SEX AND GENDER IN PEDIATRIC PAIN: PAIN RESPONSES AND PARENTAL MODELING

by

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My interest in mothers and fathers comes from having an incredible example of each. This dissertation is dedicated to my parents, David & Norma Boerner.

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ABSTRACT

Sex differences in pain are well-studied in the adult literature. Women generally demonstrate greater sensitivity and response to painful events, though the strength of the effect differs based on the type of pain and social context. The role that sex (i.e., biological/physiological characteristics of men and women) and gender (i.e., behaviours considered socially appropriate for men and women) play in children's pain has not been systematically examined. Moreover, the role of parent and child sex in parental modeling of pain behaviours on children's pain has not been investigated. The present dissertation addressed these gaps through: 1) a review of the existing literature on sex differences in children's experimental pain; 2) a laboratory-based study to examine the role of sex in the impact of parent pain expression on child pain; 3) an exploratory investigation of the role of parent and child gender in children's pain; and 4) a critical analysis of sex and gender research in pediatric pain. A systematic review and meta-analysis of sex differences was conducted of 81 studies of experimental pain in healthy children and adolescents. Overall, no sex differences were observed with the exception of cold pain intensity, where girls reported significantly higher pain intensity than boys in studies where mean participant age was >12 years, and heat pain threshold, where boys demonstrated significantly higher pain thresholds than girls. To investigate the role of sex in children's learning of pain behaviours from their parents, 168 parent-child dyads (6-8 year old children; 50% fathers, 50% sons) completed a laboratory-based study where children observed their parent's reaction to the cold pressor task, and then completed the pain task themselves. Unbeknownst to their child, parents were randomly assigned to exaggerate or minimize their facial expression, or act naturally during the pain task. Children whose parents exaggerated their expression of pain reported greater anxiety prior to completing the pain task. Additionally, girls who observed their parent exaggerating their pain response self-reported greater overall pain intensity than boys in the same condition. An exploratory analysis of gender found that anxiety in girls was predicted by their self-reported femininity. This research highlights that some sex differences may be present during childhood and adolescence, that parental modeling of pain behaviours may impact their children in a sex-specific way, and that gender is associated with certain pain-related responses.

Keywords: modeling; pediatric pain; child; parent; sex; gender

LIST OF ABBREVIATIONS AND SYMBOLS USED

Adj. R^2	Adjusted R^2 (proportion of variance explained in regression analyses)
ANOVA	Analysis of variance
AU	Action Unit (facial coding)
BSRI	Bem Sex Role Inventory questionnaire
CSRI	Children's Sex Role Inventory questionnaire
cm	Centimetre
CPT	Cold pressor task
F	F-test from ANOVA; F-ratio for regression analyses
f	Cohen's f value for effect size
FACS	Facial Action Coding System
FPS-R	Faces Pain Scale - Revised
I^2	I-squared value of heterogeneity
M	Mean
MANOVA	Multivariate analysis of variance
N	Population sample size
n	Sample size
NR	Not reported
p	P-value for testing significance
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-Analyses (guideline)
QST	Quantitative Sensory Testing
r	Pearson product-moment correlation coefficient
R^2	Proportion of variance explained in regression analyses
SD	Standard deviation
SMD	Standardized Mean Difference
t	t-value for t-tests
V	Pillai-Bartlett trace statistic for MANOVA
VAS	Visual Analogue Scale
Z	Z statistic from meta-analysis
α	Cronbach's alpha (measure of internal consistency)
β	Standardized coefficient for regression analyses
ΔR^2	R^2 change (change in proportion of variance explained in regression analyses)
η_p^2	Partial eta squared (measure of effect size)
χ^2	Chi-squared

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CHAPTER 1. INTRODUCTION

1.1 Review of relevant literature

1.1.1 An overview of pain

Pain is a complex phenomenon that impacts all people. Pain can take many forms, and is defined by the International Association for the Study of Pain as "an unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage" (IASP Task Force on Taxonomy, 1994). Pain is a multifaceted experience that involves biological factors (e.g., tissue damage, central and peripheral sensitization, endogenous pain modulation systems, genetic vulnerability to pain conditions), psychological factors (e.g., catastrophic thinking about pain, sense of control over the pain, expectations and interpretations about the pain experience, psychological responses to pain such as anxiety, depression, helpleness, acceptance, optimism, anger), and social/environmental factors (e.g., contextual factors and the environment, the presence of others, social communication through language/expressions/body posture, social hierarchy and roles, social/cultural expectations). Each of these factors interact to create the sensation of pain that the person experiences, and the expression of pain that conveys this experience to others (Craig, 2009). The function of pain serves as a signal to the individual to attend to some aspect of bodily harm, and therefore the ability to attend to the signals of pain in one's own body, as well as the pain expressions of others in the environment, is evolutionarily adaptive (T. Hadjistavropoulos et al., 2011).

1.1.2 Sex differences and gender influences on the pain experience

A discussion of sex and gender differences in the pain experience would be remiss to not begin by first clarifying the use of such terminology that has plagued the research literature with inconsistencies (National Research Council, 2001). For the purposes of the present dissertation, the terminology used will be according to the World Health Organization, where: *"'Sex' refers to the biological and physiological characteristics that define men and women. 'Gender' refers to the socially constructed roles, behaviours, activities, and attributes that a given society considers appropriate for men and women."* (World Health Organization, 2012). As such, when a study refers to differences between "males/men/boys" and "females/women/girls" and conducts direct comparisons between the two categories, they are generally referring to sex differences. When research involves measuring the extent to which an individual endorses engaging in behaviours or possessing traits that would be considered more stereotypically masculine or feminine (typically achieved through a self-report questionnaire), this would be considered a measurement of gender.

In recent years, sex differences have been one avenue through which researchers have attempted to explain the vast individual variability in the pain experience. There is a long history of sex being ignored in the literature and of basic research being conducted exclusively with male animals, likely in part due to attempts to decrease the heterogeneity in samples that was introduced by female estrous cycles (Mogil & Chanda, 2005; Mogil, 2012). This is an issue that is not exclusive to pain research with one recent review demonstrating a bias towards male subjects in numerous disciplines, most prominently in the area of neuroscience (Beery & Zucker, 2011). However, it is of particular importance in the field of pain, given the robust finding of higher prevalence of chronic pain in women for numerous pain conditions (Fillingim, King, Ribeiro-Dasilva, Rahim-Williams, & Riley, 2009). As such, it has been recommended that all pain research be conducted at least with both sexes, if not just females (Greenspan et al., 2007).

In the literature on clinical pain in adult humans, there are fairly consistent findings that women experience higher rates of chronic pain in more parts of the body, that women experience clinical pain (e.g., post-operative pain, procedural pain) as being more severe, and that women access and respond to interventions for pain differently, including a different profile of the side effects in response to analgesics (Fillingim et al., 2009; Niesters et al., 2010; Unruh, 1996). Sex differences have also been explored in responses to experimental pain, with similar findings to clinical pain with regards to pain sensitivity, though this literature is inconsistent across different pain induction methods and outcome measurements (Fillingim et al., 2009; M. Racine, Tousignant-Laflamme, Kloda, Dion, Dupuis, & Choinière, 2012a). Sex differences in clinical pain appears to change depending on developmental stage, with an emergence of sex differences observed in adolescence and with the trajectory of chronic pain prevalence differing by sex throughout middle adulthood and into later life (Blyth et al., 2001; Johannes, Le, Zhou, Johnston, & Dworkin, 2010; King et al., 2011; Sjøgren, Ekholm, Peuckmann, & Grønbaek, 2009). Numerous mechanisms have been postulated to explain the presence of sex differences in pain (Mogil, 2012), including theories examining social contributions (e.g., masculinity and femininity, gender role socialization and stereotypes(Bernardes, Keogh, & Lima, 2008; Robinson et al., 2001; Robinson et al., 2003)), psychological factors (e.g., differences in catastrophizing and coping, self-efficacy, attention (R. R. Edwards, Haythornthwaite, Sullivan, & Fillingim, 2004; Jackson, Iezzi, Gunderson, Nagasaka, & Fritch, 2002; Keogh, Hatton, & Ellery, 2000; Keogh & Herdenfeldt, 2002; Sullivan, Tripp, & Santor, 2000)), and physiological mechanisms (e.g., genetics, neurobiology, neurochemistry, pain modulation, sex hormones (Mapplebeck, Beggs, & Salter, 2016; Popescu, LeResche, Truelove, & Drangsholt, 2010)). The majority of current literature acknowledges that it is likely a complex interaction between these variables that accounts for the sex differences observed in pain, and that the most appropriate model for describing such differences would incorporate a biopsychosocial perspective.

The social basis of such differences has been receiving increasing attention in the literature as researchers begin to appreciate the multitude of social factors that may interact with physiology to create the differential experience of pain in men and women. Experimenter sex and gender influences have been shown to impact experimental pain responses (Alabas, Tashani, Tabasam, & Johnson, 2012; Vigil, Rowell, Alcock, & Maestes, 2014), as well as assessment and decision-making about pain (L. L. Cohen, Cobb, & Martin, 2014), and recent studies have demonstrated that an interaction between health care professional and patient sex influences patient reports of pain (Vigil & Alcock, 2014).

Beyond sex comparisons, gender also plays a role in the pain experiences and interactions with the health care system for both men and women. Recently, Pelletier and colleagues (2015) found seven factors (masculinity and femininity, level of stress at home, primary responsibility for and number of hours per week doing housework, personal income, and primary earner status) that were associated with sex in an adult illness population. Interestingly, half of women had an androgynous or masculine gender score, but less than a quarter of men had a feminine score. Femininity (but not female sex) was associated with health risk factors, suggesting a unique role of gender over and above sex in understanding health outcomes. With regards to pain specifically, increased femininity and decreased masculinity is associated with increased pain sensitivity (Alabas et al., 2012). Compared to the literature on sex differences, there is far less research examining the relationship between gender and pain. One factor that may have contributed to this lack of research attention is the challenges associated with measuring gender as a construct, as described below in section *1.2.3*.

1.1.3 Pain in children and adolescents

Chronic pain is a highly prevalent health problem in children and adolescents (King et al., 2011) and is associated with a host of detrimental functional and psychological problems, as well as significant economic cost to the family and society (Groenewald, Wright, & Palermo, 2015). Pain is a symptom of nearly every physical illness or disease of childhood, is an iatrogenic effect of many diagnostic procedures and treatments (e.g., allergy testing, injections of medications or anesthetics, insertion of intravenous lines, sutures, venipuncture), and is a primary concern in a number of chronic pediatric health problems (e.g., juvenile arthritis, fibromyalgia, cancer). Even generally healthy children experience pain the form of everyday pains (e.g., bumps, falls) and through routine medical procedures (e.g., immunizations).

Despite the high prevalence and morbidity associated with pediatric pain, it is a relatively new field of research (P. J. McGrath, 2011). The lack of research is likely in part because it was long believed that children and babies were not susceptible to the experience of pain (Unruh & McGrath, 2014). It is now known that not only are children capable of feeling pain, they might actually be the most vulnerable to the negative long-term effects of pain (Schmelzle-Lubiecki, Campbell, Howard, Franck, & Fitzgerald, 2007; Wollgarten-Hadamek, Hohmeister, Zohsel, Flor, & Hermann, 2011). In recent years, the research literature on pediatric pain has flourished, with increasing attention to understanding the biological, psychological, and social factors that are common between adult and child pain, as well as the developmental factors that are unique to the experience of pediatric pain (Caes et al., 2016).

1.1.4 Sex differences and gender influences in pediatric pain

Within the pediatric literature, there has been significantly less attention paid to sex differences compared to the literature on adults, though sex differences have been reported occassionally in the literature since the early years of the field of pediatric pain (Lipsitt & Levy, 1959). A consensus report released by the Consensus Working Group of the Sex, Gender, and Pain Special Interest Group of the International Association for the Study of Pain highlighted the trajectory of sex differences in pain over the course of the lifespan as being a key direction for future research (Greenspan et al., 2007).

The majority of studies in pediatric pain that have considered the impact of sex differences have conducted secondary analyses to examine potential impacts of child sex, though such studies are generally not designed or powered to look at sex differences. There has been some speculation and acknowledgement that sex might be important in pediatric pain, however this often does not extend beyond controlling for the effects of sex in analyses. The few studies that have explicitly aimed to examine sex differences have had inconsistent findings, though some report observing differences between males and females in pain responses as early as infancy (Grunau & Craig, 1987; Guinsburg et al., 2000). One finding that appears consistently is that sex differences in pain appear to emerge around the time of puberty (Finocchi & Strada, 2014; King et al., 2011; LeResche, Mancl, Drangsholt, Saunders, & Von Korff, 2005; MacGregor, Rosenberg, & Kurth, 2011; N. M. Racine et al., 2016; Schmitz, Vierhaus, & Lohaus, 2013). To date, there has been no systematic examination of the literature on sex differences in the experience and expression of pain in children.

Even smaller than the literature on sex differences in pediatric pain is the literature on gender. Only two studies to date have considered the relationship between child gender and their pain responses, with one study finding no association between child gender and pain responses (Vierhaus, Lohaus, & Schmitz, 2011), and the other finding an association between masculinity and pain, but that this relationship was stronger in boys than in girls (Myers et al., 2006). There is a clear need for further research to better understand the impact of child gender on pain responses.

1.1.5 The role of parents in pediatric pain

Parents play a critical role in their children's lives, and are frequently the individual that the child is most likely to interact with during a painful experience (Birnie, Boerner, & Chambers, 2014). Additionally, health professionals often rely on parents to assist with pain assessment and management, as they are seen as experts on their children's experience. Much of the research literature examining pain in school-age children has involved an examination of parents in some capacity, whether that be examining the role of parental behaviour on child pain (Chambers, Craig, & Bennett, 2002; Goodman & McGrath, 2003; Moon, Chambers, & McGrath, 2011) or the concordance of parent's judgments of children's pain with the child's own pain ratings (Chambers, Reid, Craig, McGrath, & Finley, 1998; Goubert, Vervoort, Cano, & Crombez, 2009; Moon et al., 2008; Zhou, Roberts, & Horgan, 2008).

Families provide an important environment within which children experience and learn about pain(Turk, Flor, & Rudy, 1987). In addition to learning experiences that occur around the child's own pain, it has been hypothesized that parental experiences of pain offers an opportunity for children to learn about appropriate responses to painful events (Levy, 2011). A recent meta-analysis demonstrated that having a parent with chronic pain has a significant impact on their children in numerous domains (K. S. Higgins et al., 2015). However, even in the absence of parental chronic pain, the everyday pain experiences of parents still contribute to their children's learning about pain.

Like many other forms of illness and disease, pain has been described to "run in families." Numerous epidemiological studies have described the aggregation of pain complaints in families (Arruda, Guidetti, Galli, Albuquerque, & Bigal, 2010; Groholt, Stigum, Nordhagen, & Kohler, 2003), and found an additive effect of parental pain whereby having two parents with chronic pain is associated with poorer outcomes than only one parent (Kaasbøll, Lydersen, & Indredavik, 2012; Lier, Nilsen, & Mork, 2014; Sherman, Bruehl, Smith, & Walker, 2013). There are certainly medical and genetic explanations that can account for large portions of this relationship, including shared exposure to harmful agents in the environment, a shared genetic predisposition to chronic pain or to a pain-inducing medical condition, or the shared genetics of pain catastrophizing (Hocking et al., 2012; Nielsen, Knudsen, & Steingrímsdóttir, 2012; Trost et al., 2015). However, social factors also play a critical role in pain, as has been demonstrated by both animal and human studies (Mogil, 2015). One example is social modeling: how individuals learn the adaptive or maladaptive pain coping behaviours that promote disability or recovery by observing the behaviour of a model in pain (Goubert, Vlaeyen, Crombez, & Craig, 2011).

The development of chronic pain involves numerous neurobiological and genetic components, many of which have been studied at length (Cservenka, Stein, Wilson, & Nagel, 2015). However, the maintenance of functional disability and psychological distress that is so impairing to many chronic pain patients (and that may in turn increase the perception of pain as being intense or disabling) could likely be prevented or interrupted by addressing maladaptive behaviour patterns, which are often a learned phenomenon (Burdette & Gale, 1988). Authors have also hypothesized that the presence of pain complaints in family members may reinforce an individual's belief that pain is subject to external, rather than internal, control (P. W. Edwards, Zeichner, Kuczmierczyk, & Zeichner, 1985). Therefore, there is a strong rationale to believe that social learning, and specifically modeling of pain behaviours, may play a role in perpetuating the behaviours that may either promote or reduce pain amongst family members (Goubert et al., 2011).

While the social influence on pain plays an important role across the lifespan, childhood in particular is a critical time when children learn about pain from others. Childhood is a period of rapid development in knowledge and understanding, and the responses of adults, peers, and (particularly in the early years) parents have a strong influence on their emerging development of pain understanding, expression and communication (Birnie et al., 2014; Palermo, Valrie, & Karlson, 2014; von Baeyer & Spagrud, 2003). Children have been theorized to learn illness behaviours through two processes: reinforcement and modeling (Merlijn et al., 2003; L. S. Walker & Zeman, 1992; Whitehead, Crowell, Heller, & Robinson, 1994). Reinforcement refers to a consequence as a result of a behaviour that will make the behaviour more likely to occur again in the future (e.g., in the context of pediatric pain, receiving special attention from a parent when displaying pain behaviours will make the child more likely to engage in those same behaviours again in the future (positive reinforcement); similarly, negative reinforcement may also occur, such as reducing the demands expected of a child when they display pain behaviours may also make the child more likely to engage in those behaviours again (Fordyce, 2015)). Modeling refers to a process of learning whereby an individual (the observer) witnesses the consequences of another individual's actions (the model), and learns how to behave when they themselves are in that situation through observation of both the behaviour and its consequences (e.g., in the context of pediatric pain, observing a parent receiving pain relief by rubbing an injured body part will make the child more likely to engage in that behaviour when they have pain themselves; (Osborne, Hatcher, & Richtsmeier, 1989)). In addition to increasing the likelihood of particular pain-related behaviours occurring, both reinforcement and modeling have been suggested to influence the pain experience by drawing an individual's attention to somatic experiences in their own bodies (Achiam-Montal & Lipsitz, 2014; Violon & Giurgea, 1984), and indeed one study found that children of patients with chronic pain were more somatically focused than children of healthy parents (Mikail & von Baeyer, 1990). Reinforcement has been widely studied in the context of the parent-child relationship. The majority of literature on parental reinforcement, also referred to as "solicitous responses" to pain, has found that it is linked to increased pain, disability, and maintenance of symptoms (Achiam-Montal & Lipsitz, 2014; Chambers et al., 2002; Claar, Simons, & Logan, 2008; Peterson & Palermo, 2004; L. S. Walker & Zeman, 1992; L. S. Walker, Claar, & Garber, 2002; Whitehead et al., 1994). While modeling is widely regarded as a critical component of the learning process, it has received less empirical attention.

