforcement pathway in predicting children’s pain outcomes during the CPT. As discussed above, these findings diverge from results of previous studies conducted in samples of children with chronic pain and their parents (Poppert Cordts et al., 2019; Schanberg et al., 2001; Stone et al., 2018) which examined outcomes including children’s pain severity and functional disability. The results of the current dissertation suggest that in children of parents with chronic pain, exposure to higher levels of parent pain interference (a proxy for parent pain behaviour) are not associated with the presence of child chronic pain, inconsistent with one hypothesized pathway in Stone and Wilson’s model; however, modeling may be associated instead with children’s coping with chronic pain (i.e. the outcomes of increased functional disability (Stone et al., 2018) and poorer psychological functioning (internalizing symptoms as in Chapter 3) as hypothesized in Stone and Wilson’s model). The parental reinforcement pathway may differentially influence children’s acute and chronic pain, or may be more evident in parents’ and children’s observed interactions during child pain experiences than in parents’ self-reported patterns of child pain reinforcement behaviour (e.g., due to retrospective reporting bias or social desirability concerns). The current dissertation provides additional clarification to this model and suggests that not all hypothesized outcomes may be impacted by the same mechanisms.

The current dissertation also examined relationships between the hypothesized child vulnerability factor of child catastrophizing about their own pain (Stone & Wilson, 2016) and child pain and internalizing outcomes. It expanded on this theory by examining child catastrophizing about parent pain and its role in predicting child outcomes beyond other forms of catastrophizing in children and their parents with chronic pain. This variable may be particularly important in children of parents with chronic pain given the threat to the attachment relationship that parental pain might represent (Mikail et al., 1994). Consistent with research examining the impact of observing others in pain (Goubert et al., 2005, 2008), exposure to parental chronic pain may result in increases in both other-oriented (e.g., catastrophizing about parent pain) and self-oriented (e.g., internalizing symptoms) distress in children. Results suggest that children's cognitions about their parents' pain may represent an important child vulnerability factor, unique from children's cognitions about their own pain, and should be further researched to determine its role in predicting child wellbeing.

While the results of the current dissertation generally support hypothesized social transmission mechanisms as hypothesized by Stone and Wilson (Stone & Wilson, 2016), alternative mechanisms that might underlie these relationships were not evaluated. The theoretical model suggests multiple pathways through which parental pain may impact children's outcomes, including genetic and neurobiological mechanisms, shared exposure to stressful environments, and broader impacts of parenting and family functioning factors. It is possible that relationships observed between parents' chronic pain, children’s pain catastrophizing, and children’s pain and internalizing outcomes could be at least partly explained by genetic factors (e.g., Trost et al., 2015). Parents with chronic pain might pass on altered pain processing to their children, making them more vulnerable to both increased pain and increased catastrophizing themselves, explaining the observed relationships through a different mechanism. Higher levels of parent and child pain catastrophizing may be indicative of an overall negative cognitive and emotion regulation style which impacts children’s outcomes, rather than the effects arising from social learning about pain and pain catastrophizing. Future research in this area should compare alternative mechanisms to clarify these relationships. Previous research suggests that both genetic and environmental factors influence the relationships between parent and child pain (Hoftun et al., 2013), but the exact pathways through which these influences occur could be further clarified in order to identify modifiable targets for potential intervention and prevention of problems with chronic pain and mental health in children of parents with chronic pain.

Additionally, the current dissertation as well as many of the previous studies testing mechanisms underlying the relationships between parent and child pain (Poppert Cordts et al., 2019; Schanberg et al., 2001; Stone et al., 2018) have employed cross-sectional designs, which do not allow for clarification of the temporal relationships between these variables. While associations in cross-sectional studies provide initial support for the possibility of a predictive pathway, research employing longitudinal designs is required to confirm the temporal direction of findings. As described by Stone and Wilson (Stone & Wilson, 2016), the relationships between the mechanisms noted in the model and child vulnerability factors (e.g., child catastrophizing about their own pain) might be bidirectional. For example, child chronic pain is associated with poorer mental health and wellbeing in parents (Palermo & Eccleston, 2009), and children’s own pain experiences could influence the way a parent with chronic pain responds to their own and their child’s pain. Children may also play a role in reinforcing parents’ pain behaviours (e.g., providing increased attention and helpfulness or reduced resistance to parental discipline when the parent is in pain). Longitudinal study designs could also clarify these relationships and provide improved understanding of the complex relationships between parent chronic pain and child pain and mental health.

5.3 Clinical Implications

The existing body of research on offspring of parents with chronic pain, including the current dissertation, clearly indicates that offspring of parents with chronic pain are a vulnerable population with respect to their own pain, health, psychological, and family outcomes. Despite these findings, interventions specifically aimed at this population have yet to be evaluated in the literature. The present dissertation identifies potential targets for future prevention and intervention programs (e.g., parental pain interference, child catastrophizing about their own and their parent’s pain, parental reinforcement of child pain); although further research is needed to establish the temporal aspects of these relationships, the findings suggest initial areas of focus for preliminary program design.

As parental chronic pain is clearly associated with increased risk for child pain and mental health problems, screening for parental chronic pain when working with children could be a helpful first step. Such screening may allow for clinicians to better support parents (e.g., referring for their own pain management services) and in turn improve outcomes for children. A brief screening tool (12 items) for parents of children with chronic pain has recently been developed which assesses parents' distress about their child's pain, behaviour in response to child pain (e.g., allowing the child to skip activities because of their pain), impact of child pain on family life, and parent health (including feeling sad/down and finding usual activities less enjoyable; Simons, Lewandowski Holley, Phelps, & Wilson, 2019). Preliminary evidence for the concurrent and discriminant validity of the screener in identifying parents who may benefit from increased intervention were found (Simons et al., 2019). Developing a similar screening tool to identify parents with chronic pain whose children may be at risk for developing problems with pain or mental health could be created and employed in various settings where health professionals work with children (e.g., primary care, child mental health clinics). Additionally, it may be helpful to screen adults seeking treatment for chronic pain for potential difficulties with pain and mental health in their children. Anecdotally, several families participating in the current dissertation studies expressed that discussing study participation revealed children's coping difficulties to their parents for the first time. When children of parents with chronic pain do not have chronic pain or clinically significant mental health symptoms themselves, their distress and potential risk factors may be missed. Screening adults with chronic pain for potential child difficulties and providing relevant psychoeducation and referral to resources may be helpful.

