

THE ROLE OF SIBLINGS IN PEDIATRIC PAIN

by

Meghan Grace Schinkel

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This dissertation is dedicated to my own sibling, Sarah, who has played such an important role in my life.

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ABSTRACT

Sibling relationships are common and have an important influence on children's development. While family factors in pediatric pain have received considerable empirical attention, this work has largely focused on parents, not siblings. The current dissertation aimed to: summarize and map the type of research that has been conducted examining siblings and pediatric pain (Paper 1); examine associations between siblings' relationship quality and their behaviours during an experimental pain task, and relations between a sibling's behaviours and a child's pain outcomes (Paper 2); and determine the extent to which parents bring siblings to pediatric appointments, and explore parents' decisions surrounding this (Paper 3). Paper 1 describes the results of a scoping review on siblings and pediatric pain, which included 35 studies. Most of the identified research on siblings and pediatric pain used quantitative methods, examined siblings in the context of chronic and disease-related pain, and focused on the genetic influence of pain conditions. Paper 2 presents a lab-based study that included a sample of 92 healthy sibling dyads between 8 and 12 years of age. Siblings completed observational and questionnaire measures of sibling relationship quality, and took turns completing the cold pressor task (CPT) with their sibling present. Siblings' behaviour during the CPT was coded, and pain outcomes were recorded. Greater levels of warmth/positivity in the sibling relationship were related to children engaging in more non-attending (e.g., distraction) and less attending (e.g., symptom talk) behaviours while completing the CPT. Greater levels of attending behaviours by the observing child were related to the sibling who was completing the CPT having a lower pain tolerance. Paper 3 summarizes a questionnaire-based study that included responses from a sample of 95 parents. Approximately 98% of parents reported having brought siblings to medical appointments. Coding of open-ended responses revealed that parents most often cited convenience as their reason for bringing siblings (83.7%), and most frequently reported allowing their children to decide the order in which siblings received an appointment/procedure (53.3%). These findings suggest that siblings, through their relationships and actions, influence how children respond to pain and need to be considered in clinical pain contexts.

LIST OF ABBREVIATIONS AND SYMBOLS USED

ANOVA	Analyses of Variance
APIM	Actor-Partner Interdependence Model
b^*	Standardized regression coefficient
CFS	Children's Fear Scale
CPRS	Child-Parent Relationship Scale
CPT	Cold Pressor Task
F	F statistic for ANOVA
FPS-R	Faces Pain Scale Revised
M	Mean
n	Sub-sample size
p	P value for the testing of significance
r	Pearson correlation statistic
SD	Standard deviation
SRQ-R	Sibling Relationship Questionnaire Revised
t	Value for t-test
χ^2	Chi-square statistic

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CHAPTER 1: INTRODUCTION

1.1 An Introduction to Pediatric Pain

Pain is a predominant issue amongst both healthy infants, children and adolescents, and those requiring clinical attention (Stevens & Zempsky, 2014). Unfortunately, children's pain is often poorly managed due to limited awareness and/or implementation of strategies for identifying and treating pediatric pain (Finley, Franck, Grunau, & von Baeyer, 2005). Pain has been defined as "a distressing experience associated with actual or potential tissue damage with sensory, emotional, cognitive and social components" (Williams & Craig, 2016, p. 2420). This definition acknowledges that pain is not only a physical event, but an individual experience influenced by multiple aspects such as one's thought processes and interpersonal environment (Williams & Craig, 2016). Children's pain can be acute or chronic. Acute pain generally lasts a short duration and terminates when medically expected (e.g., with treatment or healing) (Stevens & Zempsky, 2014). In contrast, chronic pain generally lasts or recurs over longer periods (e.g., beyond several months) and may continue past what would be medically expected (e.g., after healing) (Stevens & Zempsky, 2014). There are two types of chronic pain; persistent pain tends to be continuous, whereas recurrent pain involves repeated experiences of acute pain (e.g., recurring headaches) (Stevens & Zempsky, 2014). Experimental pain tasks, such as the cold pressor task (described in detail in section 1.7), have been used in pediatric research to systematically investigate children's pain experiences within a controlled context (K A Birnie, Caes, Wilson, Williams, & Chambers, 2014). Pain resulting from participation in these tasks is generally referred to as experimental pain.

Young children frequently experience acute pain resulting from routine injuries, illnesses, and medical experiences. For example, in a Canadian sample of children aged 9 to 13, over 90% reported having ever experienced pain from headaches, sore muscles, slivers and injections, and over 80% reported having ever experienced pain from earaches, toothaches or burns (van Dijk, McGrath, Pickett, & VanDenKerkhof, 2006). In this same sample, both boys and girls most often described pain resulting from minor bumps and cuts as being the worst pain they had felt (van Dijk et al., 2006). Procedure-related acute pain is also a common occurrence for children requiring more serious medical attention. For example, Canadian studies have found that, within a 24-hour period, approximately 78% of hospitalized children experience a minimum of one painful procedure (e.g., venipuncture) (Stevens et al., 2011), and that children's worst pain while in hospital is most often reported as being due to a medical procedure (Kathryn A Birnie, Hons, et al., 2014). Many adolescents also experience recurrent pain, with longitudinal data of Canadian adolescents identifying rates as high as 31.8% for headaches, 25.8% for backaches and 22.2% for stomachaches occurring at least once per week (Stanford, Chambers, Biesanz, & Chen, 2008). Further, a systematic review of pediatric chronic pain reported a median point prevalence rate of 18% for children and adolescents with general or nonspecific recurrent pain (King et al., 2011). Therefore, acute and chronic pain are clearly predominant issues amongst children and adolescents.

Appropriate assessment and management of children's pain is important, as studies have shown that the nature of infant's and children's pain experiences (e.g., during procedures) can influence their perceptions of, and reactions to, pain later on (Kennedy, Luhmann, & Zempsky, 2008; Taddio & Katz, 2005). As such, a growing and

diverse field has emerged focused on pediatric pain (Caes et al., 2016). Although research in the field has primarily examined the assessment, treatment, and characterization of children's pain, exploration of the role of family and parent factors has been a consistent focus (Caes et al., 2016). Families are a central aspect of children's pain experiences (Birnie, Boerner, & Chambers, 2014). Families, particularly parents, have been extensively studied in the context of children's acute and chronic pain, with research examining the role of parents in child pain assessment and intervention, and children's pain-related outcomes (e.g., distress, disability) (Birnie et al., 2014). However, research on families in pediatric pain has frequently failed to include siblings. The current dissertation builds on our knowledge of the importance of families in pediatric pain by considering the role of siblings.

1.2 Theoretical Basis

Although there are no known theories explicitly focused on siblings and children's pain, family systems theory (Bavelas & Segal, 1982) and family models in pediatric pain (Palermo & Chambers, 2005; Palermo, Valrie, & Karlson, 2014) together provide a strong conceptual framework for the current dissertation. Family systems theory speaks to the value of understanding individuals within the context of the broader family, and offers a framework from which one can approach doing so (Bavelas & Segal, 1982). According to this theory, the family is composed of interrelated individuals who bi-directionally impact one another (Bavelas & Segal, 1982). Communication, via behaviour, is at the centre of the family system, and both reflects and perpetuates the relationships between members (Bavelas & Segal, 1982). The theory acknowledges that,

based on the goal of the research, concentrating on subsystems within the family can be beneficial (Bavelas & Segal, 1982).

Narrowing in on pain more specifically, frameworks have been developed to describe the interaction between child and family variables in pediatric pain (Palermo & Chambers, 2005; Palermo et al., 2014). Although focused on parents, these frameworks view children's experience of pain as being bi-directionally related to individual factors pertaining to the child and other family members (i.e., the parent as described in these models), as well as wider dyadic and familial factors (Palermo & Chambers, 2005; Palermo et al., 2014). Further, these frameworks point to the importance of examining the association between a family member's behaviour, and a child's pain experience, within the milieu of broader familial influences (Palermo & Chambers, 2005; Palermo et al., 2014).

These theoretical frameworks highlight the importance of families in understanding children's behaviour and pain experiences. Siblings are a valuable familial subsystem to examine to expand our knowledge of the role of families in children's responses to pain. Further, as suggested by the families and pain frameworks (Palermo & Chambers, 2005; Palermo et al., 2014), to comprehensively understand potential associations between sibling behaviours and children's pain experiences, one should also consider how these behaviours may be influenced by broader dyadic factors, such as relationship quality. From a methodological perspective, given the proposed importance of behaviour in the family system (Bavelas & Segal, 1982), examination of siblings' behaviours directly may be a valuable approach to studying siblings' influence in pediatric pain. Lastly, a common theme across these theories is the suggested bi-

directional nature of relations between variables of interest (Bavelas & Segal, 1982; Palermo & Chambers, 2005; Palermo et al., 2014). Therefore, a dyadic approach to analyzing data, which permits examination of relations between both siblings, may offer unique insights.

1.3 Families and Pediatric Pain

As noted above, a considerable body of evidence has evolved pointing to the importance of families in pediatric pain. Of most relevance to the current dissertation is the research examining the influence of family relationships/functioning on children's pain experiences, as well as research on the association between parent behaviours and children's pain outcomes.

The quality of relationships amongst family members and general family functioning have been found to be influential in children's responses to both chronic and acute pain. One study that examined adolescents with chronic pain found that parents who reported more distress in their relationship with their child had children who reported more psychological distress (Logan, Guite, Sherry, & Rose, 2006). Further, when parents reported lower levels of distress in their relationship with their child, suggesting a more positive relationship quality, their child's self-report of pain intensity and disability were more strongly related (Logan et al., 2006). A systematic review of studies exploring family functioning in the context of pediatric chronic pain found that greater levels of dysfunction in the family (e.g., conflict) was generally related to children experiencing more disability due to their chronic pain (Lewandowski, Palermo, Stinson, Handley, & Chambers, 2010).

A recent study explored family functioning in children's acute, experimental pain. Self-reported poorer family functioning by children ages 8 to 12-years was related to children reporting higher levels of catastrophizing (e.g., ruminating or magnifying) about their pain (Birnie, Chambers, Chorney, Fernandez, & McGrath, 2017). Further, although not all aspects of family functioning related to children's behaviours in a consistent manner, greater observed family cohesion during a conflict discussion task was related to children doing more "other talk" (e.g., non-pain talk), while less family cohesion was related to children engaging in more symptom complaints while completing the cold pressor task with an observing parent (Birnie et al., 2017). Taken together, poorer relationship quality and functioning within the family seems to have a negative influence on children's responses to pain, such as greater levels of distress or more pain-focused thoughts and behaviours.

A considerable amount of research has explored relations between parent behaviours (i.e., what parents say and do) and children's responses to acute pain. In general, parent behaviours that focus attention on a child's pain experience (referred to in the current dissertation as "attending behaviours"), such as reassurance, are thought to promote poorer outcomes for children, whereas behaviours that remove attention from a pain experience (referred to in the current dissertation as "non-attending behaviours"), such as humour or non-procedure talk, are thought to promote better outcomes (Birnie et al., 2014). Studies using experimental pain tasks, including the cold pressor and water load tasks, have demonstrated that parents' use of attending behaviours (e.g., reassurance, symptom talk, sympathy) are related to children engaging in more symptom complaints, self-reporting more pain, and having lower pain tolerance scores (Moon, Chambers, &

McGrath, 2011; Schinkel, Chambers, Caes, & Moon, 2017; Walker et al., 2006).

Conversely, parental use of non-attending behaviours (e.g., distraction, humour) during experimental pain has been related to children engaging in fewer symptom complaints, reporting lower pain-related unpleasantness and having higher pain tolerance scores (Moon et al., 2011; Walker et al., 2006).

Studies have similarly explored relations between parent behaviours and children's pain experiences in clinical acute pain contexts. Parental use of attending behaviours, such as reassurance or empathy, during child immunizations has been related to children expressing more distress and fear (Cohen, Manimala, & Blount, 2000; Manimala, Blount, & Cohen, 2000). Parental distraction during acute procedural pain has been related to children reporting less fear, and expressing less fear, pain, and distress during the procedure (Manimala et al., 2000; McCarthy et al., 2010). Distraction has been recommended as an approach to use with children to decrease the acute pain and distress associated with needles, based on a systematic review and meta-analysis (Kathryn A. Birnie, Noel, et al., 2014). Further, children have reported parental distraction as having a more beneficial effect than attending behaviours during pain, suggesting children recognize it as a helpful strategy, although parents did not similarly endorse this (Walker et al., 2006).

The research summarized above clearly demonstrates the importance of family relationships and parent behaviours in pediatric pain. It is unknown whether the same findings demonstrated in the parent-child pain literature also apply to siblings. Therefore, examining these factors amongst siblings may have theoretical, empirical and clinical relevance.

1.4 The Importance of Siblings

Research on children underscores the importance of sibling relationships. Approximately 80% of children have siblings, and for many this represents an ongoing relationship throughout their lives (Dunn, 2000). The sibling relationship has been described as a particularly salient environment for young children to learn and grow, as its obligatory nature reduces the social risk associated with attempting new skills (Howe & Recchia, 2014). Further, similarities in daily activities and behavioural functioning have been documented in some sibling dyads, lending support to the occurrence of observational learning in some sibling relationships (Whiteman, McHale, & Crouter, 2007).

There is compelling evidence for both the positive and negative impact siblings have on children's development. For example, reviews of the literature on sibling relationships have illustrated how siblings can foster cognitive and social skills, such as language abilities, empathy, emotional competence, perspective taking and conflict resolution (Brody, 2004; McHale, Updegraff, & Whiteman, 2012). Alternatively, conflictual sibling relationships and negative modeling have been related to poorer functional outcomes, such as engaging in risky activities and negative social behaviours (Brody, 2004; McHale et al., 2012). These reviews have also described how siblings can have indirect effects on one another through their impact on the familial and social environments to which their siblings are exposed (Brody, 2004; McHale et al., 2012). It has been suggested that sibling relationships may be particularly impactful during the childhood years, based on findings that both warmth/closeness and conflict tend to decline amongst siblings in adolescence indicating a more neutral relationship

(Buhrmester & Furman, 1990). It is clear from this research that siblings are influential across many aspects of children's lives. Thus, it is reasonable to consider that sibling relationships, perhaps particularly during childhood, may also be influential in pediatric pain experiences.

1.5 Siblings and Chronic Illness

Further support for examining siblings in pediatric pain comes from research illustrating the impact children's chronic health issues more broadly can have on siblings. A recent meta-analysis demonstrated that siblings of children with chronic health issues (e.g., cancer, diabetes) are vulnerable to psychological difficulties (Vermaes, van Susante, & van Bakel, 2012). These siblings experience greater internalizing and externalizing issues than siblings of healthy children, with siblings of children with more severe chronic illnesses (i.e., requiring more intensive treatment, higher mortality) having poorer functioning on these outcomes (Vermaes et al., 2012). Health-related quality of life is impacted in siblings of children with chronic illnesses; a systematic review demonstrated evidence for both poorer and improved quality of life relative to siblings of healthy children (Limbers & Skipper, 2014), suggesting impacts are not always uniformly negative. Concurrent with the findings regarding psychological functioning, quality of life was more adversely affected amongst siblings of children with more severe illnesses (Limbers & Skipper, 2014). Siblings of children with chronic illness may benefit from treatment targeting psycho-social issues. For example, siblings of pediatric cancer patients endorsed lower levels of anxiety and depression following participation in an eight-week, cognitive-behavioural based group program for siblings of children with cancer (Barrera, Chung, & Fleming, 2005).

The impact of having a sibling with a chronic health issue extends to healthy sibling's academic and everyday experiences. One systematic review explored the influence of pediatric chronic illness (primarily cancer) on healthy siblings' school functioning, and found evidence for psychological issues in the school context, including post-traumatic stress symptoms and attentional difficulties (Gan, Lum, Wakefield, Nandakumar, & Fardell, 2017). Healthy siblings were also found to experience academic challenges, a decline in school attendance/participation, and poorer social functioning, although some support for positive social outcomes was also described (Gan et al., 2017). A recent qualitative study illustrated the challenges and rewards siblings of children with chronic health issues experience in their daily life (Woodgate, Edwards, Ripat, Rempel, & Johnson, 2016). Healthy youth described a desire to take part in activities with their sibling with complex health issues, as well as the sense of importance and responsibility their relationship with their sibling imbued (Woodgate et al., 2016). However, siblings also reported experiencing resentment due to the additional forethought required to engage in activities (e.g., travelling), or inability to do so, because of their sibling's needs (Woodgate et al., 2016). Similar themes were noted in a qualitative study examining the experience of siblings of children with cancer within the context of a sibling group intervention (Neville et al., 2016). For example, siblings reported experiencing conflictual feelings (e.g., joy and worry), no longer engaging in enjoyed activities (e.g., due to changing family priorities), and attempting to care for and amuse their sibling with cancer (Neville et al., 2016). Thus, it is evident that children's health status and health-related experiences have a wide-reaching influence on siblings, both positively and negatively.

1.6 Siblings and Pain

Although much less developed than the body of work on parents, research in the field of pediatric pain has begun to specifically consider siblings. A scoping review of the literature on siblings and pediatric pain is provided in Chapter Two; however, a brief overview is provided here.

Anecdotal evidence suggests that siblings may be present when children are in pain and, given the high frequency with which children experience various forms of pain (see section 1.1), sibling presence during child pain is potentially a common occurrence. One study found that, in a sample of 6 to 8-year-old children, 27% identified a sibling as someone they knew who often experiences pain, and this percentage was greater than that of children who identified a parent (Boerner, Chambers, McGrath, LoLordo, & Uher, 2017). This suggests that young children are aware of their siblings' pain experiences (Boerner et al., 2017). Children may also have a desire to help their sibling when they experience pain. A qualitative study found that children reported assisting their parent with managing their siblings' post-operative pain, such as by giving their sibling medication or a pacifier (Kankkunen, Vehvilainen-Julkunen, & Pietila, 2002). One case study described including an older sibling, along with their mother, in an intervention targeting cancer-related procedural pain and anxiety (Barrera, 2000). This case study reported that the older sibling effectively implemented behavioural/cognitive-behavioural strategies (e.g., distraction) during their younger sibling's cancer-related procedure, resulting in positive outcomes for both siblings (Barrera, 2000). This finding offers some preliminary support for the positive role siblings may play during children's acute pain

experiences; however, research specifically examining sibling behaviours during pediatric acute pain is needed.

When children experience chronic pain, this can have a broad influence on their sibling's well-being. Relative to siblings of healthy children, siblings of children with chronic pain conditions experience poorer psychological and social functioning (Engstrom, 1992; Guite, Lobato, Shalon, Plante, & Kao, 2007). Higher family stress is related to siblings of chronic pain patients experiencing more functional disability and somatic symptoms themselves (Guite et al., 2007). Qualitative studies have described the negative impact chronic pain can have on siblings' relationship (Gorodzinsky et al., 2013), and play with one another (Britton & Moore, 2002). Children and adolescents have also reported disruptions to family activities as a result of their sibling's chronic pain (Britton & Moore, 2002; Gorodzinsky et al., 2013). However, benefits to having a sibling with chronic pain have also been reported, such as a greater sense of compassion (Britton & Moore, 2002), or an enriched sibling relationship (Gorodzinsky et al., 2013). Interestingly, these findings are similar with those described above regarding the impact of chronic illness more generally on siblings' psycho-social functioning and daily experiences. It is clear from this literature that siblings are cognizant of, and attempt to be responsive to, one another's pain. Further, prolonged pain experiences can influence siblings' relationships, interactions, and functioning.

1.7 Overview of Methodology

The purpose of the dissertation was to explore siblings in the context of pediatric pain. To do so, a multi-method approach was used. The sections below present an overview of, and rationale for, the methodological approaches used in this dissertation.

The section begins by providing an overview of scoping review methodology. This approach was used in the current dissertation to provide an overview of the literature on siblings and pediatric pain (see Chapter 2). Detailed information on the methodology used in Chapter 3 is also provided, including a review of the cold pressor task, pain assessment in children, observational coding, and dyadic data analysis.

1.7.1 Scoping Reviews

A scoping review is a methodology for providing a broad overview of the literature on a specific topic (Arksey & O'Malley, 2005; Levac, Colquhoun, & O'Brien, 2010; Peters et al., 2015), and is a particularly well-suited approach for reviewing fields that are less established (Levac et al., 2010; Peters et al., 2015). Scoping reviews have been described as differing from systematic reviews in that they tend to be directed by less narrow research questions, and often include studies using a range of methodologies (Arksey & O'Malley, 2005; Levac et al., 2010; Peters et al., 2015). Further, since they are more concerned with providing a wide-ranging summary than an evidence-based answer to a precise question, scoping reviews commonly do not undertake quality assessment of the included research nor synthesize study findings (Arksey & O'Malley, 2005; Peters et al., 2015).

Five key steps have been proposed for conducting a scoping review, and these include: 1. Identifying the research question, 2. Identifying the relevant studies, 3. Selecting studies for inclusion, 4. Charting the data, and 5. Collating, summarizing and reporting the results (Arksey & O'Malley, 2005, p.8-9; Levac et al., 2010, p.4). One frequent purpose for conducting a scoping review is to determine the gaps within a

current area of research (Arksey & O'Malley, 2005). Based on the findings, researchers can provide suggestions for continued research on the topic (Levac et al., 2010).

1.7.2 The Cold Pressor Task

The cold pressor task (CPT) is one of the most frequently used experimental pain tasks with pediatric samples, and typically involves the child placing their hand into a tub of cold water (K A Birnie, Caes, et al., 2014; von Baeyer, Piira, Chambers, Trapanotto, & Zeltzer, 2005). Children are generally instructed to leave their hand in the water for as long as they are able, until it becomes too painful, affording young participants control over the pain experience (von Baeyer et al., 2005). The CPT is considered an ethical means of examining pain in children, posing minimal risk, with adverse events being highly uncommon (Kathryn A Birnie, Noel, Chambers, von Baeyer, & Fernandez, 2011). Further, children generally indicate having positive experiences, and feeling they have contributed in aiding others through their participation in studies using the CPT (Kathryn A Birnie et al., 2011). This task has been reported to be used with healthy and clinical (e.g., chronic pain) populations of children and adolescents across the age span, as early as 3-years-old (Kathryn A Birnie, Petter, Boerner, Noel, & Chambers, 2012).

A primary benefit of the CPT is that it allows researchers to study pediatric pain in a more standardized manner, and address hypotheses that would not be realistic to examine in everyday or clinical contexts (K A Birnie, Caes, et al., 2014; von Baeyer et al., 2005). Further, published guidelines for using the CPT with children exist to assist researchers in developing and implementing cold pressor studies (von Baeyer et al., 2005). Of relevance to the current dissertation, one of the identified strengths of the CPT is that it represents a particularly useful method for examining the role of family factors

and behaviours in pediatric pain (K A Birnie, Caes, et al., 2014). Although the CPT's relevance to everyday or clinical pains has been less researched (K A Birnie, Caes, et al., 2014; von Baeyer et al., 2005), it is considered to be most clinically applicable to the experience of acute pain, such as surgical or needle pain (von Baeyer et al., 2005). Providing some support for this assertion is the consistency in findings between studies examining the relationship between parent behaviours and children's acute pain experiences in experimental and clinical contexts (see section 1.3; e.g., Cohen et al., 2000; Manimala et al., 2000; McCarthy et al., 2010; Moon et al., 2011; Schinkel et al., 2017).

1.7.3 Pain Assessment in Children

Assessing children's pain levels is an important task in pediatric pain research. It is recognized that a child's experience of pain is composed of several relevant dimensions, and thus it has been recommended that assessment involve measurement of multiple aspects of pain, including pain intensity and pain-related emotions (P. J. McGrath et al., 2008; von Baeyer, 2014). There is evidence that, by the age of 8, children are capable of differentiating between intensity and affect when providing pain ratings (Goodenough et al., 1999). Commonly measured outcomes in pediatric studies using the cold pressor task include pain tolerance (i.e., the duration of time the child leaves their hand in the water), pain intensity and pain-related affect (Kathryn A Birnie et al., 2012; von Baeyer et al., 2005).

Self-report has been described as the key means of measuring pain in young children (von Baeyer, 2014). Faces scales are one tool for obtaining self-report measurements. These scales generally require a child to choose from a series of pictures

the face that best matches their experience (von Baeyer, 2006). Line-drawing faces scales have been recommended over photographs since they are neutral with regard to gender and ethnicity (von Baeyer, 2006). Further, they are simpler from a cognitive perspective than visual analogue or numeric rating scales (von Baeyer, 2006, 2014). However, it is important to be cognizant of the developmental skills required for a child to self-report their pain, even using faces scales, such as receptive language, symbolic processing, matching, and interoception (Besenski, Forsyth, & von Baeyer, 2007). It has been suggested that children are generally able to self-report their pain using a faces scale by the age of 4 (von Baeyer, 2006). Based on the literature described above, in the current dissertation, line-drawing faces scales were used to obtain self-report ratings of children's pain intensity and fear, and pain tolerance was additionally recorded, to provide a comprehensive assessment of children's cold pressor pain. Specifically, the *Faces Pain Scale-Revised* ("Faces Pain Scale-Revised," 2001; Hicks, von Baeyer, Spafford, van Korlaar, & Goodenough, 2001) was used to measure pain intensity, and the *Children's Fear Scale* (McMurtry, Noel, Chambers, & McGrath, 2011) was used to measure children's pain-related fear, as these measures have demonstrated evidence for validity in children as young as age 4 and 5, respectively. Further, the *Faces Pain Scale-Revised* was the suggested measure for children ages 4 – 12 in a systematic review of pain intensity measures based on its psychometrics (Stinson, Kavanagh, Yamada, Gill, & Stevens, 2006).

1.7.4 Observational Measures

Observational measures provide a means of quantifying behaviours, which may be verbal or non-verbal (J. Chorney & McMurtry, 2014). The use of an observational

measure typically entails viewing an individual(s) engaging in behaviours in a context of interest, and then documenting pre-determined behaviours, as defined in a coding system, in a reliable manner (Bakeman & Gottman, 1997). Observational measures can contribute to the breadth of studies by providing a richer estimate of the construct of interest than may be obtained through self-report alone (Bakeman & Gottman, 1997). In pediatric psychology, observational coding systems provide an ideal means of addressing hypotheses regarding specific behaviours children may engage in, or their interactions with others (J. M. Chorney, McMurtry, Chambers, & Bakeman, 2015). Guidelines have been published to assist pediatric researchers in creating and modifying observational coding systems (J. M. Chorney et al., 2015). Laboratory based tasks provide a context in which to use observational measures that have been described as offering a balance between experimental control and external validity (Wysocki, 2015). Within the context of laboratory tasks, observational measures were used in the current dissertation to assess sibling behaviours during pain and sibling relationship quality.

1.7.4.1 Observational assessment during the cold pressor task.

Observational measures have been widely used in the field of pediatric pain to assess children's pain behaviours (J. Chorney & McMurtry, 2014). Behavioural measures have been developed for the purpose of serving as a pain assessment tool (von Baeyer & Spagrud, 2007), as well as a means of examining interactions during pain, particularly between parents and children (Bai, Swanson, & Santacroce, 2017). The latter purpose was of interest in this dissertation. In laboratory settings, the cold pressor task has been used to examine the behaviour of both children and observers during pain (e.g., Kathryn A. Birnie et al., 2017; Moon et al., 2011; Schinkel et al., 2017). Using the guidelines

developed by Chorney and colleagues (J. M. Chorney et al., 2015), existing coding systems were modified in the current dissertation to measure siblings' behaviours during the cold pressor task (an overview of this coding system is provided in Chapter 3, and detailed descriptions of the behaviour codes are provided in Appendix B).

1.7.4.2 Observational assessment of sibling relationship quality.

Iturralde and colleagues developed a problem-solving interaction task and corresponding coding system to measure children's sibling relationship quality (Iturralde, Margolin, & Spies Shapiro, 2013). This task and coding system were used in the current dissertation to provide an observational measure of sibling relationship quality. Briefly, siblings were required to build a tower using craft materials, and their observed relationship positivity and negativity during the task were subsequently coded (Iturralde et al., 2013). A more in depth description of this task and observational measure is provided in Chapter 3, and detailed descriptions of the behaviour codes are provided in Appendix B.

1.7.5 Dyadic Data Analysis

Family researchers in the field of pediatric pain have been encouraged to begin using more complex statistical approaches, such as structural equation modelling, to address more innovative research questions (Palermo & Chambers, 2005). The current dissertation used a multi-informant approach, collecting data from both siblings in a dyad. Structural equation modelling, using the actor-partner interdependence model, was used to analyze the resulting dyadic data. This approach controls for the non-independence of the dyadic data, and permits the examination of relations between

variables within (i.e., actor effects) and between (i.e., partner effects) members of a dyad (Cook & Kenny, 2005).

1.8 Overview of Dissertation Papers

Building on the research described above, the current dissertation sought to provide a comprehensive and novel examination of the role of siblings in pediatric pain. Broadly, the dissertation aimed to: 1) offer an account of the current state of the literature on siblings in pediatric pain, 2) examine the role of sibling relationships and behaviours in children's experience of acute pain, and 3) explore the nature of, and issues relevant to, the presence of siblings in pediatric medical contexts. Three separate papers are presented here, each with unique objectives, that together seek to meet these three overarching goals.

Chapter 2 presents a scoping review of the research conducted to date on siblings in pediatric pain, which was informed by the approach described by Arksey and O'Malley (2005) and Levac and colleagues (2010). The objective of the scoping review was to summarize the characteristics of the research studies conducted thus far examining siblings in pediatric pain, with a goal of identifying gaps in the current literature and relevant directions for future investigation. To provide a comprehensive overview of the topic, the review included pediatric studies examining siblings in a range of pain contexts (acute, chronic, experimental) using a variety of methodologies (qualitative, quantitative). Information was extracted relating to the included studies' methodology, sample, outcome assessment, and focus.