Social modeling is a process component of Social Learning Theory, originally developed by Albert Bandura (1977). Social Learning Theory describes the reciprocal interaction between personal characteristics and the individual's environment, whereby learning can occur not only through direct trial-and-error experience, but also vicariously through observing the behaviour and consequences of others. Learning from other's modeled behaviour is not simply rote imitation, as it involves selectively attending to, extracting, and integrating information that is relevant to the observer (Bandura, 1977). While reinforcement is involved in the social modeling process, Bandura describes reinforcement as an antecedent to learning (i.e., anticipating a reinforcing outcome motivates an individual to attend to a model) rather than the learning occurring as a consequence of reinforcement (Bandura, 1977).

Social learning has been well-investigated in the context of other aversive experiences, such as anxiety, where the published literature suggests that modeling is one of the primary pathways through which fear develops (Muris, Steerneman, Merckelbach, & Meesters, 1996). From an evolutionary perspective, the tendency to engage in social learning for an aversive experience like pain is very adaptive, as the individual may learn what to avoid to keep the pain from occurring, what to do to help the pain get better, and how to elicit help from others all without having to directly experience the pain themselves.

Epidemiological studies have reported that the presence of a “pain model”, usually an immediate family member with chronic pain, is associated with increased pain complaints in the individual, and chronic pain patients report more pain models than controls (Lester, Lefebvre, & Keefe, 1994; Merlijn et al., 2003; Turkat, Kuczmierczyk, & Adams, 1984; Violon & Giurgea, 1984; Zeichner, Widner, Loftin, Panopoulos, & Allen, 1999). There are a number of limitations with the literature on the impact of parental modeling of pain behaviours that restricts the ability to draw conclusions. Such studies do not take into account the genetic or biological contributions of pain conditions, and therefore are unable to draw conclusions regarding the relative influence of genetics/biology and social learning environment. The majority of studies that have examined the relationship between parental chronic pain conditions and the development of pain in their children have relied on adult participants to self-report their memories of pain models in their childhood, and only a small minority of studies have compared children's reports to collateral reports from their parents (Merlijn et al., 2003). Participant's memories of their exposure to pain models in childhood are likely highly influenced by experiences and knowledge acquired through adolescence and adulthood, and may not accurately represent their childhood experience (Turk et al., 1987). Bruehl and colleagues (2005) compared adult children's reports of their parent's chronic pain history with the parents' own self-reports, and found high sensitivity to detecting chronic pain in their parents, but only modest levels of specificity, with offspring generally overestimating their parent's chronic pain. Interestingly, reports of paternal chronic pain history appeared to be more reliable than maternal history, and offspring tended to provide more reliable reports for an opposite-sex parent (i.e., males provided more reliable reports of maternal history and females provided more reliable reports of paternal history (Bruehl et al., 2005)). Another study found that females tended to demonstrate increased sensitivity in accurate reporting of familial headache history than males (Ottman, Hong, & Lipton, 1993). Such issues highlight the importance of research that examines the consequences of observing modeled pain behaviour in vivo (as it occurs) to more rigorously examine this relationship, rather than relying on retrospective reports, as well as the importance in considering the sex of the reporter and the pain model. Research using experimental design offers one means by which this can be achieved.

The first experimental study that attempted to examine the relationship between parent and child pain behaviours was by Thastum and colleagues (1997), who compared the responses of a small sample of parents and children to the cold pressor task. They concluded that there was a relationship between parent and child pain intensity and tolerance, but the design of the study (primarily designed to compare the responses of healthy children and parents to children with juvenile arthritis and their parents) limited any conclusions as to whether this association was due to a shared sensitivity to pain or to learning factors. Additionally, despite the fact that data was collected from mothers or fathers, the small sample size precluded any analysis of sex-based differences in the association between parent and child pain.

Goodman and McGrath (2003) expanded on this work by experimentally manipulating parent behaviour and observing outcomes of parental modeling in real-time. In this study, mothers of 10-14 year-old children were randomly assigned to minimize their display of pain during a pain task, exaggerate their display of pain during a pain task, or were given no specific instructions about how to act (control condition). Children observed their mother complete the pain task, and then completed the pain task themselves. Results indicated that children whose mothers exaggerated their expression of pain reached their pain threshold sooner than children in the control condition. Additionally, children whose mothers minimized their display of pain also displayed decreased facial expressions in response to the experimental pain task. This study provided the first experimental evidence that even a brief exposure to maternal modeling of pain behaviours had an impact on the children's own behavioural reaction to the pain experience. No sex differences were observed based on the sex of the child in this study, however, as will be described in the following section, the fact that all pain models were mothers may have limited the extent to which the modeled behaviour had an impact on their child.

1.1.5 The relevance of child and parent sex and gender in the context of pain

There is clearly a strong theoretical and empirical basis to support social modeling as an important aspect of the process of behavioural learning. However, as individuals are exposed to numerous potential models every day, it is important to consider what might make a particular model more salient to an observer, and in turn, what might make the model's behaviours more likely to be integrated into the behavioural repertoire of the child. Several factors have been theorized to be implicated in this process, including the characteristics of the model and observer (e.g., status, competence, dependency, relationship between model and observer), and the value of the behaviour to the observer (Bandura, 1977; Rosekrans, 1967), with a strong rationale for parental modeling of behaviours being particularly salient to their children (Goubert et al., 2011)..

One factor theorized to be important for children in determining which models to attend to is sex. From a very early age, children have been shown to be aware of their sex, as well as the typical behaviours and attributes associated with a particular sex (Maccoby, 1988). There is a strong rationale that this characteristic would be used by children to determine the relevance of a particular model to themselves and that social learning is an important process in development of gender-congruent behaviour (Bussey & Perry, 1982; Mischel, 1966; Perry & Bussey, 1979).

One of the seminal studies of social modeling, the classic "Bobo doll" experiment by Albert Bandura and colleagues, described that there was a greater influence of observing male model on boy's and girl's aggressive behaviour than a female model (Bandura, Ross, & Ross, 1961). However, when verbal aggression was studied separately from physical aggression, children appeared to be more influenced by a model that was the same sex as them. The authors suggested that both the sex of the model, as well as the extent to which the behaviour in question is “gendered” (i.e., associated with a particular sex), should be considered as relevant. An experimental study by Perry and Bussey (1979) found that children were more likely to imitate models that are considered to behave in a way that is appropriate for the child’s sex, but that while girls will imitate any model who displays typically feminine behaviour, boys will only imitate models who are both male and displays typically masculine behaviours.

In the anxiety literature, a questionnaire-based study by Muris and colleagues (1996) found that mothers expressed more fears in the presence of their children than fathers. Additionally, they found there was a significant relationship between maternal expressions of fear and children’s own fearfulness, but no such relationship was observed with fathers. The authors refer to the phenomenon of “social referencing” in explaining that young children look to their mothers for emotional information in ambiguous circumstances (Muris et al., 1996). Another study found that both boys and girls who observed a maternal model prior to dental treatment showed a reduction in heart rate compared to children who observed a paternal model, whose heart rate was more comparable to a control group (Farhat-McHayleh, Sabbagh, & Souaid, 2007). As pain and anxiety are highly related (Rhudy & Meagher, 2000) and similar sex differences with behavioural expression of pain has been observed (i.e., women express more pain than men (Keogh, 2014)), it is likely that a similar relationship may be present in pain. Unfortunately, the focus on maternal-child relationships in the pain context has, to date, prevented an exploration of the role of fathers in social modeling of pain behaviours.

In the context of the pain experience, gender socialization theories have suggested that through modeling and reinforcement, children learn gender-appropriate pain responses, and that identifying strongly with a particular gender role is associated with more gender-conforming pain behaviours (Myers, Riley, & Robinson, 2003; Pool, Schwegler, Theodore, & Fuchs, 2007). While the majority of research has only examined the impact of maternal pain history on their children's own pain experiences, the limited literature examining this relationship in fathers has had mixed findings (Evans & Keenan, 2007; Jones, Silman, & Macfarlane, 2004; Kaasbøll, Ranøyen, Nilsen, Lydersen, & Indredavik, 2015; Sherman et al., 2013). In the broader illness literature, children have been demonstrated to be at greater risk for negative outcomes when a same-sex parent is seriously ill with this relationship particularly relevant for boys (Barkmann, Romer, Watson, & Schulte-Markwort, 2007). Pothmann and colleagues (1994) found that children reported a higher incidence of headache in their same-sex parent (i.e., boys reported a higher incidence of headache in their fathers than mothers and vice versa for girls); despite the fact the overall rates of headache were higher amongst mothers than fathers. Deubner (1977) also found that the presence of maternal headache was associated with equal increased risk of migraine in girls and boys, while the risk of migraine was greater for boys when fathers experienced migraines. Evans and colleagues (2010) presented data from qualitative interviews of children with chronic pain and their mothers that suggested that the impact of parental pain models might be sex-specific, both concerning the sex of the child and of the parent (i.e., pain and disability in girls was associated with the presence of a maternal pain model, while pain and disability in boys was associated with paternal pain models). Taken together, this evidence suggests that the impact of a model of pain/illness behaviour may be sex-linked, and the father-son relationship may be of particular importance. However, these findings could also be explained by biological transmission, rather than psychosocial factors. Little experimental research has directly examined sex-specific effects of exposure to a model of illness behaviour.

In the adult literature, there has been some evidence for sex-specific effects of pain models. Edwards and colleagues (1985) found that pain models had a greater impact on women than men. Similarly, Fillingim and colleagues (2000) found that having a significant family history of pain was related to both increased sensitivity to experimental pain and increased reports of recent pain complaints in women, but not men. The authors hypothesized that women were more attuned to pain in others, making them more likely to be influenced by the modeling of pain behaviours of the people around them (Fillingim et al., 2000). However, it has also been argued that the increased prevalence of chronic pain in women, the presence of menstrual pain, and the fact that it is more socially acceptable for women to outwardly express their pain, means that girls are more likely to be exposed to a relevant (i.e., same-sex) pain model than boys(Hermann, 2007; Koutantji, Pearce, & Oakley, 1998). Experimental research has also shown that the sex of the model can impact pain responses, finding that observing a male model was associated with a greater nocebo hyperalgesia response, irrespective of the sex of the observer, which the authors suggested may have been due to a male model of pain being perceived as a more credible source than female models (Świder & Babel, 2013). However, these findings must be taken with the consideration that sex differences in pain generally are a robust finding in the adult literature, while the pediatric literature is less consistent (Fillingim et al., 2009). Within the child literature, Goodman and McGrath (2003) reported no differences based on child sex, yet all children were only exposed to maternal models of pain.

It is important to consider parents when examining sex differences in children's pain, as literature from the field of social psychology and child development suggest that parents play an important role in their child's gender development and learning of gender-appropriate behaviours (Langlois & Downs, 1980). Gender roles influence how a child learns how to behave during a painful experience (i.e., learning to respond in a manner consistent with others of their sex), and to develop interpretations and expectations about pain in themselves and other people. Fathers have been shown to demonstrate differential treatment to their children on the basis of the child's sex more than mothers, particularly with boys, and to encourage more gender-typed activities (Lytton & Romney, 1991). Gender roles and beliefs held by parents are likely transmitted to their children in the form of differential reinforcement and punishment of sex-typed behaviours, and such gender roles are thought to be stricter for boys than girls (Lytton & Romney, 1991; Williams, Goodman, & Green, 1985). Parents have been shown to provide more physical comfort to girls in response to everyday pains (Fearon, McGrath, & Achat, 1996), and expect boys to learn to tolerate pain more than girls (Kankkunen, Vehviläinen- Julkunen, Pietilä, & Halonen, 2003).

Related to the lack of sex difference research in pediatric pain is the dearth of literature on fathers. The vast majority of studies examining the influence of the family in pediatric pain have exclusively examined mothers. The few studies that have collected samples of fathers have noted numerous differences between mothers and fathers. Fathers have been shown to be more accurate than mothers in rating their child's pain (Moon et al., 2008), catastrophize less than mothers about their child's pain (Hechler et al., 2011), and fathers of children with chronic pain report fewer physical and psychological complaints than mothers (Evans & Keenan, 2007). Fathers have been shown to exhibit a lower priority for pain control than mothers during highly intense pain (Caes, Vervoort, Eccleston, & Goubert, 2012), use more criticism and less symptom-focus talk than mothers (Moon et al., 2011), engage in more discouraging responses to their children's pain and less illness-encouraging behaviour than mothers (Goubert, Vervoort, De Ruddere, & Crombez, 2012; L. S. Walker & Zeman, 1992), fidget more in the presence of their children's pain (Schinkel, Chambers, Caes, & Moon, 2016), have different beliefs than mothers about their children's pain management (e.g., believed that children should learn to tolerate and cope with pain on their own; (Kankkunen et al., 2003)), and the presence of a father has been associated with better pain improvement in their children when accompanying them to the emergency department (Johnston et al., 1998). Additionally, a recent study of parental opioid administration demonstrated that not only did parent sex impact preferences and practices, there was also an interaction of parent sex with child sex, whereby fathers were less likely to withhold opioids for their female children compared to male children (Voepel-Lewis, Zikmund-Fisher, Smith, Zyzanski, & Tait, 2015).

In spite of such differences, the majority of the published literature focuses on samples that consist primarily of mothers, likely due to ease of recruitment. Such a trend makes it difficult to draw conclusions about the role of fathers in pediatric pain, or how the sex differences in adult pain impact the children of these adults. It is critical to include fathers in research for the reasons above, and to reflect the unique and increasing role that fathers play in their children's lives and development (Kochanska & Kim, 2013; Lewis & Lamb, 2003; McWayne, Downer, Campos, & Harris, 2013).

1.2 Methodological considerations in the present dissertation

The use of experimental pain in children and measurement of pain and gender will be explored to outline some of the methodological and ethical issues that are relevant to the study of sex and gender differences and their impact on parental modeling of pain behaviours.

1.2.1 Use of experimental pain in children

While experimental pain has been used in the adult literature for decades, experimentally inducing pain in children is a practice that requires special consideration, given the vulnerable nature of the population (P. A. McGrath, 1993). In recent years, experimental pain induction has been widely used in children to examine issues related to pediatric pain that require experimental control or manipulation that cannot be feasibly or ethically conducted in a clinical setting (Birnie, Caes, Wilson, Williams, & Chambers, 2014). Experimental pain tasks also offer, in many cases, the examination of a novel pain experience that is less impacted by previous experience and expectations than other types of pain (e.g., immunizations) that children have experienced before. A commonly used method of experimental pain induction is the cold pressor task (CPT), which involves the child immersing their hand into a bath of cold water until the experience becomes too painful. The cold pressor task has well-established guidelines for its use (von Baeyer, Piira, Chambers, Trapanotto, & Zeltzer, 2005), and has been used in experimental settings in children as young as 3 years old (Birnie, Petter, Boerner, Noel, & Chambers, 2012). The CPT is widely regarded as being a task that is safe and has a low risk of adverse events, as the temperature of the water and the maximum immersion time means that the experience of cold poses no physiological risk to the child and that the discomfort experienced dissipates quickly upon removal of the hand from the water (Birnie, Noel, Chambers, von Baeyer, & Fernandez, 2011). Additionally, the task is considered ethically acceptable, as children have full control over when to stop the task, and have generally reported very positive experiences completing the task (Birnie et al., 2011). Preliminary research has suggested that the experience of cold pressor pain may be somewhat similar to the pain and anxiety provoked by needle procedures (Boerner et al., E-pub ahead of print) and that CPT outcomes are related to functional impairment (Tsao, Glover, Bursch, Ifekwunigwe, & Zeltzer, 2002).

1.2.2 Measurement of the pain experience

The measurement of the pain experience has long been a methodological struggle for pain researchers and clinicians. As pain is an individual and largely internal experience that is not associated with a specific physiological indicator or test, self-report is considered an important aspect of pain measurement and is often referred to as the "gold standard" (von Baeyer, 2009). However, there are significant challenges associated with measuring pain in children, including challenges in understanding terminology and numerical representations of the pain experience, challenges in communication, and understanding of anchors, to name a few (von Baeyer, 2009). A significant body of the pediatric pain literature has been dedicated to the study of pain assessment in children (Caes et al., 2016) and developing tools to assist children in providing a self-report that is appropriate to their developmental level. The *Faces Pain Scale – Revised*(Hicks, Baeyer, Spafford, van Korlaar, & Goodenough, 2001) is one example of such efforts to devise a developmentally appropriate self-report measure of pain, which has been found to have strength in its psychometric properties (Stinson, Kavanagh, Yamada, Gill, & Stevens, 2006). Due to the challenges in obtaining children's self-report of pain, parents are often asked to provide a proxy report of their child's pain. As parents are intimately familiar with their child's typical behaviours and responses, and are generally involved in caregiving when their child experiences pain, they are thought to be the most reliable source of information after the child themselves. However, previous research has suggested that there is often disagreement between parents and children's ratings of children's pain (Zhou et al., 2008).

