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The Interpersonal Context of Co-occurring Posttraumatic Stress Disorder Symptoms and Pediatric Chronic Pain: The Role of Parent Responses

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The Interpersonal Context of Co-occurring Posttraumatic Stress Disorder Symptoms and Pediatric Chronic Pain: The Role of Parent Responses

by

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Abstract

Introduction: Posttraumatic stress disorder (PTSD) symptoms occur at elevated rates among youth with chronic pain and their parents and are associated with worse youth pain outcomes. Interpersonal factors, such as parent distress and protective behaviours, have been posited as key mechanisms likely to influence the persistence of pain and PTSD symptoms in youth. Parent responses to youth pain have been shown to be dynamic over time, pointing to the importance of examining the interpersonal context of pediatric chronic pain using an ecological momentary assessment (EMA) approach. **Method:** The current study adopted a multi-method approach to examine the role of parent protective responses in the relationship between parent and youth PTSD symptoms and youth chronic pain outcomes, among a sample of youth with chronic pain and their parents. Eighty-four youth with chronic pain (70% female, M_{age} = 14.2 years), recruited from a tertiary level pediatric chronic pain program, and one of their parents, participated. At baseline, youth and one of their parents participated in a diagnostic clinical interview of internalizing mental health symptoms. Youth also completed self-report measures assessing pain intensity, pain interference, pain unpleasantness, and PTSD symptoms. Parents completed self-report measures of their own PTSD symptoms and protective responses to youth pain. Following baseline assessment, youth and parents completed daily electronic assessments of pain and parent protective responses for 7 days. Finally, youth and parents participated in a lab-visit, where parents and youth engaged in a discussion task following youth completion of the cold pressor task. **Results:** Findings revealed that higher parent PTSD symptoms predicted a stronger daily association between parent protective behaviours and youth pain unpleasantness, but not youth pain intensity or interference. However, parent protective responses measured with a static questionnaire, parents' stop tendency following an experimental pain task, and pain

attending talk during a discussion task following the experimental pain task, were not associated with youth or parent PTSD symptoms or youth pain outcomes. **Conclusion:** These findings suggest that parent protective responses to youth pain may be an interpersonal mechanism through which parent PTSD impacts children's chronic pain symptoms. Additionally, EMA methodology may be a more ecologically valid approach to studying pediatric chronic pain.

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Chapter 1: Introduction

Chronic pain, defined as pain occurring constantly or frequently for 3 months or more (Task Force on Taxonomy of the International Association for the Study of Pain, 1994), is prevalent among youth (King et al., 2011) and can lead to persistent pain problems into adulthood (Walker, Sherman, Bruehl, Garber, & Smith, 2012). Chronic pain in adolescence is also linked to increased risk of developing internalizing mental health disorders (anxiety, PTSD, depression) into adulthood (Noel, Groenewald, Beals-Erickson, Gebert, & Palermo, 2016). In addition, early life trauma (e.g., childhood abuse) is associated with increased risk of developing pain problems later in life (Davis, Luecken, & Zautra, 2005; Finestone et al., 2000; Powers et al., 2014). This is critical, given that up to 30% of youth experience one or more traumatic events by 16 years of age and up to 13% of those children develop posttraumatic stress symptoms (Copeland, Keeler, Angold, & Costello, 2007).

Posttraumatic stress disorder (PTSD) is the development of characteristic symptoms following exposure to actual or threatened death, serious injury, or sexual violence by either directly experiencing the traumatic event, repeated exposure to details of traumatic events, witnessing the event as it occurs to others, or learning the event happened to a close relative or friend (American Psychiatric Association, 2013). PTSD and chronic pain have been found to co-occur with high frequency in adults (Asmundson, Coons, Taylor, & Katz, 2002) and, for the first time, the prevalence of this co-occurrence was recently investigated in youth (Noel, Wilson, et al., 2016). Specifically, Noel et al. (2016) found that 32% of youth with chronic pain reported clinically elevated PTSD symptoms as compared to pain-free peers (8%). Moreover, parents of youth with chronic pain were also found to experience clinically elevated PTSD symptoms (20%) as compared to parents of youth without chronic pain (1%) (Noel, Wilson, et al., 2016).

Among youth with chronic pain *and* their parents, higher PTSD symptoms were related to worse youth pain outcomes (Noel, Wilson, et al., 2016). Thus, PTSD symptom elevations are common in youth with chronic pain and linked to worse pain and functioning in this already vulnerable group of youth.

Although the prevalence of co-occurring PTSD and chronic pain symptoms in youth and their parents have now been demonstrated, empirical research is only beginning to investigate the potential mechanisms underlying the relationships between parent and youth PTSD symptoms and youth chronic pain outcomes. In a cross-sectional examination, child catastrophic thinking was shown to mediate the relationship between parent and child PTSD symptoms and child chronic pain outcomes (Neville, Soltani, Pavlova, & Noel, 2018), demonstrating the potential importance of cognitive-affective factors of both parents and children in this co-occurrence. Conceptual models of PTSD and pain, as well as their co-occurrence, posit that this relationship should be viewed within an interpersonal context (Goubert et al., 2005; Holley, Wilson, Noel, & Palermo, 2016; Nelson, Cunningham, & Kashikar-Zuck, 2017). Specifically, parent responses to youth pain (e.g., protectiveness) have been proposed as a mechanism through which parent and youth PTSD symptoms may influence youths' chronic pain (Holley et al., 2016). In pediatric chronic pain populations, parent solicitous and protective responses towards their child's pain (i.e., parental reinforcement of and attention to pain behaviors, such as keeping the child home from school) have been shown to be robust predictors of worse child pain outcomes (Claar, Simons, & Logan, 2008; Guite, McCue, Sherker, Sherry, & Rose, 2011). Parent PTSD symptoms, such as hypervigilance and attentional bias to threat, may lead parents to be more aware of and distressed by their child's pain cues, and vulnerable to engaging in pain promoting/protective behaviours towards their child's pain. Indeed, parents who are more

distressed have been shown to engage in more solicitous and protective behaviours (i.e., positive or negative reinforcement for pain complaints) when their child is in pain, which serves to increase child pain and disability (Langer, Romano, Mancl, & Levy, 2014). Youth PTSD symptoms may also signal threat to parents and elicit these protective parental responses, which inadvertently exacerbates pain complaints.

Despite being implicated in models of the co-occurrence of PTSD symptoms and pediatric chronic pain, the role of parent protective behaviours in this relationship has not yet been examined. The purpose of the current study is to investigate the influence of parent protective responses in the parent PTSD-youth pain and youth PTSD-youth pain relationships, among a cohort of youth with chronic pain.

1.1 Pediatric Chronic Pain

The distinction between acute and chronic pain is traditionally recognized as a designated interval of time since the onset of pain, typically 3 months, and pain that persists beyond the expected period of healing (Task Force on Taxonomy of the International Association for the Study of Pain, 1994). Chronic pain is a highly prevalent health concern in children and adolescents, affecting 11- 38% of youth (King et al., 2011) and research suggests that this prevalence has grown over the past several decades (Sillanpaa & Anttila, 1996). The economic burden of pediatric chronic pain is estimated to be over $15 billion dollars CAD annually in total incremental health care expenditures, exceeding costs of childhood asthma and obesity (Groenewald, Wright, & Palermo, 2015). Chronic pain may include persistent (i.e., ongoing) or recurrent (i.e., episodic) pain and may develop from an underlying health condition, such as juvenile rheumatoid arthritis, or may be the disorder in and of itself, such as headaches or complex regional pain syndrome (Friedrichsdorf et al., 2016). Epidemiological research

suggests that the most frequent pediatric chronic pain conditions are migraine headaches, abdominal pain, and musculoskeletal pain (Goodman & McGrath, 1991), with the prevalence of most pain types being generally higher among girls and with increasing age (King et al., 2011).

Although many children are able to cope in the face of chronic pain and resume daily activities, approximately 3-5% of children with chronic pain are severely impacted, demonstrating moderate to high levels of pain-related disability, regardless of pain intensity (Hechler, Dobe, & Zernikow, 2010; Huguet & Miro, 2008). Moreover, greater pain severity is related to worse quality of life, increased school absences, and increased likelihood of visiting a health care professional and using pain medication (Huguet & Miro, 2008). Chronic pain has a profound impact on the daily functioning of youth and is associated with frequent school absences, sleep disturbance, emotional distress, and reduced participation in peer activities (Palermo, 2000). Pediatric chronic pain also impacts the broader family. Parenting a child with chronic pain is associated with anxiety, depressive symptoms, and role stress (Eccleston, Crombez, Scotford, Clinch, & Connell, 2004). Families of children with chronic pain have also been shown to have poorer family functioning as compared to healthy controls, with higher levels of family dysfunction associated with increased pain-related disability in youth (Lewandowski, Palermo, Stinson, Handley, & Chambers, 2010).

1.2 Pediatric Chronic Pain and Comorbid Mental Health Conditions

In addition to functional impairment and disability, high rates of internalizing mental health disorders are found in children with chronic pain (Coffelt, Bauer, & Carroll, 2013). Indeed, Coffelt et al. (2013) found that comorbid mental health disorders were common in a sample of children admitted to a tertiary-level pediatric hospital with chronic pain, with 28% of children experiencing a co-morbid mood disorder and 18% of children experiencing anxiety and

panic disorders. Moreover, chronic pain in adolescence is linked to greater lifetime risk of internalizing mental health disorders (anxiety [including PTSD] and depression) (Noel, Groenewald, et al., 2016; Shelby et al., 2013). In a longitudinal study of 8-17 year-old children with functional abdominal pain, Shelby et al. (2013) showed that not only was risk of current anxiety disorders higher in patients with chronic abdominal pain as compared to controls (30% vs 12%), but risk of lifetime anxiety disorders was also higher (51% vs 20%). Importantly, this study suggested that, regardless of whether or not pain resolves, chronic pain increases a child's vulnerability to developing anxiety disorders into adulthood (Shelby et al., 2013). A study by Noel et al. (2016) showed similar findings using a nationally representative sample of 14,790 adolescents with a variety of chronic pains, including headaches, stomach-aches, muscle and joint pain. Lifetime rates of anxiety (including PTSD) and depressive disorders were higher in individuals with a history of chronic pain in adolescence (21.1% and 24.5%, respectively) as compared to those without a history of adolescent chronic pain (12.4% and 14.1%, respectively) (Noel, Groenewald, et al., 2016).