The paper presented in Chapter 3 examined the relation between siblings' relationship quality and behaviours during the cold pressor task, as well as the relation between an observing sibling's behaviours and a child's pain outcomes. Methodological

limitations have been noted across family research in pediatric pain, as well as research on sibling relationships more broadly, and within health and pain. Specifically, research in these fields have tended to employ questionnaires to assess outcomes, with minimal use of observational approaches (Alderfer et al., 2010; Noller, 2005; Palermo & Chambers, 2005; Palermo et al., 2014). Further, the need to utilize multiple informants when assessing outcomes pertaining to family experiences has been repeatedly identified (Alderfer et al., 2010; Gorodzinsky et al., 2013; Knecht, Hellmers, & Metzling, 2015; Limbers & Skipper, 2014; Palermo & Chambers, 2005). This study was designed to address these limitations. Further, it addressed one of the gaps identified in the scoping review by investigating siblings in an acute pain context using an experimental task.

The study involved sibling dyads between the ages of 8 and 12 completing self-report and observational measures of sibling relationship quality, as well as taking turns participating in the cold pressor task with their sibling present. Pain intensity, tolerance, and pain-related fear were measured for each child, and the behaviour of the observing and participating sibling during the cold pressor task was coded as attending, non-attending, and coping/encouragement. Based on the work exploring family and parent factors described in section 1.3, it was hypothesized that greater positivity/warmth in the sibling relationships would be related to siblings engaging in more behaviours that removed attention from the pain/task (e.g., distraction), while more negative relationships would be related to siblings engaging in more pain/task focused behaviours (e.g., symptom talk) during the cold pressor. Further, it was hypothesized that behaviours by the observing sibling that were focused on the pain/task (e.g., reassurance) would be related to poorer pain outcomes for the child completing the cold pressor, whereas

behaviours removing attention from the pain/task would be related to better pain outcomes. Structural equation modelling, including the actor-partner interdependence model, was conducted to analyze the dyadic data.

The paper presented in Chapter 4 examined the extent to which siblings attend various pediatric medical appointments. It was hypothesized that most parents would report having brought siblings to their children's medical appointments. This study also investigated parents' perspectives on why they bring siblings to medical appointments and how they decide which child receives an appointment/procedure first; the relationship between child age and sex, and parent-child and sibling relationship quality and parents' responses to these questions was also examined. The sample consisted of the parents of the siblings who participated in the study described in Chapter 3. Parents completed a questionnaire which included closed and open-ended questions regarding sibling presence at pediatric appointments; responses to the open-ended questions were subsequently coded. Parents also completed self-report measures of their children's sibling relationship quality and their relationship with each of their children. In addition to providing insight into factors relating to the presence of siblings in medical contexts more broadly, this study also identified clinical settings that may be particularly relevant for examining relations between sibling variables and children's pain experiences.

CHAPTER 2: A SCOPING REVIEW ON THE STUDY OF SIBLINGS IN PEDIATRIC PAIN

The manuscript based on this study is presented below. Meghan Schinkel, under the supervision of Dr. Christine Chambers, was responsible for developing the research question, search strategy (in consultation with librarians) and data extraction manual, and applying for funding. She contributed substantially to the study search and screening, and data extraction. She was responsible for analyzing the findings and writing the current manuscript. The manuscript was reviewed by the co-authors, and their feedback incorporated, prior to submitting the manuscript. This chapter represents an accepted manuscript of an article published by Taylor & Francis in the *Canadian Journal of Pain*. The manuscript was accepted for publication on October 27, 2017, and is available online at <http://www.tandfonline.com/doi/full/10.1080/24740527.2017.1399053>. The current reference for this manuscript is:

Schinkel, M.G., Chambers, C.T., Hayden, J.A., Jordan, A., Dol, J., & Higgins, K.S. (2017). A Scoping Review on the Study of Siblings in Pediatric Pain. *Canadian Journal of Pain, 1*(1), 199-215.

2.1 Abstract

Background: Sibling relationships are longstanding across an individual's life, and are influential in children's development. The study of siblings in pediatric pain is, although in early stages, a growing field. **Aims:** This scoping review sought to summarize and map the type of research available examining siblings and pediatric pain, to identify gaps and directions for future research. **Methods:** Studies were identified based on a search of PubMed, CINAHL, PsycInfo, Embase, and Web of Science (up to November 2016). We extracted data about study methods, the sample, outcome assessment, and the influence/relationships investigated. **Results:** Thirty-five studies were included. Most studies used quantitative methods ($n = 28$), and participants typically comprised children (i.e., aged 6-12; $n = 24$) and adolescents (i.e., aged 13-18; $n = 18$). The majority of studies examined siblings in the context of chronic and disease-related pain ($n = 30$). While quantitative studies primarily focused on the genetic influence of pain conditions ($n = 18$), qualitative and mixed methods studies typically focused on exploring the impact of siblings with and without pain on one another ($n = 2$), and the impact of pain on the broader dyadic relationship/functioning ($n = 4$). **Conclusions:** Sibling research in pediatric pain has been primarily focused on the biological/physical components of pain using quantitative approaches. Conducting more studies using qualitative or mixed-methods designs, incorporating multiple assessment measures (e.g., observational, self-report) and multiple perspectives (e.g., siblings, health professionals) may provide an opportunity to gain richer and more comprehensive information regarding the experience of siblings.

Key words: siblings, pediatric pain, families, children, scoping review

2.2 Introduction

A strong body of research has been developed in the field of pediatric pain over the past few decades exploring various aspects of children's pain (Caes et al., 2016). In particular, research has moved beyond examining only child and adolescent functioning to exploring the wider context of families in pediatric pain. Within chronic pain, models have been developed to elucidate the interplay between family-related variables and children's experiences of pain and functioning (Palermo & Chambers, 2005; Palermo et al., 2014). Studies have also focused on examining parental behaviour in the context of children's acute procedural pain (e.g., Manimala, Blount, & Cohen, 2000; Martin, Chorney, Cohen, & Kain, 2013; McMurtry, C.M., McGrath, P.J., Chambers., 2006; McMurtry, Chambers, McGrath, & Asp, 2010). Laboratory based studies have added further insight into the influence of familial variables on children's pain experiences. For example, experimental pain tasks (e.g., the cold pressor task) have been widely utilized to explore family factors in pediatric pain, such as the influence of parent behaviours (e.g., Moon, Chambers, & McGrath, 2011; Schinkel, Chambers, Caes, & Moon, 2017; Walker et al., 2006), parental social modeling (Boerner et al., 2017) and family functioning (Kathryn A. Birnie et al., 2017). Despite a plethora of family-focused research across multiple domains of pediatric pain, siblings have received relatively little attention in the pediatric pain literature. However, siblings are also important family members for children. The majority of children have a sibling (Dunn, 2000), and research on siblings has outlined the impact siblings exert on one another with regard to developmental outcomes, and psycho-social and behavioural functioning (Brody, 2004; McHale, Updegraff, & Whiteman, 2012 provide reviews). Further, research on pediatric chronic

health issues suggests a sibling's experience of illness can adversely influence children's functioning in several areas, such as psychological symptoms (Vermaes et al., 2012), quality of life (Limbers & Skipper, 2014), and academic participation and performance (Gan et al., 2017).

Although limited research exists exploring siblings in pediatric pain, there have been a growing number of both quantitative and qualitative studies focused on the topic. For example, quantitative studies have revealed differences in psycho-social functioning between siblings of healthy children and those with chronic pain conditions, with siblings of pediatric pain patients experiencing poorer functioning, such as anxiety, depression and social difficulties (Engstrom, 1992; Guite et al., 2007). Qualitative studies have begun to illustrate the nature of young siblings' relationships and everyday life when one experiences chronic pain (Britton & Moore, 2002; Gorodzinsky et al., 2013). This work highlights the influence pain can have on siblings in terms of their personal mental health and relationship with one another, and suggests that continued research on the topic has potential to make a valuable contribution to our understanding of the role of families in pediatric pain.

Unlike the more narrow focus of a systematic review, scoping reviews aim to broadly summarize and map research in a given field (Arksey & O'Malley, 2005; Levac et al., 2010; Peters et al., 2015). They are often conducted when a goal is to determine areas in need of further research (Arksey & O'Malley, 2005). In contrast to systematic reviews, scoping reviews tend to include studies using a wider array of methods (published or unpublished) (Arksey & O'Malley, 2005; Levac et al., 2010; Peters et al., 2015), and generally focus on describing the literature rather than synthesizing findings to

determine effectiveness or the strength/direction of impact (Arksey & O'Malley, 2005; Peters et al., 2015). Scoping reviews are considered a useful approach for research areas that are still developing (Levac et al., 2010; Peters et al., 2015), or where the research is varied (Peters et al., 2015). The existing body of research on siblings and pediatric pain is both limited and varied in terms of focus, methodology, and discipline, thus indicating a scoping review is the appropriate method for reviewing this area. No known reviews have been conducted on the topic to date. Therefore, the field could benefit from a summary of the work that has been conducted; this may help to identify gaps in the field, stimulate further research and provide direction moving forward.

In order to provide an overview of the literature to date, the objective of the scoping review was to summarize and map the type of research that has been conducted examining siblings and pediatric pain. Specifically, the review sought to address the question, "What are the characteristics of research studies that have explored the role of siblings in pediatric pain?" This was undertaken with a goal to identify gaps in the literature and directions for future research.

2.3 Methods

The methodological approach was informed by current guidelines for conducting scoping reviews (Arksey & O'Malley, 2005; Levac et al., 2010; Peters et al., 2015; The Joanna Briggs Institute, 2015).

2.3.1 Search Strategy

A search of the electronic databases PubMed, CINAHL, PsycInfo, Embase, and Web of Science was conducted on November 8, 2016. These databases were selected in order to try and identify relevant studies across disciplines. The search included a

combination of terms, formatted for each database, related to siblings (e.g., sibling, sister, brother), pain (e.g., chronic pain, experimental pain, needle) and children (e.g., child, pediatrics). The pain terms included keywords related to chronic pain, experimental pain and procedural pain, and were informed by keywords used in recent reviews in pediatric pain (Boerner KE, Birnie KA, Caes L, Schinkel M., 2014; Higgins et al., 2015). The child terms represented a validated search strategy for identifying pediatric focused studies (Leclercq, Leeflang, Van Dalen, & Kremer, 2013). Development of the search terms and identification of appropriate databases also involved consultation with librarians, who have expertise in conducting searches for reviews, and discussion amongst the co-authors. See section 2.10 (Supplemental Materials) for the complete search terms used formatted for each database. Additional relevant articles known to the authors based on their knowledge of the literature were also identified for subsequent screening. An additional search was conducted of the electronic databases used in the original systematic search, up to the date of the original search, of the included conference abstracts to ensure no subsequent published manuscripts based on the abstracts had been missed.

2.3.2 Eligibility Criteria

To be eligible for inclusion, studies had to be pediatric-focused, which was defined as including a sample composed of children ages 0-18 (Bai et al., 2017; Boerner KE, Birnie KA, Caes L, Schinkel M., 2014; Caes et al., 2016), and/or adults reporting on children, or adult retrospective studies (i.e., adults reflecting on their experiences as children). Additionally, both siblings and pain or a pain condition had to be of primary interest, as identified in the title and/or abstract. Studies examining siblings in the context

of experimental, acute, chronic, or procedural pain were all included. All studies available up until the date of the search that were written in English and reported empirical data or synthesized data using any methodological design, either published or unpublished, were included.

Studies were excluded if they described families broadly with siblings not being a specific focus, or included healthy siblings only as a healthy control group. Case studies reporting on more than one sibling who had the same illness (e.g., a case study on a genetic condition), studies referring to pain in an emotional sense (e.g., depression), and studies focused on cancer related pain were excluded. Lastly, articles that were commentaries (i.e., opinion or reaction/reflection based publications) or letters to the editor were excluded.

2.3.3 Study Selection

All identified studies were imported into and screened using *Covidence* (“Covidence,” 2017), which is an online screening and data extraction tool designed to help facilitate the review process (see Figure 1 for a flow chart outlining the study selection process). First, MS and JD separately completed a title and abstract screen of all the identified studies ($n = 11590$). Discrepancies were resolved by consensus between the two co-authors. For all studies that passed this initial screening stage ($n = 176$), the full text was retrieved and reviewed separately by MS and JD, and discrepancies were again resolved by consensus. Two additional studies were excluded during the data extraction phase due to not reporting on a pediatric sample.

2.3.4 Data Charting

A comprehensive data extraction manual, which provided descriptions of the extraction categories, was developed and underwent several rounds of review by the team of authors prior to charting the data. The full extraction manual is included in Appendix B. To summarize, we collected: 1) descriptive information about the article, including the name of the study, the authors, publication year, geographic location of the study (or if not listed, the location affiliation of the first author), the discipline affiliation(s) of all authors, and the type of study (published research, dissertation, case study, or conference abstract); 2) information regarding the methodology used (qualitative, quantitative, or review; methodological sub-categories were included within each); 3) information about the study sample, including the age of the children (Baby/toddler = < 2 years old; Preschool = 2-5 years old; Child = 6-12 years old; Adolescent = 13-18 years old; or Not specified) (Caes et al., 2016), whether adults were included in the study (parents, health professionals, teachers, or adults reporting retrospectively), the type(s) of pain population(s)/context of interest in the study (acute/procedural, chronic/disease related, or experimental - e.g., the cold pressor task; subcategories were included within each), and whether children with a comorbid/other condition of interest (e.g., pain being studied in a group of children with a comorbid, non-pain related condition) or healthy children (e.g., healthy siblings, healthy children experiencing experimental or acute/procedural pain) were included in the sample; 4) the type(s) of outcomes assessed (demographic variables, quality of life, mental health/psychosocial functioning, adaptive functioning/disability, sibling relationship quality, parent-child relationship quality, parent marital relationship quality, family functioning, pain or somatic symptoms, genetic vulnerability, and Juvenile Arthritis disease features), how outcomes were measured (questionnaire or

survey, observational measures, focus groups, interviews, health records/medical results, pain assessment tools), and who reported on outcomes (parent, healthy/pain free sibling, sibling with pain/condition, health professional) and whether they were reporting on themselves, others (e.g., a parent reporting on their child, a child reporting on their sibling), or having their behaviour observed; and lastly, 5) the influence/relationships investigated in the study in relation to siblings (the impact of the sibling experiencing pain on the healthy/pain free sibling, impact of healthy/pain free sibling on sibling experiencing pain, bidirectional, impact of/relationship between two siblings with pain/condition on one another, mediating impact of parents or family, the impact of pain on the broader dyadic relationship or functioning, genetic influence). Following initial data extraction, it was determined that “Juvenile Arthritis disease features” should be an option under the outcomes assessed category, and “genetic influence” (i.e., studies examining siblings within the context of genetic vulnerability for pediatric pain conditions) should be an option under the influence/relationship investigated category. Therefore, these options were subsequently added and relevant studies were re-categorized.

The data was charted in *Microsoft Excel*, and primarily involved indicating the option(s) for each extraction category that best characterized the study. Data from studies could be extracted as falling into more than one option within each category. Data charting was completed for all included studies independently by two co-authors (MS and either JD or KH, who each charted data for half of the studies). Data charting files were compared between reviewers, and discrepancies were resolved by consensus.

2.3.5 Summarizing the Results

Microsoft Excel was used to calculate descriptive statistics (e.g., totals, percentages), and to create figures to summarize the data. Descriptive information of all included studies were examined together. The studies were then split based on methodology (quantitative, qualitative, mixed methods), and more detailed results (e.g., participant characteristics, outcomes) were examined separately within each of the methodology categories.

2.4 Results

2.4.1 Descriptive Information

Thirty-five studies were included in the review, representing a total of 21810 subjects (note: 8 studies reported the sample size as the number of participating families). See section 2.10 (Supplemental Materials) for descriptive information (e.g., publication year, discipline) of the included studies. The majority of included studies were published research studies ($n = 21$), with the remainder comprising conference abstracts ($n = 12$) or dissertations ($n = 2$) (Table 1). No subsequent published manuscripts based on the included conference abstracts were identified in the search. Most of the research papers (or studies) were classified as quantitative ($n = 28$), although some qualitative studies have been conducted ($n = 5$). Additionally, two studies were mixed methods, utilizing both quantitative and qualitative methodology. We did not identify any reviews conducted in the field as of the date of the search (Table 1).

2.4.2 Quantitative Studies

Methods.

Experimental/quasi-experimental ($n = 10$) and non-experimental methods ($n = 12$) were used in a similar number of studies, with fewer studies using a cross-sectional

design ($n = 6$). Almost all studies were classified as including a descriptive component (i.e., reporting descriptive findings; $n = 23$). Although longitudinal and measurement (e.g., questionnaire development) were included as options, no studies were extracted as falling into these categories.

Sample.

Most studies included participants in the child ($n = 18$) or adolescent ($n = 11$) age categories, with fewer studies including preschool aged children ($n = 9$) or babies/toddlers ($n = 4$) (Figure 2). It should be noted that seven studies did not specify the age of their pediatric sample. Adults also often participated in the quantitative studies. Most studies included parents ($n = 19$), three studies also included health professionals (M. B. Moroldo et al., 2004; M. Moroldo, Tague, Shear, Glass, & Giannini, 1997; Svensson, Larsson, Bille, & Lichtenstein, 1999), and one study included teachers (Scherder, Rommelse, Bröring, Faraone, & Sergeant, 2008). None of the examined studies included adults who reported retrospectively on their childhood (Figure 2).

With regard to the type of pain examined, almost all studies focused on chronic or disease related pain ($n = 24$), with the most common pain sample being Juvenile Arthritis/Rheumatic Diseases ($n = 6$). Four studies were focused on acute/procedural pain (Badiee, Nassiri, & Armanian, 2014; M L Campbell-Yeo et al., 2012; Marsha L Campbell-Yeo et al., 2014; Scherder et al., 2008), and only one examined pain in the context of an experimental task (Scherder et al., 2008) (Table 2). In addition, three studies included a sample of children with co-morbid or other conditions of interest (J. J. Field, Macklin, Yan, Strunk, & DeBaun, 2008; Scherder et al., 2008; Wong et al., 2011). Most studies ($n = 21$) included healthy children in their sample.

Outcomes.

Most of the quantitative studies examined demographic variables (e.g., socioeconomic status; $n = 19$). Following demographics, the most frequently assessed outcomes were genetic vulnerability ($n = 18$) and pain or somatic symptoms (e.g., pain severity, condition related symptoms; $n = 17$). Mental health/psychosocial functioning was also of interest in a number of studies ($n = 8$) (Figure 3). Many studies assessed outcomes that did not fall into one of the extraction categories, and these were varied in nature such that they could not be meaningfully categorized (e.g., malaria history (Campbell et al., 2009), co-sleeping with a parent, sibling or pet during first year of life (Miller et al., 2015)).

Outcome Assessment.

Most studies relied on questionnaires or surveys to assess outcomes ($n = 20$), followed by health records or medical results ($n = 11$). Only two studies utilized observational measures (Badiee et al., 2014; M L Campbell-Yeo et al., 2012) and only one study used pain assessment tools (Scherder et al., 2008) (Figure 4). Half of the studies ($n = 14$) used parent report to assess outcomes. Within these studies, 93% of parents reported on others and 43% reported outcomes on themselves. Healthy/pain free siblings ($n = 8$) and siblings with pain ($n = 11$) reported on outcomes in less than half of the studies. Within both categories, most children reported on themselves. Of the three studies that used health professionals to report on outcomes, all reported on others (Table 3).

Influence/relationships investigated.

The majority of quantitative studies were focused on siblings in the context of a genetic influence/vulnerability for a pediatric pain condition ($n = 18$). This was followed by studies examining the impact of/relationship between two siblings with pain/a condition on one another ($n = 6$) (Figure 5). No studies examined the mediating impact of parents or family (e.g., examining how parent mental health mediates the impact of child chronic pain on a healthy sibling).

2.4.3 Qualitative Studies

Methods.

Interviews ($n = 3$) (Britton & Moore, 2002; Gordon, 2013; Gorodzinsky et al., 2013), a qualitative questionnaire ($n = 1$) (Britton & Moore, 2002), and focus groups ($n = 1$) (Akobeng et al., 1999) were used to obtain data in the qualitative studies, with three studies also using “other” means (e.g., drawings (Moscato, Calabrese, Moscato, & Ribaud, 2009)). To analyze the qualitative data, one study reported using inductive content analysis (Gordon, 2013), one grounded theory (Britton & Moore, 2002), and one the Delphi coding procedure (Gorodzinsky et al., 2013). Two studies reported using “other” qualitative analytic approaches (e.g., describing qualitative findings from a projective test (Moscato et al., 2009)).

Sample.

Aligning with the quantitative studies, most qualitative studies included participants within the child ($n = 4$) and adolescent ($n = 5$) age ranges. Parents were also included in three of the qualitative studies (Akobeng et al., 1999; Britton & Moore, 2002; Gordon, 2013), and health professionals were included in one study (Moscato et al., 2009) (Figure 2). All of the qualitative studies were focused on chronic/disease-related

pain, with the specific disease of interest varying across studies (Table 2). No studies included a sample of children with co-morbid or other conditions of interest, but all studies had healthy children included in the sample.

Influence/relationships investigated.

Three of the qualitative studies were focused on the impact of pain on the broader dyadic relationship or functioning (Gordon, 2013; Gorodzinsky et al., 2013; Moscato et al., 2009), with two studies also focused on the bidirectional impact of siblings with pain and healthy/pain-free siblings on one another (Gordon, 2013; Gorodzinsky et al., 2013). One study was focused solely on the impact of the sibling with pain on the healthy/pain free sibling (Akobeng et al., 1999), and one study was classified as “other”, and was focused on the general experiences of families of children with Juvenile Idiopathic Arthritis (Britton & Moore, 2002) (Figure 5).

2.4.4 Mixed Methods Studies

Methods.

Within the two mixed methods studies, one used an experimental/quasi-experimental (Valkenburg, Tibboel, & van Dijk, 2015) and the other a non-experimental design (Wutzke, 1999), with both including a descriptive component. To analyze the qualitative data, one study reported using thematic analysis (Wutzke, 1999) and the other study did not clearly specify their approach, but reported using qualitative questions to obtain data (Valkenburg et al., 2015).

Sample.

Both studies included participants in the child and adolescent age ranges ($n = 2$). Parents were included in both studies, with one study also including teachers (Wutzke,

1999) (Figure 2). One of the studies was focused on chronic pain (Wutzke, 1999), while the other examined pain in the context of an experimental task (Valkenburg et al., 2015) (Table 2). One of the studies included a sample of children with a co-morbid or other condition of interest (Valkenburg et al., 2015), and both included healthy children in the sample.

Outcomes.

Demographics were assessed in both studies, with the following outcomes additionally being assessed in either one of the two studies: mental health/psycho-social functioning (Wutzke, 1999), adaptive functioning/disability (Valkenburg et al., 2015), sibling relationship quality (Wutzke, 1999), parent-child relationship quality (Wutzke, 1999), and pain/somatic symptoms (Valkenburg et al., 2015). Both studies also assessed outcomes that fell in the “other” category (e.g., reaction time of non-dominant hand (Valkenburg et al., 2015), general experience of having a sibling with juvenile rheumatoid arthritis (Wutzke, 1999)).

Outcome Assessment.

Outcomes were measured using questionnaires in both studies, with one study additionally using observational measures, pain assessment tools and “other” measurement tools (Valkenburg et al., 2015), and the other study additionally using interviews (Wutzke, 1999). Parents (reporting on others) and healthy/pain free siblings (self-report) provided information in both studies, with a sibling with pain/a pain condition additionally reporting on themselves in one of the studies (Valkenburg et al., 2015). A teacher also provided information in one of the studies (Wutzke, 1999).

Influence/relationships investigated.

One of the studies focused on the impact of the sibling with pain on the healthy/pain free sibling as well as the impact of pain on the broader dyadic relationship or functioning (Wutzke, 1999). The impact investigated in the second study was extracted as falling into the “other” option, and was focused on pain in children with Down syndrome and their siblings (Valkenburg et al., 2015) (Figure 5).

2.5 Discussion

This scoping review sought to summarize and map the research conducted to date on siblings and pediatric pain, with an aim to identify gaps in the literature and directions for future research. Limited research on the topic was identified. Only 60% of the 35 included studies were published research studies, suggesting the field is still developing, and that more research is needed. Regarding methodology, most identified studies were quantitative, using either experimental/quasi-experimental or non-experimental designs. A small group of qualitative studies have also been conducted, and they varied in terms of their approach to obtaining data and analyzing findings. Only two mixed-methods studies were identified. Therefore, the field has taken a primarily quantitative approach to understanding siblings and pediatric pain, with less focus thus far on obtaining qualitative information regarding participants’ perspectives and experiences, or using complimentary mixed-methods approaches.

The findings pertaining to the sample characteristics were generally consistent across methodology type. Concurrent with the broader pediatric pain literature (Caes et al., 2016), participants were most often in the child and adolescent age groups. However, a notable number of studies included preschool aged children or babies/toddlers (combined $n = 13$). Therefore, research examining siblings in pediatric pain is generally

well distributed across the pediatric age span. Parents were typically included in the studies examined, suggesting information pertaining to, or reported by, parents have been valued in the field thus far. Chronic and disease-related pain was the predominant context in which research has examined siblings in pediatric pain, with less attention paid to the potential role of siblings in acute pain experiences. Further, most studies included healthy children in their sample, likely reflecting an inclusion of healthy siblings of chronic pain patients.

Within the quantitative studies, the most commonly assessed outcomes were genetic vulnerability and pain or somatic symptoms, suggesting a focus thus far on the biological or physical components of pediatric pain. Mental health and/or psychosocial functioning were also assessed in several studies, indicating that research on siblings has also examined, albeit to a lesser extent, psychological factors related to pain. Questionnaires and surveys were the predominant means of assessing outcomes for quantitative studies. Parents were a primary source of information, with parents reporting on outcomes in half of the studies. Children (i.e., healthy siblings or siblings with pain) also provided information, although less frequently (less than half of the included studies). Therefore, research findings have been primarily based on parent, questionnaire-report, with less focus on obtaining children's perspectives or garnering information from behavioural observation.

The findings for the influence/relationships investigated amongst the quantitative studies revealed that most studies were interested in siblings in the context of a genetic influence/vulnerability for a pain condition. However, a difference was noted across the methodology types. Unlike quantitative studies, qualitative and mixed methods studies

more often focused on the impact of children on their sibling, and the impact of pain on the broader dyadic relationship or functioning. Therefore, much of our understanding of siblings' functioning and broader experiences come from a qualitative perspective, with limited supporting quantitative data on these topics.

2.5.1 Identified Gaps and Directions for Future Research

As evidenced by the limited numbers of studies in specific areas, gaps were noted regarding the methodology, samples, outcome assessment, and the outcomes and influence/relationships investigated, suggesting some relevant directions for continued research.

First, in terms of methodology, limited qualitative studies exist focusing on siblings and pediatric pain. Given that it is a relatively new field, conducting more qualitative research with patients, families and clinicians may serve as a means for identifying predominant issues and concerns from the perspectives of those who are most impacted. Further, qualitative methodology typically involves encouraging participants to provide detailed, non-directed responses to open-ended questions on specific topics. Therefore, qualitative studies may offer more in-depth and detailed information regarding specific aspects of individuals' personal experiences than that which may be obtained through quantitative methods alone (e.g. questionnaires). This richer understanding could also contribute to the development of theoretical models regarding how siblings may impact, and be impacted by, children's pain experiences. Topics of importance identified through qualitative studies could then be further explored using qualitative, quantitative or mixed-methods designs. Consistent with limitations identified in sibling research (Alderfer et al., 2010; Noller, 2005) and family research in pediatric pain (Palermo &

Chambers, 2005; Palermo et al., 2014), no longitudinal studies were identified in the current review. Longitudinal designs could usefully be conducted to answer potentially important research questions, such as the impact of pediatric chronic pain on siblings across developmental stages, or the impact of viewing a sibling undergo a painful procedure on a healthy child's subsequent experience. Further, no measurement studies were identified. In order for the field to grow, more validated self-report and observational measures pertaining to siblings and pediatric pain will need to be developed.

Second, regarding the samples used, almost all studies regardless of methodology concentrated on siblings in the context of chronic or disease-related pain. This is certainly a valuable area for continued research. However, attention should also be given to siblings in the context of acute procedural pain (e.g., surgery, needles) or everyday pains (e.g., illness, injuries). Approximately 98% of parents report bringing siblings to their child's medical appointments, with 85% specifically reporting bringing siblings to needle procedures (see Chapter 4), supporting the relevance of exploring the influence of siblings in these acute pain settings. Quantitative and qualitative designs could be used to answer any number of relevant research questions, such as siblings' impact on children's procedural pain or distress, or children's responses to their siblings' common pains at home. Only two studies were identified that examined experimental pain. Experimental pain tasks offer a more standardized approach to studying pediatric pain (Kathryn A Birnie, Caes, Wilson, Williams, & Chambers, 2014), and thus have the potential to provide valuable insight into sibling factors relevant to chronic or acute pain. For example, using a standardized experimental pain task, researchers could compare child responses to a pain stimulus with or without a sibling present. Differences in child

responses could then be attributed to the presence of the sibling with a greater degree of confidence than could be afforded in a more unpredictable clinical context. Guidelines exist that could be used to assist researchers in identifying the most appropriate pain task for the research question at hand (see Birnie et al., 2014).

Third, regarding outcome assessment, most quantitative studies on siblings and pediatric pain used questionnaires to assess outcomes. A frequent dependence on questionnaires, and need to begin to use other forms of outcome assessment, has been noted both in sibling research (Alderfer et al., 2010; Noller, 2005) and in research on families in pediatric pain (Palermo & Chambers, 2005; Palermo et al., 2014). Research on siblings and pain could begin to use other assessment measures, such as observational measures or pain assessment tools, to provide richer and more comprehensive information. Further, very few studies included health professionals. Incorporating health professionals is likely valuable as they could offer a unique perspective on the outcomes of interest. They may also have insight into other important topics for research on siblings, relevant to chronic or acute pain, based on their experiences working with families.