Self-reports and proxy reports of pain are clearly subject to numerous types of bias. As such, researchers have attempted to address this by using a multi-modal examination of pain that combines self-report with behavioural measures of pain. The Social Communication Model of Pain states that the internal experience of pain is encoded in the form of behaviours that are observable to others that allow them to draw inferences about the internal experience of an individual with pain (Craig, 2009). The study described in Chapters 3 and 4 attempted to measure these encoded behaviours in the form of pain tolerance and facial expression to complement the self-reported ratings provided by the children. Additionally, by asking parents and children to provide proxy ratings of each other's pain allowed an examination of how an individual's expressed pain behaviours were being decoded by the observer.

1.2.3 Measurement of adult and child gender

Gender is a construct that many researchers have struggled with how to measure, as the definition of gender itself is controversial and constantly evolving. There was a large increase in interest in gender research in the 1970s-1990s, during which a number of measures were developed (Thompson & Bennett, 2015). This raises an issue with the current relevance of such measures, as typical gender roles and perceptions of masculinity and femininity have shifted since that time (Colley, Mulhern, Maltby, & Wood, 2009; Seem & Clark, 2006). Additionally, there are a number of different definitions of gender that may influence how it is measured (Oliffe & Greaves, 2012). The majority of gender measures used in psychological research tend to measure conformity with masculine and feminine stereotypical behaviour, but this differs greatly based on numerous factors such as cultural and societal stereotypes for a particular region as well as current trends. One of the most widely used gender measures, the *Bem Sex Role Inventory,* has been widely used to predict numerous outcomes, including health behaviours, but relies on norms that were collected in the 1970s (Bem, 1974). Recently there has been an increase in the development of measures that assess very specific aspects of gender roles (e.g., as they relate specifically to the pain experience (Robinson et al., 2001)), or either masculinity *or* femininity(Levant, Richmond, Cook, House, & Aupont, 2007; Thompson & Bennett, 2015). However, there are few measures that allow for a broad examination of both masculinity and femininity within the same measure, and that has been validated in the same sample.

1.3 Outline of dissertation papers

The present dissertation aimed to address important gaps in the literature on sex and gender differences in pediatric pain and examine the impact that sex may have on children's learning of pain behaviours via parental modeling. To accomplish this, a systematic review and meta-analysis was conducted, as well as a laboratory-based experimental study looking at sex differences, an exploratory analysis of the experimental study results in the context of gender, and a critical analysis and discussion of the current state of the literature on sex and gender research in pain across the lifespan. These studies are presented as four separate papers in this dissertation (Chapters 2-5). Chapter 6 will provide an overall discussion of the results, their integration with the existing literature, applications to theoretical and clinical settings, and recommendations for future research.

1.3.1 Aims and hypotheses of Chapter 2

The first study, as described in Chapter 2, is a systematic review and meta-analysis of the literature on healthy children's responses to experimental pain tasks. Experimental pain and healthy children specifically were selected to reduce the numerous confounding factors implicated in clinical pain. Studies of healthy/community-based samples of children 0-18 years who underwent a laboratory pain task were included, and data regarding pain outcomes in response to the pain task were extracted. The available literature on sex and gender differences in these pain outcomes were systematically reviewed, and data from all studies (regardless of whether the results of sex-based comparisons were reported in the manuscript; authors were contacted to obtain data split by sex) were pooled to conduct a meta-analysis of sex differences in healthy children's experimental pain outcomes across research groups and experimental methodologies. The meta-analysis is also conducted separately for children and adolescents when sufficient data was available to examine the impact of developmental factors on sex differences in pain. As numerous studies have postulated that sex differences emerge around the time of puberty (King et al., 2011; LeResche et al., 2005), it was expected that no sex differences would be present in child samples, but that they would be reported in samples of adolescents. It was hypothesized that when sex differences were present, they would be in the same direction as those observed in the adult literature (i.e., girls would exhibit greater sensitivity to pain than boys (Fillingim et al., 2009)).

1.3.2 Aims and hypotheses of Chapter 3

The second study, as described in Chapter 3, was a laboratory-based experimental investigation of the impact of observing a parent's behavioural response (exaggerated facial expression, minimized facial expression, or natural reaction) to a painful event on their child's own pain experience and whether differences were present based on parent and child sex. This study strategically addressed the lack of research regarding the role of fathers in pediatric pain (Phares, Lopez, Fields, Kamboukos, & Duhig, 2005), and provided additional understanding of the psychosocial factors involved in the aggregation of pain behaviours in families through a controlled experimental manipulation and multi-method examination of the pain experience. This experimental design provides a significant contribution to a field of literature that has been historically limited by the use of questionnaire-based methods. It was expected that children whose parents minimized their display of pain would experience less pain and anxiety themselves than controls, that children whose parents exaggerated their reaction to the pain would experience more pain and anxiety than controls, and that exploratory analyses of facial expressions would show that children's behavioural responses to the pain would be similar to the display of their parent (Goodman & McGrath, 2003). As previous research has suggested that the impact of pain models is greater on females than males, it was hypothesized that girls would be more influenced by observing their parent's behaviour than boys (P. W. Edwards et al., 1985; Fillingim et al., 2000; Świder & Babel, 2013). Additionally, children observing a same-sex parent were hypothesized to be more influenced by observing their behaviour than children in sex-unmatched dyads (Kanfer, Duerfeldt, Martin, & Dorsey, 1971).

1.3.3 Aims and hypotheses of Chapter 4

Chapter 4 describes an exploratory analysis of the gender-related variables measured in the study described in Chapter 3, and their impact on children's pain experiences. It was expected that gender would offer a unique contribution to our understanding of the pain response, beyond sex differences, and that the directions of such differences would be similar to the previous research in adults (Alabas et al., 2012; Applegate et al., 2005; Otto & Dougher, 1985).

1.3.4 Aims and hypotheses of Chapter 5

Chapter 5 is an editorial on the subject of sex and gender research in pain across the lifespan, discussing strengths, limitations, and conclusions of the research conducted to date, as well as suggestions for future research directions.

CHAPTER 2. SEX DIFFERENCES IN EXPERIMENTAL PAIN AMONG HEALTHY CHILDREN: A SYSTEMATIC REVIEW AND META-ANALYSIS

The manuscript prepared for this study is presented below. Readers are advised that Katelynn Boerner, under the supervision of Dr. Christine Chambers, was responsible for devising the research questions and search strategy, conducting the database searches, developing the inclusion/exclusion criteria and overseeing the screening of abstract results, developing the data extraction system and overseeing data extraction, and contacting study authors for missing data. She was the lead on data analysis and interpretation, with the support of her co-authors, and wrote the initial draft of the manuscript. Prior to submission, she received and incorporated feedback from the study's co-authors. The manuscript underwent peer-review and was accepted for publication, following one revision that Katelynn Boerner led the response to, in *PAIN* on January 30, 2014. The full reference for this manuscript is:

Boerner, K.E., Birnie, K.A., Caes, L., Schinkel, M., & Chambers, C.T. (2014). Sex differences in experimental pain among healthy children: A systematic review and meta-analysis. *Pain*. *155*(5), 983-993. doi: 10.1016/j.pain.2014.01.031

Please note that a corrigendum addressing two errors in the above manuscript was submitted on April 18, 2016, and is included in Appendix B.

2.1 Abstract

Sex differences in response to experimental pain are commonly reported in systematic reviews in the adult literature. The objective of the present research was to conduct a systematic review and meta-analysis of sex differences in healthy children's responses to experimental pain (e.g., cold pressor, heat pain, pressure pain) and, where possible, to conduct analyses separately for children and adolescents. A search was conducted of electronic databases for published papers in English of empirical research using experimental pain tasks to examine pain-related outcomes in healthy boys and girls between 0 and 18 years of age. Eighty articles were eligible for inclusion and were coded to extract information relevant to sex differences. The systematic review indicated that, across different experimental pain tasks, the majority of studies reported no significant differences between boys and girls on pain-related outcomes. However, the meta-analysis of available combined data found that girls reported significantly higher pain intensity compared to boys in studies where the mean age of participants was greater than 12 years. Additionally, a meta-analysis of heat pain found that boys had significantly higher tolerance than girls overall, and boys had significantly higher heat pain threshold than girls in studies where the mean age of participants was 12 years or younger. These findings suggest that developmental stage may be relevant for understanding sex differences in pain.

Keywords: sex differences; experimental pain; children; meta-analysis

2.2 Introduction

Sex differences represent a rapidly growing body of literature in the areas of biology, medicine, and neuroscience, as researchers attempt to illuminate the mechanisms that underlie differences between men and women (Cahill, 2006). According to the World Health Organization, sex refers to the biological and physiological distinctions between women and men. This can be contrasted with gender, which is defined as a psychosocial construct that embodies the attributes, behaviours, and roles that a given society considers to be acceptable for men and women (World Health Organization, 2012).

Sex differences are commonly reported in adult pain, with numerous reviews providing evidence of greater prevalence rates of acute and chronic pain among women, with women also demonstrating greater sensitivity to experimental pain tasks, though the strength of this effect differs between pain modalities, outcome measures, and time points, and is considered to be a controversial phenomena (Fillingim et al., 2009; Mogil, 2012; M. Racine et al., 2012a). The abundance of literature on adult sex differences in pain has allowed researchers to explore mechanisms through which pain differs in men and women, including both biological and psychosocial mechanisms (R. R. Edwards et al., 2004; Fillingim et al., 2009; Keogh et al., 2000; Popescu et al., 2010). Such research has important implications with regards to the assessment and treatment of pain in adults, such as recent advances in theories of "personalized pain management" through research on the differential analgesic responding of men and women (Niesters et al., 2010). Due to developmental factors it is inappropriate to generalize adult findings to pediatric populations, and the literature on sex differences in children's pain is comparatively sparse.

Epidemiological studies of chronic pain in childhood suggest that prevalence of chronic pain is greatest among adolescent girls, with the emergence of sex differences in chronic pain conditions seen around the time of pubertal development (King et al., 2011). These findings are concordant with speculation from the adult literature that sex hormones are one of the mechanisms through which sex differences in pain perception and responding are explained (Aloisi & Bonifazi, 2006; Gupta, McCarson, Welch, & Berman, 2011). Given the complexity of the numerous factors implicated in the development of chronic pain, a systematic review of research on sex differences in healthy children's pain is needed to fully understand and explore potential mechanisms. Experimental pain provides a starting point for such examinations, controlling for many of the confounding factors that complicate interpretations of results in studies of clinical pain. Prior reviews have only provided narrative descriptions of select studies of sex differences in experimental pain among children and adolescents (Keogh, 2012; Moon & Unruh, 2014). The primary objectives of the present study were to: (1) systematically review the existing literature on sex differences in responses to experimental pain in healthy children, and (2) meta-analyze data from published studies on experimental pain in boys and girls to provide a further investigation of sex differences beyond those statistics reported in published articles. Additionally, where possible, meta-analyses were to be conducted separately for children (participant mean age less than 12 years) and adolescents (participant mean age of 12 years or older). Finally, an additional objective was to examine the reporting practices of sex and gender in the studies included in the review.

2.3 Methods

Search method

A search was conducted of key electronic databases (PsycINFO, EMBASE, CINAHL, PubMed) from the inception of databases through November 2012. The basic structure of the search strategy was as follows: [((pediatric) OR child) OR adolescent] AND [pain] AND [(((((((experimental pain) OR cold pressor) OR quantitative sensory test) OR water load) OR heat pain) OR thermal pain) OR pressure pain) OR exercise task], searching primarily titles and abstracts of these key databases, using truncations as appropriate for the database (e.g., child*, adolescen*, quantitative sensory test*). Keywords were chosen to capture the population age range of interest, studies that included pain as an outcome, and to focus the search specifically on studies including an experimental pain task.

Eligibility criteria

Eligibility criteria required that included articles be: (1) An empirical investigation using an experimental pain task to examine pain-related outcomes (pain intensity, pain tolerance, pain threshold, pain affect, facial activity in response to pain, or physiological responses to pain); (2) Published in manuscript form in English; (3) Studies using community/healthy samples of children between 0 and 18 years of age only (or a healthy control group included in studies of clinical populations); (4) Studies that included both boys and girls. Experimental pain tasks were defined as any task that was intended to induce pain for which a pain-related outcome was measured.

Screening for eligibility, coding, and requests for missing data

The initial search revealed 519 unique abstracts, once duplicates were removed. Each abstract was reviewed by two co-authors (K.E.B. and K.A.B.) to determine eligibility. If eligibility could not be determined from the abstract, the full article was examined. A total of 440 abstracts were excluded for the following primary reasons: participants did not complete an experimental pain task (*n*=33, 7.5%), the study did not measure any pain-related outcomes (*n*=8, 1.8%), the abstract was not published in manuscript form (e.g., dissertations, book chapters, conference abstracts, *n*=46, 10.5%), the article was not published in English (*n*=8, 1.8%), the study was conducted with a clinical sample and did not include a healthy control group (*n*=69, 15.7%), the study included individuals outside of the 0-18 years of age range (*n*=254, 57.7%), the study sample was composed of only boys or only girls (*n*=8, 1.8%), the study was conducted with animals (*n*=14, 3.2%).

Therefore, from the initial search, 79 articles were identified as being eligible. Each of the 79 articles were read and data was extracted by a study author (K.E.B., K.A.B., L.C., or M.S.) using an author-created coding form that documented sample characteristics, details of the experimental pain tasks performed, and details related to any pain-related outcomes measured (including mean and standard deviation of the pain outcome for both boys and girls, as well as the results of any statistical tests conducted to examine sex differences). During coding, three additional articles were identified as being eligible for inclusion, as they were referenced in the paper as reporting on additional results from the same study sample (Noel, Chambers, McGrath, Klein, & Stewart, 2012b; Tsao et al., 2004; Vervoort, Caes, Trost, Notebaert, & Goubert, 2012). These three articles were also coded and included in the study, resulting in a total of 82 articles coded for inclusion. See Figure 2.8.1 for a study flowchart employing the PRISMA model (Moher, Liberati, Tetzlaff, Altman, & The PRISMA Group, 2009).

Coding sheets were examined to identify missing data. Authors were contacted and asked to supply data for any article that did not include the following: age range of participants, mean age of participants, mean and standard deviation for boys and girls separately for any pain outcome. When applicable, data was requested for baseline/control experimental pain tasks (i.e., tasks that did not involve an intervention or experimental manipulation) and for healthy/community samples only. Two attempts were made to contact the corresponding author of each paper where data was missing. Based on author responses, two articles that had originally been included in the review (Bruehl, Dengler-Crish, Smith, & Walker, 2010; Vederhus et al., 2012) were excluded, as it was revealed that the sample fell outside of the 0-18 year old age range. This resulted in a final total of 80 articles included, reporting on 81 separate studies, as one article reported on two studies with separate samples (Vierhaus et al., 2011).

Overlapping samples

Every attempt was made to avoid the inclusion of overlapping samples in the review, as this would involve an over-representation of a subset of children. If it was unclear whether samples were overlapping, authors were emailed to confirm this information. Where it was known that samples were overlapping (i.e., >1 study included in the review that reported on the same sample of children), the authors of the present review went back to the first published study from that sample and worked forward chronologically through multiple publications reporting on the same sample of children, making note of outcomes the first time that full data was reported (e.g., means and standard deviations of pain outcome for boys and girls separately, and statistics regarding sex differences). If full data was not available from any of the studies involved in the overlapping sample, the authors were contacted and asked to provide data about the first chronological incidence of reporting. Where it was unclear whether samples were overlapping, the authors were contacted and asked to indicate whether multiple publications reported on the same sample of children. If authors did not respond, the studies were assumed to represent different samples of children and were treated as such in the review.

Data Analytic Approach

Information from data extraction coding sheets were entered into SPSS 20, and information from the systematic review was summarized using descriptive statistics. Due to the low number of studies included in the systematic review, results were combined across different experimental pain tasks.Sufficient data was available to conduct meta-analyses separately for cold pressor pain, heat pain, and pressure pain. Data needed to be available from at least two studies to conduct a meta-analysis for a particular pain outcome. All data suitable for pooling was analyzedwith RevMan 5.2 software using a fixed-effects analysis (unless otherwise indicated), as heterogeneity across studies was not observed or was low for each outcome (The Cochrane Collaboration, 2012). Heterogneity was calculated using the I^2 statistic, with 0-40% interpreted as heterogeneity that might not be important, 30-60% taken as moderate heterogeneity, and 75-100% representing considerable heterogeneity (J. P. T. Higgins & Green, 2011). For each study, the standardized mean difference and a 95% confidence interval was calculated. In studies where the same pain task was administered more than one time and the results of each trial were reported separately (or when the same pain outcome was measured more than one time within a pain task and data was reported for each time point), only data from the first trial/measurement was included. Note that when the same pain task was administered under different conditions but the order was counterbalanced across participants (Moon et al., 2011), a pooled mean was taken of pain outcomes across both conditions, as it could not be determined which pain task was administered first for each participant. The following formulas were used to pool means and SDs: pooled mean= [(mean1 x N1) + (mean2 x N2) / (N1 + N2)] and pooled SD =square root of [($SD1^2$)(N1-1) + ($SD2^2$)(N2-1)] / N1 + N2 -2. A pooled mean was also calculated for studies that reported results of the same pain task performed at multiple body locations (e.g., pressure pain measured at the neck and shoulder).

Given that many studies of sex differences in adult pain have speculated about the role of sex hormones in the development of sex differences in pain, the meta-analysis was also conducted separately for studies in which the mean age of participants was greater than or equal to 12 years of age, and those studies in which the mean age of participants was less than 12 years of age. This age was chosen as the cut-off as it represents the age at which many girls and boys have entered puberty, and as such (in the absence of measures of pubertal status) provides a proxy for the emergence of sex hormones(Parent et al., 2003). Note that this approach was only taken for cases in which data was available for at least two studies in each age group. If the mean age of participants was not available because it was not reported or because the data suitable for pooling was from a subset of participants rather than the entire sample, categorization was determined by the age range of participants or the mean age of participants in the entire sample.