For decades, PTSD and chronic pain have been found to be highly co-morbid in adults (Asmundson et al., 2002) and yet the prevalence of PTSD symptoms in youth with chronic pain has only recently been demonstrated (Noel, Wilson, et al., 2016). Noel et al. (2016) examined PTSD and pain symptoms in cohorts of youth with versus without chronic pain, and their parents. Findings revealed that youth with chronic pain and their parents reported *higher* rates of clinically elevated PTSD symptoms as compared to pain-free peers and their parents. Moreover, among youth with chronic pain, both youth *and* parent PTSD symptoms were linked to worse youth pain (Noel, Wilson, et al., 2016). This study demonstrated the importance of investigating PTSD at the symptom level, even if parents or children do not meet criteria for a clinical

diagnosis. Given that children with co-morbid chronic pain and clinically significant anxiety have been shown to be less likely to respond to cognitive behavioural therapy for chronic pain (Cunningham et al., 2016), understanding how and why co-occurring mental health disorders/symptoms influence chronic pain outcomes and functioning is crucial. Moreover, Noel et al. (2016) suggested that not only are children's own mental health symptoms critical to consider in the co-occurrence of PTSD and chronic pain, but symptoms of parents may be equally important targets in assessment and intervention to improve youth chronic pain trajectories. Indeed, this interpersonal context is considered core in conceptual models of co-occurring PTSD and pediatric chronic pain (Holley et al., 2016). However, to date, the role of parent behaviours in this co-occurrence has not been examined.

1.3 Conceptual Models of the Co-occurrence of Pain and PTSD

Although PTSD symptoms have been found to be highly prevalent among youth with chronic pain and their parents, there is little understanding of the modifiable mechanisms underlying this comorbidity that influence children's response to pain. Theoretical models of potential mechanisms underlying the co-occurrence of PTSD and chronic pain in adults posit that shared vulnerabilities and/or mutually maintaining factors (e.g., attentional biases toward threatening stimuli and anxiety sensitivity) contribute to the development and maintenance of both conditions (Asmundson et al., 2002).

A recently proposed conceptual framework for the *pediatric* period, highlighting potential factors underlying the association between PTSD symptoms and chronic pain in youth, uniquely underscores the importance of interpersonal influences (Holley et al., 2016). Specifically, Holley et al. (2016) proposed parent distress (including their own PTSD symptoms), parent behaviours (protectiveness), and parent-child interactions as interpersonal

mechanisms likely to influence the persistence of pain and PTSD symptoms in youth. For example, parent distress may increase parents' risk of engaging in maladaptive parent protective responses (e.g., removing them from pain-inducing activities), which encourage children's avoidance behaviour and pain interference. Similarly, Nelson et al. (2017) recently presented a conceptual framework adapted from the biopsychosocial model of pain, to understand the role of adverse childhood experiences (ACEs) in pediatric chronic pain, underscoring the potential impact of the broader family environment on chronic pain symptoms. Like the model of co-occurring PTSD and chronic pain proposed by Holley et al., (2016), this model proposed family factors (e.g., parental overprotective behaviours, parent-child communication styles and parental coping) as important to investigate in the relationship between ACEs and chronic pain in youth. Indeed, following a traumatic event (e.g., natural disaster), lack of parental support, family conflict, and parental overprotectiveness have been associated with increased PTSD symptoms in youth (Bokszczanin, 2008). Parents can influence children's chronic pain symptoms through reinforcement of pain behaviours (e.g., allowing the child to avoid activities due to pain) and psychological responses (e.g., distress) (Asmundson, Noel, Petter, & Parkerson, 2012). Likewise, relationships between parent and child responses in the face of pain are likely bidirectional, reciprocally influencing each other over time (Asmundson et al., 2012).

1.4 Parent Factors in Pediatric Chronic Pain

It is well established that pain should be viewed in the interpersonal context (Craig, 2009; Goubert et al., 2005). Research has made important advances in understanding the profound influence of parents and family on children's chronic pain symptoms (Palermo & Chambers, 2005), as well as the impact of parenting a child with chronic pain (Palermo & Eccleston, 2009). Palermo and Chambers' (2005) framework of parent and family factors in children's chronic

pain integrates two important theoretical models: Operant-behavioural theories (i.e., parent social reinforcement) and family systems theories (i.e., the broader family environment). This framework posits that individual parenting factors (e.g., parenting reinforcement/solicitousness) should be considered within the context of dyadic variables (e.g., parent-child interactions) and the broader family environment (e.g., family functioning), emphasizing the reciprocal and complex relationships between pain, parent, and child factors (e.g., child gender, developmental status and emotional symptoms) interacting at all levels of the model. It follows that, as posited by Holley et al. (2016), the influence of parent and family factors on the co-occurrence of PTSD symptoms and chronic pain in youth are critical to understand.

1.4.1 Parent Responses to Pain

Social learning theory (Bandura, 1977) is often implicated in models of pediatric pain, wherein children may learn through observing another's response to pain (e.g., parental modeling) (Goodman & McGrath, 2003) as well as from the parent's responses towards the child's pain (e.g., parental reinforcement for child pain behaviours) (Walker & Zeman, 1992). The influence of parental modelling of pain behaviours has been demonstrated experimentally, wherein children observed their mother complete an experimental pain induction cold water task and then subsequently completed the task themselves (Goodman & McGrath, 2003). Children whose parents were instructed to exaggerate their pain behaviour during the task demonstrated a lower pain threshold compared to a control and minimize condition (Goodman & McGrath, 2003). As well as mere observation of a behaviour, social learning theory posits that behaviour leading to a positive consequence is more likely to be learned and maintained (Bandura, 1977). Thus, a child may be more likely to engage in pain behaviour if both exposed to a model of the behaviour (i.e., vicarious learning through a parent) and if this behaviour results in a positive

outcome (i.e., operant learning such as parent attention or avoidance of a pain inducing activity). Although evidence of modeling is limited, children with unexplained pain (currently understood as “Primary Pain Disorders”) (Schechter, 2014) have been shown to be more likely to identify models of pain behaviour in their environment and more positive reinforcement from pain behaviour compared to children with explained pain (Osborne, Hatcher, & Richtsmeier, 1989).

Parent responses to their child in the face of pain have been found to lessen or exacerbate children’s pain symptoms (Palermo & Eccleston, 2009). For example, Walker et al. (2006) found that, during an experimental pain inducing task with children with recurrent abdominal pain, children’s symptom complaints doubled when parents were trained to use pain attending talk (e.g., What does it feel like?) and reduced by half when parents were trained to use non-attending talk (e.g., What would you like to do this evening?). Parent protective responses, characterized by attending to pain symptoms and provision of special privileges, are also associated with an array of maladaptive child outcomes, including increased somatic symptoms, functional disability (Claar et al., 2008; Langer, Romano, Levy, Walker, & Whitehead, 2009), and poor school attendance and functioning (Logan, Simons, & Carpino, 2012). Higher parental distress has been linked to the tendency to restrict their child’s activity (Caes, Vervoort, Eccleston, Vandenhende, & Goubert, 2011), and to worse child pain intensity and disability (Logan & Scharff, 2005; Ross et al., 1993). Moreover, parental protectiveness has been shown to mediate the relationship between parent pain-specific distress and child functional disability (Sieberg, Williams, & Simons, 2011). Teaching parents behavioural strategies for child adaptive functioning is a critical component of cognitive behavioural therapy for pediatric chronic pain (Eccleston et al., 2014). However, more recent approaches to the management of chronic pain also importantly target parent distress (Palermo et al., 2016).

Parents who are enduring distress when faced with their child in pain, may be more vulnerable to engaging in self-oriented maladaptive behavioural responses (e.g., keeping the child home from school) in an effort to reduce their own distress (Simons, Goubert, Vervoort, & Borsook, 2016). Indeed, parents of healthy children and children with chronic pain, who report higher levels of distress have been shown to exhibit a greater tendency to want to stop their child's pain during an experimental pain induction task (i.e., stop tendency), which is a measure of parent protective response (Caes et al., 2011). Parenting a child with chronic pain is associated with significant role stress, anxiety and depressive symptoms (Eccleston et al., 2004), which may increase parents' vulnerability to engaging in these maladaptive parenting behaviours. The Pediatric Fear-Avoidance Model of chronic pain explains the bidirectional nature of relationships between parent behaviours (e.g., protectiveness), parent affective and psychological responses (e.g., distress), and child psychological responses (e.g., fear, catastrophizing) in encouraging and/or maintaining child avoidance behaviour, which serves to fuel child pain and disability (Asmundson et al., 2012). Taken together, parent and child responses to pain may interact in a cyclical manner that perpetuates and maintains child fear, avoidance and disability, and ultimately pain chronicity.

Lab-based observation of parent behaviours has been largely limited to healthy (i.e., without chronic pain) samples undergoing experimental acute pain, which may lack validity in chronic pain samples. In a study comparing experimental pain responses in children with and without chronic pain, children with chronic pain were found to be significantly more likely to tolerate the cold pressor task compared to healthy children, however, pain intensity ratings did not differ between the two groups (Tsao, Evans, Seidman, & Zeltzer, 2012). Thus, it is possible that for children with chronic pain, the cold pressor task is less distressing. Moreover, most

research on parent responses to child chronic pain has relied on single report of pain symptoms and parent behaviours, with an underlying assumption that symptoms and responses remain consistent over time. Connelly et al. (2017) recently extended this work by applying an innovative smart-phone based momentary assessment methodology to investigate the *dynamic* influence of parent responses on children's pain. Children with juvenile idiopathic arthritis and their parents completed e-diaries 3 times per day for 30 days. This study found that parent protective responses to child pain predicted a subsequent increase in child pain intensity and interference. In addition, parents engaged in *more* protective behaviours when/after their child reported more pain symptoms (Connelly, Bromberg, Anthony, Gil, & Schanberg, 2017). Moreover, parents tended to engage *less* in protective responses at times when they reported higher levels of positive affect (Connelly et al., 2017). Additionally, parent responses to child pain varied more within caregivers than between them, demonstrating that parent responses are dynamic over time and underscoring the importance of examining these responses using a daily, ecologically-valid methodological approach. This study demonstrates that parents' own emotional state has a significant influence on their responses to child pain, supporting the importance of investigating parent affect/mental health in examinations of child chronic pain. Indeed, previous research has shown that a reduction in parent distress following cognitive-behaviour-based intervention for parents of youth with chronic pain leads to fewer parent protective responses as well as decreases in child pain intensity (Noel, Alberts, et al., 2016). The ecological momentary assessment (EMA) methodology used by Connelly et al. (2017) has not yet been used to examine parent responses and child pain outcomes among children with idiopathic chronic pain or the impact of parent and child mental health on these relationships.