The findings regarding the source of information for outcome assessment amongst the quantitative studies suggest that most studies did not use a multi-informant approach. As noted above, half of the studies used parent report, while less than half of the studies included children themselves (i.e., healthy siblings or siblings with pain) to provide information on outcomes. This finding has both empirical and theoretical implications. As recommended for family research in pediatric pain (Palermo & Chambers, 2005), future research on siblings should incorporate the perspectives of multiple family

members when possible, including all relevant caregivers and siblings, to obtain a complete picture of the issue of interest. This is important because studies on siblings of children with health issues, including pain, have noted discrepancies between reports within family members (Gorodzinsky et al., 2013; Guite et al., 2007; Limbers & Skipper, 2014). From a theoretical perspective, the need for theory-guided studies on siblings and pediatric pain has been noted (Jaaniste, Phipps, Lang, & Champion, 2013). Research in this area would be wise to capitalize on the well-developed models that already exist on families and pediatric pain (see Palermo & Chambers, 2005; Palermo et al., 2014). However, these models view relations between family and child variables relevant to pediatric pain as being bi-directional (Palermo & Chambers, 2005; Palermo et al., 2014). Therefore, a multi-informant approach to the study of siblings is warranted to build on our existing theoretical understanding of families and pediatric pain.

Lastly, the outcomes and influence/relationships investigated in the included studies suggest a strong focus thus far, particularly amongst quantitative studies, on genetic factors related to pediatric pain conditions. Pediatric health issues more broadly can influence the functioning and experiences of healthy siblings across a number of domains (e.g., psychological and social functioning, daily life, academics) (Alderfer et al., 2010; Gan et al., 2017; Limbers & Skipper, 2014; Vermaes et al., 2012; Woodgate et al., 2016), pointing to the relevance of further exploring these variables in siblings of chronic pain patients. An examination of the results of the quantitative (Engstrom, 1992; Guite et al., 2007) and mixed-methods (Wutzke, 1999) studies included in the current review that examined the impact of chronic pain on healthy siblings' psycho-social functioning revealed some convergent findings, and point to a generally negative

influence. Specifically, as noted in the introduction, two quantitative studies similarly found that siblings of children with chronic pain conditions have significantly more social/peer difficulties, and greater anxiety and depression compared to control groups of siblings of healthy children (Engstrom, 1992; Guite et al., 2007). Further, although no control group was included, a mixed-methods dissertation found that seven of the ten healthy siblings of children with juvenile rheumatoid arthritis included in the study were identified (based on self, parent, and/or teacher report) as having significant difficulties on a measure of psycho-social functioning (e.g., regarding internalizing behaviours, externalizing behaviours) (Wutzke, 1999). Thus, continued research is needed that focuses on other potentially important factors related to siblings and pediatric pain, such as psycho-social and adaptive functioning, quality of life, family functioning and relationships, or social determinants of health. Research focused on a broader array of outcomes amongst both healthy siblings and those with chronic pain conditions will provide a more comprehensive account of the influence siblings may have in both chronic and acute pain contexts. Further, although 18-years was used as the upper age limit for studies included in the current review, examining sibling relationships in the context of pediatric pain during older adolescents/early adulthood would be a valuable direction for future research, as sibling impacts may differ as older adolescents leave the family home.

2.5.2 Limitations

There are several limitations to the current scoping review that should be noted. Although the search strategy was developed to capture all potentially relevant studies, it is possible that some relevant studies were missed. Further, the scoping review did not differentiate between studies based on their sample size or type (i.e., published research,

abstract, dissertation), and the quality of the included research studies or the potential strength of their findings was not assessed. This may be particularly important to consider given the high number of included studies that were not published research (e.g., conference abstracts with no identified corresponding peer-reviewed manuscript), and therefore may not have been exposed to the same level of scrutiny as standard peer review. Further, stakeholder consultation has been suggested as a step that could be undertaken when conducting a scoping review (Arksey & O'Malley, 2005; Levac et al., 2010). Given that the results of the review confirmed that the field is still in early stages of development and continued research is needed across many areas, it was decided that formally conducting a stakeholder consultation would not add significant value.

However, the review findings and potential interpretations were formally discussed amongst the co-authors, which include individuals engaged in family research in pediatric pain. Engaging stakeholders, including clinicians and families, in study design and implementation of research focused on siblings and pediatric pain will be valuable as the field progresses. Once the literature is more developed and there are more established findings, stakeholders may be able to comment on these findings in relation to their experiences and offer suggestions for new research questions (Levac et al., 2010).

2.5.3 Conclusion

The findings of this scoping review suggest that research on siblings in pediatric pain is a growing field. Although some areas emerged as being further developed than others, such as research using quantitative methods, and studies focused on genetics and chronic/disease-related pain, continued research is needed across many domains.

Theoretical models on families and pediatric pain (see Palermo & Chambers, 2005;

Palermo et al., 2014) could be applied to research on siblings to provide both a theoretical foundation, as well as ideas for relevant research questions. As the field develops, the role of siblings should be more explicitly incorporated into these family models. Although the best research design will be informed by the question of interest, a mixed methods approach using multiple informants will likely yield the most meaningful information. Validated tools relating to siblings and pediatric pain, including both observational and self-report measures, need to be developed to adequately address relevant research questions. Once sufficient research exists examining specific research questions or outcomes pertaining to siblings, conducting a systematic review and meta-analysis will be an important next step in developing an evidence base. For example, genetic factors pertaining to pediatric pain conditions or psycho-social outcomes of siblings of children with chronic pain would be meaningful topics for systematic reviews once the literature is more developed. It is hoped the findings of this review can be used as a guide for researchers interested in furthering the understanding of siblings in pediatric pain.

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<http://doi.org/10.1111/cch.12345>

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2.8 Figures

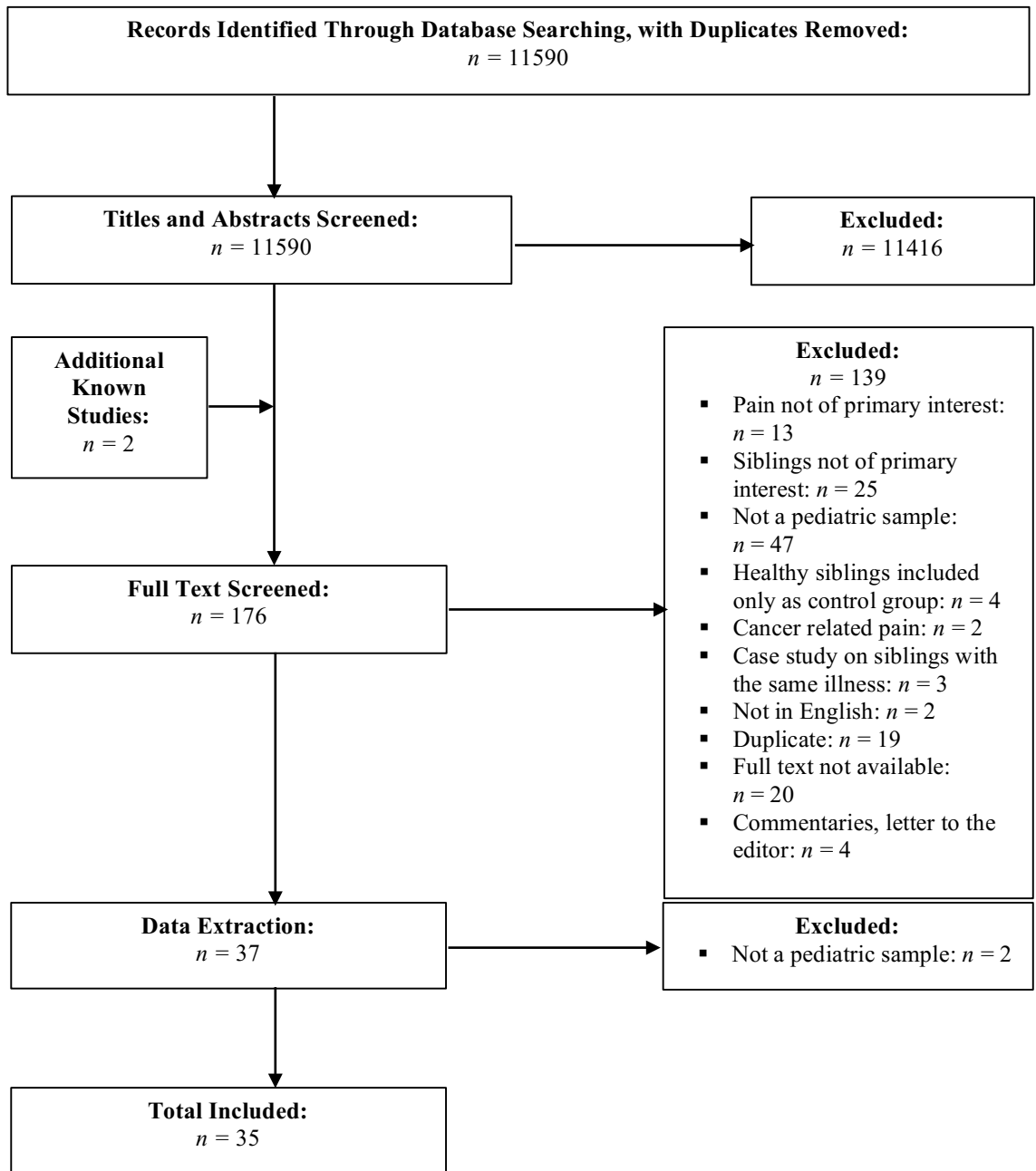


Figure 2.1. Flow chart outlining the study selection process.

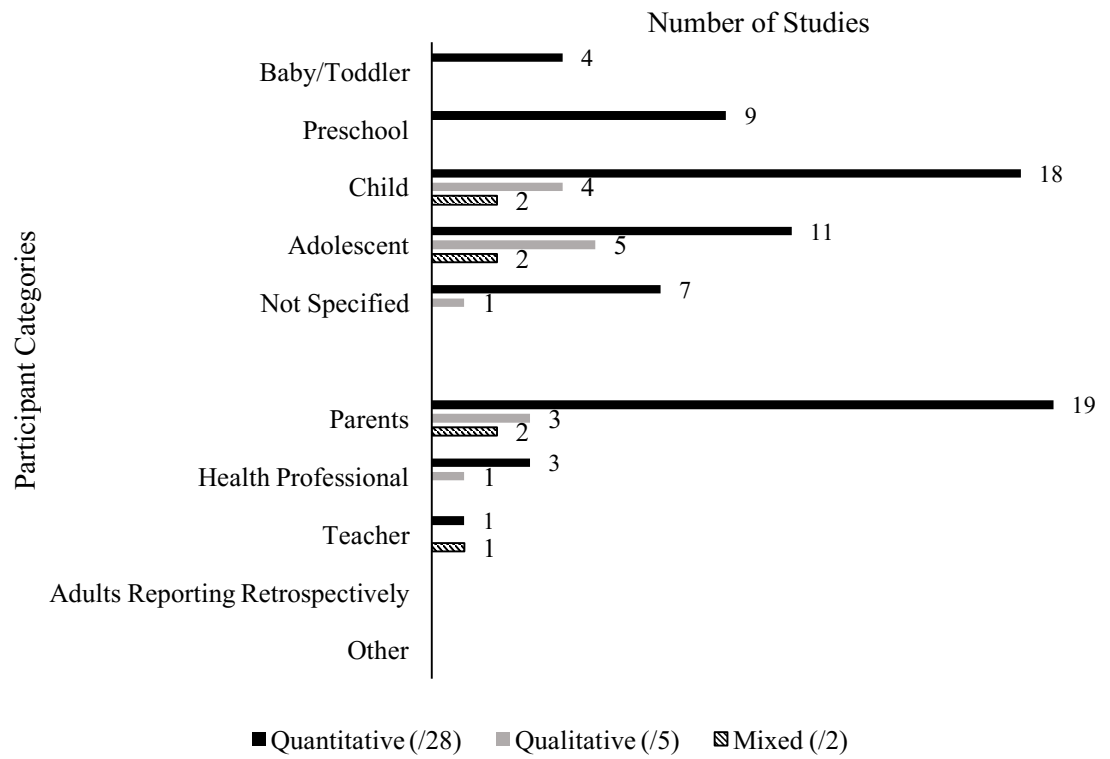


Figure 2.2. Participant characteristics of included studies by methodology type across age of pediatric sample and adult involvement.

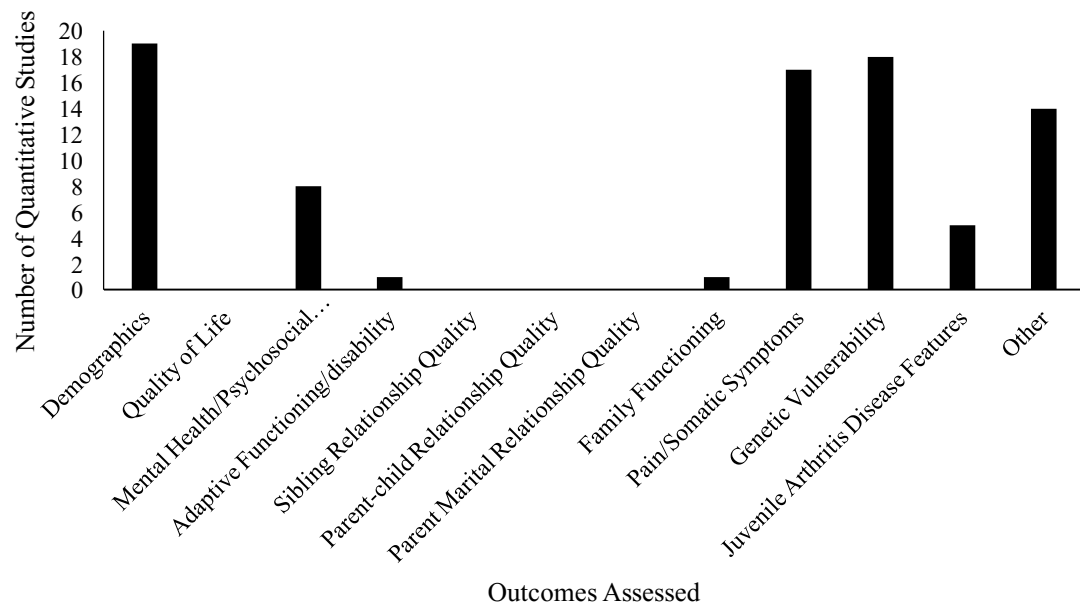


Figure 2.3. Number of quantitative studies assessing extracted outcomes.

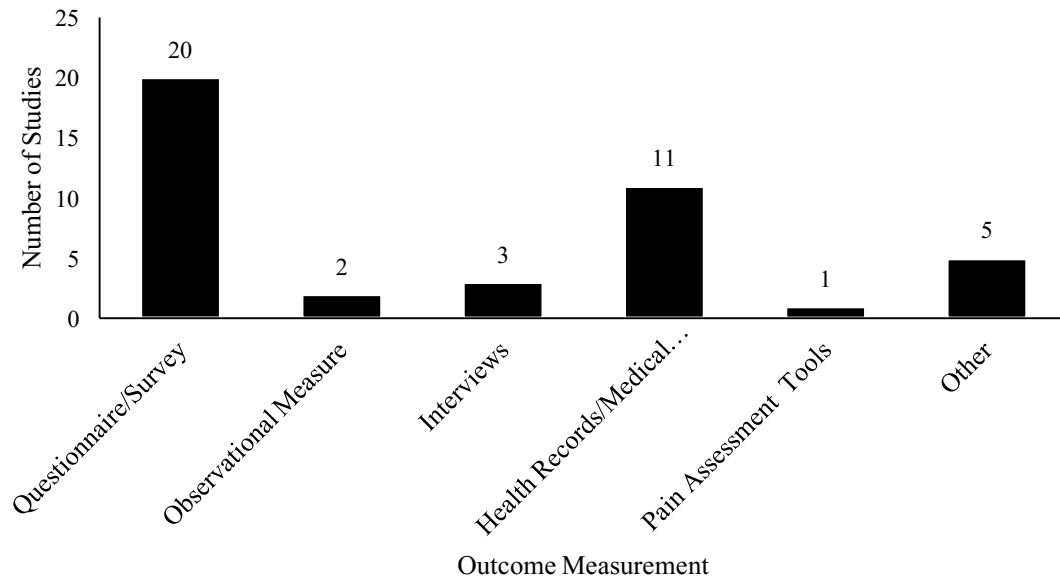


Figure 2.4. Type of outcome measurement used across quantitative studies.

Note: No identified studies met inclusion criteria for the “Focus Group” extraction option.

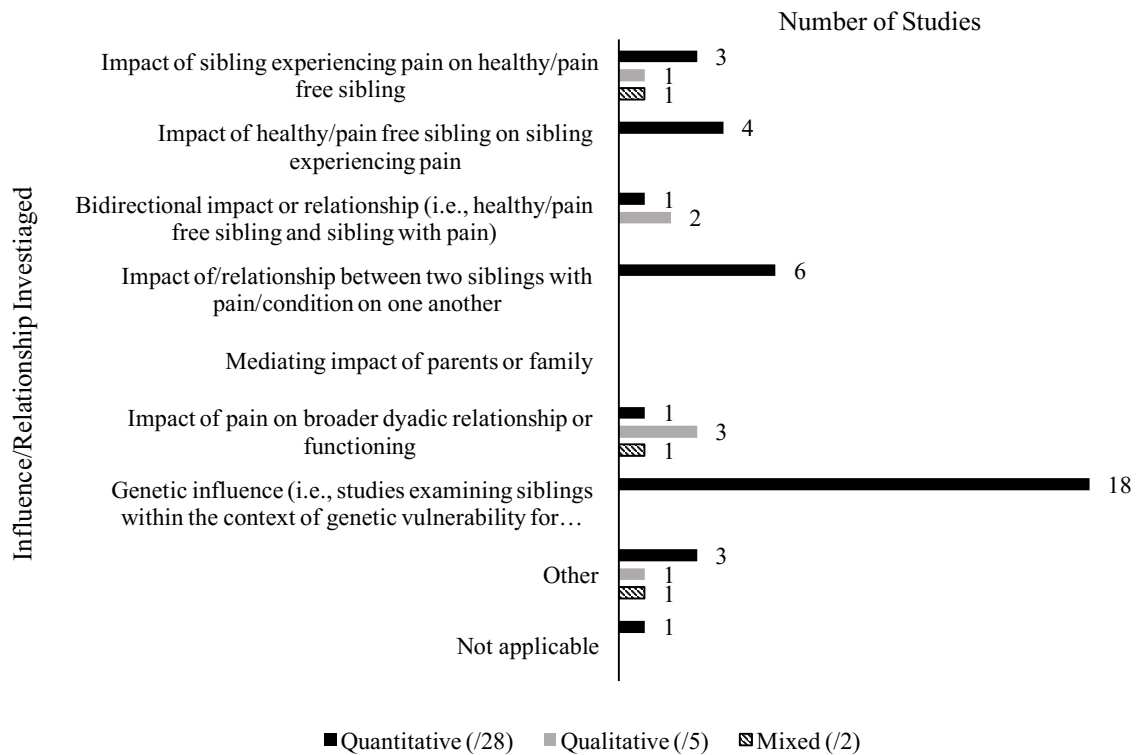


Figure 2.5. The influence/relationship investigated regarding siblings in pediatric pain by methodology type.

2.9 Tables

Table 2.1

Type of Study and Methodology Used Across the Included Studies

Study	Study Type			Methodology	
	Published research study	Dissertation	Conference Abstract	Qualitative	Quantitative
(Badiee et al., 2014)	X				X
(Barton et al., 2013)			X		X
(Britton & Moore, 2002)	X			X	
(M L Campbell-Yeo et al., 2012)	X				X
(Campbell et al., 2009)			X		X
Marsha L Campbell-Yeo et al., 2014)	X				X
(Champion et al., 2012)	X				X
(Champion et al., 2013)			X		X
(Chan et al., 2013)			X		X
(El-Metwally et al., 2008)	X				X
(J. J. Field et al., 2008)	X				X
(Filocamo et al., 2011)			X		X
(Flynn et al., 2010)			X		X
(Gordon, 2013)		X		X	
(Gorodzinsky et al., 2013)	X			X	
(Guite et al., 2007)	X				X
(Gunalan et al., 2012)			X		X
(Kofman et al., 2013)			X		X
(Lee et al., 2012)			X		X
(McOmber & Shulman, 2009)			X		X
(Mikkelsson, Kaprio, Salminen, Pulkkinen, & Rose, 2001)	X				X
(Miller et al., 2015)	X				X

Study	Study Type			Methodology	
	Published research study	Dissertation	Conference Abstract	Qualitative	Quantitative
(M. Moroldo et al., 1997)	X				X
(M. B. Moroldo et al., 2004)	X				X
(Moscato et al., 2009)			X	X	
(Prahald et al., 2000)	X				X
(Säilä et al., 2001)	X				X
(Scherder et al., 2008)	X				X
(Ståhl et al., 2013)	X				X
(Svensson et al., 1999)	X				X
(Valkenburg et al., 2015)	X			X	X
(Wong et al., 2011)			X		X
(Wutzke, 1999)		X		X	X
(Akobeng et al., 1999)	X			X	
(Engstrom, 1992)	X				X

Table 2.2

Type of Pain Examined Across Included Studies by Methodology Type

Pain Type	Number of Studies		
	Quantitative (/28)	Qualitative (/5)	Mixed (/2)
Acute/procedural	4	0	0
Needle/Immunization	0		
Blood Draw	4		
Post-operative	0		
Other	1		
Chronic/Disease Related	24	5	1
Chronic Pain	0	0	0
Irritable Bowel	1	0	0
Disease/Syndrome			
Inflammatory Bowel Disease	1	2	0
Migraine/Headache	2	2	0
Juvenile Arthritis/Rheumatic	6	1	1
Diseases			
Abdominal Pain	3	1	0
Back Pain	3	0	0
Musculoskeletal	2	1	0
Growing Pains	4	0	0
Sickle Cell Disease	2	0	0
Other	1	1	0
Experimental	1	0	1
Cold Pressor	0		0
Quantitative Sensory Testing	0		1
Water Load Task	0		0
Other	1		0

Table 2.3

Sources of Information for Outcome Assessment Across Quantitative Studies

Informant	Number of Studies	% Within Category
Parent	14	
Self-report		43
Reporting on others (e.g., children)		93
Behaviour observed		0
Healthy/Pain Free Sibling	8	
Self-report		100
Reporting on others (e.g., sib w/ pain)		0
Behaviour observed		0
Sibling(s) with Pain/Condition	11	
Self-report		82
Reporting on others (e.g., healthy sib)		0
Behaviour observed		18
Health Professional	3	
Self-report		0
Reporting on others (e.g., children)		100
Behaviour observed		0
Other	1	

2.10 Supplemental Materials

Compete Search Terms

Sibling Terms	Pain Terms	Child Terms
<ul style="list-style-type: none"> • Siblings • Sibling • Twins • Twin • Sister • Brother • Multiple Birth Offspring 	<p>Chronic Pain Terms (Higgins et al., 2015)</p> <ul style="list-style-type: none"> • Pain* • Fibromyalgia • Irritable bowel syndrome • Arthrit* • Osteoarthritis* • Headache* • Migraine* • Neuralgi* • Neuropath* • Complex regional pain syndrome • Chronic Pain • Arthritis • Osteoarthritis • Headache • Migraine • Neuralgia • Peripheral Nervous System Diseases <p>Experimental Pain Terms (Boerner et al., 2014)</p> <ul style="list-style-type: none"> • experimental pain • cold pressor • quantitative sensory test • water load • heat pain • thermal pain • pressure pain 	<p>Leclercq et al. (2013)</p> <ul style="list-style-type: none"> • Infan* • newborn* • new-born* • perinat* • neonat* • baby • baby* • babies • toddler* • minors • minors* • boy • boys • boyfriend • boyhood • girl* • kid • kids • child • child* • children* • schoolchild* • schoolchild • school child • adolescen*

	<ul style="list-style-type: none"> • exercise task <p>Procedural Pain Terms</p> <ul style="list-style-type: none"> • Needle • Surgery • Puncture • Operation • Blood draw 	<ul style="list-style-type: none"> • juvenil* • youth* • teen* • under*age* • pubescen* • pediatrics • pediatric* • paediatric* • peadiatric* • school • school* • prematur* • preterm*
<p>Pub Med search format:</p> <p>"siblings"[MeSH] OR sibling*[tw] OR "Twins"[MeSH] OR Twin*[tw] OR sister*[tw] OR brother*[tw] OR "Multiple Birth Offspring"[MeSH]</p>	<p>Pub Med search format:</p> <p>Pain*[tiab] OR Fibromyalgia[tiab] OR Irritable bowel syndrome[tiab] OR Arthrit*[tiab] OR Osteoarthritis*[tiab] OR Headache*[tiab] OR Migraine*[tiab] OR Neuralgi*[tiab] OR Neuropath*[tiab] OR Complex regional pain syndrome[tiab] OR Pain[MeSH:NoExp] OR Chronic Pain[MeSH] OR Fibromyalgia[MeSH:NoExp] OR Irritable Bowel Syndrome[MeSH:NoExp] OR Arthritis[MeSH:NoExp] OR Osteoarthritis[MeSH:NoExp] OR Headache[MeSH:NoExp] OR Migraine[MeSH:NoExp] OR Neuralgia[MeSH:NoExp] OR Peripheral Nervous System Diseases[MeSH:NoExp] OR Complex Regional Pain Syndromes[MeSH:NoExp] OR Needle[tiab] OR Surgery[tiab] OR Puncture[tiab] OR Operation[tiab] OR Blood draw[tiab] OR experimental</p>	<p>Pub Med Search Format:</p> <p>Infan* OR newborn* OR new-born* OR perinat* OR neonat* OR baby OR baby* OR babies OR toddler* OR minors OR minors* OR boy OR boys OR boyfriend OR boyhood OR girl* OR kid OR kids OR child OR child* OR children* OR schoolchild* OR schoolchild OR school child[tiab] OR school child*[tiab] OR adolescen* OR juvenil* OR youth* OR teen* OR under*age* OR pubescen* OR pediatrics[MeSH] OR pediatric* OR paediatric* OR peadiatric* OR school[tiab] OR school*[tiab] OR prematur* OR preterm*</p>

	pain[tiab] OR cold pressor[tiab] OR quantitative sensory test[tiab] OR water load[tiab] OR heat pain[tiab] OR thermal pain[tiab] OR pressure pain[tiab] OR exercise task[tiab]	
<p>Cinahl search format:</p> <p>MH "siblings" OR TX sibling* OR MH "Twins" OR TX Twin* OR TX sister* OR TX brother* OR MH "Multiple Birth Offspring"</p>	<p>Cinahl search format:</p> <p>TI Pain* OR AB Pain* OR TI Fibromyalgia OR AB Fibromyalgia OR TI Irritable bowel syndrome OR AB Irritable bowel syndrome OR TI Arthrit* OR AB Arthrit* OR TI Osteoarthritis* OR AB Osteoarthritis* OR TI Headache* OR AB Headache* OR TI Migraine* OR AB Migraine* OR TI Neuralgi* OR AB Neuralgi* OR TI Neuropath* OR AB Neuropath* OR TI Complex regional pain syndrome OR AB Complex regional pain syndrome OR MH Pain OR MH Chronic Pain OR MH Fibromyalgia OR MH Irritable Bowel Syndrome OR MH Arthritis OR MH Osteoarthritis OR MH Headache OR MH Migraine OR MH Neuralgia OR MH Peripheral Nervous System Diseases OR MH Complex Regional Pain Syndromes OR TI Needle OR AB Needle OR TI Surgery OR AB Surgery OR TI Puncture OR AB Puncture OR TI Operation OR AB Operation OR TI Blood draw OR AB Blood draw OR TI experimental pain OR AB experimental pain OR TI cold pressor OR AB cold pressor OR TI quantitative sensory test OR AB quantitative sensory test OR TI water load OR AB water load OR TI heat pain OR AB heat pain OR TI thermal pain OR AB thermal pain OR TI pressure pain OR AB pressure pain OR TI exercise task OR AB exercise task</p>	<p>Cinahl search format:</p> <p>Infan* OR newborn* OR new-born* OR perinat* OR neonat* OR baby OR baby* OR babies OR toddler* OR minors OR minors* OR boy OR boys OR boyfriend OR boyhood OR girl* OR kid OR kids OR child OR child* OR children* OR schoolchild* OR schoolchild OR TI school child OR AB school child OR TI school child* OR AB school child* OR adolescen* OR juvenil* OR youth* OR teen* OR under*age* OR pubescen* OR MH pediatrics OR pediatric* OR paediatric* OR peditric* OR TI school OR AB school OR TI school* OR AB school* OR prematur* OR preterm*</p>