Based on the results of the current dissertation supporting the role of parental modeling of pain behaviours in children's distress, increased supports for parents with chronic pain aimed at reducing their levels of pain interference may improve not only parents' own health and coping, but also their children's outcomes. Increasing access to these services and screening parents to discuss intervention options are important steps in improving adult and child health. In addition to the mechanisms studied in the current dissertation, poorly managed parent pain may impact children through other pathways, such as exposure to increased family stress (Stone & Wilson, 2016). Inadequate pain management plays a role in opioid misuse (Groenewald, Law, Fisher, Beals-Erickson, & Palermo, 2019), which may have downstream effects on children, further highlighting the need to support parents in managing their own pain.

Parent reinforcement of child pain behaviours is another potential intervention target identified by the current dissertation. Research in child chronic pain has included teaching parents about the role of their responses to child pain in predicting children's outcomes and has generally been successful in changing parent responses (e.g., Sieberg et al., 2017). In a study of parents and children who participated in an intensive, interdisciplinary treatment program for child chronic pain (including parents-only groups), decreased parent use of protective and monitoring behaviours in response to child pain was associated with decreases in child functional disability over time (Pielech et al., 2018). Future research should examine the effectiveness of providing similar education to parents with chronic pain. For example, many multidisciplinary chronic pain management programs for adults incorporate psychoeducational groups aimed at improving patients' pain self-management, including information on topics such as the gate control theory of pain, pacing strategies, pain acceptance, and communicating about pain with partners (Du et al., 2011). Incorporating into these groups information about responding to child pain, talking about own chronic pain with children, and modeling of pain behaviours may be helpful and would meet needs that parents with chronic pain and young adult offspring of parents with chronic pain have identified in qualitative research (Umberger et al., 2015).

Child and parent catastrophizing about their own and one another's pain have been associated with children's pain and internalizing outcomes in the context of parental chronic pain. Pain catastrophizing has been the target of interventions and examined as a process measure in predicting changes in pain outcomes in treatment studies (Schütze et al., 2018). Parent pain catastrophizing has been shown to be related to parents' use of protective responses to their children's chronic pain (Wilson & Fales, 2015; Wilson et al., 2014) and the impact of parent reinforcement of child pain behaviour has been shown to be moderated by child catastrophizing about their own pain (Williams et al., 2011). Reducing parents' catastrophizing may positively impact children's outcomes through multiple pathways (e.g., decreasing reinforcement responses, decreasing parent modeling of maladaptive pain coping). Reducing child catastrophizing about their own and their parents' pain may also be helpful. A recent meta-analysis of studies examining interventions for reducing pain catastrophizing in adults found evidence for medium strength effects of cognitive-behavioural therapy, acceptance and commitment therapy, and multimodal treatment in reducing pain catastrophizing (Schütze et al., 2018). Qualitative research with parents with chronic pain and their young adult children has similarly identified correcting cognitive distortions, such as catastrophizing, as an important need for being able to cope with parental chronic pain (Umberger et al., 2015). The authors suggest that interventions supporting skills in this area could improve children's coping with parental chronic pain. Future research should examine intervention programs for improving pain catastrophizing about own and others' pain in children and parents and their effects on improving child outcomes.

Given the numerous potential targets for intervention for improving pain and mental health outcomes in children of parents with chronic pain identified in the current dissertation (parental pain interference, child and parent catastrophizing about their own and each others' pain, parent reinforcement of child pain behaviours) and suggested in previous research (e.g., parent-child communication about pain, attachment, emotional availability (Donnelly & Jaaniste, 2016; Umberger et al., 2013, 2015), intervention programs addressing multiple targets and tracking process measures and outcomes over time would likely be helpful. Research designs that compare such treatment to active controls (e.g., general support groups) or waitlist controls and examine the temporal relationships between variables over time should be incorporated. Research suggests that group treatment with a variety of therapeutic modalities can be effective for children with chronic pain and their parents (Huestis et al., 2017; Kanstrup et al., 2016) and that goal agreement between parents and children is predictive of better child outcomes (Fisher, Bromberg, Tai, & Palermo, 2016). Treatment groups that incorporate both parents and their children and support parent-child pairs in working together on relevant goals may be helpful.

An important aspect of intervention design for any group is involvement of end users in the development process (i.e. incorporating patient-oriented research and user-centered design principles; Baek, Boling, & Frick, 2008; Minogue, Cooke, Donskoy, Vicary, & Wells, 2018). Qualitative research has begun to examine the needs of parents with chronic pain and their children (Umberger et al., 2015). Future studies should combine the empirical evidence gathered thus far regarding predictors of child outcomes in the context of parental chronic pain with lived experience of parents with chronic pain and their children in order to develop relevant, accessible intervention programs which address topics that are not only evidence-based but also important to potential end users of the programs. Patient-oriented research methodology has been shown to improve implementation of research findings (Brett et al., 2014) and these principles should be drawn on to develop intervention programs for parents with chronic pain and their children. Qualitative research with parents with chronic pain and young adults of parents with chronic pain has identified several areas to be addressed in interventions, some of which are consistent with empirical research (e.g., need for support in correcting cognitive distortions such as catastrophizing about pain) and others which have not been directly examined in the context of parental chronic pain (e.g., need for support in improving parent-child communication about chronic pain; Umberger et al., 2015). Some research has examined interventions for improving communication in families where a parent has chronic pain (Swift, Hocking, Dickinson, & Jones, 2018), but these studies have generally examined communication between parents with pain and their partners, and have not evaluated the impacts of such interventions on children's outcomes. Future research should use well-established methods for patient-oriented research (Hoddinott et al., 2018) to work in partnership with parents with chronic pain and their children to create relevant, accessible, and effective interventions for this vulnerable population.

5.4 Key Strengths and Limitations

5.4.1 Systematic Review and Synthesis Methods

A key strength of the current dissertation was the use of comprehensive systematic review and synthesis strategies to examine overall patterns of results across existing literature on outcomes in offspring of parents with chronic pain. The use of a systematic search reduced bias in identifying as many relevant publications as possible. Meta-analysis methodology allows for the statistical combination of results across studies, reducing bias as compared to narrative reviews (which may use strategies such as descriptive summaries and vote counting to summarize study results) and allowing for conclusions to be made based on much larger samples than individual studies would allow (Littell et al., 2008). The incorporation of multiple study designs (population-based, clinical, qualitative) was a strength, as qualitative research revealed additional insights beyond the results of quantitative studies and may have been missed with more narrow inclusion criteria. Meta-ethnography was used to synthesize qualitative results, and is a well-established synthesis method with clear guidelines and an interpretive basis that was consistent with the qualitative research designs used in individual studies included in the review (Atkins et al., 2008; Britten et al., 2002; Paterson, 2012). Coding for the meta-ethnography was completed by two independent coders in order to increase trustworthiness of the results (Krefting, 1991).