Like parent distress, parent PTSD symptoms, such as intrusive thoughts and hyperarousal, may impede parents' ability to generate and engage in adaptive responses to pain, making them more prone to engaging in protective behaviours. Parents with elevated PTSD symptoms may also display selective attention to threat and hypervigilance, thereby increasing their sensitivity to children's pain cues and their likelihood of interpreting pain as highly threatening. This would, in turn, fuel maladaptive responding characterized by avoidance and reinforcement for pain (e.g., protective behaviours). Alternatively, children with elevated PTSD symptoms may be hypervigilant to their own pain symptoms and express their pain to parents more often and intensely. Indeed, higher PTSD symptoms in youth with chronic pain have been associated with increased child pain catastrophizing (Neville et al., 2018), which has been associated with increased tendency for children to verbally and non-verbally express their pain (Vervoort et al., 2008). If parents are more likely to respond with protective behaviours in response to children's increased reporting of pain complaints (Connelly et al., 2017), which are likely to be most pronounced when parental and child PTSD symptoms are high, then this would be expected to exacerbate child pain symptoms. Children's anxiety symptoms have also been found to moderate the relationship between parental protective behaviours and children's functional disability (Claar et al., 2008). Thus, it is likely that youth with PTSD symptoms may be more vulnerable to the effects of these maladaptive parent responses. Nevertheless, despite being strongly implicated as an underlying mechanism in models of PTSD and chronic pain as well as their co-occurrence, parent behaviours have not yet been empirically investigated. Better understanding the role of parents in this co-occurrence may inform future interventions to improve pain trajectories and mental health symptoms in these vulnerable youth.

The research discussed herein supports the need to adopt a multi-method approach to investigating youths' chronic pain, which is a strength of the current study. This is the first study to examine the influential role of parent behaviours (assessed using momentary daily assessments, questionnaires, and observed lab interactions) in the relationship between youth and parent PTSD symptoms and youth pain outcomes among a sample of youth with chronic pain and their parents.

1.5 Research Aims and Hypotheses

Aim 1: To examine the role of parent and youth PTSD symptoms in the daily relationship between protective parent responses and youth pain outcomes. Hypothesis 1: Higher parent and youth levels of PTSD symptoms assessed at baseline will predict stronger daily associations between parent protective behaviours and youth pain outcomes (i.e., pain intensity, interference and unpleasantness).

Aim 2: To examine the role of parent protective responses assessed in the laboratory setting in the co-occurrence of parent and youth PTSD symptoms and youth pain outcomes. Hypothesis 2: Greater parent protective responses (i.e., parent stop tendency, pain attending talk following experimental pain induction with youth, static questionnaire-based parent report of protective responses) will mediate the relationships between parent and youth PTSD symptoms and youth chronic pain outcomes (i.e., pain intensity, interference and unpleasantness).

Chapter 2: Methods

The current study is part of the Pain and Mental Health in Youth (PATH) Study, a larger longitudinal research project investigating internalizing mental health disorders in a cohort of youth with chronic pain and biopsychosocial factors (e.g., cognitive biases, sleep disturbance, neurobiological factors) underlying the co-occurrence of PTSD and pediatric chronic pain. The

PATH Study is approved by the University of Calgary's Human Research Ethics Board (REB15-3100). The current masters project utilized data collected at baseline, 7 days, and the subsequent lab visit. Three-month follow-up data was not included.

2.1 Participants

One hundred and ninety-three youth with chronic pain and one of their parents were recruited from the Vi Riddell Outpatient Pain Rehabilitation Program, including Headache, Abdominal Pain and Complex Pain clinics, at the Alberta Children's Hospital (ACH). Of those, 64 declined participation and 39 were not eligible. Thus, 90 youth and one of their parents consented to participate in the study, however, six parent-youth dyads withdrew prior to beginning participation. The current sample includes 84 youth with chronic pain (70% female, M_{age} = 14.2 years [SD=2.29], range = 10-18 years) and one of their parents (94% mothers). Youth were eligible for the study if they were between 10-18 years of age, were identified as having chronic pain (pain $\geq$ 3 months) without an underlying disease (e.g., juvenile arthritis or cancer) by a provider in the pain clinics and reported ongoing pain upon recruitment screening. Exclusion criteria included being unable to read/speak English, diagnosis of a severe cognitive impairment, developmental disorder, schizophrenia spectrum or other psychotic disorders, the inability to access the internet, and/or presence of a serious chronic health condition.

Youth were referred to, and/or enrolled in, the headache (56%), complex pain (43%), and abdominal pain (1%) clinics. Youth reported experiencing the most pain in their stomach (19%), head (63.1%), muscles and joints (27.4%), legs (16.7%), chest (10.7%) and other (25%). Youth reported an average pain intensity level in the past week of 5.59 out of 10 (SD = 1.92) and average pain duration of 3.67 years (SD = 3.40). Sociodemographic data are presented in Table 1.

2.2 Procedure

Parents were first approached by a member of the pain clinic staff for permission to be contacted by the research team. The clinical staff of the Vi Riddell Pain Program provided the contact information of new patients, as well as patients who had received care in the chronic pain clinics within the last year, to study team members at the Alberta Children's Pain Research lab. Participants who had previously participated in a Clinical Outcomes Study (REB14-0162) and who consented to be contacted about future studies were also contacted by research staff. Families who showed interest in the study were screened for eligibility over the phone. At this time, research staff addressed any questions the family had and either conducted or scheduled a consent conference phone call with both the parent and youth. During the consent call, the research staff explained the information included in the consent form, including details of the study and limits of confidentiality, and scheduled the lab visit. Parents and youth consented/assented using an online consent form and completed questionnaires using Research Electronic Data Capture (REDCap), a secure online data collection tool, a secure online web-based application, for online completion (Harris et al., 2009). Hardcopy, written informed consent was also obtained at the time of the lab visit. For a timeline of the current study see Figure 1.

2.2.1 Diagnostic Assessment of PTSD

At baseline, one week prior to the lab visit, youth completed a diagnostic assessment of internalizing mental health disorders over the phone with a researcher using the Schedule for Affective Disorders and Schizophrenia for School Aged Children (Kiddie-SADS). Clinician-administered interviews are the gold standard for assessing clinical diagnoses in youth and were used to characterize youth PTSD at the diagnostic level. These diagnostic interviews were

administered by trained clinical psychology graduate students. Rates of youth PTSD diagnoses were expected to be lower as compared to symptom elevations from questionnaire data. Diagnostic clinical interviews were, thus, planned to describe the sample in terms of diagnostic status. Based on research and conceptual models (e.g., Holley et al., 2016; Noel, Wilson, et al., 2016), PTSD symptoms were used to investigate aims and hypotheses of the current study.

2.2.2 Ecological Momentary Assessment

For 7 consecutive days prior to a lab visit, youth completed daily electronic assessments of their pain intensity, pain interference, and pain unpleasantness. Electronic daily diaries have been demonstrated to be feasible among children with chronic pain, as well as have been shown to increase compliance and accuracy of daily pain reporting compared to paper formats (Palermo, Valenzuela, & Stork, 2004). Parents completed daily assessments of their own protective responses to their child's pain. Similar to Connelly et al. (2017), this provided a measure of daily behaviour in the more naturalistic setting of the home. Secure links to the REDCap surveys were emailed or texted (depending on parent and youth preference) to parents and youth at 6pm each evening.

2.2.3 Laboratory Assessment

One week following the baseline assessment, youth and one of their parents visited the Alberta Children's Pain Research Lab located in the Vi Riddell Pain & Rehabilitation Center at Alberta Children's Hospital. Prior to the lab visit, youth and one of their parents completed self-report measures via REDCap assessing pain intensity and interference, PTSD symptoms, and parent responses to youth pain. During the lab visit, youth participated in the cold pressor task, which is an ethically acceptable experimental pain task commonly used in pediatric research (Birnie, Noel, Chambers, von Baeyer, & Fernandez, 2011). During this task, youth submersed

their hand in cold 10ºC water, cooled by an electric-apparatus, for a maximum of 4 minutes while their parent observed via video in an adjoining room. Youth were instructed to leave their hand in the water for as long as they could, even if it became uncomfortable. They were also told that they could remove their hand from the water at any time if it became too uncomfortable or hurt too much, thus giving them control over the pain stimulus (considered critically important for ethical acceptability). Youth were not told about the 4-minute time limit prior to the task. Use of uninformed ceilings, such as this, have been shown to provide greater variability in hand exposure to water and are recommended for measurements of pain tolerance (Birnie, Petter, Boerner, Noel, & Chambers, 2012; von Baeyer, Piira, Chambers, Trapanotto, & Zeltzer, 2005). The researcher was seated behind the youth during the cold pressor task to monitor participation while minimizing possible examiner influence. Immediately following the cold pressor task, parents were asked how much they wanted to stop their child's participation in the cold water task (i.e., stop tendency) as a measure of their tendency to respond protectively during the task. The parent and youth were then reunited in the observation room where the task took place, and were instructed to talk as they normally would about the cold water task. This conversation was audio recorded and transcribed verbatim for subsequent analysis. These controlled parent-youth interactions following evoked pain were coded for pain attending vs non-attending utterances based on a coding scheme developed by Walker et al. (2006) and extended by Vervoort et al. (2011) and Moon et al. (2011). A random sample of 20% of the transcripts were coded by a second coder to determine interrater reliability using intra-class correlation (Walker et al., 2006).

By incorporating both a controlled, lab-based discussion and EMA, this study uniquely captured both processes as they unfold in daily life and controlled snapshots of parent-youth interactions around acute pain in the lab. As a token of appreciation, youth and parents each

received a gift card after the Kiddie-SADS interview (worth $20 and $10, respectively). Following the lab visit, the parent and youth also each received a $25 gift card.

2.3 Measures

2.3.1 Baseline Measures

Demographic information. Parents reported on their own and their child's sex, ethnicity, child's age, and household annual income.

Pain characteristics – youth. Youth completed the valid and reliable Pain Questionnaire (Palermo et al., 2004). Youth reported on which part of their body they experience the most pain as well as how long their pain problem has been present in years and months. Youth reported their average pain intensity in the past 7 days using an 11-point numeric rating scale (0 = "no pain", 10 = "worst pain possible") and their average pain unpleasantness using a 5-point Likert scale from "Not at all" to "Very much."