<p>PsycINFO search format:</p> <p>DE "siblings" OR TX sibling* OR DE "Twins" OR TX Twin* OR TX sister* OR TX brother* OR DE "Multiple Birth Offspring"</p>	<p>PsycINFO search format:</p> <p>TI Pain* OR AB Pain* OR TI Fibromyalgia OR AB Fibromyalgia OR TI Irritable bowel syndrome OR AB Irritable bowel syndrome OR TI Arthrit* OR AB Arthrit* OR TI Osteoarthritis* OR AB Osteoarthritis* OR TI Headache* OR AB Headache* OR TI Migraine* OR AB Migraine* OR TI Neuralgi* OR AB Neuralgi* OR TI Neuropath* OR AB Neuropath* OR TI Complex regional pain syndrome OR AB Complex regional pain syndrome OR DE Pain OR DE Chronic Pain OR DE Fibromyalgia OR DE Irritable Bowel Syndrome OR DE Arthritis OR DE Osteoarthritis OR DE Headache OR DE Migraine OR DE Neuralgia OR DE Peripheral Nervous System Diseases OR DE Complex Regional Pain Syndromes OR TI Needle OR AB Needle OR TI Surgery OR AB Surgery OR TI Puncture OR AB Puncture OR TI Operation OR AB Operation OR TI Blood draw OR AB Blood draw OR TI experimental pain OR AB experimental pain OR TI cold pressor OR AB cold pressor OR TI quantitative sensory test OR AB quantitative sensory test OR TI water load OR AB water load OR TI heat pain OR AB heat pain OR TI thermal pain OR AB thermal pain OR TI pressure pain OR AB pressure pain OR TI exercise task OR AB exercise task</p>	<p>PsycINFO search format:</p> <p>Infan* OR newborn* OR new-born* OR perinat* OR neonat* OR baby OR baby* OR babies OR toddler* OR minors OR minors* OR boy OR boys OR boyfriend OR boyhood OR girl* OR kid OR kids OR child OR child* OR children* OR schoolchild* OR schoolchild OR TI school child OR AB school child OR TI school child* OR AB school child* OR adolescen* OR juvenil* OR youth* OR teen* OR under*age* OR pubescen* OR DE pediatrics OR pediatric* OR paediatric* OR peadiatric* OR TI school OR AB school OR TI school* OR AB school* OR prematur* OR preterm*</p>
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<p>Embase Search Format:</p> <p>"siblings"/exp OR sibling* OR "Twins"/exp OR Twin* OR sister* OR brother* OR "Multiple Birth Offspring"/exp</p>	<p>Embase Search Format:</p> <p>Pain*:ti,ab OR Fibromyalgia:ti,ab OR “Irritable bowel syndrome”:ti,ab OR Arthrit*:ti,ab OR Osteoarthritis*:ti,ab OR Headache*:ti,ab OR Migraine*:ti,ab OR Neuralgi*:ti,ab OR Neuropath*:ti,ab OR “Complex regional pain syndrome”:ti,ab OR ‘Pain’/exp OR ‘Chronic Pain’/exp OR ‘Fibromyalgia’/exp OR ‘Irritable Bowel Syndrome’/exp OR ‘Arthritis’/exp OR ‘Osteoarthritis’/exp OR ‘Headache’/exp OR ‘Migraine’/exp OR ‘Neuralgia’/exp OR ‘Peripheral Nervous System Diseases’/exp OR ‘Complex Regional Pain Syndromes’/exp OR Needle:ti,ab OR Surgery:ti,ab OR Puncture:ti,ab OR Operation:ti,ab OR “Blood draw”:ti,ab OR “experimental pain”:ti,ab OR “cold pressor”:ti,ab OR “quantitative sensory test”:ti,ab OR “water load”:ti,ab OR “heat pain”:ti,ab OR “thermal pain”:ti,ab OR “pressure pain”:ti,ab OR “exercise task”:ti,ab</p>	<p>Embase Search Format:</p> <p>Infan* OR newborn* OR new-born* OR perinat* OR neonat* OR baby OR baby* OR babies OR toddler* OR minors OR minors* OR boy OR boys OR boyfriend OR boyhood OR girl* OR kid OR kids OR child OR child* OR children* OR schoolchild* OR schoolchild OR “school child”:ti,ab OR “school child*”:ti,ab OR adolescen* OR juvenil* OR youth* OR teen* OR under* NEXT/1 age* OR pubescen* OR ‘pediatrics’/exp OR pediatric* OR paediatric* OR peadiatric* OR school:ti,ab OR school*:ti,ab OR prematur* OR preterm*</p>
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<p>Web of Science Search Format:</p> <p>"siblings" OR sibling* OR "Twins" OR Twin* OR sister* OR brother* OR "Multiple Birth Offspring"</p>	<p>Web of Science Search Format:</p> <p>Pain* OR Fibromyalgia OR Irritable bowel syndrome OR Arthrit* OR Osteoarthritis* OR Headache* OR Migraine* OR Neuralgi* OR Neuropath* OR Complex regional pain syndrome OR Pain OR Chronic Pain OR Fibromyalgia OR Irritable Bowel Syndrome OR Arthritis OR Osteoarthritis OR Headache OR Migraine OR Neuralgia OR Peripheral Nervous System Diseases OR Complex Regional Pain Syndromes OR Needle OR Surgery OR Puncture OR Operation OR Blood draw OR experimental pain OR cold pressor OR quantitative sensory test OR water load OR heat pain OR thermal pain OR pressure pain OR exercise task</p>	<p>Web of Science Search Format:</p> <p>Infan* OR newborn* OR new-born* OR perinat* OR neonat* OR baby OR baby* OR babies OR toddler* OR minors OR minors* OR boy OR boys OR boyfriend OR boyhood OR girl* OR kid OR kids OR child OR child* OR children* OR schoolchild* OR schoolchild OR school child OR school child* OR adolescen* OR juvenil* OR youth* OR teen* OR under*age* OR pubescen* OR pediatrics OR pediatric* OR paediatric* OR peadiatric* OR school OR school* OR prematur* OR preterm*</p>
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Table Representing Descriptive Information for Included Studies

	Number of Studies
Publication Year	
1992	1
1997	1
1999	3
2000	1
2001	2
2002	1
2004	1
2007	1
2008	3
2009	3
2010	1
2011	2
2012	4
2013	7
2014	2
2015	2
Country	
Australia	10
Canada	5
Finland	4
Ghana	1
Iran	1
Italy	1
Sweden	2
The Netherlands	2
United Kingdom	2
United States	8
Unknown	2
Discipline	
Medicine	19
Psychiatry	5
Psychology	7
Nursing	2
Physiotherapy	1
Other	14
Not Listed	10

Note: For “Discipline”, no studies were extracted as “Genetics” or “Occupational Therapy”.

**CHAPTER 3: A DYADIC ANALYSIS OF SIBLINGS' RELATIONSHIP
QUALITY, BEHAVIOURAL RESPONSES, AND PAIN EXPERIENCES DURING
EXPERIMENTAL PAIN**

The manuscript based on this study is presented below. Meghan Schinkel, under the supervision of Dr. Christine Chambers, was responsible for developing the research questions, methodology and analytic approach, obtaining ethical approval, and applying for funding. She developed the study protocol, contributed substantially to data collection and relevant coding, and oversaw staff and volunteers who were involved with the study. She was responsible for conducting analyses, and writing the current manuscript. The manuscript was reviewed by the co-authors, and their feedback incorporated, prior to submission. The manuscript was submitted to *PAIN* on August 16, 2017, reviews were received on October 5, 2017, and the manuscript was resubmitted on January 1, 2018. The current reference for this manuscript is:

Schinkel, M.G., Chambers, C.T., Corkum, P., & Jacques, S. (under review). A Dyadic Analysis of Siblings' Relationship Quality, Behavioural Responses, and Pain Experiences during Experimental Pain. *PAIN*.

3.1 Abstract

Research on family factors in paediatric pain has primarily focused on parents; the role of siblings has been largely ignored. This study examined whether sibling relationship quality was related to siblings' behaviours during experimental pain, and whether the behaviours of an observing sibling were related to children's pain outcomes. Ninety-two sibling dyads between 8-12 years old completed both observational and questionnaire measures of sibling relationship quality. Children took turns completing the cold pressor task (CPT) in a counterbalanced order with their sibling present. Pain outcomes (intensity, fear, tolerance) were recorded for each sibling, and the behaviour of the observing and participating siblings during the CPT were coded as attending, non-attending, and coping/encouragement. Structural equation modelling, using the actor-partner interdependence model, was conducted to analyse the dyadic data. While participating in the CPT with their sibling present, greater levels of warmth and positivity in the sibling relationship were related to children engaging in more non-attending behaviours and less attending behaviours. Greater levels of attending behaviours by the observing child was related to the sibling having a lower pain tolerance, and greater levels of coping/encouragement behaviours by the observing child was related to the sibling reporting greater pain intensity and fear during the CPT. Children with warmer/positive sibling relationships were more likely to respond to acute pain by shifting the focus away from their pain experience (e.g., through distraction) when a sibling was present. Pain-focused behaviours by an observing sibling are related to greater child pain and fear during experimental pain.

Key words: siblings, family factors, pediatric pain, experimental pain, dyadic analysis

3.2 Introduction

Approximately 80% of individuals have a sibling (Dunn, 2000), and siblings are important in each other's lives. Family systems theory highlights the importance of considering the context of family relationships, and conceptualizes members as having a bi-directional influence on one another (Bavelas & Segal, 1982). Reviews have documented the difficulties experienced by siblings of children with chronic health issues (e.g., mental health, quality of life, somatic symptoms) (Knecht et al., 2015; Limbers & Skipper, 2014; Vermaes et al., 2012). In the field of paediatric pain, extensive work has focused on the role of family factors (K. Birnie, Boerner, et al., 2014; Palermo & Chambers, 2005; Palermo et al., 2014). However, this research has primarily concentrated on parents, not siblings.

Siblings are influential in children's development across a number of domains (Brody, 2004; McHale, Updegraff, & Whiteman, 2012 offer reviews). Development of emotion and behaviour regulation skills are posited to be central components of positive child sibling relationships (Kramer, 2010). A longitudinal study found that highly positive sibling relationships minimized child internalizing difficulties following acute life stressors, and this was true irrespective of the age difference and sex combination of siblings (Gass, Jenkins, & Dunn, 2007). Sibling relationship quality or sibling behaviours may influence how healthy children emotionally and behaviourally respond to the potentially distressing experience of acute pain, although no known studies have explored this.

Studies that have examined family functioning and parent behaviours as they relate to children's acute pain experiences are relevant to understanding the potential

influence of siblings' relationship quality and behaviours. One study found that less family cohesion predicted higher levels of symptom complaints, while more cohesion was related to greater "other talk" (e.g., non-pain focused talk) by children completing the cold pressor with their parent present, providing some support for their predicted association between poorer family functioning and greater child talk attending to the pain experience (Kathryn A. Birnie et al., 2017). Parent behaviours that draw attention to a child's pain experience, such as discussing symptoms or reassurance, are related to children expressing or reporting more pain in experimental contexts (Moon et al., 2011; Schinkel et al., 2017; Walker et al., 2006), and more fear during clinical pain (Manimala et al., 2000). Conversely, parent behaviours that draw attention away from children's pain experiences, such as distraction or humour, are related to children expressing less experimental (Moon et al., 2011; Walker et al., 2006) and clinical (Manimala et al., 2000; McCarthy et al., 2010) pain or distress.

Drawing from the family and parent research, the current study sought to examine siblings during paediatric acute pain. The study used a controlled experimental pain task (von Baeyer et al., 2005), a dyadic approach to data analysis (Cook & Kenny, 2005), and built on gaps in sibling research by using self-report and observational measures (Alderfer et al., 2010; Noller, 2005) completed by both children (Alderfer et al., 2010; Knecht et al., 2015). The objectives were to: 1) examine relations between sibling relationship quality and the behaviours children engage in while observing their sibling's pain and experiencing pain themselves during the cold pressor task; 2) examine relations between an observing sibling's behaviour and a child's pain outcomes. It was hypothesised that: 1) greater positivity/warmth in the sibling relationship would relate to

siblings engaging in more behaviours that remove attention from the pain/task (e.g., distraction), while more negative relationships would relate to more pain/task-focused behaviours (e.g., symptom talk) by siblings; 2) pain/task-focused behaviours would relate to poorer pain outcomes, whereas behaviours removing attention from the pain/task would relate to better pain outcomes.

3.3 Methods

The current chapter is focused on sibling relationship quality, and children's behaviours and pain outcomes during the cold pressor task. A second, separate brief report using the same sample describes findings regarding parent perspectives on sibling presence at paediatric medical appointments (see Chapter 4).

3.3.1 Participants

The study included a sample of 92 sibling dyads between the ages of 8–12-years-old, and one of their parents/guardians (80 women, 12 men). The older siblings (identified based on parent report) had a mean age of 10.84 years ($SD = 1.05$; 44 boys, 48 girls) and the younger siblings had a mean age of 8.73 years ($SD = 0.88$; 51 boys, 41 girls). The majority of children were biological siblings ($n = 86$), although step-siblings ($n = 3$), half-siblings ($n = 2$), and adoptive siblings ($n = 1$) also participated. In terms of sibling dyad composition, the sample included 23 dyads composed of two sisters ($n = 4$ twin girls), 26 of two brothers, 21 of an older sister and younger brother, 17 of an older brother and younger sister, and 5 sets of boy/girl twins. Most children were identified by their parents as being White (older sibling: $n = 79$, younger sibling: $n = 77$), followed by Biracial (older sibling: $n = 5$, younger sibling: $n = 5$), Asian (older sibling: $n = 1$, younger sibling: $n = 2$), Arab (older sibling: $n = 2$, younger sibling: $n = 2$), Black (older sibling: n

= 2, younger sibling: $n = 2$), Native/Aboriginal (older sibling: $n = 1$, younger sibling: $n = 2$), or other (older sibling: $n = 2$, younger sibling: $n = 2$). Most parents reported that the siblings lived together 100% of the time ($n = 89$; $n = 2$ reported 90%, and $n = 1$ reported 50%), and parents reported a mean of 2.87 ($SD = 1.14$, range = 2 - 7) children in their immediate family.

Parents had a mean age of 40.83 years ($SD = 4.26$; range = 29-55), and most reported being married (77.20%). Over half of the parents reported having completed a university degree or graduate school/professional training (66.30%). Reported annual household income ranged from \$10 - \$25,000 (2.20%) to greater than \$150,000 (22.80%). Most parents identified as White ($n = 82$), followed by Asian ($n = 2$), Arab ($n = 2$), Black ($n = 2$), other ($n = 2$), Native/Aboriginal ($n = 1$), and Biracial ($n = 1$). Most parents reported that the family normally spoke English at home ($n = 89$; French: $n = 2$; other: $n = 1$).

Participants were recruited from the community using advertisements and social media (e.g., Facebook), and by contacting potential participants from the research centre's and the hospital's research registries. To be eligible for the study, both siblings had to be between the ages of 8-12-years-old, and could be either biological siblings, including identical or fraternal twins, step or half-siblings, or adoptive siblings, and needed to live together at least 50% of the time. Children had to be fluent in English, typically developing, generally healthy, and not have any hearing or vision impairments that were not corrected for by glasses or hearing aids. Further, children could not have any conditions that could increase their risk for an adverse outcome during the cold pressor task (e.g., cardiovascular disease) (von Baeyer et al., 2005), and could not have

previously participated in a cold pressor task study. These criteria were evaluated by parent report during recruitment screening. For a parent/guardian to be eligible for the study they had to be a primary caregiver to both participating children (biological, step, or adoptive mother or father, or legal guardian), lived with both children at least 50% of the time, and be fluent in English. Of the 95 families enrolled in the study, three were excluded from analyses due to not speaking English during the study tasks ($n = 1$) and research assistant error ($n = 2$).

3.3.2 Procedure

The study took place at the IWK Health Centre, and was approved by the Research Ethics Board. Participation involved one visit to the research centre. Informed consent and assent were obtained from parents and children respectively at the outset of the study. Parents were in a separate room from their children for the duration of the study, and completed a demographics questionnaire and a measure of their children's sibling relationship quality. Children completed a tower-building task and the cold pressor task in a counterbalanced order. Following these tasks, children were placed in separate rooms and completed a measure of their sibling relationship quality with a research assistant present. At the end of the study, parents and children were brought back together for debriefing. Children were each provided with a certificate and \$20.00, and parents were given \$15.00 to thank them for participating in the study and to cover transportation costs.

Tower-building task.

The children completed a tower-building task originally developed by Iturralde and colleagues for use with siblings designed to capture normal sibling interactions

(Iturralde et al., 2013). The children were provided with a set of everyday non-sturdy craft materials (e.g., paper, tape, straws, rubber bands), and were instructed to build a tower as tall as they could. They were told that they would have 15 minutes to complete their tower, and that their tower would be compared to other siblings who had completed the task. Children were provided a warning when they had 2 minutes left. The siblings were video and audio recorded, and their behaviour was coded to provide an observational measure of sibling relationship quality. Significant associations between coded relationship quality during this task and self-reported relationship quality have been documented in siblings aged 8-17, lending support for the validity of this task (Iturralde et al., 2013). Similar associations were found in the current study, and the results are reported below.

Cold pressor task.

The children took turns completing the cold pressor task (CPT) in a counterbalanced order (older vs. younger sibling) with their sibling present. Siblings were not informed of the order until immediately before the CPT. The cold pressor task is frequently used, and allows researchers to investigate pain in children in an ethical and standardized manner that lends itself particularly well to the examination of family factors and interactions (K A Birnie, Caes, et al., 2014; Kathryn A Birnie et al., 2011). As recommended, children were left alone during completion of the CPT, although they were monitored through a live video feed to ensure their safety (K A Birnie, Caes, et al., 2014). During the CPT children were instructed to, when told by a research assistant over an intercom, submerge their non-dominant hand into the water up to the place where their wrist bends. They were told to keep their hand in the water for as long as they could, even

if it was uncomfortable, until it became too uncomfortable or hurt too much. If a child had not removed their hand after 4 minutes, they were instructed to do so by a research assistant over an intercom, but were not informed of the time limit in advance. The water was maintained at $10^{\circ}\text{C} \pm 1^{\circ}\text{C}$. This procedure is in keeping with published guidelines for use of the CPT with children (von Baeyer et al., 2005). Observing siblings were seated across from the child participating in the CPT, and were not given direct instructions regarding how to interact with one another. The task was video and audio recorded, and the behaviour of the siblings was subsequently coded.

Immediately following each CPT, the child who completed the CPT was asked to provide ratings of their pain intensity (“most” pain) and pain-related fear. Children were turned away from their sibling when they provided these ratings, and a research assistant distracted the sibling who had been observing to minimize their influence on children’s ratings. The procedure was repeated in the same manner for the child completing the CPT second.

3.3.3 Measures

Self-reported sibling relationship quality.

In order to assess the quality of the children’s sibling relationship, each child and the parent completed respective versions of the *Sibling Relationship Questionnaire – Revised* (SRQ-R) (Furman & Buhrmester, 1985). This measure was rated as well-established in a review of family measures in paediatric psychology, suggesting acceptable psychometrics (Alderfer et al., 2008). Forty-eight items were rated on a Likert-type scale from “Hardly at all” (1) to “Extremely Much” (5). Because some participating families were composed of more than two children, parents and children

were reminded to complete the measure based on the children/sibling that was participating in the study with them. The composite scores for warmth/closeness and conflict were of interest in the current study. Following recommended scoring procedures (Furman & Buhrmester, 1985; Furman, personal communication), a composite score for warmth/closeness was derived by averaging seven scale scores (each calculated by averaging 3 items) for intimacy, prosocial behaviour, companionship, similarity, admiration of and by siblings, and affection (i.e., a total of 21 items). A composite score for conflict was derived in the same manner, and was composed of quarrelling, antagonism, and competition (i.e., a total of 9 items). Higher scores represent higher levels of the composite. As recommended, if a participant skipped more than one item on a scale, the scale score was treated as a missing value (Furman, personal communication). In the case of a missing scale score, the composite score was calculated as the average of the completed scales ($n = 1$ parent, $n = 2$ older siblings, $n = 2$ younger siblings).

Behaviour coded sibling relationship quality during the tower-building task.

The tower task videos were observed and coded using *Noldus The Observer XT Version 10* (“Noldus the Observer XT,” 2010). A revised version of the coding system developed by Iturralde and colleagues (2013) for use with the tower-building task was used in the current study to provide an observational measure of sibling relationship quality, and is available from the corresponding author. The use of the overall dyadic positivity and negativity variables, and the respective codes composing these variables, were consistent with Iturralde and colleagues (2013). However, because detailed information regarding the coding system was not available, the information available was expanded on to create operational definitions for the codes, and specific examples were

added. The process for revising the coding system followed the steps outlined by Chorney and colleagues (J. M. Chorney et al., 2015) and was informed by a review of relevant literature, viewing a sample of videos, and discussion amongst the coders and co-authors.

Sibling positivity was composed of four codes (Iturralde et al., 2013): 1) Validation: comments/behaviours that were meant to be reinforcing (e.g., “that’s a good idea”), 2) Engagement: the child’s engagement in the task (e.g., “this is fun”, task-oriented behaviours), 3) Warmth: demonstrations of affection or enjoyment of the sibling (e.g., “you’re the best”), and 4) Cooperation: comments/behaviours indicating the siblings were working together (e.g., “let’s both fold the straws”). Sibling negativity was composed of three codes (Iturralde et al., 2013): 1) Criticism of Sibling: comments/behaviours that were critical of the sibling themselves (e.g., “you’re annoying”), 2) Criticism of Task Ideas or Behaviour: criticisms of the siblings’ task-related ideas/behaviours (e.g., “that’s a dumb idea”), and 3) Conflict: verbal or physical conflict between the siblings (e.g., “I’m telling Mom if you keep doing that”). A score was provided for each sibling for the validation, engagement, warmth, criticism of sibling, and criticism of task ideas/behaviour codes, and a score for the dyad was provided for the cooperation and conflict codes.

Although a global coding approach in which behaviours were rated on a scale based on frequency and intensity was used by Iturralde and colleagues (2013), a presence/absence coding approach was used in the current study as it was found to be more reliable. Codes were scored as present or absent across 5-minute intervals during the 15-minute task. Iturralde and colleagues (2013) rated codes at 5-minute intervals, and

it was felt to be an appropriate interval length given the frequency and duration of the coded behaviours (Bakeman & Gottman, 1997; J. M. Chorney et al., 2015). The videos were viewed three times (Iturralde et al., 2013): on the first pass, the behaviours were just observed without providing scores, on the second pass, the behaviour of the older sibling was coded, and on the third pass the behaviour of the younger sibling and the dyadic behaviours were coded. A total score was calculated for each sibling dyad for positivity and negativity by summing the relevant codes across the intervals.

All participants were coded by the first author, and a second research assistant reliability coded approximately 20% of the participants (randomly identified) at regular intervals (i.e., approximately every 10 participants) (J. M. Chorney et al., 2015). Percent agreement is a useful measure of inter-rater reliability because reliability can be inferred exactly from the values, and is appropriate to use when the probability of rater guessing is low (McHugh, 2012). Hence, given that coder training was informed by published guidelines (J. M. Chorney et al., 2015), minimizing the probability of guessing, percent agreement was deemed an acceptable measure of inter-rater reliability (McHugh, 2012). Mean percent agreement was 87.04% for sibling positivity, and 77.04% for sibling negativity, suggesting acceptable reliability (Stemler, 2004).

Consistent with the approach used by Iturralde and colleagues (2013), to investigate the convergent validity of the coding system, correlations were examined between parent, older sibling, and younger sibling self-reported relationship quality and observed sibling positivity and negativity. The only significant relation discovered was between observed negativity and younger sibling reported warmth/closeness, $r = -.23$, $p < .05$, providing some support for the validity. However, as suggested by Iturralde and

colleagues, the questionnaire and tower building task may in part assess different aspects of sibling relationship quality (Iturralde et al., 2013). Additionally, the content validity is supported by the process taken to revise the coding system described above (J. M. Chorney et al., 2015).

Pain outcome measures reported during the cold pressor task.

Children rated their “most” pain during the CPT using the *Faces Pain Scale-Revised* (FPS-R) (“Faces Pain Scale-Revised,” 2001; Hicks et al., 2001). The FPS-R is a valid measure of pain intensity for children ages 4-12-years-old (Hicks et al., 2001). The scale is composed of 6 faces representing “no pain” (0) to “very much pain” (10). Children were asked to point to the face that showed how much pain or hurt they felt when it hurt the most when their hand was in the water.

Children were also asked to rate their pain-related fear during the CPT using the *Children’s Fear Scale* (CFS) (McMurtry et al., 2011). This measure was found to be reliable and valid in a sample of children aged 5-10-years-old (McMurtry et al., 2011). The scale is made up of 5 faces ranging from “not scared at all” (0) to “the most scared possible” (4). Children were asked to choose the face that showed how scared they were during the CPT.

Pain tolerance was scored as the duration of time in seconds each child left their hand submerged in the water during the CPT, up to a maximum of 240 seconds.

Coded behaviours during the cold pressor task.

The cold pressor videos were observed and coded using *Noldus The Observer XT Version 10* (“Noldus the Observer XT,” 2010). A coding system was used in the current study to capture the behaviours of both the sibling observing and the sibling participating

in the cold pressor task, and is available from the corresponding author. The coding system was modified from existing coding systems for parent and child behaviours during the cold pressor task (Moon et al., 2011; Schinkel et al., 2017), and a subsequent version modified to capture adolescent friendship interactions (Forgeron et al., 2017). It should be noted that these manuals were themselves informed by the Child-Adult Medical Procedure Interaction Scale Revised (R. Blount et al., 1997) and/or Short Form (R. L. Blount, Bunke, Cohen, & Forbes, 2001), which have been considered well-established and approaching well-established respectively, based on their psychometric properties (Bai et al., 2017). Due to its relevance to coding siblings, behaviours identified in the tower-building task coding system (Iturralde et al., 2013), as well as knowledge of the relevant literature and viewing a sample of observations, were also used to inform the development of operational definitions and examples in the current coding system. The modification process to develop the current coding system followed the process described by Chorney and colleagues (2015).

The behaviour of the child observing their sibling completing the CPT was coded across 12 categories of behaviour, and the behaviour of the child completing the CPT was coded across 10 categories, including humour, procedure related talk/attending behaviours and symptom focused talk and behaviours (see Tables 1 and 2 for a list of behaviours with examples). Behaviours were coded for both the first and second cold pressors each sibling dyad completed. Behaviours were coded as present or absent across 5-second intervals throughout the duration of the cold pressor (i.e., the last coded interval was the interval in which the child removed their hand from the water). An interval of 5-seconds was identified as most appropriate based on the frequency and duration of the

observed behaviours (Bakeman & Gottman, 1997; J. M. Chorney et al., 2015). In addition, it has been previously successfully used when coding behaviour during the CPT (Schinkel et al., 2017).

Similar to the tower-building task, the videos were viewed three times with behaviours just being watched on the first pass. The behaviours of the observing sibling were coded on the second pass, and the behaviours of the participating sibling were coded on the third pass. A proportion score was calculated for each behaviour (intervals rated present/total intervals coded), given the variability in the length of time of the CPT across participants (see Tables 3 and 4 for descriptive statistics). For analyses, behaviours were grouped into attending behaviours, non-attending behaviours, and coping/encouragement by calculating a mean proportion score of the relevant sub-codes (see Tables 1 and 2 for the sub-codes composing each behaviour group). The grouping of sub-codes was informed by previous studies examining behaviour during the CPT (Kathryn A. Birnie, Chambers, Chorney, Fernandez, & McGrath, 2016; Moon et al., 2011), and modified for the current coding system.

All participants were coded by one research assistant, and coder training was again informed by recommended guidelines (J. M. Chorney et al., 2015). Reliability coding was completed by the first author on approximately 20% of randomly identified participant videos, at regular intervals throughout the coding process (i.e., approximately every 10 participants) (J. M. Chorney et al., 2015). Because some behaviours were frequently coded as absent, Kappa was used as a more conservative estimate of inter-rater reliability for this system (Stemler, 2004). Kappa coefficients ranged from .84-1.0 for

coding of the observing sibling, and .82-1.0 for coding of the participating sibling, suggesting acceptable reliability (Bakeman & Gottman, 1997; Landis & Koch, 1977).

3.3.4 Analyses

Structural equation modelling was used to test the primary hypotheses. The analyses on self-reported sibling relationship quality and CPT behaviours, and observer CPT behaviours and pain outcomes were examined with the actor-partner interdependence model (APIM), using structural equation modelling (Cook & Kenny, 2005). The advantage of this model is that it accounts for the non-independence of the dyadic sibling data (Cook & Kenny, 2005). The APIM examines actor effects (i.e., the relation between two different variables for the same individual in a dyad) and partner effects (i.e., the relation between a variable for one individual and a different variable for the other dyad member) (see Figure 1) (Cook & Kenny, 2005). In the current study, an example of an actor effect would be a significant relation between a child's own self-reported sibling relationship quality and their own behaviours during the CPT, whereas an example of a partner effect would be a significant relation between a child's self-reported sibling relationship quality and their sibling's behaviours during the CPT. Dyads were treated as distinguishable, and MLR estimation was used to account for any non-normality. Dyads were distinguished based on the order in which the child participated in the CPT, such that "Sibling 1" was the child who completed the CPT first (and observed second), and "Sibling 2" was the child who completed the CPT second (and observed first) (see Figure 2).

3.4 Results

3.4.1 Order Effects

Although appropriate counterbalancing was used thereby controlling for any systematic variation due to order effects (A. Field, 2009), task order was explored to see if any differences existed. The impact of task order (i.e., cold pressor vs. tower-building task first) and cold pressor order (Sibling 1 vs. Sibling 2) on cold pressor pain outcomes (i.e., pain intensity, fear and tolerance) and coded behaviours (i.e., attending, non-attending, and coping/encouragement) was examined using a series of 2 (Between: task order) x 2 (Within: cold pressor order) mixed Analyses of Variance (ANOVA). A significant interaction between task and cold pressor order was discovered for coping/encouragement behaviour by the observing sibling, $F(1,90) = 4.19, p < .05$. However, follow up paired samples t tests revealed no significant differences between the first and second observing sibling's coping/encouragement behaviours when they completed the cold pressor task first, or the tower-build task first (although this difference was close to reaching significance, $p = .05$, and likely driving the interaction). No other significant main effects or interactions were discovered, suggesting that the order of study tasks did not significantly influence any cold pressor pain outcome or behaviour scores.

Independent samples t tests were conducted to examine the impact of task order (i.e., cold pressor vs. tower-building task first) on siblings' coded dyadic scores for positivity and negativity during the tower-building task. Although no significant differences were discovered for negativity scores, a significant difference between siblings' positivity score was discovered, $t(90) = 2.14, p < .05$, with siblings who completed the cold pressor task first having higher coded positivity ($M = 14.09, SD = 2.53$) than siblings who completed the tower-building task first ($M = 12.89, SD = 2.83$).

3.4.2 Objective #1: Relations Between Sibling Relationship Quality and Siblings' CPT Behaviours

Child reported sibling relationship quality.

The results of the APIM analyses examining relations between children's self-reported sibling relationship quality (i.e., warmth/closeness and conflict composite scores based on the *Sibling Relationship Questionnaire – Revised* (Furman & Buhrmester, 1985); see Table 5 for descriptive statistics) and coded observing and participating behaviours during the CPT (i.e., proportions of attending behaviours, non-attending behaviours, and coping/encouragement) are presented here (see Table 7). A significant actor effect was discovered for the first sibling's reported warmth/closeness and their own participating non-attending behaviours; greater levels of warmth/closeness in the sibling relationship reported by the sibling who participated in the CPT first was related to them engaging in greater proportions of non-attending behaviours themselves while completing the CPT. A marginally significant partner effect was found for the first sibling's reported conflict and the second sibling's participating coping/encouragement behaviours; greater levels of conflict in the sibling relationship reported by the first child was related to their sibling (i.e., the sibling who participated in the CPT second) engaging in lower proportions of coping/encouragement behaviours while they were completing the CPT. No other significant actor or partner effects were discovered.