2.4 Results

Published accounts of sex differences in pain outcomes

Results of the systematic review are presented for each pain outcome measure summarized across experimental pain tasks. Note that several studies (n=25, 30.9%) had children complete more than one different type of experimental pain task, and results from statistical tests of sex differences were included for each unique pain task, even if it was performed on the same sample of children. Tables 2.7.1 through 2.7.4 provide the results of the systematic review separately by experimental pain induction method. Note that the following studies eligible for inclusion in the systematic review conducted additional experimental pain tasks (e.g., fabric prickliness test, ischemic pain, brush allodynia, manual palpation, dynamic mechanical allodynia, tactile pain sensitivity) but did not conduct statistical tests examining sex differences in healthy children and therefore are not included in Tables 2.7.1-2.7.4: (Bar-Shalita, Vatine, Seltzer, & Parush, 2009; Chaves, Nagamine, de Sousa, de Oliveira, & Grossi, 2010; Hirschfeld et al., 2012; Schmelzle-Lubiecki et al., 2007; S. M. Walker et al., 2009; Wollgarten-Hadamek et al., 2011).

Pain intensity. Of the pain tasks where it was reported that statistical tests of sex differences in pain intensity were conducted (*n*=21 pain tasks from 18 unique studies), 90.5% reported no sex differences, and 9.5% indicated girls reported significantly higher levels of pain intensity than boys.

Pain threshold. Of the pain tasks where it was reported that statistical tests of sex differences in pain threshold were conducted (*n*=16 pain tasks from 9 unique studies), 68.8% reported no sex differences, and 31.2% indicated that boys had a significantly higher pain threshold than girls.

Pain tolerance. Of the pain tasks where it was reported that statistical tests of sex differences in pain tolerancewere conducted (*n*=16 pain tasks from 16 unique studies), 75% reported no sex differences, 12.5% indicated that girls had a higher pain tolerance than boys, and 12.5% indicated that boys had a higher pain tolerance than girls.

Pain affect. Of the pain tasks where it was reported that statistical tests of sex differences in pain affect were conducted (*n*=7 pain tasks from 5 unique studies), 85.7% reported no sex differences, and 14.3% indicated that girls reported greater pain affect than boys in response to an experimental pain task.

Facial activity in response to pain. Of the pain tasks where it was reported that statistical tests of sex differences in facial activity in response to pain were conducted (*n*=8 pain tasks from 8 unique studies), 75% reported no sex differences, and 25% indicated that boys displayed greater facial activity in response to pain than girls.

Physiological responses to pain. Of the pain tasks where it was reported that statistical tests of sex differences in physiological responses to pain were conducted (*n*=6 pain tasks reporting on 9 measures of physiological responses from 4 unique studies[1]), 88.9% reported no sex differences, and 11.1% indicated that boys had a greater physiological response (blood pressure) to pain than girls.

Meta-analysis of sex differences in cold pressor pain

When full data was not available in the published manuscript, authors were contacted and requested to provide data for the meta-analytic portion of this research. Of the 49 requests for data sent, 9 responses (18.4%) were received indicating that the data was not available, and 27 responses (55.1%) provided additional data. When this was combined with the data available from published mansucripts, data for meta-analysis was available from 33 separate samples, with a combined total of 2109 unique participants (1069 girls and 1040 boys).

Pain intensity. Data from 19 studies (published in 18 separate articles) were entered into the meta-analysis, which compared self-reported pain intensity during the cold pressor taskin a total of 628 girls and 633 boys (Chambers et al., 2002; Dufton, Dunn, Slosky, & Compas, 2011; Goffaux et al., 2008; Goodman & McGrath, 2003; Jaaniste, Hayes, & von Baeyer, 2007; Moon et al., 2008; Moon et al., 2011; Myers et al., 2006; Noel et al., 2012b; Piira, Taplin, Goodenough, & Von Baeyer, 2002; Piira, Hayes, Goodenough, & von Baeyer, 2006; Thastum et al., 1997; Thastum, Zachariae, & Herlin, 2001; Trapanotto et al., 2009; Tsao et al., 2002; Vervoort, Goubert, & Crombez, 2009; Vervoort et al., 2011; Vierhaus et al., 2011). Pain intensity was measured using a variety of self-report tools, including the Faces Pain Scale (original (Bieri, Reeve, Champion, Addicoat, & Ziegler, 1990) and revised (Hicks et al., 2001) versions), numerical rating scales, visual analogue scales, and the Coloured Analogue Scale (P. A. McGrath et al., 1996). This analysis revealed a standardized mean difference (SMD) of 0.10 [-0.01, 0.21] and an I^2 of 0%, indicating no observed heterogeneity. While the mean self-reported pain intensity of girls was greater than boys, this effect was not significant ($Z = 1.76$, $p = .08$).

The meta-analysis was repeated to separately examine studies for which the mean age of participants was greater/equal to or less than 12 years of age. For studies with a mean age of less than 12 years, data from 12 studies were entered into the meta-analysis, with a total of 302 girls and 303 boys (Chambers et al., 2002; Dufton et al., 2011; Goffaux et al., 2008; Jaaniste et al., 2007; Moon et al., 2008; Moon et al., 2011; Noel et al., 2012b; Piira et al., 2002; Piira et al., 2006; Thastum et al., 1997; Trapanotto et al., 2009; Tsao et al., 2002). This analysis revealed a SMD of 0.01 [-0.15, 0.17], an I^2 of 0%, and no significant differences between boys and girls on self-reported pain intensity (Z= 0.08, $p = .93$).

However, a significant effect was present in the studies with a mean age of equal to or greater than 12 years, in which seven studies (from six published articles) were entered into the meta-analysis, with a total of 321 girls and 330 boys (Goodman &

McGrath, 2003; Myers et al., 2006; Thastum et al., 2001; Vervoort et al., 2009; Vervoort et al., 2011; Vierhaus et al., 2011). This analysis revealed a SMD of 0.19 [0.03, 0.34], an I^2 of 33%, and a significant difference in which girls reported significantly greater pain intensity in response to the cold pressor task than boys ($Z = 2.35$, $p = .02$).

Pain threshold. Data from six studies were entered into the meta-analysis, which compared pain threshold in a total of 154 girls and 149 boys (Dahlquist et al., 2007; Dahlquist et al., 2009; Dahlquist et al., 2010; Thastum et al., 1997; Thastum et al., 2001; Trapanotto et al., 2009). This analysis revealed a SMD of 0.12 [-0.11, 0.35] and an I^2 of 15%, indicating low heterogeneity. This effect was not significant ($Z = 1.02$, $p = .31$), indicating no significant differences in pain threshold during the cold pressor task between boys and girls. As all but one study had a mean age less than 12 years old, the meta-analysis was not conducted separately for different age groups.

Pain tolerance. Data from 18 studies (published in 17 separate articles) were entered into the meta-analysis, which compared pain tolerance in a total of 628 girls and 600 boys (Chambers et al., 2002; Dahlquist et al., 2007; Dahlquist et al., 2009; Dahlquist et al., 2010; Dufton et al., 2011; Jaaniste et al., 2007; Law et al., 2011; Moon et al., 2011; Myers et al., 2006; Piira et al., 2002; Piira et al., 2006; Thastum et al., 1997; Thastum et al., 2001; Trapanotto et al., 2009; Tsao et al., 2002; Vierhaus et al., 2011; Weiss, Dahlquist, & Wohlheiter, 2011). This analysis revealed a SMD of 0.04 [-0.07, 0.16] and an I^2 of 0%, indicating no observed heterogeneity. This effect was not significant ($Z = 0.72$, $p = .47$), indicating no difference between boys and girls on pain tolerance during the cold pressor task.

The meta-analysis was repeated to separately examine studies for which the mean age of participants was greater/equal to or less than 12 years of age. For studies with a mean age of less than 12 years, data from 14 studies were entered into the meta-analysis, with a total of 366 girls and 342 boys (Chambers et al., 2002; Dahlquist et al., 2007; Dahlquist et al., 2009; Dahlquist et al., 2010; Dufton et al., 2011; Jaaniste et al., 2007; Law et al., 2011; Moon et al., 2011; Piira et al., 2002; Piira et al., 2006; Thastum et al., 1997; Trapanotto et al., 2009; Tsao et al., 2002; Weiss et al., 2011). This analysis revealed a SMD of 0.07 [-0.08, 0.22], an I^2 of 0%, and no signficant differences between boys and girls on pain tolerance during the cold pressor task (Z= 0.93, p = .35).

Similar results were seen in the meta-analysis of studies in which participant mean age was greater than 12 years, in which four studies (from three published articles) were included, with a total of 262 girls and 258 boys (Myers et al., 2006; Thastum et al., 2001; Vierhaus et al., 2011). This analysis revealed a SMD of 0.00 [-0.17, 0.18], an I^2 of 0%, and no signficant differences between boys and girls on pain tolerance during the cold pressor task (Z= 0.03, p = .98).

Pain affect. Data from nine studies were entered into the meta-analysis, which compared self-reported pain affect in a total of 308 girls and 327 boys (Chambers et al., 2002; LeBaron, Zeltzer, & Fanurik, 1989; Moon et al., 2011; Myers et al., 2006; Noel et al., 2012b; Thastum et al., 1997; Thastum et al., 2001; Trapanotto et al., 2009; Verhoeven, Goubert, Jaaniste, Van Ryckeghem, D. M. L., & Crombez, 2012). Pain affect was measured using several self-report tools, including the Facial Affective Scale (P. A. McGrath, de Verber, & Hearn, 1985), the Children’s Fear Scale (McMurtry, Noel, Chambers, & McGrath, 2011), and numerical rating scales and visual analogue scales for “pain discomfort” or “pain unpleasantness.” This analysis revealed a SMD of 0.02 [-0.13, 0.18] and an I^2 of 0%, indicating no observed heterogeneity. This effect was not significant (Z = 0.29, p = .77), indicating no difference between boys and girls on self-reported pain affect during the cold pressor task.

The meta-analysis was repeated to separately examine studies for which the mean age of participants was greater/equal to or less than 12 years of age. For studies with a mean age of less than 12 years, data from six studies were entered into the meta-analysis, with a total of 183 girls and 207 boys (Chambers et al., 2002; LeBaron et al., 1989; Moon et al., 2011; Noel et al., 2012b; Thastum et al., 1997; Trapanotto et al., 2009). This analysis revealed a SMD of 0.02 [-0.13, 0.18], an I^2 of 0%, and no signficant differences between boys and girls on pain affect during the cold pressor task (Z= 0.29, p = .77).

Similar results were seen in the meta-analysis of studies in which participant mean age was greater than 12 years, in which three studies were included, with a total of 125 girls and 120 boys (Myers et al., 2006; Thastum et al., 2001; Verhoeven et al., 2012). This analysis revealed a SMD of 0.08 [-0.17, 0.33], an I^2 of 0%, and no signficant differences between boys and girls on pain affect during the cold pressor task (Z= 0.65, p = .52).

Facial expression of pain. Data from six studies were entered into the meta-analysis, which compared facial expressions of pain in a total of 118 girls and 127 boys (Chambers et al., 2002; Goodman & McGrath, 2003; Larochette, Chambers, & Craig, 2006; Moon et al., 2008; Vervoort et al., 2009; Vervoort et al., 2011). Scores for facial expression in response to pain were coded in each study using the Child Facial Coding System (Chambers, Cassidy, McGrath, Gilbert, & Craig, 1996) or the Facial Action Coding System (Ekman & Friesen, 1978) (note that how facial expression scores were calculated differed across studies in that some studies reported a score based on all facial action units, while others calculated a score based on only those facial action units that have been identified as corresponding to expressions of pain). This analysis revealed a SMD of 0.00 [-0.26, 0.25] and an I^2 of 0%, indicating no observed heterogeneity. This effect was not significant ($Z = 0.03$, $p = .98$), indicating no difference between boys and girls on facial expressions in response to the cold pressor task.

The meta-analysis was repeated to separately examine studies for which the mean age of participants was greater/equal to or less than 12 years of age. For studies with a mean age of less than 12 years, data from three studies were entered into the meta-analysis, with a total of 81 girls and 82 boys (Chambers et al., 2002; Larochette et al., 2006; Moon et al., 2008). This analysis revealed a SMD of 0.08 [-0.22, 0.39], an I^2 of 0%, and no signficant differences between boys and girls in facial expressions in response to the cold pressor task ($Z= 0.53$, $p = .60$).

Similar results were seen in the meta-analysis of studies in which participant mean age was greater than 12 years, in which three studies were included, with a total of 62 girls and 70 boys (Goodman & McGrath, 2003; Vervoort et al., 2009; Vervoort et al., 2011). This analysis revealed a SMD of 0.02 [-0.33, 0.36], an I^2 of 40%, and no signficant differences between boys and girls on facial expression in response to the cold pressor task ($Z= 0.09$, $p = .93$).

Physiological reaction. Data from four studies were entered into the meta-analysis, which compared physiological reactions to the cold pressor task in a total of 154 girls and 149 boys (Chambers et al., 2002; Dufton et al., 2011; Goffaux et al., 2008; Moon et al., 2008). In each of the included studies, physiological reactions were measured using participant heart rate. This analysis revealed a SMD of 0.09 [-0.24, 0.41] and an I^2 of 0%, indicating no observed heterogeneity. This effect was not significant ($Z = 0.52$, $p = .60$), indicating no significant differences in heart rate in response to the cold pressor task between boys and girls. As all included studies had a mean age less than 12 years old, the meta-analysis was not conducted separately for different age groups.

Meta-analysis of sex differences in experimental heat pain

Meta-analyses were conducted for pain intensity, tolerance, and threshold for experimental heat pain. There was insufficient data available to conduct analyses for pain affect, facial expression, or physiological responses.

Pain intensity. Data from three studies were entered into the meta-analysis, which compared self-reported pain intensity in a total of 154 girls and 155 boys during experimental heat pain(Caes, Vervoort, Trost, & Goubert, 2012; Goffaux et al., 2008; Myers et al., 2006). Pain intensity was measured using numerical rating scales and visual analogue scales. This analysis revealed a SMD of 0.07 [-0.15, 0.30] and an I^2 of 0%, indicating no observed heterogeneity. No significant differences between boys and girls on self-reported pain intensity during heat pain were observed ($Z = 0.63$, $p = .53$). As there were less than two studies in each age group, the meta-analysis was not conducted separately for different age groups.

Pain threshold. Data from three studies were entered into the meta-analysis, which compared pain threshold in a total of 179 girls and 183 boys (Blankenburg et al., 2010; Blankenburg et al., 2011; Goffaux et al., 2008). This analysis revealed a SMD of -0.31 [-0.52,-0.11] and an I^2 of 0%, indicating no observed heterogeneity. This effect was significant ($Z = 2.96$, $p = .003$), indicating that boys had a significantly higher heat pain threshold than girls.

As two of the included studies reported means and standard deviations separately by age group, it was possible to conduct a meta-analysis to separately examine study samples for which the mean age of participants was greater/equal to or less than 12 years of age. For studies with a mean age of less than 12 years, data from three studies were entered into the meta-analysis, with a total of 104 girls and 107 boys (Blankenburg et al., 2010; Blankenburg et al., 2011; Goffaux et al., 2008). This analysis revealed a SMD of -0.34 [-0.61,-0.07], an I^2 of 0%, and a significant difference in which boys had significantly higher heat pain threshold than girls ($Z = 2.46$, $p = .01$).

Similar results were seen in the meta-analysis of studies in which participant mean age was greater than 12 years, in which data from two studies were included, with a total of 75 girls and 76 boys (Blankenburg et al., 2010; Blankenburg et al., 2011). This analysis revealed a SMD of -0.27 [-0.60, 0.05], and an I^2 of 0%. While the mean pain threshold of boys was greater than the mean pain threshold of girls, this effect was not significant ($Z = 1.68$, $p = .09$).

Pain tolerance. Data from two studies were entered into the meta-analysis, which compared pain tolerance in a total of 152 girls and 148 boys (Caes et al., 2012; Lu et al., 2005). Note that as heterogeneity was high in this comparison (I^2=91%) a random effects model was used. This analysis revealed a SMD of -1.26 [-2.29, -0.23] with a significant effect ($Z = 2.40$, $p = .02$), indicating that boys had significantly higher tolerance of heat pain than girls. As there were less than two studies in each age group, the meta-analysis was not conducted separately for different age groups.

Meta-analysis of sex differences in experimental pressure pain

Meta-analyses were conducted for pain intensity and threshold for experimental heat pain. There was insufficient data available to conduct analyses for pain tolerance, pain affect, facial expression, or physiological responses.

Pain intensity. Data from two studies were entered into the meta-analysis, which compared self-reported pain intensity in a total of 164 girls and 160 boys undergoing experimental pressure pain (Myers et al., 2006; Vervoort et al., 2008). Pain intensity was measured using numerical rating scales and visual analogue scales. This analysis revealed a standardized mean difference (SMD) of 0.17 [-0.5, 0.39] and an I^2 of 0%, indicating no observed heterogeneity. No significant differences between boys and girls on self-reported pain intensity during pressure pain were observed ($Z = 1.51$, $p = .13$). As there were less than two studies in each age group, the meta-analysis was not conducted separately for different age groups.

Pain threshold. Data from two studies were entered into the meta-analysis, which compared pain threshold in a total of 81 girls and 62 boys (Metsahonkala et al., 2006; Vervoort et al., 2008). Note that as heterogeneity was high in this comparison (I^2=78%) a random effects model was used. This analysis revealed a SMD of -0.35 [-1.08,0.39] and this effect was not significant ($Z = 0.93$, $p = .35$), indicating no significant differences in pain threshold during experimental pressure pain between boys and girls. As there were less than two studies in each age group, the meta-analysis was not conducted separately for different age groups.

Reporting practices of sex and gender variables

Of the 81 included studies, 41 studies (50.6%) had reported results of statistical tests examining sex differences for at least one pain-related outcome (note that this included studies that merely reported that sex differences were or were not present, even if the authors did not include numerical values of the statistical test conducted or the mean values of the different groups). Nine studies (11.1%) reported entering sex as a covariate in their analyses. Two studies (2.5%) reported using a validated measure of child gender (e.g., the *Children's Sex Role Inventory* (Boldizar, 1991)) to examine the relationship between child gender and pain outcomes (Myers et al., 2006; Vierhaus et al., 2011).