Pain interference - youth. Pain interference was assessed using the Patient-Reported Outcomes Measurement Information System (PROMIS-25) Profile pain interference subscale. The four items of the Pain Interference subscale are rated using a 5-point Likert scale from "never" to "almost always" (Irwin et al., 2012). A total score of pain interference is obtained by summing responses. This total is subsequently transformed into standardized T-scores used for analyses. Internal consistency for the baseline measure of youth pain interference was excellent ($\alpha = .82$).

PTSD diagnoses - youth. The Kiddie-SADS-Present and Lifetime Version (K-SADS-PL) was administered to assess internalizing mental health disorders (i.e., PTSD) at the diagnostic level. The K-SADS-PL is a semi-structured clinical interview used to diagnose current and lifetime history of psychopathology in children ages 6-18 years, according to the

Diagnostic and Statistical Manual of Mental Disorders- 5th Edition (DSM-5) criteria. Trained clinical psychology students administered the K-SADS-PL by interviewing both the parent and youth separately. The K-SADS-PL consists of an unstructured Introductory Interview, a Diagnostic Screening Interview, a Supplement Completion Checklist, applicable Diagnostic Supplements, the Summary Lifetime Diagnoses Checklist, and the Children's Global Assessment Scale. Where discrepancies take place between the parent and youth report, clinical judgment is used to complete the summary lifetime diagnosis. All interviews were audio-recorded. The K-SADS-PL has been shown to have good reliability and predictive validity (Kaufman et al., 1997). The use of a gold standard diagnostic clinical interview to assess PTSD is important to provide a prevalence rate of PTSD at the diagnostic level in this sample of youth with chronic pain.

PTSD symptoms - youth. PTSD symptoms in youth were assessed using the 27-item self-report Child PTSD Symptom Scale (CPSS-5), which assesses PTSD symptoms according to DSM-5 criteria (Foa, Johnson, Feeny, & Treadwell, 2001). This questionnaire asks youth to identify a scary or upsetting incident (e.g., a car accident) and subsequently rate 20 symptoms on a 5-point Likert scale from 0 = "not at all" to 4 = "6 or more times a week/almost always". Seven impairment items are then rated as present or absent. Items are summed to yield a total symptom severity score. Higher scores represent higher levels of PTSD symptoms and impairment. The previous version of the scale (CPSS-4) demonstrated excellent reliability and validity (Nixon et al., 2013). The scale has been previously used to assess PTSD symptoms in children with chronic pain (Noel, Wilson, et al., 2016). Internal consistency was excellent ($\alpha = .95$) in this sample.

PTSD symptoms – parent. PTSD symptoms of parents of youth with chronic pain were assessed using the 20-item self-report PTSD Checklist for DSM-5 (PCL-5) (Weathers et al.,

2013). This measure asks parents to identify of the worst event that has ever happened to them and subsequently rate how much 20 symptoms bothered them in the past month on a 5-point Likert scale, from 0 = "not at all" to 4 = "extremely". A total score of PTSD symptom severity is obtained by summing item responses with higher scores representing higher levels of symptom severity. The PCL-5 has been found to have excellent reliability and validity (Blevins, Weathers, Davis, Witte, & Domino, 2015), has been used in previous research with parents of children with chronic pain (Noel, Wilson, et al., 2016), and has a suggested clinical cut-off score of 33 ("U.S. Department of Veteran Affairs. PTSD: National Center for PTSD. PTSD Checklist for DSM-5 (PCL-5). Available at: http://www.ptsd.va.gov/professional/assessment/adult-sr/ptsd-checklist.asp. Accessed June 28, 2018,"). Internal consistency was excellent (α = .94) in this sample.

Parent reported protective responses. Parent protective responses to youth pain were measured using the protect scale of the Adult Responses to Children's Symptoms (ARCS) (Van Slyke & Walker, 2006). Using a 5-point scale ranging from "never" to "always," parents reported on the frequency with which they engage in various behaviours (e.g., "Give your child special privileges") when their child has pain. The ARCS has been shown to be reliable and valid in samples of youth with chronic pain (Noel, Alberts, et al., 2016). Based on recent research (Noel, Palermo, et al., 2015) examining the factorial validity of the ARCS separately in caregivers of children versus adolescents with chronic pain, the current study used this newly derived ARCS scoring (for details regarding scoring see Noel, Palermo, et al., 2015). Responses are averaged to provide a subscale score, with higher scores indicative of greater parent protective behaviours. Internal consistency for the ARCS protect scale reported at baseline was good (α = .87 for caregivers of children and .75 for caregivers of adolescents).

2.3.2 Laboratory Based Measures

Parent pain attending talk. The parent-youth discussion task transcripts were coded for utterances based on a coding scheme developed by Walker et al. (2006) and extended by Vervoort et al. (2011) and Moon et al. (2011). Accordingly, parent utterances were coded for 1) Pain Attending Talk (i.e., utterances that focus on the child's pain), 2) Pain Non-Attending Talk (i.e., utterances that do not focus on the child's pain) and 3) Other talk (i.e., utterances that are inaudible or about technical aspects of the cold pressor task (Moon, Chambers, & McGrath, 2011; Vervoort et al., 2011; Walker et al., 2006). This coding scheme has been previously used in research involving a parent-child discussion task following the cold pressor task with healthy children (Vervoort et al., 2011). Following procedures used by Vervoort et al. (2011), the number of utterances coded under Parent Pain Attending Talk were divided by the total number of parent utterances to provide a proportion score of Parental Pain Attending Talk used in analyses. Parent pain attending talk is a measure of protectiveness as, by definition, parent protective responses are positive or negative pain attending and reinforcing behaviours. The intra-class correlation reliability coefficient between coders was adequate at .70 (Vervoort et al., 2011).

Parent stop tendency. Immediately following observation of their child completing the cold pressor task, parents completed the Parental Stop Tendency Scale (Caes et al., 2011). Parents were asked to rate how much they wanted to stop the cold pressor task on an 11-point numeric rating scale, from 0 = "not at all" to 10 = "a lot". Using this scale, parents of healthy children who report greater distress have been shown to demonstrate a greater tendency to want to stop their child's pain inducing activity, which may simulate parents' tendency towards protective behaviours (i.e., restricting the child's activity) (Caes et al., 2011). Immediately

following the cold pressor task, parents also reported on how anxious, worried and upset they felt on an 11-point numeric rating scale, from 0 = "not at all" to 10 = "Extremely". Additionally, parents reported on how much pain they thought their child felt during the cold water task on an 11-point numeric scale, from 0 = "no pain" to 10 = "worst pain possible".

Youth pain. Youths' pain tolerance during the cold pressor task was measured as the duration of immersion in seconds from the time the hand was placed in the water to the time it was voluntarily withdrawn (von Baeyer et al., 2005). Immediately following the cold pressor task, youth reported their pain intensity during the task using an 11-point numeric rating scale (0 = "no pain", 10 = "worst pain possible"), and their average pain unpleasantness using a 5-point numeric scale from 1 = "Not at all" to 5 = "Very much."

2.3.3 Ecological Momentary Assessment Measures

Similar to Connelly et al. (2017), parents reported on their responses to their child's pain using the ARCS protect scale, once daily for 7 days. Internal consistencies for the ARCS protect scale reported at each time point of daily measures ranged from .73 to .92 for caregivers of children (mean α = .86), and .77 to .87 for caregivers of adolescents (mean α =.81).

Youth reported on their pain once daily for 7 days using the measures of pain intensity, unpleasantness and interference, described above. Internal consistencies for pain interference reported at each time point of daily measures ranged from .80 to .91 (mean α = .86).

Chapter 3: Statistical Analyses

Statistical analyses were conducted using Mplus version 7.1 (Muthen & Muthen, 2012) and the Statistical Package for the Social Sciences (SPSS) Version 24.

3.1 Ecological Momentary Assessment Analyses

To address hypothesis 1, that higher parent and youth levels of PTSD symptoms assessed at baseline would predict stronger daily associations between parent protective behaviours and youth pain, multilevel modeling was used to account for repeated measurements (Level 1) nested within each parent-youth dyad (Level 2). This is considered the most appropriate analysis for EMA data (Affleck, Zautra, Tennen, & Armeli, 1999). Seventy-six dyads completed up to seven repeated measurements, thus, the current sample size exceeded the basic sample size recommendations of 30 units at the Level 2 of analysis for adequate power and non-biased estimates (Maas & Hox, 2005).

Intra-class correlation coefficients (ICC) of all repeated measure variables (i.e., pain intensity, pain interference, pain unpleasantness, and parent protectiveness) were calculated to justify a multilevel modeling approach (Hox, 2010). ICC values (.03, .06, .06, .07, respectively) indicated that most of the proportion of total variance of these variables was due to within-person differences (Marcoulides & Schumacker, 2009).

Multilevel path analysis is ideal for analyzing complex models because it allows for all outcomes to be correlated at each point in time and because it allows for the simultaneous estimate of multiple path coefficients in one overall model (Lleras, 2005). Following this, a multilevel path analysis was conducted using Mplus version 7.1 (Muthen & Muthen, 2012), in which a 2-level path model was estimated. Specifically, to investigate the daily associations between predictors and youth outcomes, the between-person (i.e., level 2) x within-person (i.e., level 1) part of the 2-level path model was specified (Maas & Hox, 2005). In other words, the level-1 variables (i.e., youth pain intensity, youth pain unpleasantness, youth pain interference, and parent protectiveness) were decomposed into within- and between-person level relationships (Zhang, Zyphur, & Preacher, 2009). In this part of the model, the daily associations between

parent protectiveness and youth pain intensity, youth pain unpleasantness, and youth pain interference were predicted. To investigate the direct effect of stable parent/youth predictors (i.e., PTSD) on youth pain outcomes, the between-person part of the 2-level path model, reflecting the direct relationship of parent PTSD symptoms and youth PTSD symptoms on youth pain intensity, youth pain unpleasantness, and youth pain interference, was specified. In addition, the cross-level interaction effect of parent PTSD symptoms on the daily association between parent protectiveness and youth pain intensity, youth pain unpleasantness, and youth pain interference was specified. Finally, the cross-level interaction effect of youth PTSD symptoms on the daily association between parent protectiveness and youth pain intensity, youth pain unpleasantness, and youth pain interference was specified. Maximum likelihood was used with robust standard errors as estimator in the path analysis. Parameters were standardized estimates to facilitate the interpretation of our results (Hox, 2010).