Observed sibling relationship quality.

The results of the structural equation modeling analyses examining relations between siblings' observed relationship quality (i.e., dyadic scores for coded positivity and negativity during the tower-building task; see Table 5 for descriptive statistics) and

coded observing and participating behaviours during the CPT (i.e., proportions of attending behaviours, non-attending behaviours, and coping/encouragement) are presented here (see Table 8). Greater levels of observed positivity were significantly related to the sibling who participated in the CPT second engaging in lower proportions of attending behaviours, and greater proportions of non-attending behaviours while completing the CPT. Further, greater levels of observed negativity were significantly related to the sibling who participated in the CPT first engaging in greater proportions of attending behaviours, and the sibling who participated in the CPT second engaging in lower proportions of attending behaviours while completing the CPT. No other significant effects were discovered.

3.4.3 Objective #2: The Relation Between The Observing Sibling's CPT Behaviours and The Participating Sibling's Pain Outcomes

The results of the APIM analyses examining relations between the children's coded behaviours while observing the CPT (i.e., proportions of attending behaviours, non-attending behaviours, and coping/encouragement) and the participating sibling's pain outcomes (i.e., self-reported pain intensity scores ["most" pain] based on the FPS-R ("Faces Pain Scale-Revised," 2001; Hicks et al., 2001), self-reported pain-related fear scores based on the CFS (McMurtry et al., 2011), and pain tolerance scores; see Table 6 for descriptive statistics) are presented here (see Table 9). Significant negative partner effects were discovered for both observing siblings' proportions of attending behaviours and their participating siblings' pain tolerance scores; greater attending behaviours by the child who was observing their sibling do the CPT was related to lower pain tolerance scores for their participating sibling, and this effect was significant in both the first and

second cold pressors that sibling dyads completed. A significant partner effect was found for the second sibling's observing non-attending behaviours and the first sibling's pain-related fear; greater proportions of non-attending behaviours by the second sibling (i.e., the sibling who observed the CPT first) while watching their sibling do the CPT was related to greater pain-related fear ratings reported by their participating sibling (i.e., the sibling who completed the CPT first). Significant partner effects were also discovered such that greater proportions of coping/encouragement behaviours by the second sibling (i.e., the first observer) while watching their sibling do the CPT were related to higher pain intensity and pain-related fear ratings reported by their participating sibling (i.e., the sibling who completed the CPT first). Significant actor effects were discovered for the observing sibling's coping and encouragement; greater proportions of coping/encouragement behaviours by the second sibling (i.e., the first observer) while observing the CPT was related to their own higher self-reported pain intensity ratings, and greater proportions of coping/encouragement behaviours by the first sibling (i.e., the second observer) while observing was related to their own higher self-reported pain-related fear ratings. No other significant actor or partner effects were discovered.

3.4.4 Post-hoc Analyses

Sex and Age

Post-hoc analyses were conducted to investigate whether sibling sex and the age difference between sibling pairs predicted the behaviours children engaged in during the cold pressor task. A series of multiple regression analyses were conducted with sibling 1 sex, sibling 2 sex and sibling age difference entered together as predictor variables, and the coded behaviours for the first and second cold pressor as the respective dependent

variables (i.e., observing and participating attending, non-attending, and coping/encouragement behaviours). The model was significant for the first sibling's participating attending behaviours, adjusted $R^2 = .084$, $F(3,88) = 3.76$, $p < .05$. Sibling 1 sex (i.e., the child's own sex) was the only significant predictor, *standardized* $\beta = -.34$, $p < .01$. An examination of the means suggest that boys who participated in the CPT first engaged in a greater proportion of attending behaviours ($M = .47$, $SD = .12$) while completing the CPT than girls who participated in the CPT first ($M = .37$, $SD = .15$). The model was also significant for the first sibling's participating non-attending behaviours, adjusted $R^2 = .12$, $F(3,88) = 5.02$, $p < .01$. Sibling 1 sex (i.e., the child's own sex) was the only significant predictor, *standardized* $\beta = .39$, $p < .001$. An examination of the means suggest that girls who participated in the CPT first engaged in a greater proportion of non-attending behaviours ($M = .083$, $SD = .095$) while completing the CPT than boys who participated in the CPT first ($M = .024$, $SD = .048$). The model was not significant for any of the other behaviours investigated.

Sibling Validation and CPT Behaviours

Given the purported importance of validation in the pain context (Edmond & Keefe, 2015), exploratory correlational analyses were conducted between the validation sub-code and the CPT behaviours that were identified in the structural equation modelling analyses as being significantly related to sibling positivity (see above). Although validation was not significantly related to the second sibling's participating attending behaviours, it was significantly related to the second sibling's participating non-attending behaviours, $r = .30$, $p < .01$.

3.5 Discussion

The current study examined whether sibling relationship quality was related to siblings' behaviour proportion scores during the cold pressor task. As hypothesised, greater observed positivity during the tower task was related to the second sibling engaging in more non-attending behaviours while participating in the CPT, and greater levels of the first sibling's self-reported warmth/closeness was related to themselves engaging in more non-attending behaviours while completing the CPT with their sibling present. Further, more observed positivity during the tower task was related to the second sibling engaging in less attending behaviours while completing the CPT. Therefore, positive/warmer sibling relationships related to children engaging in less pain-oriented behaviours, and more behaviours that shifted the focus away from their pain (e.g., distraction). Children with better sibling relationships may engage in natural conversation and humour more often, and thus be more likely to use "non-attending behaviours" as a potentially helpful strategy while in pain with their sibling present. Gass and colleagues similarly suggested that positive sibling relationships may reduce the negative impacts of acute life stressors through promoting the use of distraction amongst siblings during times of stress (Gass et al., 2007).

Greater levels of observed negativity were significantly related to the first sibling engaging in more attending behaviours, and the second sibling engaging in less attending behaviours while participating in the CPT. The effect was as expected for the first sibling; when a child is experiencing pain with a sibling with whom they have a more negative relationship, they may be more likely to attend inward to their current experience. However, the effect was not as hypothesised for the sibling who completed the CPT second. Siblings who have more negative relationships may respond in different ways to

the same experience. Supporting this interpretation, Whiteman, McHale and Crouter (Whiteman et al., 2007) found that adolescent siblings who try to be different from each other have significantly poorer relationship intimacy than siblings who try to be similar.

Sibling relationship quality only significantly related to children's behaviours while participating in the CPT, not while observing. The children's experience as a "participant" may have been more salient than their experience as an observer, and thus more likely to relate to their relationship quality. This result is similar to Birnie and colleagues' finding that family functioning was only significantly related to children's behaviours during the cold pressor, not parents' observing behaviours (Kathryn A. Birnie et al., 2017). Some family factors may have stronger relations with pain behaviours than observer behaviours during acute pain.

The relation between behaviour proportion scores of an observing sibling and children's pain outcomes were also investigated. Greater attending behaviours by the child who observed was related to their sibling having lower pain tolerance scores, and this partner effect was significant for both siblings. Further, greater coping and encouragement behaviours by the second sibling (i.e., first observer) while observing was related to their sibling reporting higher pain intensity and pain-related fear. These findings are consistent with the hypothesis that behaviours focused on the pain/task would be related to poorer pain outcomes, and with studies examining associations between parent behaviours and child pain outcomes (e.g., Manimala et al., 2000; Moon et al., 2011; Schinkel et al., 2017; Walker et al., 2006). Although the findings regarding "coping/encouragement" behaviours may seem counterintuitive, these behaviours draw attention to the pain experience. In some studies procedure related praise is indeed

considered an attending behaviour (Kathryn A. Birnie et al., 2016; Moon et al., 2011). Pain-focused behaviours themselves may be relevant to poorer pain outcomes for children irrespective of who (parent, sibling) engages in them.

Greater proportions of non-attending behaviours by the second sibling (i.e., first observer) while observing their sibling during the CPT was significantly related to sibling reports of greater fear. This finding is contrary to the hypothesis, and is inconsistent with studies looking at parent behaviour (e.g., Manimala et al., 2000; McCarthy et al., 2010; Moon et al., 2011; Walker et al., 2006). One interpretation is that non-attending behaviours relate to children's pain outcomes differently when it is a sibling who engages in them rather than a parent. It is likely that most sibling relationships are more equal in terms of power relative to a parent-child relationship, and this difference in roles may be particularly salient during expressions of emotions. Children may not have distracted with the intent of reducing their sibling's fear, and their non-attending behaviours may not have been interpreted by the sibling in pain as an act of support. Further, children may not have engaged in these behaviours effectively (McCarthy et al., 2010). Given that this finding represents a relation and directionality cannot be assumed, observing children may also have responded to their sibling's higher levels of fear by attempting to draw their attention away from the pain experience.

More significant partner effects for the relation between observing sibling behaviours and pain outcomes were found for the first cold pressor (i.e., the behaviours of the first observing sibling relating to pain outcomes of the first participating sibling). Both children's lack of experience with the task may have made the observing sibling's behaviour more likely to relate to their sibling's pain outcomes. However, statistical path

comparisons of the partner effects for the first and second cold pressor were not conducted, so this observed difference should be interpreted with caution.

Although hypotheses were not made regarding actor effects for relations between sibling observing behaviours and their own pain outcomes, significant findings emerged in a manner consistent with the observed partner effects. Greater proportions of coping/encouragement behaviours by the second child while observing was related to their own higher pain intensity ratings, and greater proportions of coping/encouragement by the first sibling while observing was related to their own higher fear ratings. Children who endorse higher pain and fear may be more likely to engage in coping and encouragement behaviours while observing their sibling in pain, perhaps to regulate their own emotions.

More research is needed to support clinical recommendations regarding sibling behaviour during child pain. However, the current findings indicate that clinicians should view present siblings as active members of a child's pain experience. Parents should be aware that their children's sibling relationship quality may influence how they respond to acute pain if a sibling is present. The current findings suggest that, if siblings have a positive relationship, a child may be more inclined to engage in behaviours that remove attention from their pain experience (e.g., distraction). Similar to parent advice, the results tentatively suggest that present siblings should be encouraged to minimize engaging in behaviours that bring attention to the pain experience. If research can also identify sibling behaviours that relate to reductions in child pain, siblings have the potential to contribute to paediatric pain management. Incorporating siblings into pain

management plans could educate children on strategies for managing their own and others' pain.

There are several limitations that should be noted. The results represent associations between the variables of interest, and thus causation or directionality of the effects cannot be assumed. Further, many effects were significant for only one sibling, or one combination, in the dyad. Therefore, possibly other, more critical, sibling/behaviour related variables were not captured in the current study. Although, as described earlier, considerable efforts were taken to minimize bias, behavioural coding was conducted by individuals who were aware of the study objectives. Further, the tower-building task coding system was modified from the original version to maximize reliability. The cold pressor represents a controlled and ethical method for studying pain in children (Kathryn A Birnie et al., 2011), and is well suited for investigating family interactions (K A Birnie, Caes, et al., 2014). However, its applicability to daily or clinical pain remains understudied (K A Birnie, Caes, et al., 2014). Lastly, although analyzing the data in disaggregated form was considered most appropriate, the consequent greater analyses may have heightened the probability of a Type 1 error (Holmbeck, Li, Schurman, Friedman, & Coakley, 2002).

Continued research on siblings in paediatric pain is needed to build on the current limitations, and address novel questions. Using sequential coding and analytic approaches (J. M. Chorney et al., 2015) could elucidate richer information such as the sequence of siblings' behaviours or differences in timing across behaviours; factors relating to this (e.g., pain tolerance) could be examined. Future sibling research could explore other potentially important variables, such as child temperament or disposition (Harper, Penner,

Peterson, Albrecht, & Taub, 2012) and pain catastrophizing (Kathryn A. Birnie et al., 2016; Vervoort et al., 2011). Exploring sibling validation in the pain context will also be an important area for future research (Edmond & Keefe, 2015). Post-hoc analyses in the current study only identified a child's own sex (i.e., not their sibling's sex) as a significant predictor of CPT behaviour. However, future research should continue to explore differences between various compositions of siblings (e.g., same versus opposite sex). Comparing cold pressor behaviours of healthy siblings and sibling dyads in which one has chronic pain may provide insight into the impact of chronic pain on sibling interactions and pain responses. Further, studies could examine how sibling behaviours influence a child's pain outcomes in a clinical context (e.g., immunizations). Lastly, task order impacted siblings' positivity during the tower-building task, with siblings who completed the CPT first demonstrating greater positivity than siblings who completed the tower-building task first. The pain task may have promoted a sense of empathy amongst siblings, thus enhancing positivity. Future work could explore the impact of undergoing painful experiences with a sibling on sibling empathy or resiliency.

Siblings have been vastly understudied despite their potential contribution to children's clinical pain management. The current findings establish the relevance of sibling relationship quality and behaviours in paediatric acute pain and, concurrent with the broader developmental research (Brody, 2004; McHale et al., 2012), demonstrate how siblings can exert both positive and negative influences on one another. The current study expands our conceptualization of the importance of family factors in children's pain experiences (Palermo & Chambers, 2005; Palermo et al., 2014), and highlights the

potential role siblings may play in how children learn to interpret and respond to acute pain experiences across development.

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3.8 Figures

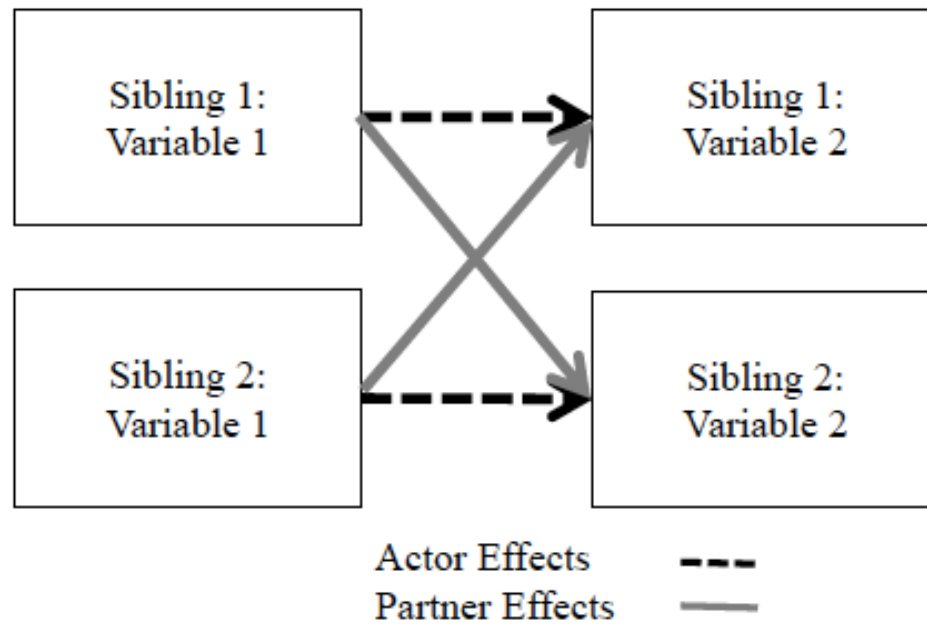


Figure 3.1. General depiction of the actor-partner interdependence model for sibling dyads (Cook & Kenny, 2005).

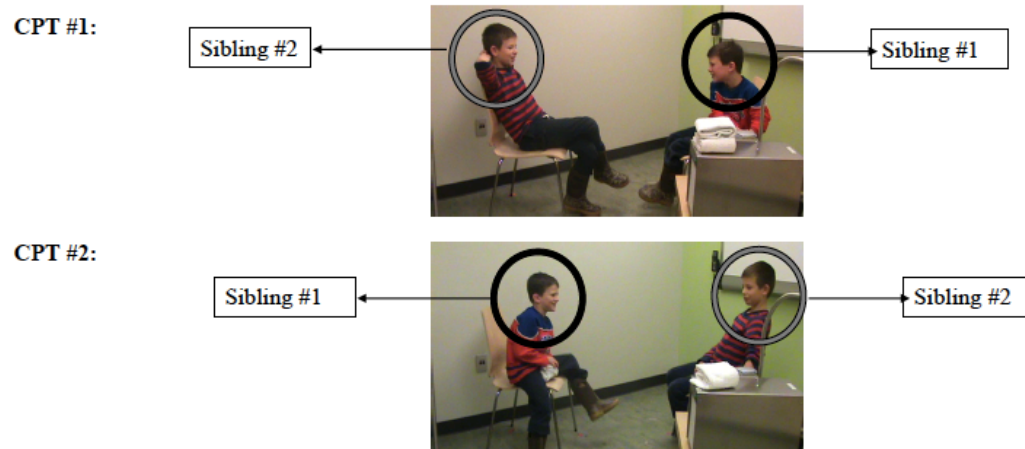


Figure 3.2. Illustration of how sibling dyads were distinguished for analyses. The siblings pictured here are completing the cold pressor task. Consent was obtained for use of these photos.

3.9 Tables

Table 3.1

Behaviour Codes and Examples for the Sibling Observing the CPT

Grouping	Behaviour	Example
Attending	Sympathy, comfort & reassurance	“Don’t worry, you’re going to be fine”; holds sibling’s hand
	Procedure related talk/attending behaviours	“The water looks like it’s bubbling”
	Symptom focused talk & behaviour	“How bad does it hurt?”
	Symptom & coping related self-reflections and predictions	“I bet it’s going to hurt a lot when I do it”
Non-attending	Distraction/Non-procedure related talk	“Lets ask Mom if we can get pizza for dinner”
	Humour	Child tells a joke
Coping/ Encouragement	Coping commands & behaviours	“Move your fingers around to keep them warm”
	Procedure related praise/encouragement	“You’re doing such a good job”
Other	Competition/light hearted teasing	“Lets see who can do it longest”
	Criticism & conflict	“You suck at this”
	Fidgeting & restlessness	Tapping feet; swinging legs
	Other	“Huh?”

Table 3.2

Behaviour Codes and Examples for the Sibling Participating in the CPT

Grouping	Behaviour	Example
Attending	Procedure related talk/attending behaviours	“The machine is noisy”
	Symptom focused talk & behaviour	“It’s freezing cold”
	Resistance	“I can’t do this any more”
Non-attending	Distraction/Non-procedure related talk	“Play a game with me”
	Humour	Child makes a silly face
Coping/ Encouragement	Coping talk, requests & behaviours	“I’m keeping my hand really still so I can’t feel the cold as much”
	Self procedure & symptom related praise/encouragement/positive reframing	“I’m being braver than I thought I would”
Other	Competition/light hearted teasing	“I bet that you won’t last as long as me when you do it”
	Criticism & conflict	“Stop it, you’re going to get us in trouble”
	Other	“Hmm”

Table 3.3

Descriptive Summary of the Observing Sibling's Behaviours During the CPT

Behaviour	1 st CPT			2 nd CPT		
	Mean Proportion	Standard Deviation	Range	Mean Proportion	Standard Deviation	Range
Sympathy, comfort & reassurance	.05	.13	.00 - 1.00	.06	.15	.00 - 1.00
Procedure related talk/attending behaviours	.27	.28	.00 - 1.00	.33	.29	.00 - 1.00
Symptom focused talk & behaviour	.18	.17	.00 - .75	.12	.14	.00 - .50
Symptom & coping related self-reflections and predictions	.02	.07	.00 - .54	.06	.13	.00 - .75
Distraction/Non-procedure related talk	.10	.18	.00 - .89	.07	.15	.00 - .80
Humour	.17	.24	.00 - 1.00	.17	.23	.00 - 1.00
Coping commands & behaviours	.01	.05	.00 - .33	.02	.06	.00 - .37
Procedure related praise/encouragement	.01	.05	.00 - .40	.02	.06	.00 - .37
Competition/light hearted teasing	.01	.03	.00 - .14	.04	.09	.00 - .33
Criticism & conflict	.02	.07	.00 - .42	.03	.07	.00 - .33
Fidgeting & restlessness	.20	.25	.00 - 1.00	.21	.30	.00 - 1.00
Other	.04	.09	.00 - .67	.05	.09	.00 - .44

Note. Proportion scores were calculated for each behaviour (intervals rated present/total intervals coded).

Table 3.4

Descriptive Summary of the Participating Sibling's Behaviours During the CPT

Behaviour	1 st CPT			2 nd CPT		
	Mean Proportion	Standard Deviation	Range	Mean Proportion	Standard Deviation	Range
Procedure related talk/attending behaviours	.81	.22	.20 - 1.00	.78	.22	.10 - 1.00
Symptom focused talk & behaviour	.44	.28	.00 - 1.00	.44	.30	.00 - 1.00
Resistance	.02	.06	.00 - .33	.02	.05	.00 - .25
Distraction/Non-procedure related talk	.05	.13	.00 - .67	.06	.11	.00 - .58
Humour	.05	.10	.00 - .50	.06	.11	.00 - .50
Coping talk, requests & behaviours	.03	.10	.00 - .75	.03	.10	.00 - .75
Self procedure & symptom related praise/encouragement/positive reframing	.03	.08	.00 - .37	.04	.09	.00 - .50
Competition/light hearted teasing	.00	.01	.00 - .10	.01	.04	.00 - .25
Criticism & conflict	.01	.02	.00 - .12	.01	.04	.00 - .35
Other	.02	.04	.00 - .22	.03	.06	.00 - .33

Note. Proportion scores were calculated for each behaviour (intervals rated present/total intervals coded).

Table 3.5

Descriptive Summary of Reported and Observed Sibling Relationship Quality

Sibling Relationship Quality Measure	Mean	Standard Deviation	Range
Reported Warmth/Closeness			
Parent	3.46	0.60	1.90 – 4.52
Older sibling	3.29	0.68	1.57 – 4.90
Younger sibling	3.14	0.77	1.19 – 4.81
Reported Conflict			
Parent	2.81	0.71	1.00 – 4.78
Older sibling	3.03	0.78	1.33 – 4.56
Younger sibling	2.84	0.87	1.00 – 4.89
Observed Positivity	13.49	2.73	8.00 – 19.00
Observed Negativity	5.76	2.78	0.00 – 13.00

Note. Scores for reported warmth/closeness and conflict, based on the *Sibling Relationship Questionnaire – Revised* (Furman & Buhrmester, 1985), could range from 1 – 5 with higher scores indicating higher levels of the composite. Scores could range from 0 – 21 for observed positivity, and from 0 – 15 for observed negativity based on coding during the tower-building task, with higher scores indicating higher levels of the construct.

Table 3.6

Descriptive Summary of Cold Pressor Pain Outcomes

Pain Outcome	Mean	Standard Deviation	Range
1 st CPT			
Pain Intensity	4.72	2.53	0 - 10
Pain-related Fear	0.75	0.85	0 - 4
Tolerance	89.04	87.75	8 - 240
2 nd CPT			
Pain Intensity	5.00	2.52	0 - 10
Pain-related Fear	0.64	0.81	0 - 3
Tolerance	81.68	86.45	9 - 240

Note. Scores could range from 0 - 10 for pain intensity (FPS-R (“Faces Pain Scale-Revised,” 2001; Hicks et al., 2001)), from 0 – 4 for pain-related fear (CFS (McMurtry et al., 2011)), and from 0 – 240 for pain tolerance.

Table 3.7

Actor and Partner Effects of Child Self-reported Sibling Relationship Quality and Siblings' Coded Observing and Participating CPT Behaviours

		Observing Attending	Observing Non- attending	Observing Coping/ Encouragement	Participating Attending	Participating Non- attending	Participating Coping/ Encouragement
SRQ-R Warmth/ Closeness							
	Actor						
	Sib 1– Sib 1	.010	.130	.113	-.201	.227*	.084
	Sib 2– Sib 2	.096	-.135	.112	-.054	.081	-.096
	Partner						
	Sib 1–Sib 2	-.238	.168	.036	-.037	-.033	.098
	Sib 2–Sib 1	-.046	-.009	-.091	-.101	.018	-.020
SRQ-R Conflict							
	Actor						
	Sib 1–Sib 1	-.026	.063	.086	.030	-.121	-.127
	Sib 2–Sib 2	-.078	.096	.024	-.010	.103	.020
	Partner						
	Sib 1–Sib 2	.106	.134	.082	.009	.104	-.141+
	Sib 2–Sib 1	.097	.085	.089	.098	.051	.049

Statistics represent standardized regression coefficients (b^*), and indicate the direction of the effect (i.e., negative numbers represent negative relationships between the variables of interest);

* $p < .05$

+ $p = .05$

Table 3.8

Effects of Observed Sibling Relationship Quality During the Tower-building Task and Siblings' Coded Observing and Participating CPT Behaviours

		Observing Attending	Observing Non-attending	Observing Coping/ Encouragement	Participating Attending	Participating Non-attending	Participating Coping/ Encouragement
Observed Positivity	Sibling 1	.009	.167	-.063	-.092	.137	.079
	Sibling 2	.031	.123	.104	-.218*	.309**	.042
Observed Negativity	Sibling 1	.051	-.008	.182	.240*	.008	-.087
	Sibling 2	.182	-.076	.070	-.227*	.145	.079

Statistics represent standardized regression coefficients (b^*), and indicate the direction of the effect (i.e., negative numbers represent negative relationships between the variables of interest).

* $p < .05$

** $p < .01$

Table 3.9

Actor and Partner Effects of the Observing Sibling's Coded CPT Behaviours and the Participating Sibling's Pain Outcomes

		Pain Intensity	Pain-related Fear	Pain Tolerance
Observing Attending				
	Actor			
	Sib 1– Sib 1	.024	.104	.008
	Sib 2– Sib 2	.000	.076	-.121
	Partner			
	Sib 1–Sib 2	.053	.105	-.151*
	Sib 2–Sib 1	-.147	.007	-.258**
Observing Non-attending				
	Actor			
	Sib 1– Sib 1	.101	-.146	.032
	Sib 2– Sib 2	.147	.128	-.146
	Partner			
	Sib 1–Sib 2	.075	-.011	.079
	Sib 2–Sib 1	.041	.250*	.068
Observing Coping/ Encouragement				
	Actor			
	Sib 1– Sib 1	.119	.217*	-.051
	Sib 2– Sib 2	.190*	-.090	-.019
	Partner			
	Sib 1–Sib 2	.126	.044	-.059
	Sib 2–Sib 1	.270**	.174*	-.044

Statistics represent standardized regression coefficients (b^*), and indicate the direction of the effect (i.e., negative numbers represent negative relationships between the variables of interest)

* $p < .05$

** $p < .01$

CHAPTER 4: PARENTS' PERSPECTIVES ON SIBLING PRESENCE AT PEDIATRIC MEDICAL APPOINTMENTS

The manuscript based on this study is presented below. Meghan Schinkel, under the supervision of Dr. Christine Chambers, was responsible for developing the study questions, methodology, and protocol, and obtained ethical approval and applied for study funding. She contributed substantially to data collection and coding, and oversaw staff and volunteers who were involved with the study. She was responsible for developing the analytic plan, conducting analyses, and writing the current manuscript. The study co-authors (Chambers, C.T., Corkum, P., & Jacques, S) reviewed the manuscript, and their feedback was incorporated. This manuscript will be submitted for publication.

4.1 Abstract

The study examined sibling presence at pediatric medical appointments. Ninety-five parents of 8-12-year-old siblings completed questionnaires examining sibling presence at medical appointments, and sibling and parent-child relationship quality. Almost all (97.9%) parents reported having brought siblings to medical appointments, and most often cited convenience as their reason (83.7%). Parents frequently reported allowing their children to decide which sibling would receive an appointment/procedure first (53.3%). Parent-child conflict and closeness were related to parents' reasons for bringing siblings to medical appointments, and child age was related to parents' responses for how they decide the order in which their children receive appointments/procedures.

4.2 Introduction

Having a sibling is a common (Dunn, 2000) and impactful experience for children and youth (Brody, 2004; McHale et al., 2012). When a sibling has a chronic illness, this can affect children's well-being, such as increasing susceptibility for psychological issues (Vermaes, van Susante, & van Bakel, 2012) and academic struggles (Gan et al., 2017). Yet siblings may also exert influences on healthy children's more routine medical experiences, such as regular check-ups and vaccinations. Understanding more about sibling presence in various medical contexts is important, as research with adults suggests that the presence of companions has both beneficial and detrimental influences on medical appointments (Laisaar-Powell et al., 2013).

Based on suggested immunization schedules, receiving a needle is at minimum a yearly occurrence for children, beginning in infancy (Centres for Disease Control and Prevention, 2017). Further, studies examining children and youth's access of health care services have reported yearly visit rates of 2.1 and 3.0 for physician appointments, and 0.15 and 0.29 for emergency visits (Chen & Escarce, 2006; Manos, Cui, MacDonald, Parker, & Dummer, 2014). Because parents often accompany children to medical appointments (Brown, Brett, Stewart, & Marshall, 1998), parents may also bring siblings along. Indeed, one study that examined accompaniment to family doctor appointments found that 10.5% of individuals who were accompanied came with a sibling (Brown et al., 1998). However, this sample included both children and adults.

Children are unique from adults and research focused on sibling presence specifically for pediatric settings is warranted. Unlike adults, sibling companions who are children likely have less control over their attendance and factors related to their

involvement in medical appointments, as parents are responsible for making these decisions. It is possible that parents' reasons for bringing siblings (e.g., necessity versus choice) could influence parents' and siblings' behaviour during pediatric appointments. Further, children may be more likely to simultaneously play the roles of both sibling companion and patient within the same appointment (e.g., when both receive influenza vaccinations). This may leave parents faced with having to make decisions regarding which child should receive the appointment/procedure first. It is possible that parents' decisions regarding sibling order could positively or negatively influence child responses to a medical procedure. Given its potential to influence a child's health care experience, understanding parents' reasons and variables that may be related to their decisions around sibling presence and order of procedure is important.