Limitations of the use of these techniques in the present dissertation included the lack of quality evaluation of the included primary studies, due to heterogeneity in study designs precluding the use of a consistent evaluation tool across included studies, and the small number of studies included in some individual meta-analyses. The lack of quality evaluation may have meant that studies with low-quality research designs were included in the analyses with equal weight as other more rigorous studies, decreasing the accuracy of the results. Additionally, this review and synthesis was completed three years ago. Review of newer research in this area found by re-running the systematic search suggests that study designs and conclusions continue to be similar as in the original review, suggesting that the review is still up to date according to a decision-making framework for determining when to update reviews (Garner et al., 2016). Moderators of the relationship between parental chronic pain and offspring outcomes were not examined in the current dissertation, which could have skewed the results (Rosenthal & DiMatteo, 2002). Several additional limitations of meta-analysis methodology have been identified in the literature (Gurevitch, Koricheva, Nakagawa, & Stewart, 2018; Littell et al., 2008; Rosenthal & DiMatteo, 2002). Strong meta-analysis methodology cannot overcome problems with study quality and bias in the primary research literature (e.g., publication bias, over- or underrepresentation of particular populations in primary studies; Gurevitch et al., 2018; Rosenthal & DiMatteo, 2002). Further primary studies are needed to clarify several research questions associated with parental chronic pain and offspring outcomes as discussed above, which limits the conclusions of the meta-analyses. Decisions about which outcomes are conceptually similar enough to combine in meta-analysis are also subject to bias, potentially impacting results (Littell et al., 2008).

5.4.2 Use of the Cold Pressor Task

The use of the CPT in the current study was a key strength for several reasons. It allowed for a high level of internal control, with each child being exposed to the same pain stimulus and other contextual factors, and for parent-child dyads to be observed in vivo during the child's pain experience. Social desirability concerns were minimized by having parents and children complete the task without research assistants present, encouraging them to interact during the task as they normally would elsewhere, and with unobtrusive camera placement. Despite its strengths, there are limitations to this methodology, including regarding the generalizability of results obtained in this context to other types of pain. Parents and children may have acted differently during the CPT than they typically would during other pain experiences. Given that the task was novel to participants, they may have been less influenced by past experiences (e.g., anticipatory fear) than they would in other contexts (e.g., needle procedures). A key difference between the CPT and other forms of pain children may experience (e.g., injuries in everyday life; pain due to injections or medical procedures) is children's control over the pain; children were instructed that they could remove their hand from the CPT (and thus reduce/stop the pain) at any time. While laboratory pain tasks such as the CPT have frequently been used in pediatric pain research, little research has examined its generalizability to other types of pain. Some evidence suggests that parents and children view each other's behaviour during the CPT as only "a little different" from how they would typically act (Birnie et al., 2016). When a sample of healthy children (ages 8-14 years) were asked to compare the CPT to their most recent needle procedure, the average response was that the CPT was "a little less scary" than a needle, and between "the same" and "a little less painful" than the needle (Boerner et al., 2016). These results provide preliminary evidence that children experience the CPT as being similar to other acute pain experiences.

The completion of CPT procedures in a laboratory setting meant that parents and children had to travel to the laboratory in person to participate in the current study. While efforts were made to accommodate participants' needs as much as possible (e.g., appointments were available days, evenings, and weekends; childcare for additional children not participating in the study was provided; families were compensated for travel expenses), it is possible that the sample represents a subset of the population of parents with chronic pain and their children who were higher functioning (e.g., had better managed pain, perhaps due to increased access to pain management resources; had family and employment situations allowing flexibility for study participation). The majority of the participating parents were mothers, and most parents and children identified as Caucasian; thus, the generalizability of results of the current dissertation to samples of fathers and other racial and ethnic groups is unknown. Given that previous research has identified associations between sex, gender, race, and ethnicity and pain-related outcomes (including access to pain management; Boerner et al., 2018; Earp et al., 2019; Fillingim, King, Ribeiro-Dasilva, Rahim-Williams, & Riley, 2009; Moon et al., 2008; Shavers, Bakos, & Sheppard, 2010), it will be important to examine the relationships between parental chronic pain and child outcomes in diverse samples.

5.4.3 Observational Coding System

Parent and child verbalizations during child CPT pain were assessed using a well-established observational coding system (Child-Adult Medical Procedure Interaction Scale – Revised; CAMPIS-R; Blount et al., 1997) adapted for use in the context of laboratory pain tasks (Moon et al., 2011; Walker et al., 2006). This allowed for the collection of detailed information on parents' and children's interactions during a specific child pain experience, reducing reliance on self-report or retrospective measures. Transcripts of parent-child interactions were coded independently by trained coders with a high level of inter-rater reliability. Because the coding system only included verbalizations, additional information communicated through nonverbal behaviours may have been missed. Additionally, sequential analysis was not employed, meaning that temporal relationships between verbalizations could not be examined.

5.4.4 Use of Self-Report Measures in Parents and Children

Parents and children completed reliable, valid measures of pain-related and internalizing symptoms which had been previously psychometrically evaluated in similar samples (Crombez et al., 2003; Petter et al., 2013; Reynolds & Kamphaus, 2006; Sullivan, 1995). In the context of the CPT, the Faces Pain Scale-Revised (Hicks et al., 2001) was used to provide child- and parent-reported measures of child pain intensity. This measure is considered well-established for use in children ages 4-16 years (Cohen et al., 2008), has evidence for its validity in adults (Ferreira-Valente et al., 2011), and is commonly used for parent reports of child pain intensity (Birnie et al., 2016; Boerner et al., 2013; Spagrud et al., 2008). Several outcomes were reported on by both parents and children (e.g., child CPT pain intensity, child internalizing symptoms), allowing for multiple perspectives on each variable to be examined. This is consistent with recommendations for pain assessment suggesting that child self-reported pain intensity should be used as a primary source of information for making pain management decisions (Schiavenato & Craig, 2010; Twycross et al., 2015; von Baeyer, 2009). A limitation of the measurement design in the current study was the measurement of all constructs during one laboratory session. This cross-sectional design meant that temporal relationships between measured constructs could not be examined.