With regards to terminology use, 36 studies (44.4%) used the appropriate terminology when referring to “sex” or “gender”, according to the definitions of sex and gender set out by the World Health Organization (World Health Organization, 2012). Of the remaining studies, 29 studies (35.8%) used the term “gender” when grouping participants based on sex, 7 studies (8.6%) used the terms “sex” and “gender” interchangeably throughout the article, and 9 studies (11.1%) did not use either term at all.

2.5 Discussion

Systematic review and meta-analysis of sex differences

The results of this systematic review indicate that the majority of studies on children’s responses to experimental pain report no significant sex differences on pain-related outcomes. However, the meta-analysis of cold pressor pain intensity revealed that girls reported significantly higher pain intensity than boys when pooling data from studies that had a mean age >12 years, an age typically associated with onset of pubertal development in both boys and girls (Parent et al., 2003). While such an approach to examining age is crude, a more detailed analyses by age and/or pubertal status was not feasible with the information available. Nonetheless, this analysis offers preliminary support for the hypothesis that sex differences in experimental cold pressor pain, similar to chronic pain, emerge in adolescence and could possibly be related to the emergence of sex hormones (King et al., 2011). This is in line with findings in other areas of research, in which the emergence of sex differences of other disorders and conditions (e.g., anxiety, depression) are seen at puberty (Conley & Rudolph, 2009; Craske, 2003; Kessler, Avenevoli, & Merikangas, 2001). A more explicit examination of the role of sex hormones and pubertal stages in the development of sex differences is needed to control for other factors that could be contibuting to sex differences in adolescents (e.g., different methodologies used in research groups that study children vs. adolescents).

A meta-analysis of sex differences in response to experimental heat pain revealed that boys had significantly higher pain threshold and pain tolerance than girls, with no significant differences in pain intensity. Unlike cold pressor pain, the sex difference in heat pain threshold was still significant in studies where children had a mean age of 12 years or lower. These results should be interepreted with caution, as heat pain is not often used among children, and as such, the meta-analysis may have not been adequately powered to accurately represent the strength of sex difference effects in heat pain in children, particularly when split by age group.

In adults, pressure pain produces the largest sex differences of all experimental pain tasks (Riley, Robinson, Wise, Myers, & Fillingim, 1998). In the present meta-analysis, no sex differences were observed in experimental pressure pain. The small number of studies using heat and pressure pain limits the ability to draw conclusions regarding effects of different experimental pain tasks. Inspection of available results from the systematic review does not appear to support task-specific sex effects, though more research in this area is certainly needed. In particular, examination of possible sex differences in pain emerging at different developmental stages for different types of pain tasks are currently lacking in children (Fillingim et al., 2009; M. Racine et al., 2012a).

Reporting practices of sex and gender

In recent years, organizations such as the International Association for the Study of Pain have increasingly encouraged researchers to consider sex and gender in their research and to use appropriate terminology (Greenspan et al., 2007). Despite the majority of studies having a representative sample of both boys and girls, less than half of included studies reported tests of sex differences in pain-related outcomes. Only two of the 81 studies included in this review reported on measures of child gender and its relationship to pain outcomes. Additionally, despite increasing awareness of the distinction between sex and gender, use of terminology in reviewed articles was often inappropriate, with the most common issue being use of the term "gender" when referring to the categorical distinction between boys and girls. Appropriate reporting is critical for advancing our understanding of the role of these variables in pain.

Strengths, Limitations, & Future Directions

A strength of the present research was the use of a meta-analytic approach, which allowed for pooling of data to examine sex effects and including data from studies that otherwise were not powered to look at sex differences. Additionally, the excellent response rate of authors providing data allowed for a quantitative synthesis of a large number of studies beyond what is available in the published literature. With regards to limitations, any systematic review or meta-analysis is subject to methodological variability across studies. For example, measurement of pain outcomes occurred at different times (e.g., several studies examined pain intensity at the beginning of the pain task, while others examined worst pain upon task completion). Rules were implemented regarding data extraction to control this variability (e.g., only the first measure of pain intensity taken was used), however some methodological variability remained unavoidable (e.g., the first measure of pain intensity may have occurred at different times across studies) and may have impacted the findings (Mogil, 2012). While physiological measures of pain have been commonly reported in adult reviews of sex differences in experimental pain, these measures are often not specific to pain and should be interpreted with caution (Fillingim et al., 2009; M. Racine et al., 2012a). Finally, while the majority of meta-analyses conducted had low or no observed heterogeneity, observed heterogeneity was high for a few select analyses undertaken (heat pain tolerance and pressure pain threshold).

The division of the meta-analysis by mean age of participants has several limitations. While the mean age indicated whether the majority of children were under/over the age of 12 years, many of the studies would have included children and adolescents at various stages of pubertal development in both groups. As such, the presence of potential sex differences in those children who had undergone puberty may have been washed out because they were being considered along with pre-pubertal children. Previous research has had conflicting conclusions whether pubertal status or age is more important for understanding the development of sex differences in children's pain (LeResche et al., 2005; Schmitz et al., 2013). As pubertal status was only measured in two studies in the present review (Allen, Lu, Tsao, Worthman, & Zeltzer, 2009; Lu et al., 2005), age was used as a proxy, however, this was not ideal and future research is need to replicate our age-related findings.

The heterogeneity of methods in the included studies, as well as the wide age ranges precludes conclusive statements regarding the effects of sex on healthy children's pain experience. Future research will require studies explicitly designed to examine sex differences in various age groups across pubertal development (which may include measurement of pubertal stage and/or presence of sex hormones in addition to age). Such studies may require large samples to be able to detect the small sex difference effects presented in this review, which are similar to the small-medium effect sizes seen in adult reviews (Fillingim et al., 2009). Overall, for the cold pressor task, all standardized mean difference scores were less than 0.2 (range: 0.0-0.19), indicating very small effect sizes(J. Cohen, 1988). Heat pain tasks showed more variability in effect sizes, from small for pain intensity and threshold (*SMD* = 0.07 and -0.31, respectively), to quite large for pain tolerance (*SMD* = -1.26). Pressure pain tasks demonstrated small effect sizes for pain intensity and threshold (*SMD* = 0.17 and -.035, respectively). Researchers may consider using the effect sizes from the present study in calculating sample sizes, should they wish to examine sex differences in their own research. While many of the studies included in the present review did not report significant sex differences, this may have been due to insufficient power to detect such effects. For example, a t-test comparing boys and girls with a significance level of .05 and power of 0.8 would require close to 400 participants in each group to detect a small effect size, which is much larger than the standard sample size for experimental pain studies. Schmitz and colleagues (Schmitz et al., 2013) recently demonstrated important future directions for the field through the inclusion of large sample sizes and methodology designed explicitly to examine sex differences across pubertal development. Researchers should consider conducting similar studies looking at additional pain outcomes (e.g., pain intensity) and using different experimental pain paradigms.

With regards to future research directions, it will also be important for investigators to continue examining the impact of gender on pain responses in childhood and adolescence. A recent meta-analysis of the impact of gender roles on experimental pain responses in adults supports the role of gender schema theory in influencing differential pain responding in men and women (Alabas et al., 2012). As gender schemas are known to be incorporated and understood by children at a young age, it will be important to examine the developmental trajectory of gender influences (Myers et al., 2003). Sex differences in other psychosocial variables also deserve further investigation, such as children's pain coping styles and parental behaviour in response to pain. Finally, a similar systematic meta-analytic approach should be applied to clinical pain in children.

In summary, the majority of published accounts of sex differences in pain outcomes in healthy children reported no significant differences between boys and girls on any pain outcomes. However, the meta-analysis of available combined data found that girls reported significantly higher increased pain intensity compared to boys in studies where the mean participant age was greater than 12 years. Additionally, a meta-analysis of heat pain found that boys had significantly higher tolerance than girls, and boys had significantly higher heat pain threshold than girls in studies where the mean participant age was 12 years or younger. Researchers should continue to include analyses of both sex and gender, as well as developmental factors such as puberty, to better understand how the sex differences observed in adult pain develop from childhood.

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Table 2.7.1.

Studies examining sex differences in experimental cold pain.

Authors	Sample Size		Mean age (range)	Method (location)	Pain outcomes					
	Boys	Girls			Intensity	Threshold	Tolerance	Affect	Facial activity	Physiological responses
Allen et al. (2009) [a]	119	116	12.7 (8-18)	CPT (dominant hand)						G=B
Blankenburg et al. (2010)	88	88	NR (6-16)	QST cold pain threshold (face, hand, foot)		G<B				
Blankenburg et al.(2011) [b]	88	85	NR (7-14)	QST cold pain threshold (hands)		G=B				
Chambers et al.(2002)	60	60	9.74 (8-12)	CPT (left hand)	G=B		G=B	G=B	G<B	G=B

Authors	Sample Size		Mean age (range)	Method (location)	Pain outcomes					
	Boys	Girls			Intensity	Threshold	Tolerance	Affect	Facial activity	Physiological responses
Coldwell et al.(2002)	38	37	9.7 (8-11)	CPT (right forearm)	G=B					
Foster et al. (2003)	53	47	12.43 (8-17)	CPT (left hand)						G=B and G<B[c]
Goodman et al. (2003)	48	48	12.6 (10-14)	CPT (non-dominant hand)	G=B	G=B	G=B		G=B	
Jaaniste et al. (2007)	38	41	9.16 (7-12)	CPT (non-dominant arm)	G=B		G=B			
Larochette et al.(2006)	25	25	9.74 (8-12)	CPT (arms)					G=B	
LeBaron et al.(1989)[g]	19	18	NR (6-12)	CPT (arms)			G=B	G>B		

Authors	Sample Size		Mean age (range)	Method (location)	Pain outcomes					
	Boys	Girls			Intensity	Threshold	Tolerance	Affect	Facial activity	Physiological responses
Miller et al. (1994)	23	21	NR (8-11)	CPT (non-dominant forearm)			G>B			
Moon et al. (2008)	37	36	8.04 (4-12)	CPT (hand)	G=B				G=B	G=B
Myers et al. (2006) [a]	120	120	12.7 (8-18)	CPT (non-dominant hand)	G=B		G=B	G=B		
Piira et al. (2002)	22	31	9.08 (7-14)	CPT (dominant arm)			G=B			
Piira et al. (2006)	55	65	10.16 (7-14)	CPT (non-dominant arm)	G=B		G=B			
Trapanotto et al. (2009)	78	63	10.1 (8-12)	CPT (non-dominant arm)	G=B	G=B	G=B	G=B		

Authors	Sample Size		Mean age (range)	Method (location)	Pain outcomes					
	Boys	Girls			Intensity	Threshold	Tolerance	Affect	Facial activity	Physiological responses
Tsao et al. (2002) [d]	19	32	NR (8-10)	CPT (arms)	G=B		G=B			
Verhoeven et al. (2012) [e]	39	42	13.6 (9-18)	CPT (left hand)				G=B		
Vervoort et al.(2009) [f]	32	30	12.46 (9-15)	CPT (left hand)					G=B	
Vervoort et al.(2011) [e]	22	16	14.5 (10-18)	CPT (hands)	G=B				G<B	
Vierhaus et al.(2011) (Study #1)	53	65	12.74 (10-17)	CPT (non-dominant forearm)	G>B		G=B			
Vierhaus et al.(2011) (Study #2)	81	67	12.8 (10-17)	CPT (non-dominant forearm)	G>B		G=B			

Authors	Sample Size		Mean age (range)	Method (location)	Pain outcomes					
	Boys	Girls			Intensity	Threshold	Tolerance	Affect	Facial activity	Physiological responses
Weiss et al. (2011)	31	30	4.21 (3-5)	CPT (non-dominant hand)			G=B			
Zeltzer et al.(1989) [g]	NR	NR	NR (6-12)	CPT (arms)			G>B	G>B		

Note. In the case of studies with overlapping samples, only the first published account to meet review inclusion criteria is included. Note that while higher ratings on pain intensity indicate greater pain sensitivity, lower levels of threshold and tolerance indicate greater pain sensitivity. NR = not reported.

The following studies using experimental cold pain were included in the systematic review but did not report the results of statistical tests of sex differences in healthy children in the published manuscript: (Bar-Shalita et al., 2009; Dahlquist et al., 2007; Dahlquist et al., 2009; Dahlquist et al., 2010; Dufton et al., 2011; Feuerstein, Barr, Francoeur, Houle, & Rafman, 1982; Goffaux et al., 2008; Law et al., 2011; Meh & Denislic, 1998; Meier, Berde, DiCanzio, Zurakowski, & Sethna, 2001; Mennella, Pepino, Lehmann-Castor, & Yourshaw, 2010; Moon et al., 2011; Noel, Chambers, McGrath, Klein, & Stewart, 2012a; Noel et al., 2012b; Smith, Martin-Herz, Womack, & McMahon, 1999; Stuber et al., 2009; Thastum et al., 1997; Thastum et al., 2001)

[a] The following studies reported on results of the cold pressor task with the same sample of children: Evans, Lu, et al. (2008), Evans, Tsao, et al. (2008), Evans et al. (2009), Haas et al. (2011), Lu et al. (2007), Tsao et al. (2004), Tsao, Lu, Kim, et al. (2006), Tsao, Lu, Myers, et al. (2006), and Tsao et al. (2012).

[b] One study reported on results of a cold pain threshold test re-testing the same sample of children Hirschfeld et al. (2012).

[c] No sex differences were found on the majority of physiological outcomes in response to pain (heart rate, skin conductance, respiratory rate, EMG, or skin temperature), but boys had significantly higher blood pressure than girls.

[d] Two studies reported on results of the cold pressor task with the same sample of children: Fanurik et al. (1993) and Tsao et al.(2003).
[e] Studies reported on results from the same sample of children.
[f] One study reported on results of the cold pressor task with the same sample of children: Caes et al. (2011).
[g] One study reported on results of the cold pressor task with the same sample of children: LeBaron et al.(1989).

Table 2.7.2.

Studies examining sex differences in experimental heat pain.

Authors	Sample Size		Mean age (range)	Location	Pain outcomes					
	Boys	Girls			Intensity	Threshold	Tolerance	Affect	Facial activity	Physiological responses
Allen et al.(2009)[a]	119	116	12.7 (8-18)	Forearms						G=B
Blankenburg et al.(2010)	88	88	NR (6-16)	Face, hand, foot		G<B				
Blankenburg et al.(2011)[b]	88	85	NR (7-14)	Hands		G<B				
Lu et al. (2005)[a]	120	124	12.73 (8-18)	Forearms			G=B			
Myers et al. (2006)[a]	120	120	12.7 (8-18)	Forearms	G=B			G=B		
Vervoort et al. (2012)	32	30	13.08 (11-15)	Right wrist	G=B		G<B		G=B	

Note. In the case of studies with overlapping samples, only the first published account to meet review inclusion criteria is included. Note that while higher ratings on pain intensity indicate greater pain sensitivity, lower levels of threshold and tolerance indicate greater pain sensitivity. NR = not reported.
The following studies using experimental heat pain were included in the systematic review but did not report the results of statistical tests of sex differences in healthy children in the published manuscript:(Bar-Shalita et al., 2009; Caes et al., 2012;

Goffaux et al., 2008; Hermann, Hohmeister, Demirakca, Zohsel, & Flor, 2006; Hohmeister, Demirakca, Zohsel, Flor, & Hermann, 2009; Hohmeister et al., 2010; Meh & Denislic, 1998; Meier et al., 2001; Wollgarten-Hadamek et al., 2009; Wollgarten-Hadamek et al., 2011)

[a] The following studies reported on results of the heat pain task with the same sample of children: Evans, Lu, et al. (2008), Evans, Tsao, et al. (2008), Evans et al. (2009), Haas et al. (2011), Lu et al. (2007), Tsao et al. (2004), Tsao, Lu, Kim, et al. (2006), Tsao, Lu, Myers et al. (2006), and Tsao et al. (2012).

[b] One study reported on results of a heat pain threshold test re-testing the same sample of children: Hirschfeld et al. (2012).

Table 2.7.3.

Studies examining sex differences in experimental pressure pain.

Authors	Sample Size		Mean age (range)	Location	Pain outcomes					
	Boys	Girls			Intensity	Threshold	Tolerance	Affect	Facial activity	Physiological responses
Allen et al. (2009)[a]	119	116	12.7 (8-18)	Fingers						G=B
Blankenburg et al.(2010)	88	88	NR (6-16)	Face, hand, foot		G<B				
Blankenburg et al.(2011)[b]	88	85	NR (7-14)	Hands		G=B				
Chaves et al.(2007)	9	7	8.33 boys and 8.71 girls (7-12)	Face, hand		G=B				
Han et al.(2012)	258	247	7.9 (4-11)	Forearm		G=B				

Authors	Sample Size		Mean age (range)	Location	Pain outcomes					
	Boys	Girls			Intensity	Threshold	Tolerance	Affect	Facial activity	Physiological responses
Hogeweg et al. (1995)	33	36	11.5 boys and 11.5 girls (6-17)	Joints of the elbow, wrist, knee, and ankle, and paraspinally		G=B				
Hogeweg et al.(1996)	33	36	11.4 (6-17)	Joints of the elbow, wrist, knee, and ankle, and paraspinally		G=B[d]				
Metsahonkala et al.(2006)	22	37	NR (13)	Five cranial and neck-shoulder points and three extracephalic points		G<B				
Myers et al. (2006)[a]	120	120	12.7 (8-18)	Fingers	G=B			G=B		
Tsao et al. (2004)[a]	60	58	12.6 (8-18)	Fingers			G<B			

Authors	Sample Size		Mean age (range)	Location	Pain outcomes					
	Boys	Girls			Intensity	Threshold	Tolerance	Affect	Facial activity	Physiological responses
Vervoort et al. (2008)[e]	40	44	11.82 (9-15)	Neck, shoulder	G=B and G>B [f]				G=B	

Note. In the case of studies with overlapping samples, only the first published account to meet review inclusion criteria is included. Note that while higher ratings on pain intensity indicate greater pain sensitivity, lower levels of threshold and tolerance indicate greater pain sensitivity. NR = not reported.