There is evidence that families are more involved in activities with one another when familial relationships, including parent-child and youth's sibling relationships, are viewed as more positive (Crouter, Head, Mchale, & Tucker, 2004). Sibling sex and age are also related to children's active involvement in each other's lives (Cole & Kerns, 2001). Of particular relevance to the current study, engagement in companion activities (e.g., games, recreational activities) decreased with child age, and was higher for male sibling dyads (Cole & Kerns, 2001). Therefore, family relationship quality, child age and child sex may be relevant variables to explore within the context of sibling involvement at pediatric medical appointments.

The objective of the current study was to determine the extent to which parents bring siblings to various pediatric medical appointments, the reasons that parents provide regarding why they bring siblings to medical appointments, and how parents decide

which child receives an appointment/procedure first. It was hypothesized that most parents would report bringing siblings together to medical appointments. The relation between children's age, sex, and sibling and parent-child relationship quality, and parents' responses for why they bring siblings to medical appointments and sibling order for appointments/procedures were also examined. It was hypothesized that more positive relationships, younger child age, and male sex of a child would be related to parents being more likely to cite purposeful reasons for bringing siblings to appointments (e.g., feeling it was more convenient, their children would be supportive/positive influence) and deciding which child goes first (e.g., referencing the personality/characteristics of their children), and less likely to cite passive reasons (e.g., the appointments being booked together, allowing health professional/clinic to decide which sibling goes first). The opposite was hypothesized for more negative relationships, older child age and female sex of a child.

4.3 Methods

The participants reported on here were recruited as part of the study reported in Chapter 3 (note: the discrepancy in participant number reflects the three participants excluded from analyses in Chapter 3 remaining eligible for the current study). The current manuscript presents results pertaining to sibling presence at medical appointments. The second manuscript from this same data set, reported in Chapter 3, focuses on siblings' participation in an experimental pain task, and describes associations between siblings' relationship quality, cold pressor behaviours and pain outcomes.

4.3.1 Participants

The study included a sample of 95 primary caregivers (83 female, 12 male) of two siblings ages 8-12 years old. Participants had a mean age of 40.67 years ($SD = 4.28$; range = 29-55), and identified as White ($n = 84$), Asian ($n = 3$), Arab ($n = 2$), Black ($n = 2$), Native/Aboriginal ($n = 1$), Biracial ($n = 1$), and Other ($n = 2$). The majority reported being married ($n = 74$), followed by divorced/separated ($n = 13$), common law ($n = 4$), never married ($n = 3$), and remarried ($n = 1$). Most participants reported having a University or Graduate School/Professional Training degree ($n = 63$). The range of participants reported annual household income was between \$10 – \$25,000 ($n = 2$) to greater than \$150,000 ($n = 22$). Regarding the participants' children who were reported on in the study, older children had a mean age of 10.86 years ($SD = 1.05$, range = 8 - 12; 48 girls, 47 boys), and younger children had a mean age of 8.73 years ($SD = .88$, range = 8 - 11; 42 girls, 53 boys). In the case of twins ($n = 9$), parents were asked to identify which child was older based on who was born first. Although only two children from each family were included in the present study, participants reported a mean of 2.88 ($SD = 1.15$, range = 2 - 7) children in their immediate family.

Community recruitment strategies used included advertisements, social media (e.g., Facebook) and available research registries. To be eligible to participate, participants had to read, write, and speak fluently in English, and be a primary caregiver (biological, step, or adoptive mother or father, or legal guardian) to at least two generally healthy siblings between the ages of 8-12 years old. Further, they needed to live with both children at least 50% of the time. Their children could be biological siblings (identical/fraternal twins included), step or half-siblings, or adoptive siblings, who lived

together at least 50% of the time (see Chapter 3 for further details on the eligibility requirements of the children).

4.3.2 Procedure

This study was approved by the IWK Health Centre Research Ethics Board. Participants came to the research centre for a single visit, and provided informed consent. Participants completed a demographics measure, which included questions regarding sibling presence at medical appointments, and measures of their perception of their children's sibling relationship quality and their own relationship with each of their children. Participants were reminded to complete these measures based on their two children who were participating in the larger study. Their children participated in other study activities in a different room while participants completed these measures. Following study completion, participants received a \$15.00 honourarium for their participation. See Chapter 3 for a description of the full study procedures.

4.3.3 Measures

Sibling presence at medical appointments.

As part of a demographics measure, participants were asked questions regarding sibling presence at medical appointments. These items were developed for the purposes of the current study. First, participants were asked, "Have you ever brought more than one child in your family to a single medical appointment (i.e., brought siblings to one of your children's appointments, or booked appointments/procedures for your children together)?" If yes, participants were asked to indicate the situations in which they had brought more than one of their children (i.e., siblings together) from a list composed of: needles (e.g., immunizations, flu shot), blood work, doctor's appointment (e.g., check-up

or specialist appointment), optometrist appointment, dentist appointment, hospital visit (e.g., for surgery, medical procedure, emergency room visit), and other. Participants were also asked to respond to two open-ended questions asking: 1) “Why did you choose to bring more than one child to the appointment/procedure (e.g., convenience, you thought one sibling might be a positive influence on the other(s), etc.)? List all reasons”, and 2) “If you have booked appointments/procedures/etc. for your children together, how did you decide which sibling would have the appointment/procedure/etc. first?”. The responses to these open-ended questions were subsequently coded (see below for details).

Sibling relationship quality.

Participants completed the *Sibling Relationship Questionnaire – Revised (Parent)* (SRQ-R) (Furman & Buhrmester, 1985) to provide a measure of their perception of their participating children’s sibling relationship quality. This questionnaire was deemed to have good reliability and validity in a review of family measures in pediatric psychology (Alderfer et al., 2008). Participants rated forty-eight questions on a scale from hardly at all (1) to extremely much (5). Composite scores for warmth/closeness and conflict were of interest in the current study. Relevant scale scores were averaged to create composite scores for warmth/closeness (composed of intimacy, prosocial behaviour, companionship, similarity, admiration by sibling, admiration of sibling, and affection scales; 21 items total) and conflict (composed of quarrelling, antagonism, and competition scales; 9 items total), with greater composite scores reflecting greater levels of the composite. If more than 1 of 3 items were missing from a scale (the average of 3 items), the scale was treated as a missing score (Furman, personal communications), and composite scores were derived by averaging scores for the completed scales ($n = 1$ participant).

Parent-child relationship quality.

Participants' perspective of the quality of their relationship with their children was examined using the *Child-Parent Relationship Scale* (CPRS) (Driscoll & Pianta, 2011; Pianta, 1992). Evidence for reliability and validity of this measure with mothers and fathers has been demonstrated (Driscoll & Pianta, 2011). Participants rated 15 items on a scale from definitely does not apply (1) to definitely applies (5). Participants completed this measure twice, once for each of their participating children (randomized order of older vs. younger first). Subscale scores were created for conflict (8 items) and closeness (7 items) by summing the relevant items. Scores for the conflict subscale could range from 8 – 40, whereas scores for the closeness subscale could range from 7 – 35, with higher scores reflecting higher levels of the construct. Missing data was handled by imputing the participant's mean score for the subscale as the missing value ($n = 1$ participant, older child CPRS).

4.3.4 Coding of Open-Ended Questions

A coding manual was developed for the current study by the first author to code the responses to the open-ended questions. The written responses were reviewed, and codes were created to represent the most commonly reported answers. The coding manual consisted of definitions and examples for each code. For the first question regarding why parents chose to bring more than one child to the appointment, responses were coded across five categories including: 1) Convenience, 2) No child care, 3) Support or positive influence, 4) Appointment booked together, and 5) Other. For the second question asking how parents decided which sibling would have the appointment/procedure first, responses were coded across five categories including: 1) Children decide, 2) Reference to age, 3)

Health professional or clinic decides, 4) Personality or characteristics of the children, and 5) Other (see Table 1 for descriptions and examples of each code). Responses falling into the “Other” categories varied in nature. Responses were coded by indicating whether the response did, or did not, fall into each of the respective categories. Given that participants often listed multiple reasons, each response could be coded as falling into more than one category. Because quantitative analyses were required to address the research questions of interest (e.g., the relation between coded responses and participant variables), a quantitative coding approach was used (e.g., as opposed to identifying themes using qualitative analysis).

All responses were coded by the first author. A second research assistant reliability coded all responses. Percent agreement was used to assess inter-rater reliability, because the coding was straightforward and a detailed manual was available, making the likelihood of rater guessing low (McHugh, 2012). Overall percent agreement for the first question asking why siblings were brought together was 94.35%, and overall percent agreement for the second question asking how parents decided which sibling would go first was 96.09%, indicating acceptable reliability (Stemler, 2004).

4.4 Results

4.4.1 Sibling Presence at Medical Appointments

Overall, 97.9% of participants (i.e., 93/95 participants) reported having brought more than one child in their family to a medical appointment. With regard to the specific appointment type, participants most frequently reported bringing siblings together to dentist appointments (90.5%), followed by doctor’s appointments (89.5%), needles (85.3%), optometrist appointments (60.0%), hospital visits (44.2%), bloodwork (29.5%)

and other (3.2%). A Cochran's Q test revealed that the frequencies with which parents reported bringing siblings significantly differed by appointment type, $\chi^2(6) = 272.68, p < .001$. Follow up exact McNemar's tests revealed that the frequencies for all appointment type pairings significantly differed from one another (p 's $< .05$), except for needles and doctor's appointments, needles and dentist appointments, and doctor's and dentist appointments.

4.4.2 Coded Responses to Open-Ended Questions

Of the 93 participants who reported that they had brought more than one child to an appointment, one participant did not provide responses to the open-ended questions. Therefore, the results reported below are based on a sample of 92 participants. When asked why they had chosen to bring more than one child to the appointment/procedure, the most frequently reported answer was convenience (83.7%). This was followed by reporting the siblings to be a support or positive influence (32.6%), referencing no child care (28.3%), the appointments being booked together (15.2%), and lastly "other" (10.9%).

Regarding how they decided which sibling would go first, most participants referenced allowing their children to decide (53.3%). The personality or characteristics of the children was the next most frequently cited response (29.3%), followed by allowing the health professional/clinic to decide (23.9%), referencing age (18.5%), and lastly "other" (14.1%).

4.4.3 Relationship Between Open-ended Responses and Parent and Sibling Factors

Again, the results reported below are based on a sample of 92 participants. The relationship between sibling relationship quality (warmth/closeness and conflict based on the SRQ-R [Table 2]), parent-child relationship quality (closeness and conflict based on the CPRS [Table 2]), and children's ages (older and younger sibling) and the coded responses for each question were examined using a series of Pearson's correlations (Table 3). For the first question asking why siblings were brought to the appointment together, parent-reported conflict with both their children was positively correlated with parents citing no child care (older sibling: $r = .22, p < .05$; younger sibling: $r = .27, p < .01$). Further, parent-reported closeness with their older child was positively related to parents referencing the appointments being booked together ($r = .26, p < .05$). The older sibling's age was significantly negatively correlated with parents reporting allowing the health professional or clinic to decide which sibling would have the appointment/procedure first ($r = -.22, p < .05$). No other significant correlations were found.

The relationship between child sex (older and younger sibling) and the coded responses for each question were examined using a series of Pearson's chi-square tests (Table 4). The relationship between older child sex and referencing the personality or characteristics of their children as a factor for deciding which sibling would go first was close to reaching significance, $\chi^2(1) = 4.25, p = .066$. For participants whose oldest child was a male, 39.1% indicated deciding based on the personality/characteristics of their children, whereas for participants whose oldest child was a female, only 19.6% did. No other significant relationships emerged between older or younger sibling sex and the coded response categories.

4.5 Discussion

The current study sought to examine the extent to which parents bring siblings to pediatric appointments. As hypothesized, almost all (~98%) parents reported having ever brought siblings along to their children's medical appointments. This high percentage may in part reflect the young age of children included in the sample. Dentist appointments, doctor's appointments, and needles were reported significantly more frequently than the other appointment types (e.g., blood work), with 85% or greater of parents endorsing bringing siblings. Parents may be more likely to bring siblings in these contexts as there may be a greater likelihood that both children would concurrently require the appointment/procedure than the other appointment types (e.g., hospital visits). Fewer siblings reported at some appointment types, such as blood work and hospital visits, may also be reflective of less children requiring these appointments or fewer of these appointments being jointly booked, rather than parents' reluctance to bring siblings along. Family/office physician visits are more frequent for youth than emergency visits (Chen & Escarce, 2006; Manos et al., 2014).

The most frequently cited reason for bringing siblings to medical appointments was convenience, with over 80% of parents reporting this answer. Given the demands of parenting multiple children, and the young age of the children in the study, it makes sense that convenience is a driving reason. Close to a third of parents also identified siblings being supportive or a positive influence as a reason for bringing multiple children, suggesting they may believe siblings enhance children's medical experiences. Some evidence for positive impacts of companions on medical appointments have been found in the adult literature (Laidsaar-Powell et al., 2013). It is also possible that the examples

given in the question may have cued some parents to provide responses that fell into these code categories. However, the fact that only approximately a third of parents provided siblings being supportive as a response suggests that parents were likely answering based on their true reasons. Over half of parents reported allowing their children to decide who would receive the appointment/procedure first. Parents may believe their children will have a more positive experience if they have ownership over this decision. Alternatively, parents may not have a strong basis for making this decision, and therefore leave it up to their children. Approximately 29% of parents identified their children's personality or characteristics as influencing their decision regarding who should have the appointment/procedure first, suggesting they may view their children's individual temperament as relevant.

Greater levels of conflict in the parent-child relationship reported with both older and younger siblings was related to participants being more likely to cite no child care as a reason for bringing siblings to medical appointments. Given evidence that families with more positive parent-child relationships are more likely to do things together (Crouter et al., 2004), conversely, families with more negative parent-child relationships may be less likely to actively seek going places as a family. Rather, consistent with the hypothesis, parents may be more likely to bring siblings to appointments out of need, such as not having child care. Within siblings, greater conflict between parents and a child has been associated with greater problematic child behaviour (Lam, Solmeyer, & Mchale, 2012). Parents who have more conflictual relationships with their children may feel less comfortable leaving their children with caretakers, possibly due to behaviour concerns. It is also possible that a third variable may in part explain this relation. For example,

increased stress in the family environment could potentially relate to both conflictual parent-child relationships and a parent having limited child-care options. Contrary to the hypothesis, greater levels of parent-child closeness with their older child was related to parents being more likely to reference the appointments being booked together as their reason for bringing siblings. Given that this reason is likely reflective, at least in part, of a decision by the health care practice, this finding is difficult to interpret. Granted they know/remember the family, practices may be more likely to initiate or agree to booking appointments for siblings together when they view parent-child relationships as more positive, however research is needed to clarify this. Similarly, parents who have closer relationships with their children may feel more comfortable agreeing to, or suggesting, bringing siblings together.

Contrary to the hypothesis, as the oldest child's age increased, participants were less likely to report allowing health care professionals/clinics to decide appointment/procedure order. It is possible that as the eldest child ages parents have more opportunities to make this decision and learn from experience. Further, as children age and attend more appointments, parents may consider a greater number of factors in their decisions. Therefore, they may be less likely to defer to health care staff. There is some support for the notion that older sibling characteristics and experiences may set the stage for parenting behaviours (see Brody, 2004; McHale et al., 2012, for overviews).

Consistent with the hypothesis, parents of eldest males more often cited the personality/characteristics of their children as influencing their decision regarding appointment/procedure order than parents of eldest females, although this finding did not reach significance. Although differences were only significant between male and female

only siblings, involvement in companion activities have been found to be greater for sibling dyads that include a male (Cole & Kerns, 2001). Parents may have more chances to see how their sons' personalities influence the outcomes of "who goes first" in daily activities. Additionally, adults have a tendency to believe girls are more communicative of their pain symptoms than boys (Cohen, Cobb, & Martin, 2014). Therefore, parents may expect daughters to act in specific ways in medical contexts, whereas sons' behaviours may be considered more likely to be influenced by their personality. However, this finding should be interpreted with caution and further research is needed given that it did not reach significance.

Given that most parents find it convenient to bring multiple children to appointments, finding ways to accommodate siblings may be helpful in ensuring timely health care and positive experiences for families. This is particularly relevant given evidence that children with more siblings have lower rates of health care utilization (Chen & Escarce, 2006). Clinician guidelines for adults highlight the value of discussing the inclusion of companions, and offering ideas for supportive behaviours (Laidsaar-Powell et al., 2013). These recommendations could similarly be adapted to sibling companions at pediatric appointments. Clinicians could discuss with parents and children the preferred role of siblings, and could provide suggestions for how siblings may be helpful during the appointment. Data from the current study indicate that some parents bring siblings to medical appointments because they believe they are a source of support or a positive influence. Therefore, discussing with parents and children what specific sibling behaviours they find helpful, and encouraging these behaviours during the appointment, could potentially facilitate improved outcomes.

The current study has several limitations. First, the participants represent a convenience sample of parents who were involved in a larger study focused on siblings. Therefore, the sample may have included parents who are particularly inclined to bring siblings on outings, potentially influencing the generalizability of the findings. The sample also included a large number of participants with university level education and a high annual household income, and therefore the results may not necessarily be reflective of the general population. Further, the findings only represent the frequency with which siblings attend medical appointments; the extent of sibling involvement in these appointments was not examined. Additionally, only a small subset of potentially relevant variables were assessed in the current study, limiting the conclusions that could be drawn. The study only assessed the variables of interest (age, sex, relationship quality) for the two children who were participating in the larger study. However, despite the questionnaire instructions, it is possible that factors related to (e.g., health status, age), or experiences with, additional siblings not involved in the study influenced parents' responses to the open-ended questions. Parents were also not instructed to limit their open-ended responses to a specific time-frame. Therefore, parents' responses may not have been based solely on experiences with their children at their current ages. Lastly, given the large number of analyses conducted, the potential for Type 1 error should be kept in mind when interpreting the significant findings.

Additional research examining sibling presence at pediatric medical appointments is needed. Recent reviews in the adult literature identified both similarities and differences regarding the perceived influence of companions on medical appointments from the point of view of patients, companions and health professionals (Laidsaar-Powell

et al., 2013; Wolff & Roter, 2011). For example, both reviews identified that all three parties recognize the benefit of companions, although companions may ascribe more significance to their role than patients (Laidsaar-Powell et al., 2013), and health professionals may be more inclined to identify challenges created by companions (Wolff & Roter, 2011). The impact of accompanying siblings on children's medical visits could similarly be examined using multiple informants and outcome measures. Family variables that may be related to a more positive experience could be explored, such as sibling behaviours or characteristics. Future research may focus on determining what situational features or sibling characteristics are associated with a specific order for appointments/procedures being related to better outcomes. Examining parents' reasons for not bringing siblings to various pediatric appointments may also provide additional relevant information. Lastly, the value of being cognizant of the influence of ethical issues such as confidentiality has been acknowledged in the adult literature (Mitnick, Leffler, & Hood, 2010; Wolff & Roter, 2011), and may also be relevant to explore in pediatrics. Children's right to privacy during medical procedures and the potential impact of additional siblings, who may be distracting, on parent's and clinician's ability to promote the best quality care should be considered.

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4.8 Tables

Table 4.1

Code Descriptions and Example Quotes for Open-Ended Questions

Codes	Description	Quote
Question 1		
Convenience	Mentions that it is more convenient	<i>“Convenience to make one trip instead of several”</i>
No Child Care	References lack of child care, or associated cost	<i>“No childcare at home for other children”</i>
Support/Positive Influence	Mentions siblings being supportive of one another, or indicates they are a positive influence	<i>“Now, I bring them together b/c (because) they want to and b/c (because) my youngest is afraid of needles and she likes her sister to be there with her”</i>
Appointment Booked Together	References the office booking the appointment together, or both children being due for the appointment	<i>“Both were scheduled for the appointment”</i>
Other	Response does not fit into one of the other categories	<i>“We were en-route between a number of errands”; “Generally we do things as a family”</i>
Question 2		
Children Decide	Mentions allowing children to decide themselves by some means	<i>“I would generally ask who wanted to go first. The first one to say “me” would go”</i>
Reference to Age	Mentions children’s age as a deciding factor	<i>“We have a large family & try to stick (to) the ‘oldest goes first’ rule”</i>
Health Professional/Clinic Decides	References the health care professional or a clinic staff deciding	<i>“Usually the receptionist would decide”</i>

Codes	Description	Quote
Personality/Characteristics of Children	References children's personalities or characteristics influencing their decision	<i>"My oldest as he has not expressed as much pain/fear during appointments therefore role modeling to his younger brother that it was safe and pain free or manageable pain"</i>
Other	Response does not fit into one of the other categories	<i>"Randomly"; "Doesn't matter"</i>

Table 4.2

Descriptive Summary of Parent-reported Sibling Relationship Quality and Parent-Child

Relationship Quality

Measure	Mean	Standard Deviation
SRQ-R Warmth/Closeness	3.44	0.60
SRQ-R Conflict	2.79	0.71
CPRS Closeness		
Older Child	31.61	3.22
Younger Child	32.37	2.96
CPRS Conflict		
Older Child	17.60	7.29
Younger Child	17.20	5.80

Table 4.3

Correlations Among Sibling Factors and Coded Responses to Open-Ended Questions

Sibling Factors	Question 1 Codes				Question 2 Codes			
	Convenience	No Child Care	Support/ Positive Influence	Appointment Booked Together	Children Decide	Age	Health Professional/ Clinic	Personality/ Characteristics of Children
Older Sibling Age	-.159	-.178	.136	-.049	-.091	.018	-.223*	-.008
Younger Sibling Age	-.025	.041	.066	.120	.155	.135	-.014	-.061
Sibling Warmth/ Closeness (SRQ-R)	-.064	-.025	-.001	.031	.106	-.126	.127	.102
Sibling Conflict (SRQ-R)	.094	.123	.053	-.053	.124	-.026	-.026	-.001
Parent-Older Child Conflict (CPRS)	-.057	.223*	-.054	-.143	-.117	.046	-.048	-.031
Parent-Older Child Closeness (CPRS)	.009	-.086	-.100	.258*	.139	-.019	.132	.102

Sibling Factors	Question 1 Codes				Question 2 Codes			
	Convenience	No Child Care	Support/Positive Influence	Appointment Booked Together	Children Decide	Age	Health Professional/Clinic	Personality/Characteristics of Children
Parent-Younger Child Conflict (CPRS)	.056	.270**	-.044	-.057	.055	.188	.036	-.015
Parent-Younger Child Closeness (CPRS)	-.054	-.192	-.048	.090	.183	-.069	-.001	.016

Statistics represent Pearson's correlations (r)

* $p < .05$, ** $p < .01$

Table 4.4

Percent of Parents Reporting Coded Response Category for Open-Ended Questions by Child Sex Based on Pearson's Chi-Square Tests

Child Sex	Question 1 Codes				Question 2 Codes			
	Convenience (%)	No Child Care (%)	Support/Positive Influence (%)	Appointment Booked Together (%)	Children Decide (%)	Age (%)	Health Professional/Clinic (%)	Personality/Characteristics of Children (%)
Older Sibling Sex								
Male	76.1	30.4	37.0	19.6	50.0	15.2	19.6	39.1
Female	91.3	26.1	28.3	10.9	56.5	21.7	28.3	19.6
Younger Sibling Sex								
Male	80.8	30.8	32.7	19.2	53.8	15.4	21.2	30.8
Female	87.5	25.0	32.5	10.0	52.5	22.5	27.5	27.5

CHAPTER 5: DISCUSSION

5.1 Summary of Dissertation Papers

The goal of the current dissertation was to comprehensively study the role of siblings in pediatric pain. Through specifically exploring siblings, this dissertation sought to contribute to the existing literature on family factors in pediatric pain. This goal was addressed through three papers. Using scoping review methodology, Chapter 2 presented a summary of the current state of the literature on siblings in pediatric pain. Chapter 3 examined the relation between siblings' relationship quality and their behaviours during acute pain, and the relation between an observing sibling's behaviours and a child's pain outcomes. Finally, Chapter 4 examined sibling presence at pediatric medical appointments more broadly, and parents' perspectives on why they bring siblings to medical appointments and how they decide which child receives an appointment or procedure first.

This chapter provides a discussion of the key findings across the three papers, focusing on the findings pertaining to family relationships, and siblings' behaviours and children's acute pain experiences. A discussion of the theoretical and clinical implications of these findings is then provided. Finally, the key strengths and limitations of the dissertation are discussed, and suggestions for future research directions are provided.

5.2 Discussion of Key Findings and Integration with Existing Research

5.2.1. Family Relationships

In the current dissertation, family relationships were explored in two key areas: 1) sibling relationships as they relate to siblings' behaviours during acute pain, and 2)

sibling and parent-child relationships as they relate to sibling presence at pediatric medical appointments more broadly. This represents an important contribution to the literature on siblings in pediatric pain, as the scoping review described in Chapter 2 identified a lack of research examining relationship quality within family members in the context of siblings and pediatric pain, particularly amongst quantitative studies. The findings of the current dissertation indicate that both positive and negative aspects of relationship quality are related to siblings' behaviours in the experimental pain context, as well as parents' reasons for bringing siblings to medical appointments.

With regard to the positive dimensions of sibling relationship quality, both self-reported sibling warmth/closeness and behaviour coded positivity were found to be related to children engaging in more behaviours that shifted attention away from the pain/experience (non-attending; e.g., distraction, humour) while experiencing acute pain with a sibling present. This finding is similar to the relationship discovered between family cohesion and children's cold pressor behaviours with a parent (Kathryn A. Birnie et al., 2017). Coded positivity was additionally related to children engaging in less behaviours that were focused on the pain/experience (attending; e.g., symptom talk) while completing the CPT. As noted in Chapter 3, it is possible that siblings with stronger relationships are more inclined to engage in natural conversation, and feel more comfortable sharing humour with their sibling, and therefore engaged in these behaviours as a strategy while experiencing acute pain (Gass et al., 2007). The behaviours by the sibling completing the CPT classified as attending behaviours in the current dissertation are similar to behaviours considered to indicate distress, whereas the behaviours classified as non-attending are similar to those indicating coping during children's

procedural pain (R. Blount et al., 1997; R. L. Blount et al., 2001). Therefore, more broadly, these findings may indicate that positive/warm sibling relationships are a facilitator of resilience during pediatric acute pain, when a child's sibling is present. The findings of the study presented in Chapter 4 provide some support for this assertion, and extend it to the broader medical context. Approximately 33% of parents reported bringing siblings to medical appointments/procedures because they viewed siblings as being supportive or a positive influence. Therefore, this may indicate that some parents witness siblings as a source of resilience for their children in potentially painful or distressing medical contexts. Family functioning has been identified as a potential resource for resilience in pediatric chronic pain (Cousins, Kalapurakkal, Cohen, & Simons, 2015). The current findings may support this proposition, and extend it to sibling relationships specifically as a possible promoter of resilience for children in pediatric settings more broadly, and children's acute pain specifically.

Although parent's perspectives on the quality of their children's sibling relationship was not related to their reported reasons for bringing siblings to medical appointments, greater levels of closeness in parents' relationship with their older child was related to them more often referencing the appointments being booked together. As suggested in Chapter 4, clinicians/health care facilities may in part be responsible for this decision, and may be more inclined to book appointments for siblings together when they perceive family members as having closer relationships with one another. Therefore, the parent-child relationship may indirectly influence siblings' likelihood of being present in clinical contexts via the actions of health care professionals. This finding may support the

need for more sibling research in pediatric pain including the perspectives of health care professionals, as identified in the scoping review.

The findings regarding the relation between the negative dimensions of sibling relationship quality and siblings' behaviours during the cold pressor were less clear. Specifically, greater levels of behaviour coded negativity were related to the sibling who completed the CPT first engaging in more attending behaviours, and the sibling who completed the CPT second engaging in less attending behaviours while experiencing cold pressor pain. The finding regarding the first sibling was consistent with the hypothesis and, as suggested in Chapter 3, may reflect an inclination for children who are experiencing acute pain in the presence of a sibling to become more focused on the pain experience when the sibling relationship is negative. This finding is also consistent with the negative relationship discovered between family cohesion and children's symptom complaints during cold pressor pain in the presence of a parent (Kathryn A. Birnie et al., 2017). However, the findings regarding the second sibling were in the opposite direction. As described in Chapter 3, this may be reflective of a tendency for siblings who aim to be different from one another in daily activities (e.g., athletics, academics) to have less intimate relationships (Whiteman et al., 2007), and may demonstrate that this extends to siblings' pain behaviours. In the current study, it is unknown whether the difference discovered was a result of the child who went second actively attempting to respond differently to the pain experience than their sibling, or whether this was reflective of natural differences in how each child would have behaved regardless of their sibling. This finding may also indicate that children's acute pain responses may be more unpredictable when they are with a sibling whom they have a more negative relationship with;

clinicians should not assume that siblings undergoing the same pain experience will necessarily respond in a similar manner.

Negativity in family relationship quality was also associated with parents' reasons for bringing siblings to medical appointments in the study described in Chapter 4. Specifically, parents who reported more conflict in their relationships with both their older and younger child were more likely to describe bringing siblings to medical appointments due to no child care, possibly indicating a tendency to bring siblings out of necessity rather than an active choice. Negativity in the parent-child relationships could also reflect family stress (e.g., due to limited resources), which may drive a need to bring siblings to medical appointments. Therefore, negativity in family relationship quality may influence siblings' responses to acute pain, perhaps enhancing different patterns of behaving amongst siblings, and may also reduce parents' desire to bring siblings together to potentially painful clinical appointments. Whether these two findings are directly related is unknown, and perhaps warrants further investigation.

Taken together, these findings indicate that relationship quality amongst family member plays a role in both the pediatric contexts in which siblings are exposed to one another's potentially painful experiences, as well as the ways in which children respond to these experiences. Sibling relationship quality appears to be particularly relevant to children's behavioural responses to acute pain, possibly promoting resilience when the relationship is positive. Although findings indicate that some parents indeed view siblings as a source of support in medical contexts, parents' perception of their children's sibling relationship quality was not related to their reasons for bringing siblings to medical appointments. Rather, their own relationships with their children appears to have a more

salient influence on their reasons for bringing their children together. It is possible that since parents are likely primarily responsible for making, or agreeing to, the decision to bring siblings to medical appointments, the qualities of their own relationships may be more influential. These findings support the need identified in the scoping review for more research examining relationship quality amongst family members as it relates to siblings and pediatric pain.