5.5 Future Research Directions

Understanding the relationships between parental chronic pain and offspring outcomes, including the mechanisms underlying such relationships, is a relatively new area of research with many promising directions for future research. Multiple mechanisms have been theorized for explaining these relationships (Stone & Wilson, 2016), and research thus far has generally focused on genetic and social learning explanations (Hoftun et al., 2013; Stone et al., 2018). The current dissertation provides support for social learning mechanisms (modeling and reinforcement) in predicting internalizing and laboratory pain outcomes in children of parents with chronic pain. Most research in the area, including the current dissertation, has employed cross-sectional methods. Longitudinal studies examining temporal relationships between predictors are needed to further evaluate the theorized mechanisms underlying relationships between parent chronic pain and offspring outcomes. Additionally, few studies have compared multiple mechanisms that might explain the relationships between parental chronic pain and child outcomes (Hoftun et al., 2013). More studies employing these methods are needed in order to determine which mechanisms, or combinations of mechanisms, best explain these relationships. For example, while social learning mechanisms have been studied independently, it is not known whether genetic or other mechanisms might explain those results.

A key variable to be further explored in this area is the role of parental mental health in the context of parental chronic pain. There is a robust literature supporting the role of parental depression in predicting children’s poorer mental health outcomes (Cho et al., 2015; Goodman & Gotlib, 1999) and parental psychological health has been shown to predict unique variance in child wellbeing beyond parental chronic pain (Poppert Cordts et al., 2019). Other types of parental mental health difficulties also play a role in children’s outcomes (e.g., anxiety, posttraumatic stress; Beveridge, Neville, Wilson, & Noel, 2018; Booker, Capriola-Hall, & Ollendick, 2018; Neville, Soltani, Pavlova, & Noel, 2018). Additional research is needed to disentangle the influences of parental chronic pain and mental health on children’s outcomes.

While research thus far has generally focused on vulnerabilities in children of parents with chronic pain (Poppert Cordts et al., 2019; Schanberg et al., 2001; Stone et al., 2018), theoretical and empirical work suggests that child resilience in the face of parental chronic pain is also possible (Evans & de Souza, 2008; Stone & Wilson, 2016; Umberger et al., 2013). Identification of modifiable protective factors in children, parents, and families that can be supported may help to increase positive outcomes in children of parents with chronic pain. Acceptance-based views of chronic pain (Stone & Wilson, 2016) and open parent-child communication (Swift et al., 2018; Umberger et al., 2015) have been identified as possible targets for further exploration and potential intervention in this area.

Finally, translation of research findings into the development and evaluation of prevention and intervention programs for this vulnerable population is an important next step in this field. Numerous potential targets for intervention have been identified, and as these relationships and their underlying mechanisms are clarified, interventions focused on these targets should be developed and studied. Patient-oriented research and user-centered design principles should inform these interventions in order to ensure that they are appropriate and accessible to the target population.

5.6 Concluding Remarks

The present dissertation addressed knowledge gaps in the literature on parental chronic pain and its relationship with child outcomes using several rigorous methods. A comprehensive, mixed methods review synthesized the literature in this field and identified children of parents with chronic pain as a population vulnerable to increased pain and mental health difficulties compared to children whose parents do not have chronic pain. Building on these findings, a laboratory-based study examined social transmission pathways hypothesized to underlie relationships between parental chronic pain and child pain and internalizing outcomes in a sample of adults with chronic pain and their children (ages 8-15). Support was found for parental modeling in predicting child internalizing outcomes and for parental reinforcement in predicting child pain during the cold pressor task. In the final paper, the novel construct of child catastrophizing about parent pain was identified as a potential child vulnerability factor for pain and internalizing outcomes in children of parents with chronic pain. The laboratory-based study employed multi-method (questionnaire, observational) and multi-informant (child, parent, observer) assessment to provide rigorous examinations of these mechanisms. Taken together, the results of the dissertation contribute to clarifying the mechanisms explaining relationships between parental chronic pain and child outcomes, and identify several potential targets for intervention in this vulnerable population.

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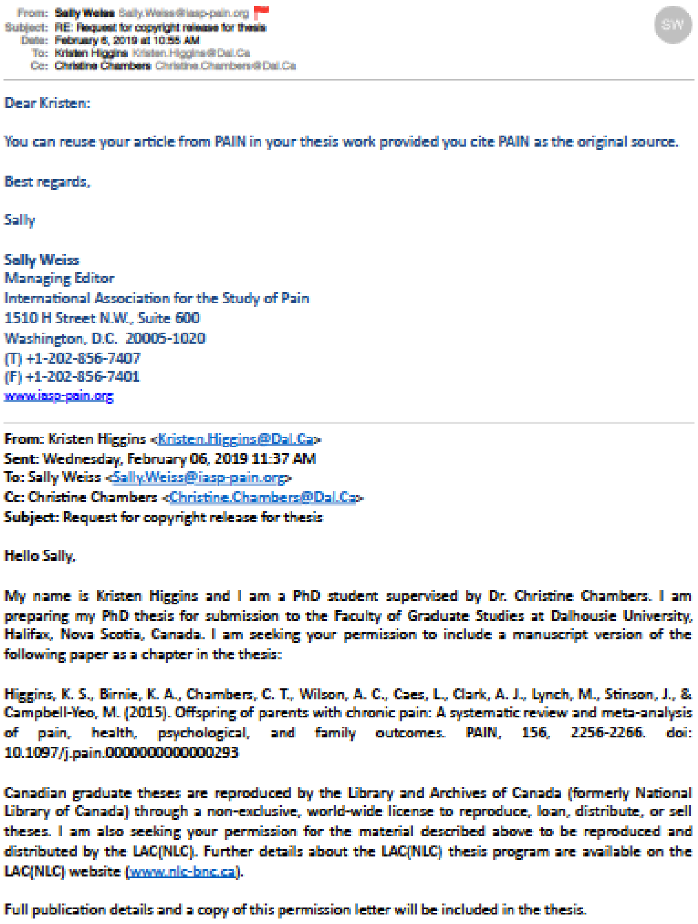
APPENDIX A: COPYRIGHT PERMISSIONS