Throughout these analyses, the Full Information Maximum Likelihood (FIML) method was relied on to reduce response bias (Duncan, Duncan, & Strycker, 2006). When using FIML, missing values (either by not having completed a full wave of data collection or just one item or one scale) are not deleted, replaced or imputed, but the missing data is handled within the analysis model. This method allows for all available information to be used to estimate the model. It is also superior to listwise deletion as no information is lost in the estimation of the analysis model. FIML estimates the population parameters that would most likely have produced the estimates from the sample data (see Collins, Schafer, & Kam, 2001).

Acknowledging the possibility of reverse causation (i.e., that youth pain could drive parent responses to their child), BIC values of our hypothesized models to BIC values of their alternative models were compared. The hypothesized models, in which parent protective

responses drove youth pain intensity (BIC = 49.29), pain interference (BIC = 164.87), and pain unpleasantness (BIC = 95.59) consistently fit the data better than alternative models, in which youth pain intensity (BIC = 174.38) pain interference (BIC = 188.88), and pain unpleasantness (BIC = 180.59) drove parent protective responses. Thus, analyses proceeded with the hypothesized models. In line with previous research on pediatric chronic pain (King et al., 2011), youth age and sex were controlled for in all analyses by estimating their association with the outcome variables of interest.

3.2 Laboratory Based Assessment Analyses

Mediation models to address hypothesis 2, that greater parent protective responses (i.e., parents' stop tendency, pain attending talk in the laboratory, and parent reported protective responses) would mediate the relationship between parent and youth PTSD symptoms and youth chronic pain outcomes, are represented in Figure 2. Statistical analyses were conducted using SPSS version 23 (IBM Corp, Armonk, NY). With a sample of 84 dyads, on the basis of previously published data (Caes et al., 2011) and power of .8, the current sample size meets that required on the basis of a priori statistical power calculations (G*Power) (Faul, Erdfelder, Buchner, & Lang, 2009). A power analysis for proposed linear multiple regression analyses with a medium effect size ($f^2 = .15$, $\alpha = .05$) suggested a total sample size of 77 dyads would provide 80% power to detect group differences and interactions in the proposed study design. Descriptive statistics were conducted to characterize the sample and calculate mean scores on key variables. Independent samples *t* tests were used to compare key variables by sex.

Cases with missing values represented less than 10% of total cases for all variables and missing data were found to be missing completely at random (Little, 1988). Thus, pairwise deletion was utilized to handle missing data in order to retain the maximum amount of available

data (Bennett, 2001). To maximize robustness of hypothesis testing and to address issues of non-normality, bootstrapping using 5000 samples was planned to be used. Bivariate Pearson correlations were conducted between the variables of interest to justify the inclusion or exclusion of these variables in subsequent mediation analyses (Noel, Rabbitts, Tai, & Palermo, 2015). Correlational analyses were conducted using two-tailed hypothesis testing. If significant correlations between key variables were found, mediation models were to be tested using the Preacher and Hayes' PROCESS macro for SPSS (IBM Corp) (Preacher & Hayes, 2004). To test for mediation, the effects of interest and their corresponding weights would be examined. In testing for mediation, the indirect effect is of primary importance, rather than the direct effects (Zhao, Lynch, & Chen, 2010). Mediation effects in the absence of significant direct effects are referred to as indirect-only mediation (Zhao et al., 2010). In line with previous research on pediatric chronic pain (King et al., 2011), youth age and sex were to be controlled for in all mediation analyses. Confidence intervals that do not contain 0 are indicative of an indirect effect, suggesting with 95% confidence that the indirect effect is not 0 (Field, 2013). Thus, mediation was considered established if the confidence interval did not contain 0.

Chapter 4: Results

4.1 Ecological Momentary Assessment

4.1.1 Descriptive Statistics

Overall, EMA variables had a range of 14.8% – 17.3% missing data across all 7 days (youth pain intensity = 15.2%; youth pain unpleasantness = 17.3%; youth pain interference = 17.3%; parent protective responses = 16.5%). Missing data also included 7.9% of parent and 11.8% of youth PTSD symptoms scales.

Table 2 reports the means and standard deviations of the EMA variables. The zero-order correlations (i.e., correlations at the individual level) of the EMA variables are reported in Table 3. Youth pain intensity was related to youth pain unpleasantness ($r = .63$, $p < .001$) and youth pain interference ($r = .50$, $p < .001$). As well, youth pain unpleasantness was related to youth pain interference ($r = .61$, $p < .001$). Parent protective responses were related to youth pain unpleasantness ($r = .26$, $p = .04$) and youth pain interference ($r = .35$, $p = .01$). Youth pain interference was related to youth PTSD symptoms ($r = .40$, $p = .001$). Parent PTSD symptoms were also related to youth PTSD symptoms ($r = .32$, $p = .01$).

4.1.2 Hypothesis Testing

The standardized results of the 2-level path analyses are represented in Figure 3 and 4. When examining the daily associations between parent protective responses and youth pain outcomes, parent protectiveness was a positive predictor of youth pain unpleasantness ($\beta = .46$, $p < .001$) and youth pain interference ($\beta = .71$, $p < .001$), but not of youth pain intensity.

When examining the daily associations between youth pain outcomes and parent protective responses as predicted by parent and youth PTSD symptoms, there was a significant positive direct effect of parent PTSD symptoms on the daily association between youth pain unpleasantness and parent protective responses ($\beta = .03$, $p = .012$), as well as a significant positive direct effect of parent PTSD symptoms on youth pain unpleasantness ($\beta = .15$, $p = .013$). There was no significant direct effect of youth PTSD symptoms on the daily association between youth pain unpleasantness and parent protective responses ($\beta = -.01$, $p = .31$), and no significant direct effect of youth PTSD symptoms on youth pain unpleasantness ($\beta = .03$, $p = .27$). There were no significant direct effects of parent ($\beta = .01$, $p = .30$) or youth ($\beta = .01$, $p = .3$) PTSD symptoms on the daily association between youth pain intensity and parent protective

responses, and no significant direct effect of parent (β = -.04, *p* = .28) or youth (β = -.01, *p* = .42) PTSD symptoms on youth pain intensity. There were no significant direct effects of parent (β = -.04, *p* = .32) or youth (β = .02, *p* = .14) PTSD symptoms on the daily association between youth pain interference and parent protective responses, and no significant direct effect of parent (β = .15, *p* = .21) or youth (β = -.06, *p* = .29) PTSD symptoms on youth pain interference.

4.2 Laboratory Based Assessment

4.2.1 Descriptive Statistics

Table 4 reports the means and standard deviations of the baseline variables of interest. Parent PTSD scores averaged 9.9 (SD = 11.68) with 2.6% of parents reporting PTSD symptoms at or above the clinical cut off of 33, which indicates clinically significant elevations in PTSD symptoms. Youth PTSD scores averaged 17.5 (SD = 17.13) with 21% of youth reporting PTSD symptoms at or above the clinical cut off of 31 (Foa, Asnaani, Zang, Capaldi, & Yeh, 2018), indicating clinically significant elevations in PTSD symptoms. The types of traumatic events reported on by parents and youth were coded by two independent coders according to previously used categories and whether or not they met DSM-5 criterion A. These results are reported in Tables 5 and 6. Diagnostic clinical interviews indicated that 7.1% of youth currently met full DSM-5 criteria for a PTSD diagnosis and 4.7% of youth met partial criteria. 9.4% of youth met DSM-5 criteria for a PTSD diagnosis in the past but no longer met criteria for the disorder, and 2.4% of youth met partial criteria for a past PTSD diagnosis. Youth age and sex were not significantly correlated with youth PTSD symptoms (*r* = .18, *p* = .12; *r* = .19, *p* = .10, respectively). Youth sex was significantly correlated with youth pain interference (*r* = .33, *p* =.002) but not pain intensity (*r* = .13, *p* = .24) or unpleasantness (*r* = .20, *p* = .07). An independent samples *t* test revealed that females reported significantly greater pain interference

than males (p = .01). Youth age was not significantly correlated with pain intensity (r = .07, p = .55), interference (r = .16, p = .17) or unpleasantness (r = .08, p = .50). During the cold pressor task, 70% of parents reported that they did not want to stop the task at all.

4.2.2 Correlational Analyses

Results of correlational analyses are shown in Table 7. Higher parent PTSD symptoms were associated with higher youth PTSD symptoms (r = .24, p = .04) and increased youth pain interference (r = .24, p = .05). Higher youth PTSD symptoms were associated with increased youth pain intensity (r = .38, p = .001), pain unpleasantness (r = .53, p < .001), and pain interference (r = .44, p <.001). Higher parent PTSD symptoms were also associated with increased youth pain interference (r = .23, p =.04). However, parent and youth PTSD symptoms were not associated with parent reported protective responses, parent stop tendency, or parent pain attending talk. Additionally, neither youth pain intensity, interference or unpleasantness were associated with parent reported protective responses, parent stop tendency, or parent pain attending talk. Parent protective behaviours (i.e., parent reported protective responses, stop tendency, pain attending talk) were not associated with youth pain during the cold pressor task (i.e., pain tolerance, intensity or unpleasantness). Youth pain during the cold pressor task was not associated with youth baseline reports of their chronic pain. Parent stop tendency was associated with how anxious (r = .53, p < .001), worried (r = .30, p = .009), and upset (r = .73, p <.001) parents reported feeling immediately following the cold pressor task, as well as how much pain they thought their child felt during the task (r = .28, p = .01). On the basis of these results, we did not proceed with further mediation analyses.

Chapter 5: Discussion

This is the first study to examine the role of parent behaviours in the co-occurrence of youth PTSD symptoms and chronic pain as well as parent PTSD symptoms and youth chronic pain outcomes. A unique contribution of this study is the multi-method investigation of parent protective responses, parent and youth PTSD symptoms, and youth pain through: 1) parent and youth report questionnaires, 2) controlled lab-based tasks (i.e., cold pressor task, discussion task), and 3) ecological momentary assessments to naturalistically investigate parent behaviours and youth pain in the home environment over time (i.e., daily diaries).