5.2.2. Sibling Behaviours and Children's Acute Pain

A second key contribution of the current dissertation to the pediatric pain literature was the examination of the relationship between an observing child's behaviour and their sibling's acute pain outcomes. The relation between parents' behaviours and children's pain outcomes is well established (see section 1.3). The study described in Chapter 3 extends this line of research to siblings, and indicates that a sibling's behaviour is related to children's acute pain in ways that are both similar and unique to parents. Further, the scoping review described in Chapter 2 identified a lack of research examining the possible role of siblings in the context of children's acute pain, and recommended more research in this area. The manuscript described in Chapter 3 directly addresses this gap.

Of particular relevance to this research objective were the identified partner effects using the actor-partner interdependence model (Cook & Kenny, 2005). A partner effect was discovered where greater levels of attending behaviours (e.g., reassurance, symptom talk) by the sibling who was observing was related to their sibling who was participating in the cold pressor having a lower pain tolerance score. This effect was significant for both siblings in their roles as observer and participant (i.e., the effect was

found in both the first and second cold pressors the siblings completed), emphasizing the strength of this finding. A partner effect was also discovered for the first cold pressor the siblings completed, where greater coping/encouragement behaviours (e.g., procedure praise) by the observing sibling was related to their participating sibling providing greater pain intensity and pain-related fear ratings following the CPT. Therefore, behaviours by an observing sibling that focus on a pain experience are generally related to worse pain outcomes for the child experiencing pain, and appear to have a broad influence on both the physical and emotional components of pain. Similar findings have been documented in the literature examining the relationship between parent behaviours and children's pain experiences in the context of experimental pain tasks (Moon et al., 2011; Schinkel et al., 2017; Walker et al., 2006), as well as in clinical acute pain settings (Cohen et al., 2000; Manimala et al., 2000). As suggested in Chapter 3, the negative influence of observer attending behaviours on children's pain appear to be consistent across family members, including both parents and siblings. Suggested mechanisms for this finding in the context of parent behaviour during pediatric acute pain include that attending behaviours, such as reassurance, communicate parental fear, perpetuate a child's distress, or signal approval for a child to engage in distress behaviours, thereby magnifying the pain experience (McMurtry, C.M., McGrath, P.J., Chambers., 2006). It is possible that similar mechanisms accounted for the current findings in siblings. When a child's sibling engages in behaviours focused on their pain experience, this may serve as form of validation from the sibling that their pain experience is indeed serious and distressing, or may signal that their sibling is afraid. This may amplify a child's experience of pain and fear and/or increase their likelihood of communicating their pain and fear (e.g., through

removing their hand from the water). It is possible that attending behaviours such as reassurance carry more weight when engaged in by a sibling, since they may be less likely to engage in these behaviours in other contexts relative to a parent. However, further research is needed exploring these mechanisms in siblings.

The findings regarding the observing sibling's non-attending behaviours were not as predicted. A partner effect was discovered where greater levels of non-attending behaviours (e.g., distraction, humour) by the first observing sibling was related to their sibling who was participating in the cold pressor providing higher pain-related fear ratings. Therefore, it appears that behaviours by an observing sibling which remove focus from the pain experience are related to some children experiencing more negative pain-related emotions in the acute pain context. This finding contradicts those in the parent-child literature, which have demonstrated a relationship between parent non-attending behaviours and better pain outcomes for children experiencing experimental (Moon et al., 2011; Walker et al., 2006) and acute procedural pain (Manimala et al., 2000; McCarthy et al., 2010). As suggested in Chapter 3, it is possible that when a sibling engages in non-attending behaviours, it functions differently than when a parent engages in these same behaviours potentially due to differences in roles and power dynamics in the sibling relationship compared to the parent-child relationship. There is evidence in the parent-child pain literature that the quality of the distraction delivered is important (McCarthy et al., 2010). It is possible that even though young siblings attempted to distract or be humorous, they may not be as effective at doing so as an adult parent may be. For example, the content of the observing child's comments or behaviours may have been distracting or humorous to themselves, but may not have been engaging to their sibling;

a parent may be more likely to keep their child's interests in mind while distracting. Therefore, siblings' non-attending behaviours may not have the same effect of removing attention from the pain experience, and thus may not alleviate children's pain-related fear. Examining the quality of distraction children provide across development and its impact on sibling pain outcomes would be an interesting avenue for future research. Given that the findings represent relations, it is also possible, as suggested in Chapter 3, that children responded to their sibling's fear by engaging in non-attending behaviours, such as distraction or humour. Siblings may also be aware of how one another generally responds to pain; children who knew their sibling tends to experience more pain-related fear may have pre-emptively engaged in more non-attending behaviours. Future studies could use sequential analysis to tease this apart. Since all significant findings discovered represented relations between sibling behaviours and poorer pain outcomes for children, it is possible that the nature of young siblings' behaviours in general may be related to children experiencing more pain and distress in acute pain settings. Further research directly comparing children's pain experiences with and without a sibling present would be needed to clarify this.

As noted in Chapter 3, significant partner effects regarding the relations between the observing sibling's behaviours and the participating sibling's pain outcomes were more often discovered for the first cold pressor the sibling dyads completed. Although this finding should be cautiously interpreted given that direct statistical path comparisons were not conducted, it may reflect the observing sibling's behaviour being more salient during the first cold pressor due to the novelty of the task. This finding may also indicate that the order in which siblings receive a painful medical procedure may be important,

given that siblings may exert a greater influence on the first child's pain experience. The results of the study described in Chapter 4 indicate that over 50% of parents report allowing their children to decide the order in which they receive an appointment/procedure. This was followed by 29% of parents describing the personality/characteristics of their children as being relevant, with a trend for parents of eldest males more often providing this response than parents of eldest females. As parents' eldest child aged, they were also less likely to describe allowing health professionals/clinics to make this decision. These findings indicate that parents' decision regarding the order for sibling appointments/procedures is influenced by several factors, however there is currently no known evidence to indicate why one sibling may be more appropriate to observe or receive a procedure first. Taken together, these findings support the need for more research directly examining what factors are related to a specific sibling order promoting better outcomes, as suggested in Chapter 4. Further examining the factors that parents are already using to guide their decisions (e.g., child personality) may be particularly relevant.

Given that sibling relationship quality was not related to children's behaviours while observing their sibling in pain, yet observing sibling behaviours were related to children's pain outcomes, identifying variables that are related to the behaviours observing siblings engage in warrants further investigation. Some parents described deciding the order in which their children received appointments/procedures based on their children's personality/characteristics or their age, and only individual factors related to the children (i.e., age and sex) were related to how parents made this decision. Although it is unknown whether parents' decision regarding order was more influenced

by factors related to the child who would be observing/waiting, or the child who would be receiving the appointment/procedure, these findings may indicate that individual factors are more relevant to the behaviours siblings engage in while observing.

Overall, the findings of this dissertation suggest that parent relationships influence siblings' presence in pediatric contexts through their relation to parents' reasons for bringing siblings to medical appointments, whereas sibling relationships are related to how children respond to acute pain in the presence of a sibling. While positive sibling relationships may promote adaptive behavioural responses to acute pain, and some parents view siblings as a source of support in medical settings, the actual behaviours children engaged in during the current study were related to their siblings experiencing poorer pain outcomes.

5.2 Theoretical Implications

To advance our understanding of siblings in pediatric pain, it has been suggested that research in the area be grounded in theory (Jaaniste et al., 2013). The current dissertation was based on the frameworks of family systems theory (Bavelas & Segal, 1982), as well as models on the role of families in pediatric pain (Palermo & Chambers, 2005; Palermo et al., 2014). As evidenced by the paucity of published research studies identified in the scoping review, more research is needed before we can fully understand how siblings may fit into our theoretical understanding of families in pediatric pain. However, the findings of the current dissertation provide a starting point.

Family systems theory asserts the importance of examining behaviour in understanding family members' influence on one another (Bavelas & Segal, 1982). In the current dissertation, associations were discovered between sibling relationships, as

measured by observed behavioural interactions, and children's behavioural responses to acute pain in the presence of their sibling, as well as between siblings' behaviours and children's pain outcomes. These findings establish that examining behaviour is a valuable, and perhaps necessary, means of understanding the role of siblings in children's pain experiences. This is an important contribution, as studies in pediatric pain that have been grounded in family systems theories have often not used behavioural observation (Palermo & Chambers, 2005).

The family models in pediatric pain identify pathways through which children's pain experiences (including physiological, emotional, and behavioural components) may be related to their family context, primarily parents, through individual factors (e.g., parent behaviour) and broader dyadic and family factors (e.g., family functioning) (Palermo & Chambers, 2005; Palermo et al., 2014). Through identifying relations at both the broader family/dyadic level (i.e., between siblings' relationship quality and their behavioural responses to pain), as well the individual level (i.e., between sibling behaviours and children's pain intensity and fear), the findings of the current dissertation demonstrate that these pathways are also applicable to understanding the role of siblings in children's acute pain experiences. However, the current findings suggest the nature of the impact may differ when examining siblings (e.g., non-attending behaviour was related to children's pain outcomes differently than that which has been observed in parents). The current dissertation also added to these theoretical models by examining the relations between individual (i.e., child age and sex) and dyadic (i.e., parent-child and sibling relationship quality) factors and siblings' presence at medical appointments more broadly. In so doing, the findings identified that these pathways are also relevant in

understanding the clinical contexts in which siblings may exert an influence on one another's pain experiences. As suggested in the scoping review, once more research has been conducted, these theoretical models should be expanded to clarify the influence of siblings on children's pain.

5.4 Clinical Implications

Given that the cold pressor task is considered most clinically relevant to acute pain (von Baeyer et al., 2005), the findings of the paper described in Chapter 3 have clinical implications regarding siblings in acute pain settings. As suggested in Chapter 3, parents and clinicians should be cognizant that, in the context of a warm and positive relationship, siblings may facilitate resiliency through making children more likely to respond to acute pain by engaging in behaviours such as distraction or humour. Observing children should be instructed to refrain from engaging in behaviours that draw attention to their sibling's pain/experience, such as reassuring their sibling. The current dissertation did not identify any sibling behaviours that were related to better pain outcomes for children. Therefore, young children may need more direction regarding the specific behaviours they should engage in, and how to engage in these behaviours, to promote better pain outcomes for their sibling. Clearly, more research is needed to identify what these behaviours may be. Once sibling behaviours have been identified that are related to better child pain outcomes, promoting their use amongst siblings may enhance the resiliency that seems to be affiliated with a positive sibling relationship.

One of the significant contributions of the study described in Chapter 4 was providing data to support the clinical relevance of understanding the potential role of siblings in children's acute pain experiences, and identifying contexts in which siblings

may likely be present. The findings of this study indicate that almost all parents bring siblings to medical appointments, most commonly dentist appointments, doctor's appointments, and needles. Therefore, understanding the influence sibling-related factors may have on children's pain or distress during dental or needle procedures, or in the context of doctor's appointments (e.g., while examining a sore throat) is clinically important. Further, replicating the study presented in Chapter 3 in a clinical setting such as needle procedures may provide further insight into the relations between sibling behaviours and relationship quality and children's pain experiences.

5.5 Key Strengths and Limitations

The current dissertation has several key strengths and limitations that should be highlighted. Firstly, the broad methodological approach of the scoping review (Arksey & O'Malley, 2005; Levac et al., 2010; Peters et al., 2015) represented an excellent means for providing an overview of the nature of the research conducted to date in the newly developing field of siblings and pediatric pain. This methodology facilitated capturing and summarizing a wide range of studies, including conference abstracts and dissertations. However, it is possible that the search strategy and exclusion criteria used resulted in some potentially relevant studies being missed or excluded. For example, pearling of the identified studies was not conducted. Studies focused on cancer related pain were excluded, as the literature on siblings of children with cancer is more developed and has been reviewed (e.g., Alderfer et al., 2010). However, examining the impact of cancer related pain specifically on siblings would be a valuable area for further investigation. Commentaries and letters to the editor were also excluded as it would have been difficult to extract meaningful data from these types of articles with the planned

extraction categories. However, it is possible that these articles may have contained relevant information. For example, commentaries could have included a review of the literature or represented the perspectives of health professionals. Although, it is unlikely that a systematic approach would have been taken to reviewing the literature or that a commentary would reflect a representative sample of health professionals. Further, as noted in Chapter 2, the quality of the included studies was not formally examined. As such, the scoping review did not permit commenting on potential gaps in the literature pertaining to the quality of the work that has been conducted on siblings and pediatric pain (Arksey & O'Malley, 2005; Levac et al., 2010). This is an important limitation given that the scoping review was conducted with an aim to identify research gaps.

Another key strength in the current dissertation was the use of observational measures, which addressed a noted gap identified in the scoping review, and in family research in pediatric pain (Palermo & Chambers, 2005; Palermo et al., 2014) and research on siblings (Alderfer et al., 2010; Noller, 2005) more broadly. The value of using observational measures in addition to questionnaires was exemplified in the fact that significant findings were discovered for children's observed and self-reported relationship quality. Therefore, both children's perception of their relationship with their sibling, as well as the observed quality of their interactions with their sibling contributed information to our understanding of children's behaviours during acute pain. Further, behavioural coding provided a novel account of children's responses to observing their sibling in pain, and experiencing acute pain themselves with a sibling present. However, it is important to bear in mind the limitations associated with the coding systems used in the current dissertation. Presence/absence coding over defined intervals was used for both

the coding of children's cold pressor behaviours and sibling relationship quality.

Although this approach provided a comprehensive estimate of all the behaviours of interest, it did not take into account the intensity of behaviours (e.g., through global ratings), or the time period or sequence of behaviours (e.g., as could be obtained through continuous recording) (J. M. Chorney et al., 2015). While the coding approach used did allow for the study objectives to be addressed, potentially interesting information regarding these other aspects of siblings' behaviours (e.g., intensity) were not able to be explored.

Strengths and limitations also exist regarding the analytic approach, and respondents used in analyses. In the study described in Chapter 3, the self-report and observational data from both siblings was examined in a disaggregated form (e.g., as opposed to creating summary scores) (Holmbeck et al., 2002). This approach is suggested when the correlations between sources are low (Holmbeck et al., 2002), as was the case between self-reported and observed sibling relationship quality. Further, this analytic approach was advantageous in that it produced distinct findings for each data source regarding the relationships between the variables of interest (Holmbeck et al., 2002), which indeed contributed important information. However, a limitation of this approach is that it may lead to a greater probability of a Type 1 error, due to the greater number of analyses required (Holmbeck et al., 2002). A large number of analyses were also conducted in the study described in Chapter 4, potentially increasing the probability of a Type 1 error. A correction for multiple comparisons, such as the Bonferroni correction, was not used in the current dissertation due to the resulting reduction in power and increase in Type II error rate (A. Field, 2013). Rather, the recommended approach of

examining only a priori hypotheses, reporting all analyses conducted, discussing how multiple findings do or do not hang together, and acknowledging the limitations associated with multiple comparisons was used in the current dissertation (Schulz & Grimes, 2005). Further, in order to minimize the number of analyses conducted, the coded behaviours for the cold pressor and tower tasks were grouped for analyses (rather than examining each behaviour separately). Regarding the respondents used in analyses, only child self-report, not parent report, of sibling relationship quality was examined in relation to the children's cold pressor behaviours in Chapter 3. This approach was advantageous in that it allowed for examination of individual actor effects and dyadic partner effects using APIM, which could not have been done using parent report given that APIM requires predictor and outcome variables for each member of the dyad. Although parent report could have been examined using structural equation modeling as was done for observed sibling relationship quality, this would have required conducting additional statistical tests which could have increased the probability of a Type I error. However, it is possible that examining parent report of sibling relationship quality could contribute meaningful information, and should be examined in future research.

As discussed in Chapter 4, a limitation of the study examining sibling presence at medical appointments was the possibility that parents' responses to the open-ended questions may have been in part influenced by experiences with their children who were not involved in the study. However, relations were only examined using variables pertaining to their two children who completed the lab-based study. As such, an additional series of correlation and chi-square analyses were conducted including only families who reported having 2 children ($n = 42$). A different pattern of results emerged.

For the question asking why siblings were brought to the appointment together, parent reported conflict with their older child was still significantly positively related to parents citing no child care, $r = .32, p < .05$. However, parent reported conflict with their younger child was no longer significantly related to citing no child care, and parent reported closeness with their older child was no longer significantly related to citing the appointments being booked together. In addition, older child sex was now significantly related to parents citing convenience, $\chi^2(1) = 4.89, p < .05$ (with parents of eldest females [95.5%] citing this response more often than parents of eldest males [70%]), and younger child sex was now significantly related to parents citing no child care, $\chi^2(1) = 7.70, p < .01$ (with parents of youngest males [30%] citing this response more often than parents of youngest females [0%]). However, it should be noted that for both these findings, two cells had expected counts less than 5, indicating this assumption had been violated. For the question asking how parents decided which child would go first, older child age was no longer significantly related to parents reporting allowing the health professional/clinic to decide, and older child sex was no longer marginally related to citing the personality/characteristics of their children. Given that parents were instructed to complete the measures based on their children who were participating in the study, as well as the loss of power when examining only two-child families, the analyses using the full sample was considered most accurate and appropriate and was therefore reported in the manuscript.

One of the primary strengths of the current dissertation was the methodological rigor afforded by the lab-based environment. The use of standardized, structured tasks, such as the cold-pressor and the tower-building tasks, provided a high degree of

experimental control. However, it is also important to recognize the limitations of this approach, particularly regarding the generalizability of the findings to typical sibling behaviours and clinical or everyday pains. It is possible that siblings would behave differently during more naturalistic interactions (e.g., while playing together at home). Siblings may also respond differently to experiencing or observing their sibling's pain in clinical contexts (e.g., procedures) or during the unanticipated pains experienced in everyday life (e.g., due to injury or illness). Further, the participants represented a sample of typically developing, generally healthy children. Although examining the role of siblings in pediatric pain amongst healthy children is still highly relevant, the findings may not generalize to sibling dyads in which one has a chronic illness or chronic pain. Replicating the current studies with clinical samples would provide meaningful insight into the potential influence of chronic illness/pain on siblings' relationships and pain behaviours. Further, it is possible that children's health status influences parent decisions around bringing siblings to medical appointments. The demographic characteristics of the sample in the current dissertation may also limit the generalizability of the findings to the general population. The sample primarily identified as white, and included a large number of families in which parents reported having a university level education and a high annual household income. The majority of participating parents were also women. This may have influenced the findings reported in this dissertation. Examining the influences of socioeconomic status, culture/ethnicity, and caregiver sex will be important in future research on siblings in pediatric pain. Further, the sample represented a group of parents who self-selected to participate in a lab-based study with two of their children. Therefore, it is possible that the families who participated in the current study may have

more positive family relationships, and children with fewer behavioural concerns.

Additionally, both children had to be between the ages of 8-12 years old; the findings may not necessarily be generalizable to families with older or younger children.

5.6 Future Research Directions

The findings of this dissertation support the relevance of further research on siblings in the field of pediatric pain. As described in Chapter 2, more research on this topic is needed, and there are a number of clinically important questions that could be explored. Specific suggestions for future research based on each of the study findings were provided in the relevant chapters. Some overarching ideas for research on siblings and pediatric pain are presented here.

Sibling researchers have been encouraged to begin studying siblings within the context of the whole family, rather than examining specific individuals or dyads (McHale et al., 2012). The current dissertation took the approach of examining one sibling dyad in a family. Although this provided a good starting point for understanding the potential influences of siblings in pediatric pain, future research on this topic would be strengthened by including multiple family members. Indeed, a need for sibling research in pediatric pain to include multiple family members was identified in the scoping review. For example, it is unlikely that young siblings are without a parent during one another's pain experiences in clinical contexts. Therefore, examining how relationships between a child's behaviours and a sibling's pain outcomes are potentially mediated by parent behaviours would be an important question for future studies. Anecdotally, several participating parents commented that they suspected their child may have behaved differently if they were with a different sibling (e.g., if they were with their older rather

than their younger brother). Further, it is probable that parents of multiple children bring more than just one sibling to medical appointments. Therefore, future research could include all relevant siblings to provide a fuller understanding of how children's experiences of chronic or acute pain are influenced by the whole family. Clearly, answering these types of questions presents both practical and analytical challenges (McHale et al., 2012).

Research on sibling relationships also illustrates the ways in which siblings can indirectly influence one another, for example through shared parents (Brody, 2004; McHale et al., 2012). Examining indirect mechanisms of sibling influence in pediatric pain would be a valuable direction for future research. For example, it's possible that a parent's prior experience with an older sibling's pain, chronic or acute, may influence how they subsequently respond to a younger sibling's pain. Further, a parent's experience bringing a subset of their children to a medical appointment may influence whether they bring additional siblings in the future. These types of questions could be examined in the context of chronic, procedure-related or everyday acute pains.

5.7 Conclusions

The current dissertation explored the role of siblings in pediatric pain, who are an understudied population in the field. The scoping review summarized the current, rather limited, literature on siblings in pediatric pain, established the need for more research, and provided recommendations for continued research on the topic. Using a standardized, lab-based environment, the cold pressor study identified sibling relationships as an important variable in understanding children's responses to acute pain in the presence of a sibling, and demonstrated that sibling's behaviours are indeed related to children's pain

outcomes. The final paper established that a high percentage of parents report bringing siblings to potentially painful pediatric medical settings and procedures. Taken together, the findings of this dissertation suggest that siblings are present in pediatric pain contexts and, through their relationships and actions, influence how children respond to pain.

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Meghan Schinkel, BSc (Hons)
Clinical Psychology PhD Student, Dalhousie University
Centre for Pediatric Pain Research, IWK Health Centre
Halifax, NS, Canada
meghan.schinkel@dal.ca<mailto:meghan.schinkel@dal.ca>

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APPENDIX B: CODING MANUALS

Instructions and detailed descriptions of behaviour codes for the cold pressor task:

- An interval sampling approach will be used (Chorney et al., 2015), in which each of the behaviour codes will be rated as present or absent across 5 second intervals throughout the duration of the cold pressor task. If a single behaviour occurs over multiple intervals, it can be coded in each interval in which it is present (i.e., a behaviour can cross multiple intervals), and multiple behaviours can be coded in each interval.
- The last coded interval is the interval in which the child removes their hand (even if this results in the full 5 seconds not being coded)
- This coding approach will provide information on the types, and relative proportions, of behaviours children are engaging in while observing and taking part in the cold pressor task.
- An interval of 5 seconds was chosen since it is a shorter duration than most of the coded behaviours and is within recommended time frames (Bakeman and Gottman, 1997, pp. 46-49). This coding approach has also previously been successfully used when coding behaviour during the cold pressor task (Schinkel et al., 2017).
- A proportion score will be calculated for each behaviour using the formula: $\frac{\# \text{ intervals with behaviour coded present}}{\text{total } \# \text{ intervals coded}}$ (Chorney et al., 2015).
- The use of proportion scores takes into account the variability in the length of time of the cold pressor task across participants (e.g., some children may leave their hand in the water for 15 seconds while others may leave it in for 200 seconds) to facilitate comparisons.
- Each cold pressor should be watched 3 times. On the first pass, the behaviours should just be observed, without providing any scores. On the second pass, the behaviour of the observing sibling should be coded, and on the third pass the behaviour of the participating sibling should be coded.
- Coding of each cold pressor (observing and participating sibling) should be done in one sitting, so make sure you leave enough time to do all 3 passes in a row.

Observing Sibling

1. Distraction/Non procedure related talk (ODis)

This code refers to comments or behaviours that are intended to distract the sibling from the pain or task. This might include discussing a topic unrelated to the task (e.g., evening plans, a past event), pointing to/discussing the cameras or paintings in the room (i.e., non-task related materials), or discussing other aspects of the study (e.g., the questionnaires, tower task, etc.). It might also include engaging in a verbal or nonverbal game (e.g., hand

games like rock, paper, scissors, repeat after me), or singing a song (if the song is intended to be funny or a joke, code as Humour [OHum]). This also includes engaging in a sibling's attempt to distract.

Examples:

- “What are you doing tonight when we get home” (*discusses plans*)
- “Look at the cameras over there”
- Points at camera, art work, etc.
- “I think that’s abstract art”
- “Why do you think there’s so much tape on the ground”
- “Lets talk about something to distract you”
- “What did you think about the Patriots losing?”
- “Do you think we’re going to have the tallest tower?”
- Sings a song
- Plays rock, paper, scissors
- Thumb war
- “Lets ask Mom if we can get pizza for dinner”

2. Humour (OHum)

This refers to any attempt (verbal or non-verbal) to be humorous or make the sibling laugh, and will usually be light hearted in tone and result in laughter/smiling. Humour can be distinguished from criticism (OCrit) based on the tone of voice, and/or whether it is received positively by the sibling.

Examples:

- Funny or silly faces
- Tells a joke
- “Is there a shark in there?”
- Makes an outrageous or sarcastic comment, e.g., “I think I’m going to drop out of school and become a celebrity”
- Sings song or dances in a way that is intended to be humorous
- Laughter (e.g., in response to sibling humour)
- Makes a funny sound

3. Competition/light hearted teasing (OComp)

This includes competitive statements that are intended to be friendly/fun/humorous as well as light hearted teasing of the sibling or of themselves. It may be done to spur the sibling on. This code can be distinguished from criticism (OCrit) in that it should be received positively by the sibling.

Examples:

- Imitating sibling’s facial expression, behaviours, comments, etc., e.g., “Oh my god this hurts so much”, shaking hands back and forth
- “Lets see how brave you really are”
- “I bet that you won’t last as long as me”

- “Come on, I lasted the whole time”
- “Your face looks so funny right now”
- “I’m going to be like crying when it’s my turn” (in teasing tone)
- “I bet I only last like a few seconds”
- “I think my time is going to be better than yours”
- “Lets see who can do it longest”
- “You’re such a baby” (in lighthearted tone)
- “You’re going to be like asking a thousand questions when it’s my turn”
- “You’re screwing it up”
- “You’re so dramatic”
- “I didn’t think you could last this long”
- “Can you take it out already”

4. Sympathy, Comfort and Reassurance (OReas)

This refers to any verbal or nonverbal displays of sympathy, comfort, or reassurance that is related to the task, the sibling’s cold/pain, or fear. Children will likely use a sympathetic tone of voice, and will do the behaviour with the intent of reducing distress or empathizing/relating with the participating sibling’s experience. These behaviours may occur within the context of a pain/task complaint from the participating sibling.

Examples:

- “You’re okay”
- “Don’t worry, you’re going to be fine”
- “They wouldn’t let us do it if it was dangerous”
- “I don’t think your hand will freeze”
- “It must hurt so much”
- “I’m right here if you need help”
- “I can hold your other hand”
- Child holds participating sib’s hand
- Child rubs participating sib’s back, leg, etc.
- Child gives participating sib a hug
- Child makes a sympathetic facial expression (e.g., in response to sibling pain complaint)
- Child moves in closer in an effort to be comforting
- Child clenches fist, grimaces, looks uncomfortable (e.g., when sib makes pain complaint, puts hand in water)
- “Yikes, that must be so cold”
- “I know it’s scary, but you don’t have to be afraid”
- “If it feels too bad you can take it out” (in sympathetic tone)

5. Coping Commands and Behaviours (OCope)

This refers to comments, suggestions, or behaviors that indicate ideas for a coping **strategy**, or models an idea for coping. This also includes questions or responses related to sibling's coping comments or behaviours, or attempts to facilitate a sibling's coping strategy (note: a specific suggestion to distract in order to cope, feel better, etc. is coded as OCope, however the ensuing conversation would be coded as ODis; general praise is coded as OProPraise).

Examples:

- Child models how to deep breath or imitates participating sib's deep breathing
- "Move your fingers around like this to keep them warm" (models moving fingers)
- "Maybe if you clench your fist it will help"
- "Maybe if I rub your other hand it will help"
- "Try giving yourself a hug"
- "If you grit your teeth you will focus your attention on your teeth"
- "Just think about how good you will feel when it's done"
- "You need to distract yourself so you don't think about the pain"

6. Procedure Related Talk/Attending Behaviours (OProTalk)

This refers to any talk or behaviours that draws attention to the cold pressor equipment or procedure. This might include discussing aspects of the procedure (e.g., putting hand in the water, using the towel, the amount of time it has been) or questions or comments about the equipment, or discussing their own previous experience with the cold pressor (discussion of previous symptoms is coded as Symptom Focused Talk [OSelfSymp]). It might also include behaviours that draw attention to the task (note the behaviours must be clear, i.e., getting up and leaning in vs just gazing at the cold pressor). Talk about their perception of the task/procedure is also included.

Examples:

- "How much time do you think it's been"
- "Where do you think they get the machine?"
- "Can you feel the bottom?"
- "You can take your hand out whenever you want to"
- "I can get the towel ready for you"
- "How do you think she knows to come back in when we're done?"
- "You're supposed to do it all the way to your wrist"
- Child pushes siblings' hand into the water up to the wrist
- Child **gets up** or **leans in** to look at the cold pressor machine
- "I don't think you're supposed to move your hand around"
- Child touches cold pressor machine
- "I felt the sides of the machine when I did it"
- "Do you think the time goes faster when you're doing it or when you're watching?"
- "You might want to roll your sleeve up more"
- "That machine is noisy"

- “The water looks like it’s bubbling”
- “This is boring”
- “This is funner than I thought it would be”
- “I think I’m going to like doing it better than watching”

7. Procedure related praise/encouragement (OProPraise)

This refers to any comments or behaviours that are meant to praise the sibling for their procedure related behaviours/performance, or encourage the sibling with regard to the cold pressor task. This can be distinguished from procedure related talk (OProTalk) by the tone of voice (i.e., if it’s encouraging vs. matter of fact).