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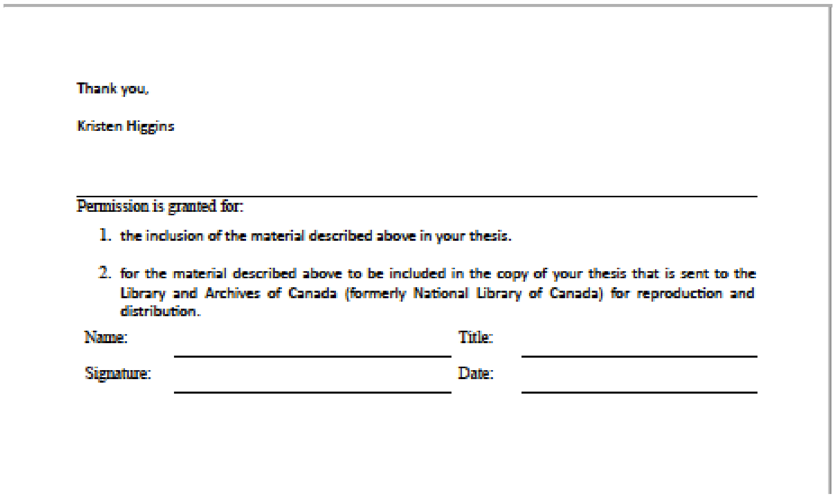

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APPENDIX B: CODING MANUAL

Rationale for Current Coding Manual

- Used Moon et al.'s (2011) coding system, which was modified from Walker et al. (2006); both are based on the CAMPIS-R
- Moon et al.'s (2011) coding scheme has been modified in the following ways for the current study:
  - Removed codes for previous cold pressor talk (not relevant to the current study)
  - Codes have been added for parent and child talk about parent pain symptoms
  - Separate code has been added for child non-symptom focused talk
- Videos of parents and children completing the cold pressor will be transcribed and their verbalizations will be coded utterance by utterance

Codes

Parent to Child:

1. Non-symptom focused talk and commands to child **(NSFC)**
2. Symptom focused talk and commands to child **(SFC)**
3. Sympathy to child **(SYMP)**
4. Procedure time talk and commands to child **(PTC)**
5. Other procedure talk and commands to child **(OPC)**
6. Humor to child **(HUM)**
7. Criticism to child **(CRIT)**
8. Procedure-related praise to child **(PRAS)**
9. Parent talk about own pain symptoms **(PPS)**
10. Other talk to child **(OTC)**

Child to Parent:

11. Cold/pain symptom talk to parent **(CPP)**
12. Anxiety talk to parent **(ANXP)**
13. Procedure time talk to parent **(PTP)**
14. Other procedure talk to parent **(OPP)**
15. Resistance talk to parent **(RESP)**
16. Child coping talk to parent **(CCP)**
17. Child talk about parent pain symptoms to parent **(CPPS)**
18. Child non-symptom focused talk to parent (**NSFP**)
19. Other talk to parent **(OTP)**

Notes:

- Any talk by the experimenter/research assistant (as well as parents' and children's talk and responses to the experimenter) are coded "**EXP**")
- Any silence of longer than 5 seconds that *follows parent speaking* is coded "**PSILEN**"

- Any silence of longer than 5 seconds that *follows child speaking* is coded "**CSILEN**"
- Nods, sighs, and other nonverbal communication that may be transcribed are not assigned a code.
- In Moon et al., a watch would beep to tell children when to place their hand in the water and when to remove it; thus, many of the verbalization examples refer to watch beeps. In the current study, this information was communicated by the experimenter over the intercom.

Parent to Child Vocalizations

1. Non-symptom focused talk and commands to child **(NSFC)**
2. Symptom focused talk and commands to child **(SFC)**
3. Sympathy to child **(SYMP)**
4. Procedure time talk and commands to child **(PTC)**
5. Other procedure talk and commands to child **(OPC)**
6. Humor to child **(HUM)**
7. Criticism to child **(CRIT)**
8. Procedure-related praise to child **(PRAS)**
9. Parent talk about own pain symptoms **(PPS)**
10. Other talk to child **(OTC)**

1. Non-symptom focused talk and commands to child (NSFC):

- ***Talk:***

Any talk by the parent **unrelated to the child's pain/cold symptoms or anxiety**. Included in this category are conversations, comments, or questions about the child's pets, siblings, parents, school, toys, etc., or about the child's plans, wants, desires, etc. that are *unrelated* to cold/pain or anxiety symptoms the child is presently experiencing. Also included are non-symptom related statements made by the parent, including praising the child (without reference to the cold pressor task or symptoms), as well as factual statements and questions about lab equipment not including the cold pressor (e.g., video cameras, smart board, towel, questionnaires, consent form) order of events, lab staff, etc. Parent sympathy in the context of a non-symptom focused conversation is included in this category. General statements or just changing the subject is coded as 1 (NSFC).

- Talk about how much time is left in the various phases of the cold pressor task (wait period and cold pressor period) is coded as 4 (PTC).
- Humor is coded as 6 (HUM).
- Criticism of the child about any topic is coded as 7 (CRIT).
- Talk about the child's past physical symptoms is coded as 10 (OTC).
- Talk about the parent's own physical symptoms (other than their chronic pain symptoms) is coded as 10 (OTC).
- Talk about the parent's chronic pain symptoms is coded as 9 (PPS)

1. "Hey kid."
2. "What do you want for dinner tonight?"
3. "Were you hungry enough to eat all of your lunch today at school?"

4. "(Little brother) Johnny is playing in the other room."
5. "Don't worry about that test at school today honey, it'll be just fine."
6. *(In response to "They are listening to us.")* "Well, it's OK." (said reassuringly)
7. "I just love that new shirt you've got onto today, what a good color for you."
8. "Still mad at me for what I said in the car? You know I was just joking around."
9. "Mmmhmmm." (parent is just responding naturally to child conversationally in context of non-symptom focused conversation, i.e., verbal nodding)
10. "Are you excited?"
11. "Are you all set to go?"
12. "10,9,8,7,6,5,4,3,2,1," (If counting is meant to distract child, not help them determine when the cold pressor task will be over)

- ***Commands:***

Any talk by the parent that gives the child directives, suggestions or advice that does not focus the child on the cold pressor procedure or the child's cold/pain or anxiety symptoms. To qualify, the directive has to be specific to instructing the child to do something unrelated to the cold pressor procedure or distracting the child's attention.