In terms of the cross-sectional questionnaire and lab-based data, 2.6% of parents and 21% of youth reported clinically elevated PTSD symptoms. Higher youth PTSD symptoms were associated with worse youth pain outcomes (i.e., pain unpleasantness, interference and intensity). Additionally, higher parent PTSD symptoms were associated with worse youth pain interference. However, parent protective responses measured with the static questionnaire, parents' stop tendency following an experimental pain task, and pain attending talk during a discussion task following the experimental pain task, were not associated with youth or parent PTSD symptoms or youth pain outcomes. The degree to which parents wanted to stop the experimental pain task was, however, related to how distressed parents reported feeling (i.e., anxious, worried and upset) and how much pain they perceived their child experienced during the task.

Assessing daily responses of parent behaviours and youth pain enabled investigation of the associations between these factors as they naturally occurred in children's home environments and in reference to their actual chronic pain complaints, using a micro-longitudinal ecologically-valid approach. This study extends previous literature using EMA methodology (Connelly et al., 2017) by examining idiopathic pediatric chronic pain in a population without an underlying disease (e.g., juvenile arthritis). When examining the daily associations between

parent protective responses and youth pain outcomes, we found that parent protective responses influenced youth pain unpleasantness and pain interference, but not pain intensity. Additionally, higher parent PTSD symptoms predicted a stronger daily association between parent protective behaviours and youth pain unpleasantness, but not youth pain intensity or interference. Conversely, higher youth PTSD symptoms did not predict a stronger daily association between parent protective behaviours and youth pain outcomes (i.e., pain unpleasantness, interference, or intensity).

Similar to the findings of Connelly et al. (2017), most of the proportion of total variance of parent behaviours was due to within-person differences, demonstrating that parent protectiveness is dynamic and variable over time. This suggests that context plays a role in how parents respond to their child's pain on a given day. Alternative models were investigated in which youth pain influenced parent responses to their child, however, these models consistently fit the data worse than hypothesized models, suggesting that parent responses are likely more strongly driving youth pain outcomes, rather than youth pain complaints eliciting maladaptive parental responding. Likewise, higher youth PTSD symptoms did not predict the daily association between parent protective behaviours and youth pain outcomes. This suggests that among youth with idiopathic chronic pain, parent psychological factors play a more important role in influencing the relationship between parent responses and child pain than children's own psychological factors.

Parents with PTSD symptoms may be hypervigilant to, or have an attentional bias towards, their children's pain cues, leading them to engage in more protective responses in the face of their child's pain (e.g., restricting activities), in turn exacerbating children's pain symptoms. Parents who catastrophize about their child's pain have been shown to be more likely

to engage in protective responses (Caes, Vervoort, Trost, & Goubert, 2012). Moreover, among parents of youth with chronic pain, higher levels of PTSD symptoms are associated with higher levels of pain catastrophizing (Neville et al., 2018). Taken together, parents with PTSD symptoms may engage in catastrophic thinking about their child's pain (which may share similar negative cognitions to PTSD symptoms such as attentional bias to threat), which may prompt protective parent responding and reinforce children's pain behaviours. Alternatively, the cognitive demands of PTSD may interfere with parents' ability to engage in the behaviours that are an integral part of psychological interventions for and recovery from pediatric chronic pain, such as encouraging the child to engage in activities despite the pain, which promote adaptive coping. The finding that parents' mental health influences the daily association between parent behaviours and youth pain is clinically relevant. Current evidence-based cognitive-behavioural therapy (CBT) for pediatric chronic pain incorporates parent components focused on reducing parental protective responses to children's pain (Eccleston et al., 2014; Fisher et al., 2014), however, they fail to address parent mental health symptoms that may be preventing required behaviour change. Importantly, higher pre-treatment parent distress has been found to predict less improvement in child pain disability over 12 months following treatment (Law et al., 2017).

Given that pain interference is a critical target in interventions for youth with chronic pain (Eccleston et al., 2014), the finding that daily parent protective behaviours predicted youth pain unpleasantness and interference, but not pain intensity, is clinically relevant and consistent with previous literature (Neville et al., 2018). It is possible that parent protective responses have a greater influence on the affective aspect of pain, and functioning of their child when in pain, rather than the magnitude of their pain (i.e., intensity). Indeed, parent protective responses reinforce reduced functioning and avoidance behaviours (e.g., letting the child stay home from

school, keeping the child inside the house). Likewise, higher parent PTSD symptoms predicted a stronger daily association between parent protective behaviours and youth pain unpleasantness (but not pain interference or intensity), pointing again to the closer alignment of parents' emotional state (i.e., PTSD symptoms) and the affective qualities of pain, particularly for children who endure long-term, high intensity average pain (5.59 out of 10 in the current sample). Future research should also examine other youth pain outcomes that parent protective responses have been shown to be associated with, such as functional disability (Claar et al., 2008; Langer et al., 2009), school attendance, and school impairment (Logan et al., 2012), which were not examined in the current study.

Conversely, cross-sectional results of the controlled lab-based task did not reveal significant relationships between parent protective responses and youth pain outcomes, nor between parent or youth PTSD symptoms and parent protective responses. These null findings call into question the ecological validity of laboratory pain tasks among youth with chronic pain. Literature involving the cold-pressor task has been largely confined to healthy children (Birnie et al., 2012). For example, among healthy children, parents who report higher levels of distress have been shown to exhibit a greater tendency to want to stop their child's pain during the cold pressor task (Caes et al., 2011). Caes et al. (2011) found similar results among children with chronic pain, however, a different, and arguably more ecologically valid, pain inducing task was used (i.e., a 2-minute walk task) as well as a different measure of stop tendency (i.e., parents were instructed to stop a videotape of their child completing the walk task when they would have wanted to tell their child to stop the task). Whereas the cold pressor task is an acute pain stimulus to a particular body part, this 2-minute task used by Caes et al. (2011) induced pain through movement, which is often avoided in response to the actual experience of chronic pain. This task

may have been more generalizable to children's chronic pain, particularly given that their sample of children predominantly suffered from hypermobility, chronic back pain, and reflex sympathetic dystrophy. Given that the current sample predominantly consisted of children with chronic headaches, the cold pressor task may not have been generalizable to children's chronic pain in this sample. Likewise, parents may not have perceived this acute pain task as threatening to their child. Indeed, parents reported a low average stop tendency (i.e., 0.71) and 70% of parents reported not wanting to stop the cold pressor task at all. Similar to Caes et al. (2011), in the current sample, parents who reported higher distress (i.e., feeling anxious, worried or upset) reported a higher stop tendency, however, stop tendency was not associated with youth baseline pain report or pain report during the task. It stands to reason that the cold pressor task may be less impactful among parents who witness their child in pain on daily basis. Unlike chronic pain, the cold pressor task is an *acutely* painful experience (specifically localized to the non-dominant hand) over which children have complete control (i.e., they can remove their hand at any time). Youth were also told what to expect of the cold water task (i.e., that they would submerse their hand in cold water that was 10 degrees Celsius). Thus, the task did not include the key elements of uncontrollability, unpredictability, or threat to the social self, which have been identified as psychological triggers of a stress response (i.e., activation of the HPA axis) (Dickerson & Kemeny, 2004), and is likely different than the threat of pain these youth experience on a daily basis. Taken together, the cold pressor task may simply not be an ecologically valid tool to use to understand processes related to chronic pain among youth with idiopathic chronic pain.

The concern of ecological validity extends to the observed parent-youth interaction. A similar discussion task and coding scheme has been previously used in conjunction with the cold pressor task among healthy children (Vervoort et al., 2011), however, this laboratory assessment

may not be an appropriate measure of parent protectiveness among children with chronic pain. Having shown the variability of parent responding over time with the EMA analyses in the current sample, a single, lab-based experiment may not elicit an accurate picture of parent behaviours towards their child's chronic pain. Youth pain reports during the cold pressor task also did not correlate with youth reports of their chronic pain at baseline. This, again, points to the benefits of employing micro-longitudinal EMA methods (i.e., repeated sampling of participants' behaviour and experience in their natural environment and in real-time) (Shiffman, Stone, & Hufford, 2008) as a novel and potentially more ecologically valid methodology in the study of pediatric chronic pain. Indeed, single measures of recalled pain over the past week, although standard in clinical and research practice, do not capture the complex and dynamic nature of pain, which is also influenced by environmental factors (Stinson, 2009). The use of real-time data capture reduces the possible effects of recall bias in pain reporting (Stinson, 2009).

Similar to previous research (Neville et al., 2018), 21% of youth in the current sample reported clinically elevated PTSD symptoms. However, fewer parents in this sample reported clinically elevated PTSD symptoms (2.6%) compared to previous research (9% and 20%) (Neville et al., 2018; Noel, Wilson, et al., 2016). Given the noteworthy trauma histories of parents in this sample (58% of parents reported traumatic events meeting DSM-5 diagnostic criterion A), the investigation of adverse childhood experiences (ACEs) may be warranted among parents of children with chronic pain. Consistent with previous research (Noel, Wilson, et al., 2016), higher youth PTSD symptoms at baseline were associated with increased youth pain intensity, pain unpleasantness, and pain interference Given that children with clinical anxiety have been shown to be less likely to respond to treatment for chronic pain (Cunningham et al., 2016), youth PTSD might also be a critical factor for predicting children most at risk for poor

pain outcomes and response to treatment. An additional contribution of this study to the literature is the assessment of youth PTSD at the diagnostic level using gold-standard diagnostic clinical interviews. These clinical interviews demonstrated that 7.1% of the current sample met criteria for a current DSM-5 diagnosis of PTSD and an additional 4.7% met partial DSM-5 criteria. Although lower than baseline questionnaire reports of clinical elevations in PTSD symptoms, this is considerably higher than the prevalence of PTSD diagnoses found in a general community sample of children, where less than 0.5% of children were found to meet criteria for a full DSM-IV PTSD diagnosis (Copeland et al., 2007).

5.1 Strengths and Limitations

A multi-method approach is a significant strength of the current study. The investigation of parent behaviours, parent and youth PTSD symptoms, and youth pain, through parent and youth report on questionnaires, controlled lab-based tasks, as well as EMA, allowed for the examination of these relationships both in a controlled, experimental environment and in children's natural day-to-day lives. Limitations of the present study should be noted. Firstly, data was collected from only one parent and the majority of parents who participated were mothers. Given the within person variability in parent protective behaviours found in this study, it is likely that in dual parent homes, both parents do not respond consistently to their child and one parents' response might buffer, or alternatively exacerbate, that of the other. Secondly, shared method variance is a concern with the same participants completing multiple measures and tasks within this study (i.e., baseline measures, EMA and lab-based tasks). Finally, the interrater reliability for coding of the parent pain attending talk in the current study was lower than previous reports (Vervoort et al., 2011).