Examples:

- “You’re so brave”
- “You’re doing such a good job”
- “I think you’ve lasted a really long time”
- “You hardly look like it hurts at all”
- “Just see if you can go a bit longer”
- “You can do it”
- “Lets see if you can last 10 more seconds”
- Sibling does fist pump, high five, thumbs up, or says “Yay”, etc. in response to sibling procedure related behaviour.
- “You’ve had pain before, you can keep going”
- “Don’t you dare take your hand out” (in light hearted/encouraging tone)

8. Symptom Focused Talk and Behaviours (OSymp)

This refers to any questions or comments about the participating siblings’ symptoms of pain, cold or fear related to the CPT, or responses to their sibling’s symptom talk. This might also include discussing how cold the water is or making comparisons regarding the water temperature.

Examples:

- “Does it hurt?”
- “How cold is it?”
- “Does it feel colder than the ocean?”
- “How bad does it hurt?”
- “Is your hand numb yet”
- “My hand felt tingly”
- “Are you nervous about keeping it in?”
- “You look like it hurts”
- “Your hand is turning red”
- “Yikes” (in response to sibling symptom complaint)
- Child feels siblings arm to see if it’s cold

9. Symptom and coping related self-reflections and predictions (OSelfSymp)

This refers to siblings predictions of how they will feel (e.g., symptoms of pain, fear) or cope during the cold pressor task, reflections regarding how they felt/cope while they did the cold pressor task, or their symptoms (e.g., fear) while watching their sibling do the task. This may involve a comparison to the sibling (note: light hearted competition is coded as OComp).

Examples:

- “I bet it’s going to hurt a lot when I do it”
- “I’m scared about having to do it next”
- “I’m scared just watching”
- Shakes hands nervously
- “Yes” (in response to “was your hand numb when you did it”)
- “Do you think I will be able to handle it like you?”
- “What if I can’t do it?”
- “I think I’ll do okay at it”
- “I think it’s going to hurt me more than you”
- “I was more scared than you are”
- “What if I can’t take it when I put my hand in?”

10. Criticism and Conflict (OCrit)

This refers to any negative comments or behaviours that are meant to criticize or put down the sibling with regard to their task behaviour, their behaviour outside of the lab, their characteristics or personality. It could also include negative facial expressions or gestures toward the sibling. Competitive statements or sarcastic comments that are harsh in tone or negative imitations of the sibling can also be included. This also refers to statements or behaviours indicating conflict or frustration between the siblings, such as commands in a harsh tone of voice, ignoring the sibling, or physical aggression.

Examples:

- “You suck at this”
- “You’re such a whiner”
- “You have no pain tolerance”
- “You look ridiculous right now”
- “You’re doing it wrong”
- “You’re always so annoying”
- “You’re being weird today”
- “You were so bad at rolling the paper for the tower”
- Imitates siblings’ facial expression/behaviours
- Makes rude face at sibling or rolls eyes in response to sibling comment
- “Whatever, I’m ignoring you”
- “I’m telling Mom you did that”
- “Stop it, you’re going to get us in trouble”
- “Ugh” (sound of frustration)

- Hitting, punching, pushing, kicking sibling’s chair, etc.
- “I’m going to last so much longer than you” (in harsh tone)
- “That’s a stupid idea” (in response to sibling’s suggestion for tower task)
- Laughing at sibling in harsh way/in response to sibling distress
- “Why are you making this go on forever” (in frustrated tone)

11. Fidgeting and Restlessness (OFid)

This includes any displays of fidgeting or restlessness by the observing sibling.

Examples:

- Swinging legs in chair
- Shifting around position in chair
- Getting up and walking around (with no obvious intent)
- Bouncing up and down in chair
- Bouncing knee
- Tapping fingers
- Tapping feet
- “Doodoodoodoo” (hums or repeats sound; not intended to be humorous or distracting)

12. Other (OOth)

This refers to any comments or behaviours that do not clearly fit into any of the other categories. This might also include comments that are inaudible, behaviours that are cut off from view, requests to repeat something, or behaviours/comments for which the intent is unclear.

Examples:

- “Hmm”
- “Okay”
- “What was that?”
- “Huh?”
- Child scratches arm

Participating Sibling

1. Distraction/Non procedure related talk (PDis)

This code refers to comments or behaviours that are intended to distract themselves from the pain or task. This might include discussing a topic unrelated to the task (e.g., evening plans, a past event), pointing to/discussing the cameras or paintings in the room (i.e., non-task related materials), or discussing other aspects of the study (e.g., the questionnaires, tower task, etc.). It might also include engaging in a verbal or nonverbal game (e.g., hand games like rock, paper, scissors, repeat after me), or singing a song (if the song is intended to be funny or a joke, code as Humour [PHum]). This behaviour may be

initiated by themselves or by their observing sibling. This also includes engaging in a sibling's attempt to distract.

Examples:

- “What are you doing tonight when we get home” (*discusses plans*)
- “Look at the cameras over there”
- Points at camera, art work, etc.
- “I think that’s abstract art”
- “Why do you think there’s so much tape on the ground”
- “Help me distract myself”
- “I need to get distracted”
- “Play a game with me”
- “What did you think about the Patriots losing?”
- “Do you think we’re going to have the tallest tower?”
- Sings a song
- Plays rock, paper, scissors
- Thumb war
- “Lets ask Mom if we can get pizza for dinner”

2. Humour (PHum)

This refers to any attempt (verbal or non-verbal) to be humorous or make the sibling laugh, and will usually be light hearted in tone and result in laughter/smiling. Humour can be distinguished from criticism (PCrit) based on the tone of voice, and/or whether it is received positively by the sibling.

Examples:

- Funny or silly faces
- Tells a joke
- “It’s sucking my hand in!” (*in humorous tone*)
- “I’m pretty sure my hand is almost frozen right off” (followed by laughter)
- Makes an outrageous or sarcastic comment, e.g., “I think I’m going to drop out of school and become a celebrity”
- Sings song or dances in a way that is intended to be humorous
- Laughter
- Makes a funny sound

3. Competition/light hearted teasing (PComp)

This includes competitive statements that are intended to be friendly/fun/humorous as well as light hearted teasing of the sibling or of themselves. This code can be distinguished from criticism (PCrit) in that it should be received positively by the sibling.

Examples:

- Imitating sibling’s facial expression, behaviours, comments, etc.,

- “I’m doing so much better than you”
- “I bet that you won’t last as long as me when you do it”
- “When it was your turn you were like oh my god!!”
- “Your face looks so funny right now”
- “You’re going to be crying when it’s your turn”
- “You only lasted like a few seconds”
- “I think my time is going to be better than yours”
- “Lets see who can do it longest”
- “I’m such a baby” (in lighthearted tone)
- “You’re so bad at helping me”
- “You’re supposed to be nice to me”
- “You look more scared than me right now”
- “I don’t care if this is on camera” (making fun of self)
- “no” (in response to “take it out already”)

4. Coping Talk, Requests and Behaviours (PCope)

This refers to comments or behaviors that indicate ideas for, or engaging in, a coping **strategy**. This may involve discussing with the observing sibling things that they are doing to help themselves cope or feel better. This also includes requests to the observing sibling for help with a coping strategy, ideas for coping strategies, etc., as well as responses related to the observing sibling’s coping related comments or behaviours (note: a specific request to distract in order to cope, feel better, etc. is coded as PCope, however the ensuing conversation would be coded as PDis; general self praise, encouragement or positively reframing is coded as OProPraise).

Examples:

- “Ya it feels a bit better when I do that” in response to sibling modeling or giving an idea for a coping strategy
- “I’m moving my fingers around” or moves hand around
- “It feels warmer when I make a fist”
- “What do you think would make it feel warmer?”
- Takes deep breaths
- Gives self a hug/rubs arm in attempt to warm up
- “Maybe if you hold my hand it will help”
- “I’m keeping my hand really still so I can’t feel the cold as much”
- “I just need to keep thinking about the \$20”
- “Distract me from the pain”

5. Procedure Related Talk/Attending Behaviours (PProTalk)

This refers to any talk or behaviours that draws attention to the cold pressor equipment or procedure (discussion of symptoms is coded PSymp). This might include discussing aspects of the procedure (e.g., putting hand in the water, using the towel, the amount of time it has been) or questions or comments about the equipment, or discussing their

previous experience observing their sibling doing the cold pressor. It might also include behaviours that draw attention to the task (note the behaviours must be clear, i.e., leaning or clearly looking in vs just gazing at the cold pressor). Talk about their perception of the task/procedure is also included.

Examples:

- “How much time do you think it’s been”
- “Where do you think they get the machine?”
- “I think I see a pump”
- “I can take my hand out whenever I want, right?”
- “Do you think I can grab the towel now?”
- “How do you think she knows to come back in when we’re done?”
- “I think the buttons are how they control the temperature”
- Child clearly examines the cold pressor machine
- “I feel like the time went faster when I was watching you”
- “When you did it he told you to take your hand out”
- “I think it’s probably been about a minute”
- “I think I need to roll up my sleeve more”
- “The machine is noisy”
- “The water looks like it’s bubbling”
- “I was supposed to use my left hand right?”
- “Yes” in response to sib asking “*did you put it in to your wrist?*”
- “Part of my arm touches the side”
- “This is boring”
- “This is actually fun”
- “I wish I had been watching first”

6. Self procedure and symptom related praise/encouragement/positive reframing (PProPraise)

This refers to any comments or behaviours that children make to praise themselves regarding their own procedure related behaviours/performance, encourage themselves in completing the cold pressor task, or attempts to positively reframe/put a positive spin on their experience/symptoms. These may be in response to procedure related praise/encouragement made by the observing sibling. This can be distinguished from procedure related talk (PProTalk) by the tone of voice (i.e., if it’s encouraging/proud/positive vs. matter of fact).

Examples:

- “I’m being braver than I thought I would”
- “I think I’m doing good”
- “I think I’ve actually kept it in for a long time”
- “I think I can keep going for a while”
- “I know I can go a bit longer”

- “I can take this”
- “I’m going to see if I can last 10 more seconds”
- Sibling does fist pump, high five, thumbs up, or says “Yay”, etc. in response to sibling procedure related praise/encouragement.
- “It’s doable”
- “Hey it’s not so bad”
- “It’s not as painful as I thought it would be”
- “It’s not as painful as gymnastics”

7. Symptom Focused Talk and Behaviours (PSymp)

This refers to comments or behaviours about the participant’s current symptoms of pain, cold, physical sensations or fear related to the CPT, or responses to their sibling’s symptom talk. This might also include discussing how cold the water is or making comparisons regarding the water temperature.

Examples:

- “Ouch!”
- “Ow ow ow ow”
- “It’s freezing cold”
- “It feels colder than the ocean”
- “It hurts so much”
- “My hand is totally numb”
- “My fingers feel tingly”
- “I’m scared I’m going to get frost bite”
- “I’m nervous to keep it in too long”
- “My hand is turning red”
- “It does feel pretty cold”
- “Yikes”
- Shakes other hand nervously
- Makes a facial expression communicating pain or fear
- “Yes” (in response to “does it hurt?”)
- “It feels sticky”
- “My legs are numb from the chair”
- “It’s starting to feel warm”

8. Resistance (PRes)

This includes any comments about wanting to take their hand out or stop the CPT.

Examples:

- “I think I have to take my hand out”
- “I can’t do this any more”
- “I want to stop so badly”
- “How did you last so long?”

- “I don’t think I can do it for as long as you”
- “Not very long” (in response to “how long do you think you will last?”)

9. Criticism and Conflict (PCrit)

This refers to any negative comments or behaviours that are meant to criticize or put down the observing sibling with regard to their task behaviour, their behaviour outside of the lab, their characteristics or personality. It could also include negative facial expressions or gestures toward the sibling. Competitive statements or sarcastic comments that are harsh in tone or negative imitations of the sibling can also be included. This also refers to statements or behaviours indicating conflict or frustration between the siblings, such as commands in a harsh tone of voice, ignoring the sibling, or physical aggression.

Examples:

- “You hardly kept your hand in at all when you did it”
- “You’re going to suck when it’s your turn”
- “You have no pain tolerance”
- “You look ridiculous right now”
- “You’re supposed to be being nice to me you know”
- “You’re always so annoying”
- “You’re being weird today”
- “You were so bad at rolling the paper for the tower”
- Imitates siblings’ facial expression/behaviours
- Makes rude face at sibling or rolls eyes in response to sibling comment
- “Whatever, I’m ignoring you”
- “I’m telling Mom you did that”
- “Stop it, you’re going to get us in trouble”
- “Ugh” (sound of frustration)
- Hitting, punching, pushing, kicking sibling’s chair, etc.
- “I’m going to last so much longer than you” (in harsh tone)
- “That’s a stupid idea” (in response to sibling’s suggestion for tower task)
- Laughing at sibling in harsh way
- “I’m going to keep it in just to make you wait” (in harsh tone)

10. Other (POth)

This refers to any comments or behaviours that do not clearly fit into any of the other categories. This might also include comments that are inaudible, behaviours that are cut off from view, requests to repeat something, or behaviours/comments for which the intent is unclear.

Examples:

- “Hmm”
- “Okay”
- “What was that?”
- “Huh?”

- Child scratches arm

Instructions and detailed descriptions of behaviour codes for the tower-building task:

- Each video should be watched 3 times. On the first pass you should just observe the interactions, without providing scores. On the second pass, the behaviour of the older sibling should be coded, and on the third pass the behaviour of the younger sibling should be coded. The dyad-level codes should also be rated on the third pass.
- Each of the codes should be rated as present or absent per 5-minute interval, across the 15-minute task (i.e., resulting in 3 scores for each code).
- A total score will be calculated for each sibling dyad for positivity and negativity by summing the codes across the intervals.

Positivity

1. Validation (Val)

This code is provided for each sibling, and includes comments or behaviours (e.g., gestures) that are meant to be reinforcing of the sibling, their efforts, or their ideas. These comments/behaviours may be related or unrelated to the tower task. This might include providing a compliment, respectfully considering a sibling's idea, or making an encouraging/supportive comment about the sibling or their ideas.

Examples:

- “that’s a good idea”
- “I think we should try it your way”
- giving sibling a high five or thumbs up/fist pump in response to an idea
- “I think that would work”
- “you did great at basketball” (or reference to some other activity/achievement)
- “you were so nice to that girl”
- “yay” (in response to sib’s idea)
- jumps up and down or nonverbal excitement (in response to sib’s idea)
- “you’re so smart”
- “wow, how did you think of that?” (in excited/supportive tone)
- “You did so good/you’re going to do good at the cold water part”
- “You’re good at cutting the paper”

2. Engagement (Eng)

This code is provided for each sibling, and refers to comments and behaviours that demonstrate motivation and diligence in working with the sibling, and enjoyment of the task. This might also include comments referring to making a plan or a strategy for building the tower, as well ideas for acting on the plan. This score should also take into account the child's level of focus on working on the tower during the segment. This code can be distinguished from co-operation in that it is more focused on the individual child's behavior (i.e., “I” behaviour and comments) as opposed to dyadic behaviour (i.e., “We” behaviours and comments).

Examples:

- Smiling while working on the task
- “This is fun”
- “Okay, lets do this” (in reference to the task)
- “I’m going to roll up all of the papers, and then tape the straws on top”
- “First I’m going to get all of our tape ready”
- “Lets get down to work, we don’t have much time”
- “How much time do you think we have left?”
- “Lets see if we can be the tallest”
- “Do you think we have the tallest tower yet?”
- Behaviours contributing to building the tower such as cutting tape, rolling paper, putting things together, etc.
- “I cut them this way so they would be taller”
- “This is awesome”
- “This tower is going to be the tallest”

Warmth (War)

This code is provided for each sibling, and refers to comments or behaviours that demonstrate affection towards the sibling or enjoyment of the sibling. This might also include apologies toward the sibling (e.g., in response to being bossy, making a negative comment, etc.), use of humour, or light hearted teasing (i.e., that is clearly intended to be funny/friendly and is received with a positive verbal or nonverbal reaction such as laughing or smiling). This category also includes positive references to engaging in activities or interacting with the sibling in daily life (i.e., evidence of being involved in one another’s lives and having a positive relationship).

Examples:

- Laughter or giggling (that is not intended to be mean and is not upsetting to the sib)
- Singing (that is not intended to be annoying or block out the sib)
- Jokes
- “you’re the best”
- “you’re so funny”
- “I’m so glad we’re doing this together”
- giving sib a hug
- patting sib on the back
- smiling at sib
- “I’m sorry, I didn’t mean it”
- “I’m so happy you were there for the cold water part”
- “you looked so funny during the cold water task” (received with laughter)
- “I can’t wait for when we go to the beach”
- “What should we buy Mom for her birthday?”

- “Do you remember when we were at Nan’s house?”
- “Do you want to play [names game] tonight?”
- “Thanks for helping me with my homework”
- Shares story about previous experience
- “it’s okay, we will try something else” (in response to sib’s idea not working)

Cooperation (Coop)

This score is provided for each sibling dyad, and refers to the sibling’s collaboration and communication during the tower task. This might include sharing task materials, deciding how to divide up tasks (this can be differentiated from bossiness by both siblings contributing to or demonstrating positive receptiveness to the division of tasks), teaching siblings how to do something, asking siblings’ opinion, or behaviours/comments reflecting that the siblings are working together. This code should also reflect the degree to which they discuss with one another throughout the task (e.g., as opposed to working silently/independently). Note, some siblings might collaborate by dividing up tasks while others might work together on the same task – the degree of positivity while working together on the task should be taken into account when assigning scores for this code. This code can be distinguished from engagement in that it is more focused on dyadic behaviour (i.e., “We” behaviours and comments) as opposed to the individual child’s behavior (i.e., “I” behaviour and comments).

Examples:

- “We’re the best team”
- “Okay, lets do that” (in response to sib’s idea)
- One sibling holds a roll of paper while the other tapes something on
- “How about you cut the tape and I will roll the paper” (received with positive behaviour or comment)
- “Maybe we can both fold the straws”
- “If you trace circles than I can cut them out”
- “We can take turns using the scissors”
- Siblings pass materials between them (e.g., trade off using tape)
- “Here let me help you with that”
- “I can show you how to roll the paper”
- “What colour paper do you want to use?”
- “Lets put these elastics on here”, followed by other sib putting on elastics
- “What can we do to make our tower taller?”

Negativity

1. Criticism of Sibling (CritSib)

This code is scored for each sibling, and refers to comments or behaviours that indicate criticism of the siblings' character. The comments and behaviours must be specific to the sibling themselves, rather than their ideas regarding the task (which is coded separately). However, criticism of sibling ideas outside of the task would be coded as criticism of sibling (e.g., criticizing sibling's ideas for an activity that night). Examples might include statements about the sibling's personality, looks, or previous or current behaviour (e.g., during previous experiment components or outside the lab). It also might include negative facial expression or body language directed at the sibling.

Examples:

- "You're stupid"
- "You never do anything right"
- "You're not fast enough"
- "You're such a baby"
- "You were so bad at the cold water task"
- "Stop being so annoying"
- "Your outfit looks dumb today"
- "You suck at [names game or activity]"
- "Why are you being so weird?"
- "I bet you you're only going to last a few seconds at the cold water" (stated in a negative way and/or received negatively)
- rolls eyes or makes smirk in response to sibling comment
- "That would be so boring" (in response to lets watch a movie tonight)

2. Criticism of Task Ideas or Behaviour (CritTask)

This code is scored for each sibling, and refers specifically to criticism of the sibling's ideas or behaviours that are related to the tower building task. This might include comments about the sibling's behaviours or ideas, or directives or behaviours that indicate the child isn't pleased with their sibling's performance (e.g., taking over doing a task that the sibling was doing). These comments will likely be negative or harsh in tone, and non-verbal behaviours will communicate a negative feeling/intent.

Examples:

- "That won't work"
- "Hurry up"
- "Do it faster"
- "We're running out of time"
- "That's a dumb idea"
- "You're not doing it right"
- "It won't be sturdy if we do that"
- "You keep forgetting about the base"

- “We’re not allowed to do that”
- “You’re not even making it taller”
- “You keep making it fall over”
- Grabs scissors from sibling and starts cutting (e.g., in response to sibling attempting to cut)
- “Here let me do it” (in negative/harsh tone)
- Nudging sibling to get them to work faster
- “You suck at that” (referring to sibling behaviour related to the tower task such as cutting, rolling paper, etc.)
- Rolling eyes or ignoring in response to a sibling’s suggestion
- “Stop doing it that way”
- “But I’m better than you at cutting”
- “No, not like that”
- laughing when sib’s idea doesn’t work

3. Conflict (Con)

This code is scored for the sibling dyad, and refers to verbal or physical conflict (e.g., aggression, arguing, threats). This code should take into account the back-and-forth nature of conflict (i.e., conflict usually is inferred based on negative comments and responses from both siblings). This might also include the siblings ignoring one another (e.g., not responding to their sibling’s comments or questions, refusing to work on the task), or siblings being bossy.

Examples:

- “Gimme that” (if this is said to take over a task, code as CritTask)
- “I’m not listening to you right now”
- “I’m ignoring you”
- “Fine I’m not doing this anymore”
- Child intentionally looks away or keeps working in response to their sibling’s question or comment
- “I hate you, you’re always mean”
- Pinching or poking
- Kicking sibling’s chair
- Punching
- Throwing materials at sibling or elsewhere
- Destroying the sibling’s work (e.g., ripping up paper)
- Grabbing items out of a sibling’s hand (if this is done to take over a task, code as CritTask)
- “I’m telling Mom if you keep doing that”
- Raising fist at sibling
- Pretending they are going to hit sibling

- “Stop that or I will hit you”
- “Urghh” (sound of frustration)
- “Stop it”
- “Don’t do that”
- “I said to put it here” (in harsh tone)
- “No, we’re doing it my way”
- Sib 1 “This sucks”, Sib 2 “So what it doesn’t have to be perfect!”
- “No, we’re supposed to be working together!”

Data extraction manual for the scoping review:

Objective: The purpose of this review is to summarize and map the type of research that has been conducted examining siblings and pediatric pain in order to identify gaps in the literature and directions for future research.

Question: What are the characteristics of research studies that have explored siblings in pediatric pain?

Inclusion and Exclusion Criteria

The first step is to determine whether or not an article should be included in the review.

Inclusion Criteria:

- Pediatric studies: the sample is composed of children ages 0-18, adults (e.g., parents, health care professionals) reporting on children, or adult retrospective studies (i.e., adults reflecting on their experiences as children)
- Siblings must be of primary interest. Specifically, the title and/or abstract must identify that the study examines siblings (e.g., they report an analysis comparing sibling functioning, siblings report on outcomes, etc.)
- Pain/a pain condition (e.g., abdominal pain, migraines) must be of primary interest. Specifically, the title and/or abstract must identify that the study examines pain or a pain condition. Studies examining experimental, acute, chronic, or procedural pain are all included
- Published & unpublished literature (e.g., dissertations, case studies, conference abstracts) are included
- All studies that report empirical data or synthesize data using any methodological design (e.g., quantitative studies, qualitative studies, reviews) are included. Note that reviews must only include studies that meet the criteria above (i.e., reviews looking at siblings and chronic illness generally – with or without pain – would be excluded).
- Studies published, or in the case of unpublished literature written, up until the date of the search.
- Written in English (Note: this was added after the search was conducted during the screening process).

Exclusion Criteria:

- Studies on families where siblings are not a specific focus (e.g., families described broadly with no specific outcomes/analyses related to siblings)
- Healthy siblings included only as a healthy control group
- Case studies reporting on more than one sibling who have the same illness (e.g., a case study on a genetic condition)

- Studies referring to pain in an emotional sense (e.g., a mental health condition such as depression being emotionally painful, trauma as an emotionally painful experience)
- Studies focused on cancer related pain
- Commentaries (i.e., opinion or reaction/reflection based publications), letters to the editor

Data Extraction:

Data should be extracted according to the categories listed below. Note that some categories include subcategories.

- 1) Name of Article: List the name of the article
- 2) Authors: List the names of the authors
- 3) Year of Publication: note the year the study was published or written (in the case of unpublished work) OR indicate not available
- 4) Geographic location of study conduct/authors: Note the country in which the study took place (or if not listed, the location affiliation of the first author)
- 5) Discipline affiliation of authors: select all relevant affiliations of listed authors
 - Medicine (e.g., anesthesia, NOT including psychiatry)
 - Psychiatry
 - Psychology
 - Nursing
 - Physiotherapy
 - Occupational Therapy
 - Genetics
 - Other
 - Not Listed
- 6) Study Type: Select the type of study
 - Published research study
 - Dissertation
 - Case study
 - Conference Abstract
- 7) Objective: List the stated objective(s) of the study (i.e., the purpose or research questions of interest)
- 8) Methodology: Select all relevant methodology used in the study (note: you can select more than one option)
 - Qualitative – If yes, indicate:
 - Thematic Analysis

- Inductive Content Analysis
- Grounded Theory
- Delphi Coding Procedure
- Interpretive Phenomenological Analysis
- Other
- Not explicitly stated
- Quantitative - If yes, indicate:
 - Descriptive study
 - Measurement study (e.g., questionnaire development)
 - Experimental/Quasi-Experimental (e.g., RCT)
 - Non-experimental (e.g., correlational, regressions)
 - Longitudinal study
 - Cross-sectional study
 - Other
- Review – If yes, indicate:
 - Systematic Review
 - Narrative Review
 - Scoping Review
 - Other

9) Participants: Record the following information about the sample. Note: if a sample includes adults reporting on children, select both the relevant adult and age range of children.

- Sample size – indicate the sample size included in the analyses. Specify if different for different participant groups (e.g., specify if 50 children and 40 parents).
- Age of children – select all that apply (if only the mean is reported, select the category that falls within the mean age)
 - Baby/toddler = < 2 years old
 - Preschool = 2-5 years old
 - Child = 6-12 years old
 - Adolescent = 13-18 years old
 - Not specified = the term child(ren)/pediatric is used without providing exact ages
- Adults – select all that apply
 - Parents
 - Health Professional
 - Teacher
 - Adults reporting retrospectively on childhood
 - Other

10) Sample Characteristics: Select the type of pain population(s) included in the study.

- Acute/procedural – specify the type(s) of acute or procedure related pain
 - Needle/Immunization
 - Blood Draw
 - Post-operative

- Other (e.g., injury, acute illness, dental procedure)
- Chronic/disease related – specify the type(s) of chronic pain/disease as stated in the study
 - Chronic Pain (i.e., the term “chronic pain” is used to describe the pain sample, without listing specific diagnoses)
 - Irritable Bowel Disease/Syndrome (IBS)
 - Inflammatory Bowel Disease (e.g., IBD, Chron’s, Ulcerative Cholitis)
 - Migraine/Headache
 - Juvenile Arthritis/Rheumatic Diseases
 - (Functional, Recurrent) Abdominal Pain
 - Back Pain
 - Musculoskeletal
 - Growing Pains
 - Sickle Cell Disease
 - Other
- Experimental
 - Cold Pressor
 - Quantitative Sensory Testing
 - Water Load Task
 - Other
- Co-morbid Conditions/Others Conditions of Interest (e.g., epilepsy, ADHD, Down Syndrome): select if the sample includes children with co-morbid conditions (e.g., pain is being studied in a group of children with a comorbid, non-pain related condition) or also includes a sample with other conditions of interest (e.g., children with a pain condition are being compared to children with a non-pain related condition)
- Healthy Children: select if the sample includes a group of healthy children (e.g., healthy siblings, healthy children experiencing experimental or acute/procedural pain)

11) Outcomes Assessed – select all of the outcomes examined.

- Demographic variables (e.g., SES, family composition; note this does not include age)
- Quality of life
- Mental health/psychosocial functioning (e.g., anxiety, depression, self-esteem)
- Adaptive functioning/disability
- Sibling relationship quality
- Parent-child relationship quality
- Parent marital relationship quality
- Family functioning (e.g., stressors, resources, daily functioning, family relationship quality)
- Pain or somatic symptoms (e.g., pain severity, condition related symptoms)
- Genetic vulnerability
- Juvenile Arthritis disease features (e.g., type, number of affected joints)(*Note: This category was added in following data extraction, and relevant studies were re-categorized)

- Other: specify

12) Outcome Measurement: Select how outcomes were measured (select all that apply):

- Questionnaire or survey
- Observational measures (e.g., behavioural coding)
- Focus Groups
- Interviews
- Health Records or Medical Results (e.g., data extracted from health records, results of medical procedures, genetic testing, etc.)
- Pain assessment tools (e.g., the Faces Pain Scale – Revised)
- Other

13) Source(s) of information: Select all informants. Note that this category refers to the participants who actually report on outcomes, such as completing questionnaires, participating in observational tasks, etc. (e.g., if the study involves parents completing measures about the healthy sibling and sibling with pain, only parents would be selected here since they are the only ones completing questionnaires).

- Parent: If yes specify
 - Self –report
 - Reporting on others (e.g., children)
 - Behavior observed
- Healthy/Pain free sibling: If yes specify
 - Self –report
 - Reporting on others (e.g., sib w/ pain)
 - Behavior observed
- Sibling(s) with pain/condition: If yes specify
 - Self –report
 - Reporting on others (e.g., healthy sib)
 - Behavior observed
- Health Professional: If yes specify
 - Self –report
 - Reporting on others (e.g., children)
 - Behavior observed
- Other

14) Impact/relationships investigated: Select the category(s) that best represent the impact(s) of pain/relationships in relation to siblings investigated in the study.

- Impact of sibling experiencing pain (chronic or acute) on healthy/pain free sibling - e.g., examining the impact of having a sibling with chronic pain on healthy sibling's functioning, mental health, etc.; this could also refer to studies examining how having a sibling with a pain condition influences a healthy child's likelihood of developing the condition themselves
- Impact of healthy/pain free sibling on sibling experiencing pain (chronic or acute) - e.g., examining how healthy children help their sibling in pain

- Bidirectional (i.e., impact of both of above, or relationships between healthy siblings and siblings with pain on outcomes)
- Impact of/relationship between two siblings with pain/condition on one another
- Mediating impact of parents or family - e.g., examining how parent mental health mediates the impact of child chronic pain on a healthy sibling
- Impact of pain on broader dyadic relationship or functioning - e.g., examining how chronic pain impacts the sibling relationship quality, examining how surgical pain influences the amount of time siblings play together
- Genetic Influence – studies examining siblings within the context of genetic vulnerability for pediatric pain conditions (*Note: This category was added in following data extraction, and relevant studies were re-categorized)
- Other: specify
- Not applicable