- ◆ A command to count is coded as a 1 (NSFC) if it is clearly meant to distract the child (usually in response to a child symptom complaint) whereas a command to count is coded as a 4 (PTC) if it is meant to help the child cope with his/her anxiety about how much time is left in the cold pressor period (usually in response to a child's question about how much time is left)

13. *(In response to "I'm scared!")* "Let's get your mind off it."
14. *(In response to "I want to take my hand out!")* "Want to talk about school to take your mind off it?"
15. *(In response to "It's way colder than the lake!")* "Well, it's better to focus on what you are going to do with the money you're getting."
16. "Let's sing a song together"
17. *In response to "Owwww!")* "Count backward from 10."
18. "Straighten your collar"
19. "Don't scratch your cut"

2. <u>Symptom focused talk and commands to child (SFC):</u>

- ***Talk:***

Any question, comment or reflection referring to the child's **current cold/pain physical symptoms, anxiety, or cold pressor-related status and resistance (including questions about how long the child will keep their hand in)**. This includes humorous statements that focus on the child's physical status. Also includes talk about the water temperature that includes terms such as "cold" or compares the temperature of the water to something obviously cold.

- ◆ Symptom focused talk in a sympathetic tone is coded as 3 (SYMP).
- ◆ Parent talk that praises the child for bravery (or generally for doing well) that is directly related to the cold pressor task is coded as 8 (PRAS).

- Straightforward or matter-of-fact talk about the temperature of the water that does not include terms such as “cold” and does not compare the temperature of the water to something obviously cold is coded as 5 (OPC).

1. “Are you OK?”
2. “Are you scared?”
3. “Does it hurt?”
4. “How does it feel?”
5. *(in response to a shriek by the child)* “I think you’re exaggerating”
6. *(in response to a complaint by the child)* “Are you just being silly?”
7. (In response to “I’m moving my hand around”) “Does that help?”
    - Note that key here is a word such as “help,” “better,” or warmer – if the response does not include one of these words, code as OTC
8. *(In response to “I’m fine”)* “Well good!”
9. “Is it worth $20?”
10. “You look like you're uncomfortable.”
11. “Is your hand about to fall off?”
12. “It doesn't hurt though?”
13. “What's the matter?”
14. “So it doesn't hurt at all?”
15. “Are you sorry you did this?”
16. “Are you going to keep it in the whole time?”
17. “How long are you going to keep it in?”
18. *(In response to “So yesterday, at school…”)* “How does it feel now?”
19. “Just take the pain”
20. (In response to “It’s not as cold as the ocean”) “No?”
21. “Is it as cold as the water in the ocean at Crystal Crescent Beach?"
22. “I think you can do this” (not in context of reassuring child – more of a “vote of confidence” that isn’t prompted by child anxiety)

- ***Commands:***

Any talk by the parent that gives the child directives, suggestions or advice that **directly focuses** the child on the cold pressor procedure or how to deal with the child's current cold/pain symptoms or anxiety. To qualify for this category, the directive has to focus the child’s attention on his or her symptoms or anxiety. It can also be a symptom-focused coping command (other than a command to engage in distraction). This category applies to *commands to cope* given to children in response to symptom or anxiety complaints. Includes suggestions by the parent that the child can stop the cold pressor task if they are too bothered/in too much pain to continue **and** directives by the parent to the child to keep their hand in the water. Also includes suggestions by the parent that the child use their imagination to re-frame the cold or encourage the child to complete the cold pressor task by imagining another situation that is cold or uncomfortable (e.g. reaching into a freezer, ice fishing, keeping very still during another medical procedure etc).

- Commands to count or other suggestions that refer to the amount of time left in the wait period and cold pressor period (usually in response to child’s questions about time left) are coded as 4 (PTC).

23. *(In response to "I want to tell you about this really cool movie we saw in school today")* "Tell me about that later - aren't we supposed to talk about how you feel?"
24. *(In response to "It hurts!")* "Take a deep breath"
25. *(In response to "It hurts!")* "Try moving your hand around"
26. *(In response to "I'm so scared!")* "Hold my hand."
27. "Talk to me."
28. "Just try to hang in there for a little longer."
29. "Just relax."
30. "Keep your hand in there Sam"
31. "Keep it in - let's see how tough you are"
32. "If it starts to bother you, take it out"
33. "You can take it out if it hurts too much"
34. "Pretend you're reaching into a freezer to get a popsicle"

3. Sympathy to child (SYMP):

Any talk, including questions, commands or reflections, by the parent that is reassuring, empathizing or apologizing in content **and/or** tone about the child's **current physical cold/pain complaints, anxiety, or the length of the wait period or cold pressor period**. **Tone** is the biggest factor in this code.

- Sympathetic talk in the course of a conversation about something **unrelated** to the child's current physical status is coded as 1 (NSFC).
- Sympathetic talk related to the child's **past** physical or chronic condition symptoms is coded as 10 (OTC).

1. *(In response to "It's so cold mom")* "You're OK."
2. "Your hand will be OK"
3. *(In response to "Is this almost over?")* "Not much longer now"; "Only a minute or so longer." (sympathetic tone)
4. *(In response to "I'm tired of sitting here")* "I'm sorry this is taking so long."
5. *(In response to "I'm scared")* "It's OK sweetie"
6. "I can imagine it must feel really uncomfortable."
7. "It must be freezing."
8. "It'll be alright, you're alright."
9. *(In response to "It's so cold!")* "I know buddy" (sympathetic tone)
10. "I was worried that it might be really cold"
11. "You're doing a great job honey" (sympathetic tone)
12. "What's wrong, buddy?" (sympathetic tone)

4. Procedure Time Talk and Commands to Child (PTC):

Any **present-focused** talk or commands to the child referring to the **amount of time** left in the wait period or cold pressor period. This category is for time talk that is **not** sympathetic in tone, it is said matter-of-factly, and it is usually in response to a question or statement by the child about how much time is left).

- Procedure time talk that is sympathetic in content and tone is coded as a 3 (SYMP).
- Commands to count and audible counting are coded as a 1 (NSFC) if they are clearly meant to distract the child (rather than help the child determine how much longer is left in the wait period or cold pressor period).
- Talk that includes mention of time or when the child will be told to take their hand out that is focused more on the rules and regulations or the sequence of events of the cold pressor task is coded as 5 (OPC).

1. *(In response to "When will it beep?")* "I don't know"
2. *(In response to "Come on – beep!")* "You just have to wait"
3. "I think it should beep soon"
4. "1 minute is a long time to wait"
5. "Not long now" (intention is not to reassure – just a general comment by the parent)
6. "You're not allowed to look at the watch – just wait"
7. "Is the beep loud?"
8. "Wouldn't it be something if we didn't hear the beep?"