5.2 Conclusions and Future Directions

This study contributes to our understanding of the co-occurrence of parent and youth PTSD symptoms and youth chronic pain, as well as the role of parent behaviours in these relationships. Parent responses to youth pain may be an interpersonal mechanism through which parent PTSD impacts children's chronic pain symptoms. Children of parents with PTSD symptoms may be most impacted by parent protective responses to their chronic pain complaints. This study has direct implications on clinical practice, highlighting the importance of addressing daily parent-youth interactions, as well as parent and youth mental health, in the assessment and treatment of pediatric chronic pain. In the current study, youth and parent PTSD symptoms were associated with worse youth pain outcomes. Currently, treatments for chronic pain do not directly address children's and parents' mental health symptoms, despite the fact that they impede responsiveness to treatment. Palermo et al. (2016) have recently demonstrated the potential of a problem-solving skills training intervention in reducing distress among parents of youth with chronic pain. Innovative approaches like this, tailoring treatment to the unique needs of parents and youth with comorbid mental health issues, may improve response to treatment and health trajectories in this vulnerable population. Finally, this study points to EMA as a novel methodology for the future study of pediatric chronic pain as it is arguably a more ecologically valid approach, capturing the dynamic nature of children's chronic pain within the natural context of the child's daily life. Future research should also consider laboratory methods using experimental pain tasks that more closely align with youths' chronic pain symptoms, and testing youth in the laboratory when they are currently experiencing their chronic pain symptoms (e.g., during a headache). Taken together, this research advances our understanding of the interpersonal context of co-occurring parent and youth PTSD symptoms and pediatric chronic

pain, as well as illuminates directions to enhance research and practice for youth with chronic pain.

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Appendix A: Tables and Figures

Table 1

Socio-Demographic Characteristics of the Sample

Socio-Demographics	N=84
Youth mean age (SD), years	14.2 (2.3)
Youth sex (% female)	70.2
Parent sex (% female)	94.0
Relationship to the youth (%)	
Biological parent	98.8
Relative	1.2
Youth ethnicity (%)	
White (Caucasian)	86.9
Two or more ethnicities	2.4
Arab/West Asian	2.4
Other	7.1
Do not want to answer	1.2
Household income (%)	
<$10,000 - $29,999	6.3
$30,000 - $59,999	12.5
$60,000 - $89,999	12.5
More than $90,000	62.5
Do not want to answer	6.3

Table 2

Descriptive Statistics for Youth and Parent Daily Variables

Variable	N	Youth report *M (SD)*	Parent report *M (SD)*
Parent PTSD symptoms (PCL-5), total	70		9.50 (10.93)
Youth PTSD symptoms, (CPSS-5), total	67	16.10 (16.18)	
Parent protective responses (ARCS protect scale), mean	76		0.43 (0.57)
Youth pain intensity, total	76	4.63 (2.40)	
Youth pain unpleasantness, total	76	1.69 (0.80)	
Youth pain interference (PROMIS), T-score	76	50.74 (9.53)	

Note. PCL-5 = PTSD Checklist for DSM-5; CPSS-5 = Child Posttraumatic Stress Disorder Scale; ARCS = Adult Responses to Children's Symptoms; PROMIS = Patient-Reported Outcomes Measurement Information System.

Table 3

Zero-order Correlations Among Variables of Interest for Daily Data

Variable	1	2	3	4	5	6
1. Parent PTSD symptoms	-	.32*	.18	.14	.17	.19
2. Youth PTSD symptoms		-	.20	.19	.17	.40**
3. Parent protective responses			-	.16	.26*	.35**
4. Youth pain intensity				-	.63***	.50***
5. Youth pain unpleasantness					-	.61***
6. Youth pain interference						-

Note. *p<.05; **p<.01, ***p<.001

Table 4

Descriptive Statistics for Youth and Parent Baseline Variables

Variable	N	Youth report *M (SD)*	Parent report *M (SD)*
Parent PTSD symptoms (PCL-5), total	79		9.9 (11.68)
Youth PTSD symptoms (CPSS-5), total	78	17.5 (17.12)	
Parent protective responses (ARCS protect scale), mean	83		1.26 (.69)
Parent stop tendency	83		0.71 (1.63)
Parent pain attending talk	76		.44 (.17)
Youth pain intensity, total	81	5.59 (1.91)	
Youth pain unpleasantness, total	81	2.04 (0.93)	
Youth pain interference (PROMIS), T-score	81	56.62 (9.45)	
Youth CPT pain tolerance, seconds	82	156.35 (93.50)	
Youth CPT pain intensity	81	4.05 (2.22)	
Youth CPT pain unpleasantness	83	2.06 (1.15)	

Note. PCL-5 = PTSD Checklist for DSM-5; CPSS-5 = Child Posttraumatic Stress Disorder Scale; ARCS = Adult Responses to Children's Symptoms; PROMIS = Patient-Reported Outcomes Measurement Information System. CPT = cold pressor task.

Table 5

Types of Traumatic Events Endorsed by Parents

Type of Event	Parent endorsement of each type of event (%)
Death	26.19
Accident	17.86
Physical illness or hospitalization	11.9
N/A	11.9
Sexual abuse	8.33
Physical abuse	7.14
Divorce	5.95
Verbal conflict/abuse	4.76
Suicide attempt	3.57
Mental illness	3.57
Natural disaster	3.57
Family related conflict	1.19
Chronic pain problem	1.19
Substance abuse	1.19
Other	1.19

Note. 58% of parents reported traumatic events that met Criterion A of DSM-5 diagnostic criteria for posttraumatic stress disorder

Table 6

Types of Traumatic Events Endorsed by Youth

Type of Event	Youth endorsement of each type of event (%)
N/A	25.00
Death	21.43
Physical illness or hospitalization	9.52
Other	8.33
Physical abuse	5.95
Fear/Anxiety	4.76
Accident	3.57
Divorce	3.57
Family related conflict	3.57
Mental illness	3.57
Substance abuse	3.57
Verbal conflict/abuse	3.57
Chronic pain problem	2.38
Pet or animal	2.38
Social difficulties	2.38
Fire	1.19
Gunpoint or fire arm	1.19
Natural disaster	1.19

Note. 29% of youth reported traumatic events that met Criterion A of DSM-5 diagnostic criteria for posttraumatic stress disorder

Table 7

Correlations Among Variables of Interest for Baseline Data

Variable	1	2	3	4	5	6	7	8	9	10	11
1. Parent PTSD symptoms	-	.24*	.14	.09	-.07	.03	.02	.23*	-.02	.09	.21
2. Youth PTSD symptoms		-	.01	-.08	-.13	.38**	.53***	.44***	.19	-.002	.03
3. Parent reported protective responses			-	.18	.07	-.16	-.04	.03	-.05	.06	-.05
4. Parent stop tendency				-	.04	.13	.05	.13	.13	.02	.04
5. Parent pain attending talk					-	-.05	.12	-.01	-.20	.04	.14
6. Youth pain intensity						-	.58**	.54**	.08	.04	.01
7. Youth pain unpleasantness							-	.66**	.08	-.11	.03
8. Youth pain interference								-	-.004	.05	.03
9. Youth CPT pain tolerance									-	-.40**	-.41**
10. Youth CPT pain intensity										-	.68**
11. Youth CPT pain unpleasantness											-

CPT = cold pressor task.
Note. *p<.05; **p<.01, ***p<.001

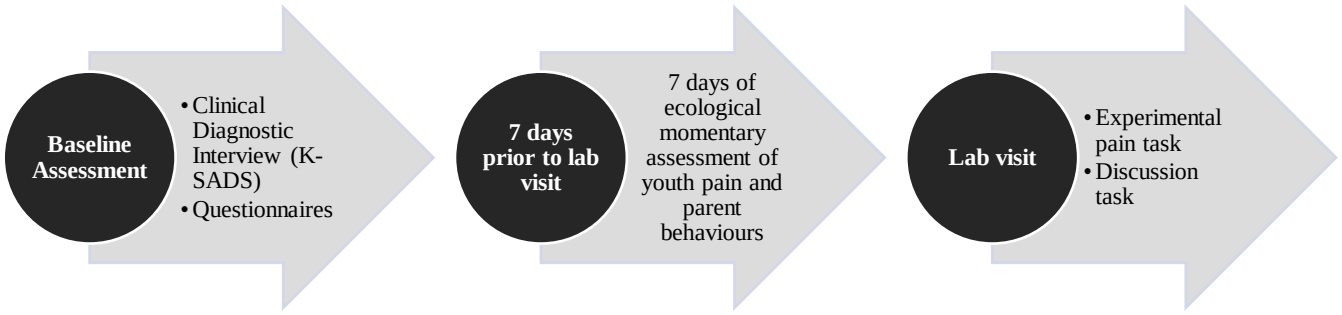

Figure 1. Timeline of the current study.