The following studies using experimental pressure pain were included in the systematic review but did not report the results of statistical tests of sex differences in healthy children in the published manuscript:(Alfvén, 1993; Anttila et al., 2002; Chaves et al., 2010; Fernandez-De-Las-Penas et al., 2010; Fernandez-de-las-Penas et al., 2011; Fernandez-Mayoralas et al., 2010; Weidenbacker, Sandry, & Moed, 1963; Wollgarten-Hadamek et al., 2011)

[a] The following studies reported on results of the pressure pain task with the same sample of children: Evans, Lu, et al. (2008), Evans, Tsao, et al. (2008), Evans et al. (2009), Haas et al. (2011), Lu et al. (2007), Lu et al. (2005), Tsao et al. (2012), Tsao, Lu, Kim, et al. (2006), and Tsao, Lu, Myers et al. (2006).

[b] One study reported on results of the pressure pain threshold test re-testing the same sample of children: Hirschfeld et al. (2012).

[c] Note that this sample conducted sex difference statistics combining both the healthy control group and the clinical sample (mean age is presented for healthy sample only).

[d] This study reported that there were no significant sex differences except for at the knee in ages 12-17 years, where G<B.

[e] One study reported on results of the pressure pain threshold test with the same sample of children: Goubert et al. (2009).

[f] Note that while no sex differences were present during the pressure pain threshold test phase (which was the result included in the systematic review, as it was the first trial of the pressure pain task), in the experimental phase girls reported a significantly higher pain intensity than boys.

Table 2.7.4.

Studies examining sex differences in other experimental pain tasks.

Pain task	Authors	Sample Size		Mean age (range)	Location	Pain outcomes	
		Boys	Girls			Intensity	Threshold
Mechanical pain							
	Bar-Shalita et al.(2009)	18	16	7.75 (NR)	Forearm	G=B [a]	
	Blankenburg et al. (2010)	88	88	NR (6-16)	Face, hand, foot	G=B	G=B
	Blankeburg et al. (2011)[b]	88	85	NR (7-14)	Hands	G=B	G=B
Wind-up ratio							
	Blankenburg et al. (2010)	88	88	NR (6-16)	Face, hand, foot	G=B	
	Blankeburg et al. (2011)[b]	88	85	NR (7-14)	Hands		G=B
Water load task							
	Walker et al. (2006a)	60	60	11.46 (8-15)	Abdomen	G=B [c]	

Note. In the case of studies with overlapping samples, only the first published account to meet review inclusion criteria is included. Note that while higher ratings on pain intensity indicate greater pain sensitivity, lower levels of threshold indicate greater pain sensitivity. Note that columns for pain tolerance, pain affect, facial activity in response to pain, and physiological responses to pain are not included in the present table, as none of these outcomes were measured for the listed pain tasks. NR = not reported.

The following studies using experimental mechanical pain were included in the systematic review but did not report the results of statistical tests of sex differences in healthy children in the published manuscript:(Hermann et al., 2006; Wollgarten-Hadamek et al., 2009; Zohsel, Hohmeister, Oelkers-Ax, Flor, & Hermann, 2006)

[a] Note that this sample conducted sex difference statistics combining both the healthy control group and the clinical sample (mean age is presented for healthy sample only).

[b] One study reported on results of a mechanical pain threshold test and wind-up ratio re-testing the same sample of children: Hirschfeld et al. (2012).

[c] Combined abdominal pain/discomfort score.

2.8 Figures

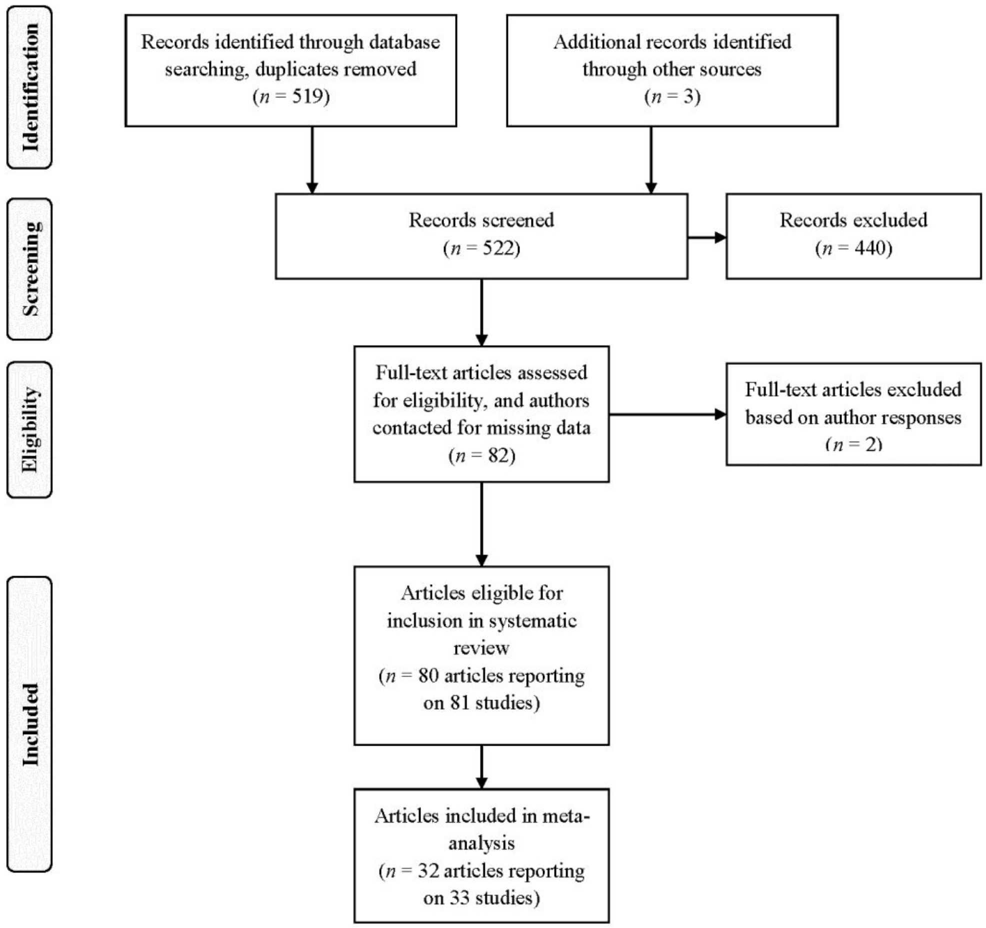

Figure 2.8.1. PRISMA flow diagram of the process of identification and screening of articles for inclusion in the systematic review and meta-analysis.

2.9 Footnotes

1 Note that one of the studies required children to participate in >1 experimental pain task (Myers et al. (2006): CPT, heat pain, pressure pain) and measured two physiological responses (salivary and blood cortisol) after the completion of all three tasks. Additionally, another included study measured physiological responses to cold pressor pain with multiple modalities ((2003): physiological responses measured using heart rate, skin conductance, respiratory rate, EMG, blood pressure, and skin temperature). Results from statistical tests of sex differences were included for each unique pain task and physiological response measure, even if it was performed on the same sample of children.

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CHAPTER 3. THE IMPACT OF PARENTAL MODELING ON CHILD PAIN RESPONSES: THE ROLE OF PARENT AND CHILD SEX

The manuscript prepared for this study is presented below. Readers are advised that Katelynn Boerner, under the supervision of Dr. Christine Chambers, applied for funding for the present project, developed the research protocol and obtained ethical approval to conduct the study (IWK Health Centre REB #1014428), oversaw recruitment of participants, and was involved in collecting data, as well as training and supervising study staff in data collection. She was responsible for all of the data analysis and the writing of the present manuscript. Prior to submission, she received and incorporated feedback from the study's co-authors. The manuscript was submitted for publication on April 18, 2016. The current reference for this manuscript is:

Boerner, K.E., Chambers, C.T., McGrath, P.J., LoLordo, V., Uher, R. (submitted). The impact of parental modeling on child pain responses: The role of parent and child sex.

3.1 Abstract

Social modeling is an important process by which pain behaviours are learned, and previous research has found parents act as model for their children's behaviour. Despite the potential for the sex of the parent and child to influence such processes, no experimental investigation to date has examined the role of sex of the model or observer in the social learning in pediatric pain. The present study recruited 168 parent-child dyads (equal numbers of father-son, father-daughter, mother-son, and mother-daughter dyads) where the children were generally healthy 6- to 8-year-olds. Unbeknownst to their child, parents were randomly assigned to exaggerate their facial expression of pain, minimize their expression of pain, or act naturally during the cold pressor task. Parents completed the cold pressor task while their child observed, and then the children completed the cold pressor task themselves. Children whose parents were in the Exaggerate condition reported higher anxiety than children of parents in the Minimize condition. Additionally, girls in the Exaggerate condition rated their own overall pain intensity during the cold pressor as being significantly higher than boys in the same condition. No child sex differences were observed in pain intensity for the Control or Minimize conditions. Parent expressions of pain impacts children's anxiety, and sex-specific effects of parental pain expression on children's own subsequent pain experience are present.

3.2 Introduction

The aggregation of pain complaints in families is a well-documented phenomenon (Groholt et al., 2003). While there are significant genetic and medical contributions to this relationship (Cservenka et al., 2015; Hocking et al., 2012), social factors, including learning of pain behaviours via modeling, have been hypothesized to play a role in explaining why pain runs in families(Goubert et al., 2011; Levy, 2011; Osborne et al., 1989; Violon & Giurgea, 1984; Zeichner et al., 1999). Through social modeling, individuals learn how to respond to painful events without directly experiencing pain themselves, and an interaction of these learned behaviours with a biological predisposition for a pain condition could impact pain and functional impairment.

Goodman and McGrath (2003) demonstrated that maternal modeling of pain behaviours impacted children's own pain outcomes. In this study, mothers were instructed to manipulate their behaviour (i.e., exaggerate their facial expression, minimize their facial expression, or act naturally) during a pain task while their child watched, and then children completed the pain task themselves. Children who observed their mother exaggerate an expression of pain reported lower pain thresholds and children who observed their mother minimize an expression of pain showed less facial pain behaviour. However, as this study only examined mother-child dyads, it is unclear the extent to which parent and child sex interact to influence the social modeling process.

Social Learning Theory posits that individuals model their behaviour after the behaviour of individuals they consider similar to them, as such models are thought to be more relevant (Bandura, 1977). As sex is one of the first individual difference variables that children are aware of and can self-identify with (Maccoby, 1988), there is a strong reason to believe that children may be more likely to model their behaviour after a same-sex individual. Children (particularly boys) have been shown to attend more to a same-sex model, increasing as children develop an understanding of gender constancy (Slaby & Frey, 1975). Maternal models have been shown to have a greater impact on children's anxiety than paternal models, though the interaction of parent and child sex has not been studied (Muris et al., 1996). Previous research has suggested sex-specific relationships between the presence of a pain model (generally the parent) and child pain and psychological outcomes (Evans et al., 2010). In adults, the presence of a pain model has been shown to have a greater impact on females than males, and while most research has failed to take into account the sex of the model (P. W. Edwards et al., 1985; Fillingim et al., 2000), one recent study found the impact to be greater on females than males regardless of model sex (Świder & Babel, 2013).

The present study aimed to understand how parent and child sex is implicated in children's pain responses after observing parental modeling of pain behaviours. In line with Goodman and McGrath (2003), it was expected that children whose parents minimized their display of pain would experience less pain and anxiety and higher pain tolerance than children whose parents exaggerated their pain display. The influence of parental modeling of pain behaviours was expected to be stronger on girls than boys, and it was expected that maternal behaviour would have a greater influence on children than paternal behaviour (P. W. Edwards et al., 1985; Fillingim et al., 2000; Muris et al., 1996). It was also expected that children in sex-matched dyads would be more influenced by their parent's behaviour than children in sex-unmatched dyads (Kanfer et al., 1971).

3.3 Methods

The data collected for this study is described in two manuscripts, with the present manuscript presenting the results related to the impact of an experimental manipulation of parent behaviour on children's subsequent pain experience and the role that parent and child sex play in this process. Additionally, an exploratory analysis with computerized facial coding was conducted to examine whether differences based on experimental condition and parent and child sex were present in children's facial expressions during the cold pressor task (CPT). The second paper (Boerner & Chambers, submitted) describes analyses related to the impact of children's self-reported gender (i.e., masculinity and femininity) on pain responses and describing the gender-typed expectations parents hold regarding their children's pain.

3.3.1 Participants

Participants were recruited from the local community using posters, mail-outs, media advertisements (e.g., newspapers), website and social media postings, as well as from our database of previous participants who had agreed to be contacted about future research studies. An *a priori* sample size calculation was conducted to determine the number of dyads to recruit, using GPower 3.1.3(Faul, Erdfelder, Lang, & Buchner, 2007). The calculation determined that a sample size of 158 was required to detect a medium effect size (f = .25) with an analysis of variance (α= .05, power = .80). To ensure an equal number of each sex pairing within each experimental group a sample size of 168 dyads was selected with 56 dyads in each of the three experimental conditions (14 each of mother-daughter, father-son, father-daughter, and mother-son dyads).

Parent-child dyads had to meet the following inclusion criteria to participate in the present study: participants had to speak/write/read English well enough to complete questionnaires (though given the young age of the children, research assistants did assist children with reading questionnaires when necessary), participants had to have no hearing or vision impairments that were not corrected for by use of glasses or a hearing aid, and participants could not have any condition that contraindicated participation in the cold pressor task (e.g., circulation problems, blood disorders, injury/frostbite to the non-dominant arm or hand, history of cardiac problems) (von Baeyer et al., 2005). Additionally, children had to be generally pain-free and healthy (i.e., children were not eligible to participate if they had developmental delays, chronic medical conditions, chronic or recurrent pains), and could not have previously participated in a cold pressor study. Parents had to be a primary caregiver of the child and live with the participating child at least 50% of the time, as previous research has suggested the impact of a parental pain model may be related to the family structure (i.e., who the child lives with primarily (Hoftun, Romundstad, & Rygg, 2013)). Prior to study enrolment, exclusion criteria for both the parent and child were assessed during a telephone screening with the participating parent. Children could only participate in the study once, with either their mother *or* their father.

178 parent-child dyads were recruited for the present study and came to the research laboratory to participate. Of these, 10 families were excluded from final data analysis for the following reasons: the child completed all other aspects of the study but refused to do the CPT (*n*=2); the study was terminated early at the family's request (*n*=1); there were substantial deviations from protocol (*n*=2); the child declined to participate during assent (*n*=2); and the parent had significant difficulty in reading/understanding the questionnaire items (*n*=3). The final sample of participants included 336 individuals (168 parent-child dyads).

3.3.2 Measures

Demographics questionnaire. Parents completed an investigator-created questionnaire that inquired about basic demographic variables for themselves and the participating child (e.g., age, sex, ethnicity), information specific to the parent (e.g., chronic pain status, health status, occupation, marital status), and descriptions of the composition of the family that the parent and child belonged to.

Pain outcome measures. A multi-method, multi-informant approach was used to measure parent and child pain during the cold pressor task.

Pain intensity (self-reported). The Faces Pain Scale-Revised (FPS-R; Hicks et al. (2001)) was used for parents and children to provide self-reported ratings of their experienced pain during the cold pressor task. The FPS-R depicts six faces of increasing intensity of pain expressions. The faces are scored from 0 to 10 in 2-point intervals. The FPS-R has been widely used as a self-report measure of pain in children and is the recommended measure for the age group in the present study (Stinson et al., 2006). In the present study, children and parents were asked to use the FPS-R to rate how much pain they felt "when it hurt the most" and to rate how much pain they felt "on average, taken all together." The "most" pain rating was always administered first.

Pain intensity (proxy report). Parents and children also provided proxy reports (i.e., ratings of the other's pain) after watching their family member complete the cold pressor task. These pain ratings were provided using the same pain scale (FPS-R) as the self-reported pain ratings as it has been suggested that using different scales for rating another's pain is associated with lower association between the individual in pain and observer's pain ratings (Zhou et al., 2008).

Parents and children were asked to rate the other's "most" and "average" pain during the task.

Pain tolerance. Pain tolerance was defined as the amount of time, in seconds, that the participant kept their hand in the water during the cold pressor task. Pain tolerance was timed by the research assistant and confirmed by review of the videotapes. As a 4-minute maximum immersion time was used for the cold pressor task, tolerance was calculated by timing the point at which the participant inserted their hand into the water until they took it out of the water, or 4 minutes, whichever came first[1].

Children's anxiety. Children rated their anxiety following observation of their parent's cold pressor task on a 10-cm Visual Analogue Scale. This scale asked children to indicate on a line how anxious/nervous they felt at that moment in time from 'Not nervous or anxious at all' to 'Most nervous or anxious'.

Facial expression of pain. Children and parent's expressions during the CPT were coded using a computerized facial coding system: Noldus FaceReader software version 6.1 with the Action Unit module. The Action Unit module assesses the frequency and intensity of twenty of the facial action units (AUs) from the Facial Action Coding System (FACS) by Ekman and Friesen(Ekman & Friesen, 1978). In addition to being well-validated for characterizing a variety of facial expressions, the Facial Action Coding System (FACS) system has been previously used to describe the facial action units that are typically present during the facial expression of pain in children (Larochette et al., 2006) and adults (Prkachin, 1992; Prkachin, 2009). The FaceReader software has been used primarily in marketing research for classifying basic emotions but has also been used in emotion and education research (Lewinski, den Uyl, & Butler, 2014). The Action Unit module is a relatively new addition to the software, and, as such, there is little prior research using this specific aspect of the program.

Default settings for FaceReader 6.1 were employed (i.e., no calibration, frame-by-frame processing) and the program coded approximately 30 frames per second. The face model 'General61' was used for the videos of parents and the 'Children' face model was used for videos of child participants. Facial action units were coded both for frequency and intensity: Frequency was coded as 0 (action unit not present in that frame of video) or 1 (action unit present in that frame of video). When action units were present, they were also coded on a scale of A (trace) to E (max) intensity, which was recoded to be on a numerical scale from 1 (trace) to 5 (max) for the purpose of analysis. Consistent with the procedure of previous facial coding studies, only action units that had a minimum of frequency of 5% occurrence were included in the final analyses (H. D. Hadjistavropoulos, Craig, Hadjistavropoulos, & Poole, 1996; T. Hadjistavropoulos, LaChapelle, Hadjistavropoulos, Green, & Asmundson, 2002; Hill & Craig, 2002; Larochette et al., 2006).