5. <u>Other Procedure Talk and Commands to Child (OPC):</u>

Any talk, questions, or commands to the child referring to aspects of the cold pressor procedure other than time (e.g., how far into the water to put hand, sitting in chair). Includes talk about rules and regulations of the procedure (often including terms like "allowed to," or "supposed to"). Also included is talk about the cold pressor apparatus (e.g., cooler, pump, aspects of the water such as smell, appearance). Includes straightforward or matter-of-fact talk about the temperature of the water that does not include terms such as "cold" and does not compare the temperature of the water to something obviously cold.

- Procedure time talk is generally coded as a 4 (PTC); however, if an utterance includes mention of time or experimenter instruction for child to remove their hand but is mainly focused on the rules and regulations or sequence of events in the cold pressor task, it is coded here – see examples 15 & 16 below.
- Any negative talk by the parent (usually accompanied by a harsh tone of voice) is coded as 7 (CRIT).
- Talk about the water temperature that is sympathetic in content and tone is coded as a 3 (SYMP).
- Talk about the water temperature that includes terms such as "cold" or compares the temperature of the water to something obviously cold is coded as 2 (SFC).

1. "Just up to your wrist."
2. "Put your hand in now."
3. "Roll up your sleeve."
4. "There you go – put your hand in."
5. "Is it in up to your wrist?"
6. "Just up to here" *(indicating on wrist)*

7. “Sit up in your chair.”
8. “Stop wiggling around – sit still.”
9. “Which hand to you start with?”
10. ‘There’s a pump in there.”
11. “The water smells like chlorine?”
12. “Am I allowed to put my hand in to try it?”
13. “How many degrees is it?”
14. “Does it feel like the water at the hotel pool?”
15. “When it beeps, that’s when I put my hand in”
16. “You’re supposed to keep it in the whole time”

6. Humor to child (HUM):

Any statement made by the parent that is clearly intended to be humorous, is primarily light-hearted in tone, and is not symptom focused. Humor is often accompanied by laughter from the person making the statement and may evoke laughter in the child. Sarcasm may be coded as humor only if lighthearted in tone or if accompanied by laughter from the recipient. Humor includes:

- Outright jokes on the "one-liner" variety.
- Laughter by the parent.
- Statements that are purely facetious, outlandish or outrageous.
- Statements that emphasize the humorous aspects of something.
- Any humorous talk about the child's symptoms is coded as 2 (e.g. “Is your hand going to fall off?”)
- Humor that is critical (i.e., sarcastic comments that are not lighthearted in tone or received with laughter) is coded as 7 (CRIT).

1. “You are being silly willy!”
2. “You're a rascal!”
3. “I’m just kidding!”
4. “See any fish in there?”
5. “Watch out for piranhas!”
6. “Don’t get sucked in!”

7. Criticism to child (CRIT):

Any negative talk by the parent **about** the child. Criticism includes negatively evaluative terms referring to the child, statements of disapproval, statements pointing out something wrong about the child or the child's behavior. Sarcastic comments that are not lighthearted in tone or received with laughter are also included. Usually, criticism is accompanied by a **harsh** tone of voice. Criticism includes:

1. “Don’t be a wuss”
2. “You’re being ridiculous”
3. “You’re behaving badly”
4. “You're sure in a bad mood today”
5. “Look at that, you've already got a stain on that new shirt”
6. “That’s not nice.”

7. “Seth!”

8. Procedure-related praise to child (PRAS):
Any talk by the parent that praises the child that makes direct reference to their performance during the cold pressor task. Also included is praise to the child about waiting patiently during the wait period.

- Praise to the child about anything unrelated to the cold pressor procedure are coded as 1 (NSFC).

1. “You’re so brave!”
2. “Boy you’re tough!”
3. “You’re doing a good job waiting patiently”
4. “I’m impressed!”
5. “You’re strong!”
6. Wow – you’re doing so well!”

9. Parent talk about own pain symptoms (PPS)
Any talk by parents about their own pain symptoms, either past or present. This could include referencing pain they are currently experiencing, referencing how they cope with their own pain, or comparing cold pressor pain to their own chronic pain.

10. Other talk to child (OTC):
Verbal behavior that does not fit any other categories. This includes verbalizations that could not be transcribed (inaudibles) and sentences that are cut off in mid-stream before the meaning can be ascertained. Also includes any talk by the parent to clarify a child statement that was not understood /heard and talk about the parent’s own physical symptoms (other than pain) and the child’s past physical symptoms. Excluded from this category are “Yes,” “No,” etc. if those comments can be coded according to the context of the conversation.

- Parent talk about their own pain symptoms (past or present) is coded 9 (PPS)

1. *(In response to “I had a headache this morning in class.”)* “You had a headache?”
2. “Let me see that cut on your arm”
3. “Those lips of yours are really chapped.”
4. “Ummmm”
5. “What did you say?”
6. “Huh?”
8. “I’m hungry”
9. “Brr! That’s cold!” (parent puts *own* hand in the cold pressor)
10. *(In response to “I’m putting my hand against the wall”)* “Why are you doing that?”

Child to Parent Vocalizations

11. Cold/pain symptom talk to parent **(CPP)**

12. Anxiety talk to parent **(ANXP)**
13. Procedure time talk to parent **(PTP)**
14. Other procedure talk to parent **(OPP)**
15. Resistance talk to parent **(RESP)**
16. Child coping talk to parent **(CCP)**
17. Child talk about parent pain symptoms to parent **(CPPS)**
18. Child non-symptom focused talk to parent (**NSFP**)
19. Other talk to parent **(OTP)**

11. Cold/pain talk to parent (CPP):

Any symptom talk related to the child's *current* physical status that is related to the cold pressor task, including talk about pain, cold, hurting, discomfort, numbness, redness of hands, or other sensations. Includes cries and screams (verbalizations that indicate distress).

- Talk about the child's **past** physical symptoms is coded as 19 (OTP).

1. "Owwh!"
2. *(In response to "Does it hurt?")* "Yes!"
3. "It's freezing!"
4. "It feels uncomfortable"
5. *(In response to "Does it sting?")* "Yes."
6. "It hurts!"
7. "It stings!"
8. "My hand is totally numb!"
9. "My fingers are bright red!"
10. "I feel like I'm getting frostbite!"
11. "Ahhh!
12. "The water feels as cold as ice"

12. Anxiety talk to parent (ANXP):

Any talk related to the child's current feelings of anxiety, fear, or apprehension (often during the wait period). **Tone** is important in this code.