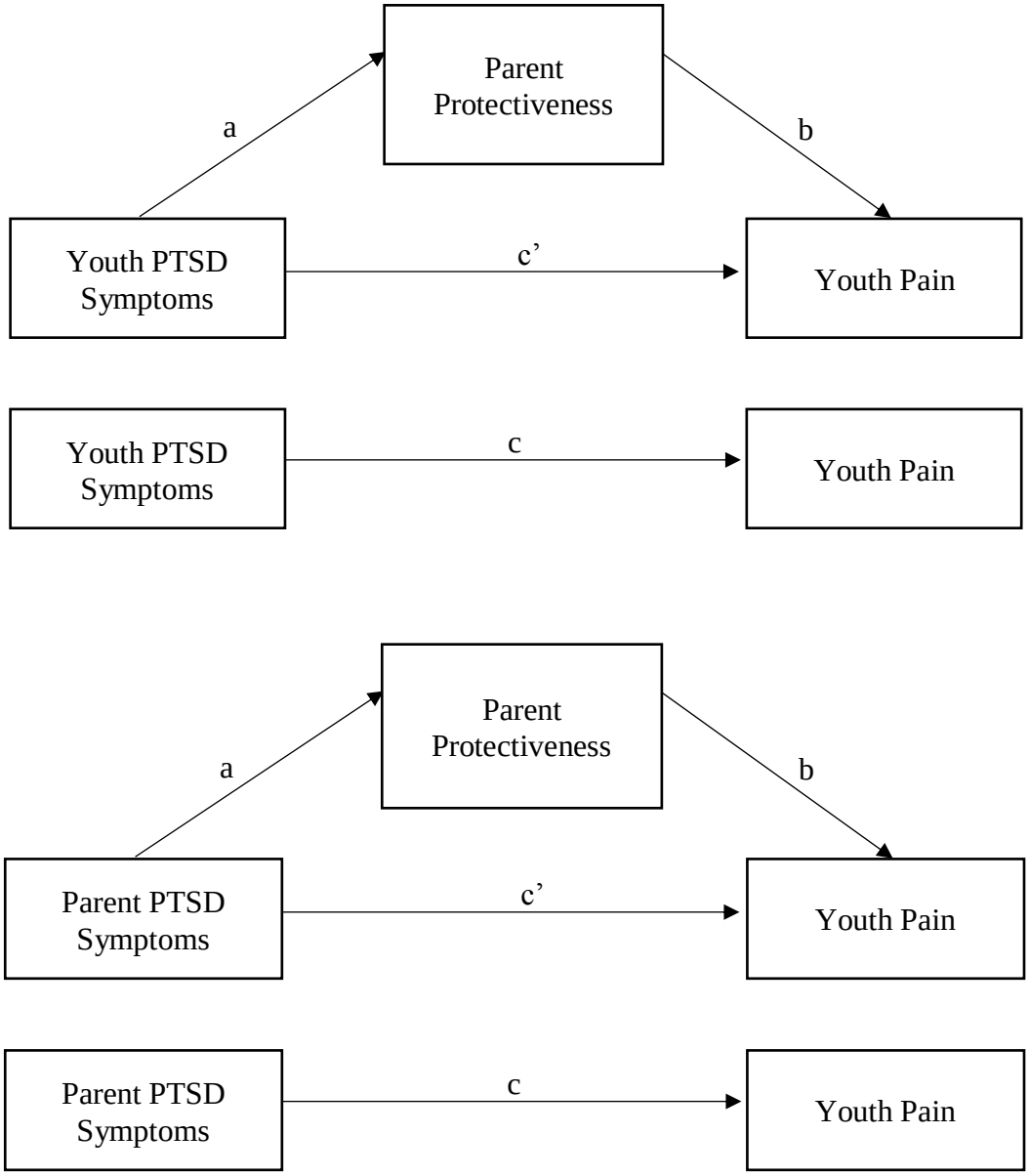

Figure 2. Graphic depictions of the hypothesized mediation models.

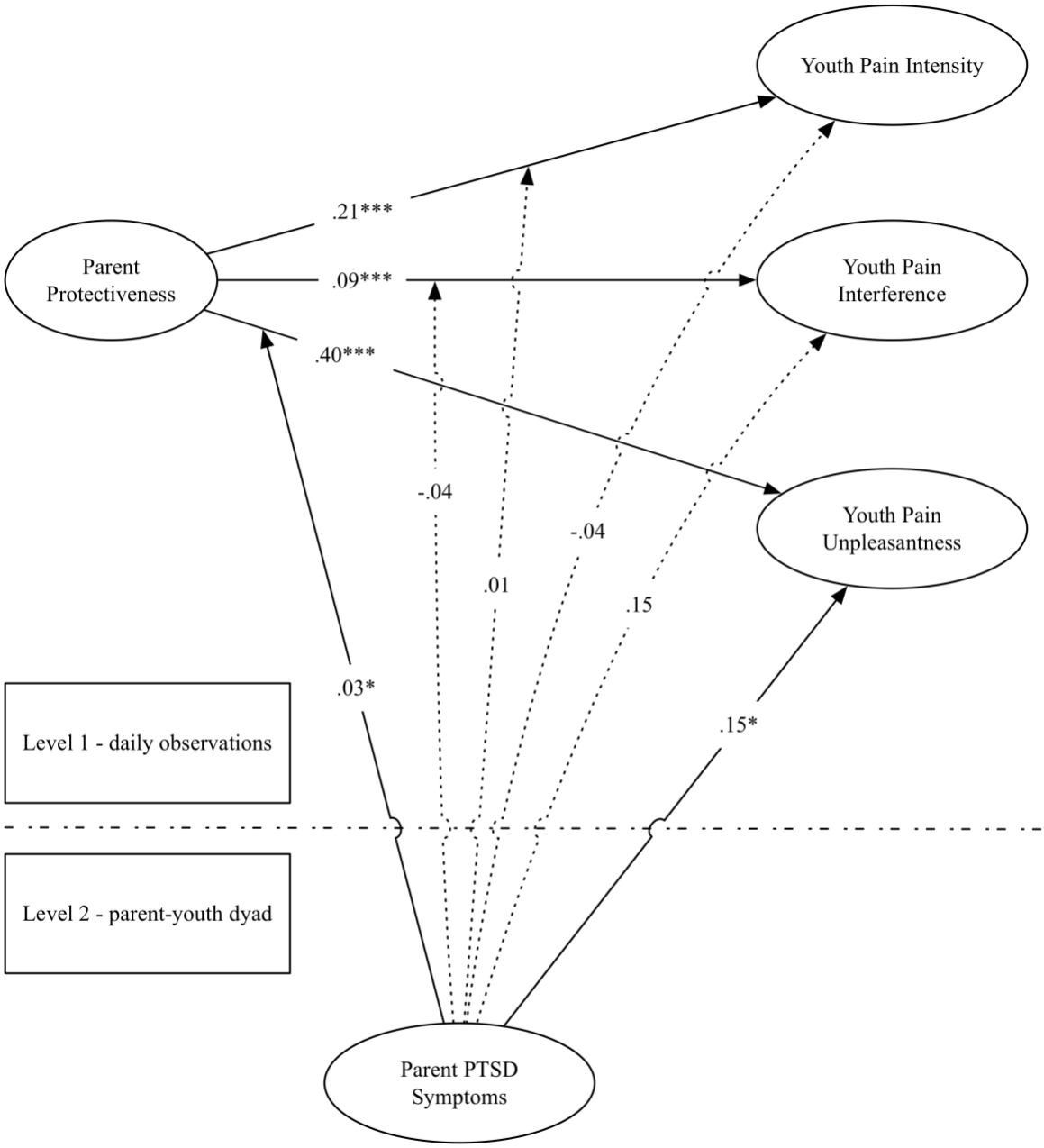

Figure 3. Standardized results of the 2-level path analysis used to test the daily association between youth pain outcomes and parent protectiveness as predicted by parent PTSD symptoms.

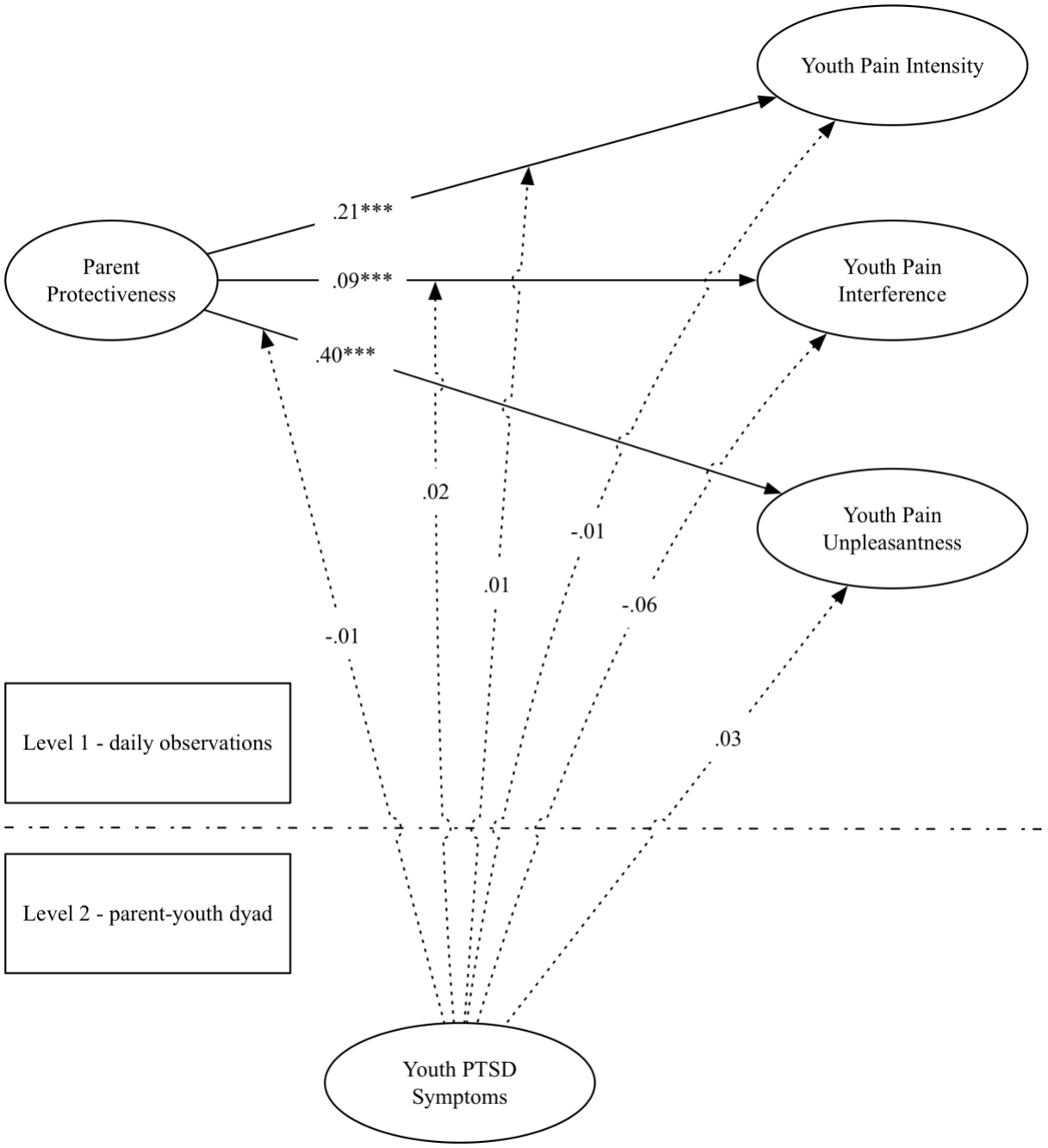

Figure 4. Standardized results of the 2-level path analysis used to test the daily association between youth pain outcomes and parent protectiveness as predicted by youth PTSD symptoms.