Facial coding was conducted for the entire duration of the CPT, but analyses focused specifically on the first ten seconds of the cold pressor task, starting the moment that the participant's hand first came in contact with the water. This time frame was chosen to capture the moment at which the expression of pain is the most salient (Craig & Patrick, 1985) and also to select a standard period of coding that could be consistent for all participants as most participants kept their hand in the water for at least ten seconds. For children (n = 5) and parents (*n* =1) who withdrew their hand within the first 10 seconds, facial coding was only conducted for the time that the participant had their hand in the water.

For many participants there were moments where the face could not be coded because of the technical issues (e.g., the camera angle could not capture the participant's face if they lowered their head). For the present analysis, any instance where the face could not be coded was considered missing data and mean scores were calculated for each action unit for each participant so that a score could be calculated regardless of the duration of the pain task or the amount of time the face was visible/able to be coded. Data was included from all participants for whom facial coding data was available (*n* = 110 children and *n* = 127 parents), i.e., the computer was able to detect their face in at least one frame of the video of for the first 10 seconds of the CPT. Additionally, facial expression data was missing from an additional two parent participants due to a technical malfunction.

Typicality of parental behaviour. As a manipulation check, children were asked to rate how typical their parent's behaviour was during the cold pressor task compared to how they usually act when they are in pain, based on a measure by Walker and colleagues (2006b). Responses were rated on a 5-point scale ranging from '*My mom/dad showed a lot more pain than usual*' (scored as 5) to '*My mom/dad showed a lot less pain than usual*' (scored as 1), with the middle option denoting '*My mom/dad acted the same as usual*' (scored as 3).

Children's self-report of pain models at home. Children completed an investigator-created questionnaire inquiring about the presence of any pain models in their lives, based on a pain model interview by Osborne and colleagues (1989).

3.3.3 Cold pressor task

The cold pressor used in the present study was an electric-cooled apparatus by Techne© (www.techne.com). The cold water bath had an 8-Litre stainless steel tank that was cooled using a dip cooler that kept the temperature of the water at 10 degrees Celsius (± 0.3°C). Of note, the dip cooler was turned off during the task itself to ensure that the research assistant who was monitoring the task via cameras from another room could hear the parent and child and monitor for safety. To prevent localized warming to the hand, the water in the cold pressor is circulated at a rate of 10 L/minute with an external pump. Parents and children used their non-dominant hand to complete the task unless they currently had a minor injury to the hand (e.g., a wart) and requested to use their dominant hand instead.

3.3.4 Procedure

Ethical approval for this study was obtained by the institutional research ethics board. Families arrived at the research centre and were introduced to both research assistants (all research assistants involved in the present study were female) and were given a brief explanation of the study procedures. Parents and children were separated and parents provided informed consent while children provided assent to a modified description of the study procedures. This was to ensure that the parent could be informed about the nature of the deception involved in their participation (i.e., manipulating their facial expressions during the pain task) without revealing the deception to their child. Once separated, parents and children were provided more specific details about their participation in the study and given the opportunity to ask questions.

After consent was obtained, parents completed a demographics questionnaire and parents and children completed measures of self-reported gender, the results of which are described elsewhere (Boerner & Chambers, submitted). Then, unbeknownst to their child, parents were randomly assigned to one of three conditions: Exaggerate (parents instructed to exaggerate their facial expression of pain during the CPT), Minimize (parents instructed to minimize their facial expression of pain during the CPT), and Control (parents were told to act they normally would during the CPT, to capture their natural reaction to the task). Randomization was stratified to ensure that there would be an equal number of mother-daughter, mother-son, father-daughter, and father-son dyads in each experimental condition. The scripts for randomization were placed in a sealed opaque envelope that was not opened by study staff until the point of randomization, so that knowledge of the experimental condition that the family was assigned to did not inadvertently influence the consent process or the questionnaires.

Instructions to parents in all conditions were provided using a script (Appendix C). Similar to the procedure used by Goodman and McGrath (2003) parents in the Exaggerate and Minimize conditions were trained on how to manipulate their facial expressions of pain by viewing a two-minute video that displayed clips of individuals (2 adult men and 2 adult women) showing exaggerated or minimized pain expressions, depending on the condition they had been assigned to. Parents in the Exaggerate condition were asked to show facial pain behaviour that would be obvious and realistic to the child, but that would not cause the child to become overly concerned about the parent's level of distress or discomfort. Parents in the Control condition watched a two-minute neutral video of how the CPT equipment is cleaned after use (script and content based on the control video used by Goodman & McGrath (2003)). After watching the video, parents were given the opportunity to ask any questions before being reunited with their child.

Parents and children were then brought back together in the room where the CPT would be completed. The parent sat next to the CPT apparatus with their child sitting across from them, approximately 24 inches away from the parent. The parent was provided with instructions for the CPT read by a research assistant (available from author upon request), and were then asked to repeat back the instructions in their own words to the research assistant to ensure comprehension. Any misunderstandings were corrected. The research assistant then left the room, and the parent completed the CPT while their children observed. Parents placed their hand up to the wrist into the cold water for up to 4 minutes (uninformed ceiling). The facial expression of both the parent and child was audio- and video-recorded during the CPT. Following completion of the CPT, parents and children privately rated the parent's worst and average pain intensity, and children also rated their own anxiety.

The parent and child then switched seats so the child sat next to the cold pressor apparatus. The research assistant provided the same instructions again and checked comprehension with the child, correcting any misunderstandings. Once the research assistant had left the room, the child then completed the cold pressor task while their parent watched, sitting across from them. Children underwent the same protocol for the CPT as the parents, in accordance with guidelines for the use of the CPT with children (von Baeyer et al., 2005). Following completion of the CPT, parents and children privately rated the child's worst and average pain intensity.

Children provided ratings about the typicality of their parent's behaviour during the CPT and reported on the presence of any pain models in their daily lives. At the completion of the study, parents and children were fully debriefed together by a research assistant (script adapted from those used by Goodman & McGrath (2003)). The research assistants left the room while the parent and child completed anonymous post-debriefing questionnaires. Each child received a junior scientist certificate for their participation and $20 to thank them for their time. Each parent received $20 to thank them for their time, and $5 to cover any transportation/parking expenses.

3.3.5 Data analysis

All analyses were conducted using IBM SPSS Statistics version 21. The above description of the facial coding procedures describes how missing data was dealt with for these analyses, and there was no data missing for any of the primary outcome measures (i.e., anxiety, pain intensity, or pain tolerance). Statistical significance tests were evaluated against the conventional alpha level of $p < .05$. No adjustments were made for multiple comparisons as the outcome variables determined *a priori* were expected to be associated with each other (as per previous findings examining pain and anxiety outcomes in the context of experimental pain). It has been previously suggested that in such cases Bonferonni adjustments are not required, and may unnecessarily increase the probability of Type II errors (Rothman, 1990; Schulz & Grimes, 2005). In line with previous suggestions (Perneger, 1998), all analyses are presented as they were conducted, with exact p-values presented to aid interpretation, and the possible interpretations of each finding are discussed.

3.4 Results

Demographics

Table 3.7.1 presents demographics data for each of the three experimental groups. There were no significant demographic differences or differences in baseline questionnaire data between the experimental groups, with the exception of child age:

children who were randomly assigned to the Exaggerate condition were significantly younger than children randomly assigned to the Control condition. For this reason, all analyses comparing children's outcomes between experimental groups were conducted including age as a covariate.

Participants were 168 healthy children (84 boys, 84 girls) between the ages of 6 years 0 months and 8 years 11 months (M = 7.14 years, SD = 0.83), and one of their parents (84 mothers, 84 fathers[2]; M_{age} = 39.59 years, SD = 5.77). The majority of participating children (78.6%, n = 132) and parents (85.1%, n = 143) identified as "White", and most parent-child dyads reporting speaking primarily English together at home (92.3%, n = 155). The majority of participating parents were married (75%, n = 126), had completed university education or graduate/professional training (62.5%, n = 105), and had an average annual household income of $75,000 or greater (60.7%, n = 102). The majority of participating dyads had more than one adult (88%, n = 147) and at least one other child (85.7%, n =144) in their immediate family.

Nineteen percent of parents (n =32) indicated that they had a chronic health condition, and 14% of parents (n=24; 12 mothers and 12 fathers) indicated that they had chronic pain, which was defined as pain of moderate to severe intensity that occurs at least once a month for 3 months in a row. The majority of parents who reported having chronic pain (n = 17, 70.8%) indicated that their child was aware of their chronic pain condition.

Manipulation checks

Child rating of parent's pain during CPT. A one-way multivariate analysis of variance (MANOVA), with experimental condition as the independent variable was conducted for children's ratings of their parent's pain during the cold pressor task (most and average pain). Using Pillai-Bartlett trace, there was a significant effect of experimental group on children's ratings of their parent's pain during the cold pressor, V= 0.33, $F(4,330)= 16.03$, $p < .001$. Post-hoc Bonferonni tests revealed that there was a significant difference between all three groups for both most and average pain (all $p < .05$). Children in the Exaggerate condition rated their parent's pain significantly higher than children in the Control condition, who rated their parent's pain significantly higher than children in the Minimize condition (see Table 3.7.2). This suggests that parents successfully manipulated their expressions in a way that resulted in their children believing that they had more (Exaggerate condition) or less (Minimize condition) pain than parents in the Control group.

Typicality of behaviour. A one-way analysis of variance (ANOVA) was conducted examining children's ratings of typicality of their parent's behaviour during the cold pressor task across the three experimental groups. There was a significant effect of the experimental condition on children's ratings of the typicality of their parent's behaviour, $F(2,164)= 13.182$, $p <.001$. Post-hoc Bonferonni tests indicated that children in the Exaggerate condition reported significantly higher scores ($M = 3.46$, $SD = 1.22$) than children in the Control condition ($M = 2.42$, $SD = 1.05$, $p = .001$) and children in the Minimize condition ($M = 2.64$, $SD = 1.12$, $p < .001$). Of note, mean scores above 3 indicate that the children generally believed their parents were showing more pain than usual and mean scores below 3 indicate that the children generally believed their parents were showing less pain than usual. Therefore, children in the Exaggerate condition reported being attuned to the fact that their parent was enhancing their expression of pain above their normal response; however, the expressions of the parents in the Minimize condition were not noted by their children as being significantly different from those in the Control condition.

Parent pain tolerance. A 3 (experimental group: Exaggerate vs. Minimize vs. Control) by 2 (child sex: son vs. daughter) by 2 (parent sex: father vs. mother) ANOVA was conducted with parent's pain tolerance (measured in seconds) as the dependent variable. There was a significant effect of experimental condition ($F(2,156)=3.972$, $p=.021$, $\eta_p^2=.048$) on parent's pain tolerance, with parents in the Minimize condition leaving their hand in the cold water for longer than parents in the Exaggerate condition (Table 3.7.3). Additionally, there was a significant effect of parent sex ($F(1,156)=5.671$, $p=.018$, $\eta_p^2=.035$) on parent's pain tolerance, with fathers demonstrating longer pain tolerance than mothers. There was no main effect of child sex ($F(1,156)=0.059$, $p=.809$) and no significant interactions.

Parent facial expression of pain. A one-way multivariate analysis of variance was conducted with the experimental condition as the independent variable and parent facial action units as the dependent variable. This MANOVA was conducted twice: once for the frequency and once for the intensity of facial actions during the first 10 seconds of the cold pressor task. Action units 1, 2, 4, 6, 7, 9, 10, 12, 14, 15, 17, 18, 23, 24, 25, and 43 were included in the final analyses, as they each had ≥5% frequency in the overall sample. Significant differences between experimental groups were observed for frequency ($V = 0.43$, $F(32,220) = 1.872$, $p = .005$)[3] and intensity ($V = 0.43$, $F(32,220) = 1.876$, $p = .005$)[4]. This suggests that the experimental condition had a significant impact on how frequent certain facial actions were and how intense their facial displays were during the initial period of the cold pressor task.

Parent self-report data

The following results should be interpreted in the context of the manipulation being parent-enacted (i.e., unlike in the child analyses, parents were aware of the experimental manipulation at the time of the cold pressor task), and that children did not have access to their parent's self-reports of pain (i.e., the ratings were conducted privately). These results are presented to provide context as to the true pain experience of the parents participating in the present study.

A 3 (experimental group: Exaggerate vs. Minimize vs. Control) by 2 (child sex: son vs. daughter) by 2 (parent sex: father vs. mother) ANOVA was conducted with parent's self-reported pain intensity as the dependent variable. For most pain intensity, there was a significant effect of experimental condition ($F(2,156)=4.559$, $p= .012$, $\eta_p^2= .055$), with parents in the Minimize condition reporting less pain than parents in the Exaggerate condition (Table 3.7.3). Additionally, there was a significant effect of parent sex ($F(1,156)=12.313$, $p= .001$, $\eta_p^2= .073$) on parent's most pain intensity, with mothers reporting higher pain intensity than fathers. Of note, when children were asked to rate their parent's most pain intensity there was a similar sex difference in that children observing their mother rated their parent's pain as being significantly higher than children observing their father, $F(1,156)=8.962$, $p= .003$, $\eta_p^2= .054$.

For average pain intensity, there was a significant effect of parent sex ($F(1,156)=13.502$, $p< .001$, $\eta_p^2= .080$), with mothers reporting greater average pain than fathers (Table 3.7.3). Additionally, there was a significant effect of parent sex by experimental condition interaction ($F(2,156)=4.108$, $p= .018$, $\eta_p^2= .050$) on parent's pain intensity, with mothers reporting significantly higher average pain than fathers in the Minimize and Exaggerate conditions, but not in the Control condition. There was no main effect of experimental condition ($F(2,156)=1.318$, $p=.271$), child sex ($F(1,156)=0.446$, $p=.505$), and no other significant interactions.

Child pre-CPT anxiety

A 3 (experimental group: Exaggerate vs. Minimize vs. Control) by 2 (child sex: son vs. daughter) by 2 (parent sex: father vs. mother) analysis of variance (ANOVA)[5] was conducted with children's self-reported anxiety as the dependent variable. Table 3.7.4 reports the means and standard deviations for each group. There was a significant effect of experimental condition on children's anxiety, with a medium-large effect size ($F(2,156)=8.649$, $p< .001$, $\eta_p^2= .100$). Pairwise comparisons revealed that children in the Exaggerate condition reported significant higher anxiety after observing their parent's cold pressor task than children in the Minimize condition. There was no effect of child sex ($F(1,156)=0.914$, $p=.340$) or parent sex ($F(1,156)=0.115$, $p=.735$), and no significant interactions.

Child pain outcomes

Child CPT pain intensity. A 3 (experimental group: Exaggerate vs. Minimize vs. Control) by 2 (child sex: son vs. daughter) by 2 (parent sex: father vs. mother) ANOVA[5] was conducted twice: once with children's self-reported *most* pain intensity as rated on the FPS-R and once with children's self-reported *average* pain intensity during the cold pressor task as rated using the FPS-R. Table 3.7.4 reports the means and standard deviations for each group. There were no significant main effects of experimental condition ($F(2,156)=0.821$, $p= .442$), child sex ($F(1,156)= 0.008$, $p = .931$), or parent sex ($F(1,156)=2.736$, $p=.100$) reported for on children's ratings of most pain during the CPT and no significant interactions were observed. When the same ANOVA was conducted with parent proxy ratings of the child's most pain during the CPT, there were also no significant main effects of interactions observed.

With regards to average pain intensity, there was no main effect of experimental condition ($F(2,156)= 0.540$, $p=.584$), child sex ($F(1,156)= 0.095$, $p= .758$), or parent sex ($F(1,156)= 2.963$, $p=.087$) on children's average pain intensity. However, there was a significant interaction between child sex and experimental condition, $F(2,156) = 3.532$, $p=.032$, $\eta_p^2= .043$. A series of t-tests conducted for each of the three experimental conditions revealed that girls in the Exaggerate condition reported significantly greater average pain intensity than boys in the Exaggerate condition, $t(54) = -2.156$, $p = .036$. No child sex differences were observed for the Control or Minimize conditions. No other significant interactions were observed for children's average pain intensity. When the same ANOVA was conducted with parent proxy ratings of the child's average pain during the CPT, no main effects and no interactions were observed.

Child CPT pain tolerance. A 3 (experimental group: Exaggerate vs. Minimize vs. Control) by 2 (child sex: son vs. daughter) by 2 (parent sex: father vs. mother) ANOVA[5] was conducted with children's pain tolerance (measured in seconds) as the dependent variable. Table 3.7.4 reports the means and standard deviations for each group. There were no significant main effects of experimental condition ($F(2,156)= 0.811$, $p=.446$), child sex ($F(1,156)=0.023$, $p= .881$), or parent sex ($F(1,156)= 0.109$, $p= .742$) on children's pain tolerance, and no interactions were observed.

Exploratory analyses of child facial expression of pain. To first provide evidence for the construct validity of the computerized facial coding for children in this study, correlations between children's facial coding scores and child self-reported pain outcomes were examined. Two 'pain expression' composite scores (one for frequency and one for intensity) were calculated for each participant by summing the mean frequency and mean intensity of each action unit over the entire duration of the CPT[6]. Note that only action units that had a minimum of 5% occurrence and have been identified in previous research as being associated with the pain expression (AUs 4, 6, 7, 9, 10, 12, 43) were included in the composite score (Craig & Patrick, 1985; Larochette et al., 2006; Prkachin, 1992). The composite score for frequency of pain-related facial actions was significantly positively correlated with children's self-reported most pain (r = .175, p= .034), indicating that, as expected, children who reported their pain as being more intense at its worst also displayed pain-related facial actions more frequently. While the composite of the intensity of pain-related facial actions was not significantly correlated with children's self-reported most pain (r = .159, p= .054), the facial action composites were also significantly negatively correlated with children's pain tolerance (r = -.246, p= .003 for frequency; r = -.241, p= .003 for intensity), indicating that, again as expected, children who tolerated the pain task for less time also displayed more frequent and intense pain-related facial actions.