1. "I'm scared"
2. "What if it hurts?"
3. "What if it's freezing Mom?"
4. "Will I get hypothermia?"
5. "Will it give me frost bite?"

13. Procedure Time Talk to Parent (PTP):

Any talk, statement or questions to the parent referring to the amount of time left in the wait period or cold pressor period. Often the talk is about when the RA will tell them to take their hand out of the water. Often impatient in tone.

- The child audibly counting following a parent command to count is coded here if it represents an attempt to determine how much more time is left in the cold pressor task (if the parent intended it as a distraction strategy, the child's counting is coded as 19 - OTP)

1. "I want it to beep!"
2. "Come on, beep!"
3. "It's taking forever to beep"
4. "2 minutes is so long!"
5. "How much longer 'till it beeps?"
6. "I want to look at the watch!"
7. "Gotta listen for the beep"

14. <u>Other Procedure Talk to Parent (OPP):</u>

Any talk, questions, or commands to the parent referring to aspects of the cold pressor procedure other than time (e.g., how far into the water to put hand). Includes talk about rules and regulations of the procedure (often including terms like "allowed to," or "supposed to"). Also included is talk about the cold pressor apparatus (e.g., cooler, pump, aspects of the water such as smell and appearance). Includes straightforward or matter-of-fact talk about the temperature of the water that does not include terms such as "cold" and does not compare the temperature of the water to something obviously cold.

- Procedure time talk is coded as a 13 (PTP).
- Talk about the water temperature that includes terms such as "cold" or compares the temperature of the water to something obviously cold is coded as 11 (CPP).

1. "I just put it up to my wrist."
2. "Do I put my hand in now?"
3. *(In response to "Is it up to your wrist?")* "Yes!"
4. "I have to keep it in for as long as I can."
5. "Which hand do I use?"
6. 'There's a pump in there."
7. "So I'm allowed to take it out when I want right?" (note that this example isn't active resistance – if it was, it would be coded 17)
8. "The water smells funny"
9. "You're not supposed to put your hand in Mom!"
10. "How many degrees did she say it was?"
11. "It feels like the water at Crystal Crescent Beach"
12. "I have my hand in the water"
13. *(In response to "keep your hand in there")* "I know!" or "I am"

15. <u>Resistance talk to parent (RESP):</u>

Any talk by the child that indicates that they want to stop the cold pressor task (i.e., they want to take their hand out before 4 minutes is up). This code **only** applies to the cold pressor period.

- Talk about cold/pain symptoms are coded 11 (CPP).
- Talk about anxiety are coded 12 (ANXP).

1. "I have to take my hand out"

2. "I can't do it!"
3. "I want to stop!"
4. "I'm going to take it out now"
5. "I can't keep it in the whole time"
6. "Can I take it out Mom?"

16. <u>Child coping talk to parent (CCP):</u>

Any talk by the child that indicates courage or attempts to soothe himself or herself verbally. Also includes the child informing the parent of things he/she is doing as part of the cold pressor procedure that are helping him/her cope with the procedure – to qualify, the child statement must include a term such as "better" "warmer" or "more comfortable."

- If the child tells the parent things he/she is doing as part of the cold pressor procedure without using a term such as "better" or "warmer," it is coded as 14 (OPP).

1. "I'm alright"
2. "I think I can go the full time"
3. "It's really not that bad"
4. "It's not that cold"
5. "I can take it"
6. "I can go all the way!"
7. "I can keep my hand in"
8. "I actually like the cold"
9. "It feels good"
10. "It's warm and soothing like hot chocolate" (using imagination to cope)
11. "I'm tough"
12. "I'm almost there"
13. "It feels better when I make a fist"
14. "I'm moving my hand around so that it feels better"

<u>17. Child talk about parent pain symptoms to parent (CPPS)</u>

Any talk by children about the parent's pain symptoms, either past or present. This could include referencing pain the parent is currently experiencing, referencing the parent's pain symptoms that have occurred in the past, or comparing cold pressor pain to their parent's chronic pain.

<u>18. Child non-symptom focused talk to parent (NSFP)</u>

Any talk by the child that is **unrelated to the child's pain/cold symptoms or anxiety**. Included in this category are conversations, comments, or questions about pets, siblings, parents, school, toys, etc., or about the child's plans, wants, desires, etc. that are *unrelated* to cold/pain or anxiety symptoms the child is presently experiencing. Also included are factual statements and questions about lab equipment not including the cold pressor (e.g., video cameras, smart board, towel, questionnaires, consent form), order of events, lab staff, etc. General statements made by the child or just changing the subject is coded as 18 (NSFP).

- Talk about how much time is left in the cold pressor task is coded as 13 (PTP).
- Talk about the child's own past physical symptoms is coded as 19 (OTP).
- Talk about the parent's chronic pain symptoms is coded as 9 (PPS).

1. "What are we having for dinner tonight?"
2. "I want to talk about something else"
3. "Today at school we played baseball"
4. "We have this painting at school"
5. "Mmhmm" (child responding to parent conversationally in context of non-symptom focused conversation, i.e. verbal nodding)
6. "I can't wait until Christmas!"

19. <u>Other talk to parent (OTP):</u>

Verbal behavior that does not fit any other categories, such as talk about the child's **past** physical symptoms or symptoms unrelated to the cold pressor and humor. Any talk about the child's general symptoms that are not related to the current procedure or being experienced currently is included in this category. Includes verbalizations that could not be transcribed (inaudibles) and sentences that are cut off in mid-stream before the meaning can be ascertained. Also includes any talk by the child to clarify an adult statement that was not understood /heard. Excluded from this category are "Yes," "No," etc. if those comments can be coded according to the context of the conversation.

- If the child tells the parent things he/she is doing as part of the cold pressor procedure using a term such as "better," it is coded as 16 (CCP).
- If the child refers to the parent's pain symptoms, this is coded as 17 (CPPS).

1. "Can we go by the store on the way home?"
2. "I'm gonna have so much money saved up!"
3. "Those are the cameras"
4. "I can't sit still!"
5. "Doo dee doo dee doo" (humming)
6. "I had a headache this morning at school"
7. *(In response to "Do you have to pee?")* "No"
8. "My hands are really dry."
9. "Don't talk about how cold it is!"
10. "I'm making a fist with my hand"
12. "You put your hand in there Dad!"
13. "Can you…"
14. "Ummmm…"
15. "What?"
16. *(In response to "Are you excited?")* "Yes"