With respect to the exploratory analyses involving the child facial expression data, two MANOVAs were conducted: one for frequency and one for intensity scores in the first ten seconds of the cold pressor task. The MANOVAs were conducted with condition, child sex, and parent sex as the independent variables, child age as a covariate, and the facial action units as the dependent variables. Action units 1, 2, 4, 6, 7, 9, 10, 12, 17, 23, 24, 25, 27, and 43 were included in the final analyses, as they each had ≥5% frequency in the sample over the entire CPT duration.

No significant differences in frequency of child facial actions during the initial period of the CPT were observed based on experimental condition (V = 0.40, F(28,170) =

1.499, $p = .062$), child sex ($V = 0.21$, $F(14,84) = 1.570$, $p = .105$) or parent sex ($V = 0.09$, $F(14,84) = 0.557$, $p = .890$), and there were no significant interactions (Table 3.7.5). However, significant differences were observed in the analyses of the intensity of facial actions in the first 10 seconds of the CPT (Table 3.7.6). There was a significant effect of condition: $V = 0.44$, $F(28,170) = 1.716$, $p = .020$. Follow-up to the MANOVA with simple contrasts (using the Control group as the reference category) indicated that action unit 23 (lip tightener) was more intense in the Exaggerate condition compared to the Control group, while action units 7 (lid tightener), 9 (nose wrinkler), 25 (lips part), and 43 (eyes closed) were more intense in the Control condition compared to the Exaggerate condition. Action unit 25 was also more intense in the Control condition compared to the Minimize condition. There was also a significant effect of child sex ($V = 0.24$, $F(14,84) = 1.877$, $p = .041$), with simple contrasts indicating that action unit 43 (eyes closed) was more intense in boys compared to girls. There was no effect of parent sex ($V = 0.12$, $F(14,84) = 0.841$, $p = .623$) and no significant interactions with respect to facial action intensity.

Pain models

Nearly half of the sample of participating children (43%; $n = 37$ boys and 36 girls) reported the presence of at least one pain model in their lives[7]. The most commonly reported pain models were friends (32%), followed by siblings (27%), and parents (16%), and seven children identified themselves when asked “Do you know anyone who has pain or hurts a lot?” Of the 93 pain models that were not the participating child themselves, half of the models were male ($n = 45$; 48.4%), and one third of the models were female ($n = 29$; 31.2%). For the remaining models, sex was not reported and/or could not be inferred from the child’s response. Seven children identified their participating parent as being a pain model, though interestingly none of those parents reported having chronic pain. Alternatively, of the 25 parents who reported having chronic pain, none of their children identified them as a pain model. In fact, half of the children of parents with chronic pain did identify a pain model, but it was not their parent with chronic pain.

3.5 Discussion

The present study builds on the previous experimental investigations of parental modeling of pain behaviours by Goodman and McGrath (2003) by examining the impact of parent and child sex. The manipulation was successful in altering children's perceptions of the amount of pain their parent was experiencing during the cold pressor task: children in the Exaggerate condition reported that their parent was experiencing more pain than children whose parents were in the Control or Minimize conditions. This manipulation had a significant impact on children's anxiety prior to completing the CPT, in that children who had just observed their parent exaggerate their response to pain reported significantly more anxiety than children who had observed their parent minimize their expression of pain. There may have been an increase in children's fear resulting from their parent's reaction to this novel task, which could be related both to the signal of threat associated with the expression of pain, and potentially the unexpected degree of intensity (i.e., children reported that their parent was showing more pain than usual). Additionally, it is possible that observing their parent minimize their reaction had a buffering effect against children's anxiety. This finding has particular relevance to the understanding of how pain-related fear may develop via observation of a modeled behaviour, which has been suggested as a mechanism in needle phobia and other procedure-related fears, and could be harnessed as a potential point for clinical intervention (Willemsen, Chowdhury, & Briscall, 2002). Future research should examine whether observing parental behaviour during pain modifies children's expectancies with regards to an impending pain experience (i.e., by measuring children's expected pain prior to and following the observation of their parent's pain), and whether this impacts children's confidence in their ability to cope with the pain.

Children's pain tolerance during the CPT did not differ based on parent and child sex, or experimental manipulation. However, when children were asked to rate their average pain intensity, girls reported significantly greater average pain intensity than boys in the Exaggerate condition. This finding suggests that, as hypothesized, observing a parent exaggerating their expression of pain had a greater impact on girl's pain intensity than boys, but that the impact of observing a natural or minimized facial expression of pain from their parent did not differentially impact boys and girls. Previous research has found that it is more socially acceptable for females to express pain, and that boys are unlikely to model behaviour that is considered to be "gender-inappropriate", which may account for the difference observed here (Fillingim et al., 2009; Keogh, 2014; Perry & Bussey, 1979; Raskin & Israel, 1981). It is also possible that girls are more likely to attend more and be influenced by social cues in their environment than boys(Bayliss, di Pellegrino, & Tipper, 2005).

Parent sex differences were observed in the directions consistent with previous research (Fillingim et al., 2009): mothers reported greater pain intensity than fathers during the CPT, and fathers tolerated the task longer than mothers. Children appeared to be aware of this difference, as children observing their mother complete the CPT rated her worst pain intensity to be higher than children observing their father complete the

CPT. Additionally, the experimental manipulation had impact on parent's pain responses, as parents in the Exaggerate condition reported their worst pain to be significantly higher and tolerated the pain for less time than parents in the Minimize condition. It is unclear whether these differences are due to the parent's interpretations of what was required (for example, even though parents were not given instructions on how long to keep their hand in the water and were only explicitly asked to manipulate their facial expressions, parents in the Minimize condition may have thought their manipulation would be more convincing if they kept their hand in the water for longer than they normally would, and vice versa for the Exaggerate condition). Similarly, even though parents completed their ratings of their own pain out of the view of the child, it is possible that they thought they were supposed to still be "in character" while completing their pain ratings, or misinterpreted the FPS-R to indicate that they were supposed to rate how much pain they were showing. Alternatively, these experimental group differences in the parent's pain experiences during the CPT could possibly also be due to an effect of the manipulation itself.

The exploratory analyses of facial expression indicated that participant's facial expressions differed between experimental groups during the initial phase of the cold pressor task, similar to the findings of Goodman and McGrath (2003). While previous studies have chosen to study only those action units that are known to be present during the expression of pain, the present study examined all action units available in the coding software with a frequency of at least 5%, in case action units that are not typically associated with pain were observed during one of the experimental manipulations (faked/exaggerated expressions of pain have been shown to be associated with activation of different action units than genuine expressions (Craig, Hyde, & Patrick, 1991;

Larochette et al., 2006)). Indeed, many of the action units on which group differences were observed in the present study were those not historically found to be associated with the pain expression. Interestingly, while both parents and children in the present study differed in facial expression based on experimental group, the specific action units on which the differences were observed were not the same between parents and children. This suggests that the impact of the manipulation influenced children's facial expressions in response to the CPT, but that children were perhaps not directly mimicking the expressions of their parents. While the results of the present study must be interpreted with caution as the use of automated coding in these samples and for these purposes is still being developed, there is a movement towards computerized facial coding in the field of pain (Bartlett, Littlewort, Frank, & Lee, 2014; Littlewort, Bartlett, & Lee, 2007; Littlewort, Bartlett, & Lee, 2009; Sikka, Dhall, & Bartlett, 2014; Sikka et al., 2015; Werner et al., 2016), with the results of the present study suggesting validity of this method, at least with regards to the relationship between the coded facial expressions and pain responses.

Nearly half of the children in this community sample reported having at least one pain model in their lives (i.e., a person they know who has pain or hurts a lot). Contrary to the hypothesis that children would frequently identify their parent as a pain model in their lives, friends and siblings were more commonly reported as a pain model. This suggests same-age peers are relevant and salient pain models for children. An alternative hypothesis is that parents actively seek to hide or minimize their pain from their children. Congruent with previous reports calling into question the reliability of retrospective offspring reports of parental chronic pain status (Bruehl et al., 2005; Ottman et al., 1993), the present study demonstrated that even when asked in childhood, offspring reports of parental chronic pain are unreliable. However, offspring reports of parental chronic pain, and not the confirmed presence of chronic pain itself, has been shown to be a significant predictor of the offspring's own pain (Bruehl et al., 2005).

The present study has a number of strengths to be considered, including the use of an experimental design to examine the social modeling process directly as it occurs. This study improved on previous pediatric pain research by recruiting a large sample that included an equal number of boys and girls, mothers and fathers. However, as children were only exposed to a model of one sex (i.e., either their mother *or* their father), it is possible that less sex-specific modeling was observed than if the child had a choice as to which sex of model to attend to (Bussey & Perry, 1982). Additionally, while the present study only examined the impact of parental behaviour on child outcomes, it is highly likely that parents modified their behaviour in response to child feedback, e.g., a child who is overtly anxious in response to their parent's pain may evoke a different parental response than a child who is not (Scarr & McCartney, 1983). Future research should examine the bidirectionality of the social learning process as it relates to pediatric pain. Finally, the present study included a sample of primarily White, high-socioeconomic status and highly educated families, which may limit the extent to which conclusions can be drawn to other populations. Future studies examining these processes in other populations and utilizing observational methods (e.g., during family visits for influenza vaccinations) would offer further insight into social modeling in real-world settings.

Overall, having a parent exaggerate their expression of pain appeared to be related to increased child anxiety and greater pain intensity in girls. Future research may examine whether the modeling of specific adaptive or maladaptive coping behaviours from the parent in pain may be modeled to the child.

3.6 Acknowledgments

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3.7 Tables

Table 3.7.1.

Demographic data by experimental condition.

	Experimental condition			
	Exaggerate (n = 56 dyads)	Minimize (n = 56 dyads)	Control (n = 56 dyads)	Group differences[a]
Child sex	Girl = 28 Boy = 28	Girl = 28 Boy = 28	Girl = 28 Boy = 28	–[b]
Child age	M = 6.99 yrs SD = 0.74	M = 7.06 yrs SD = 0.85	M = 7.37 yrs SD = 0.87	$F(2,165) = 3.362$, $p = .037$[c]
Child ethnicity				
White	n = 42	n = 45	n = 45	$\chi^2(2, N = 168) = 0.636$, $p = .727$
Other	n = 14	n = 11	n = 11	

	Experimental condition			
	Exaggerate (*n* = 56 dyads)	Minimize (*n* = 56 dyads)	Control (*n* = 56 dyads)	Group differences[a]
Parent sex	Woman = 28 Man = 28	Woman = 28 Man = 28	Woman = 28 Man = 28	-[b]
Parent age	M = 39.9 yrs SD = 6.53	M = 39.1 yrs SD = 4.95	M = 39.8 yrs SD = 5.77	$F(2,165) = 0.292$, $p = .747$
Parent ethnicity				
White	$n = 49$	$n = 47$	$n = 47$	$\chi^2(2, N = 168) = 0.376$, $p = .829$
Other	$n = 7$	$n = 9$	$n = 9$	
Parent pain status				
Chronic pain	$n = 10$	$n = 6$	$n = 8$	$\chi^2(2, N = 168) = 1.167$, $p = .558$
No chronic pain	$n = 46$	$n = 50$	$n = 48$	
% of time dyad lives together[d]				
100%	$n = 49$	$n = 48$	$n = 46$	$\chi^2(2, N = 167) = 0.833$, $p = .659$
50-99%	$n = 7$	$n = 7$	$n = 10$	

[a] Group differences were tested with one-way ANOVAs for continuous variables, and chi-squared analysis for categorical variables.

[b] No tests of group differences conducted as randomization was designed such that the sex distribution would be equal across groups.
[c] Significant difference between Exaggerate and Control conditions (p = .049).
[d] One response missing from a family in the Minimize condition.

Table 3.7.2.

Mean (SD) of children's ratings of their parent's pain by experimental condition and sex-paired dyads.

	Exaggerate				Minimize				Control			
	Mother-daughter (*n*=14)	Mother-son (*n*=14)	Father-daughter (*n*=14)	Father-son (*n*=14)	Mother-daughter (*n*=14)	Mother-son (*n*=14)	Father-daughter (*n*=14)	Father-son (*n*=14)	Mother-daughter (*n*=14)	Mother-son (*n*=14)	Father-daughter (*n*=14)	Father-son (*n*=14)
Child rating of parent's most pain intensity (FPS-R)	6.57 (3.63)	8.00 (2.35)	5.57 (3.34)	5.43 (3.37)	2.29 (2.33)	3.00 (4.06)	1.14 (1.88)	1.43 (1.99)	5.29 (3.39)	3.14 (1.70)	3.29 (2.79)	3.29 (3.29)
Child rating of parent's average pain intensity (FPS-R)	6.86 (2.57)	6.43 (2.50)	6.29 (3.02)	5.43 (3.18)	2.57 (2.87)	1.29 (1.86)	2.14 (2.41)	3.14 (3.21)	4.57 (3.80)	4.00 (2.08)	3.29 (2.43)	4.00 (3.76)

Table 3.7.3.

Mean (SD) of parent's CPT pain tolerance and self-reported pain intensity by experimental condition and sex-paired dyads.

	Exaggerate				Minimize				Control			
	Mother-daughter (*n*=14)	Mother-son (*n*=14)	Father-daughter (*n*=14)	Father-son (*n*=14)	Mother-daughter (*n*=14)	Mother-son (*n*=14)	Father-daughter (*n*=14)	Father-son (*n*=14)	Mother-daughter (*n*=14)	Mother-son (*n*=14)	Father-daughter (*n*=14)	Father-son (*n*=14)
Pain tolerance (sec)	94.64 (80.34)	69.00 (53.26)	107.71 (82.22)	105.43 (77.97)	131.29 (89.82)	121.21 (83.08)	149.86 (96.39)	159.57 (97.07)	83.50 (86.42)	114.50 (100.43)	133.43 (99.52)	150.29 (88.73)
Most pain intensity (FPS-R)	5.57 (2.62)	7.57 (1.40)	5.43 (1.99)	5.00 (1.88)	5.29 (1.68)	6.00 (2.08)	3.57 (2.85)	4.00 (2.22)	4.71 (2.67)	5.57 (1.79)	4.86 (2.18)	4.86 (2.03)
Average pain intensity (FPS-R)	4.00 (1.92)	5.43 (1.99)	3.71 (1.54)	3.00 (1.71)	4.43 (2.10)	4.43 (1.79)	2.57 (2.14)	2.57 (1.22)	3.43 (2.53)	3.71 (1.33)	3.57 (1.79)	3.71 (1.73)

Table 3.7.4.

Mean (SD) of children's pre-CPT anxiety and CPT pain outcomes by experimental condition and sex-paired dyads.

	Exaggerate				Minimize				Control			
	Mother-daughter (*n*=14)	Mother-son (*n*=14)	Father-daughter (*n*=14)	Father-son (*n*=14)	Mother-daughter (*n*=14)	Mother-son (*n*=14)	Father-daughter (*n*=14)	Father-son (*n*=14)	Mother-daughter (*n*=14)	Mother-son (*n*=14)	Father-daughter (*n*=14)	Father-son (*n*=14)
Pre-CPT anxiety [a]	3.89 (3.93)	5.49 (3.61)	5.36 (3.46)	6.37 (3.98)	2.52 (2.69)	2.79 (3.70)	2.10 (2.36)	2.91 (3.03)	4.01 (3.37)	4.24 (3.74)	4.08 (3.41)	3.19 (3.49)
Pain tolerance (sec)	102.79 (94.40)	46.64 (68.00)	55.14 (44.07)	66.07 (78.58)	52.14 (80.49)	31.29 (21.59)	53.86 (79.50)	66.21 (79.20)	64.29 (81.14)	82.00 (97.93)	44.64 (58.95)	70.14 (86.34)
Most pain intensity (FPS-R)	4.86 (4.05)	3.43 (3.63)	5.14 (3.48)	3.57 (3.44)	5.43 (3.08)	6.86 (3.74)	3.86 (3.08)	4.29 (3.75)	4.86 (3.30)	5.29 (3.81)	4.00 (3.33)	4.43 (3.69)
Average pain intensity (FPS-R)	5.43 (4.33)	3.29 (3.97)	5.29 (3.47)	3.14 (3.30)	5.00 (3.31)	7.14 (3.98)	3.71 (3.02)	4.00 (3.68)	4.57 (3.08)	5.29 (3.38)	4.43 (2.62)	4.57 (3.46)

[a]Pre-CPT anxiety measured after viewing parent's CPT and rating the parent's pain. Ratings provided on a 10cm Visual Analogue Scale, possible range of responses is from 'Not nervous or anxious at all' (0 cm) to 'Most nervous or anxious' (10 cm).

Table 3.7.5.

Mean (SD) of the frequency of children's facial action units during the first 10 seconds of the cold pressor task, by experimental condition and sex-paired dyads.

	Exaggerate				Minimize				Control			
	Mother-daughter (*n*=13)	Mother-son (*n*=8)	Father-daughter (*n*=8)	Father-son (*n*=9)	Mother-daughter (*n*=7)	Mother-son (*n*=10)	Father-daughter (*n*=9)	Father-son (*n*=8)	Mother-daughter (*n*=9)	Mother-son (*n*=9)	Father-daughter (*n*=11)	Father-son (*n*=9)
AU1 (inner brown raiser)	.089 (.128)	.098 (.217)	.112 (.306)	.102 (.186)	.327 (.464)	.137 (.226)	.150 (.330)	.000 (.000)	.003 (.004)	.002 (.006)	.223 (.395)	.204 (.305)
AU2 (outer brow raiser)	.238 (.316)	.129 (.234)	.041 (.115)	.296 (.374)	.137 (.362)	.117 (.221)	.196 (.328)	.024 (.062)	.211 (.334)	.200 (.260)	.270 (.396)	.244 (.428)
AU4 (brow lowerer)	.363 (.456)	.616 (.454)	.379 (.403)	.386 (.392)	.654 (.458)	.576 (.425)	.549 (.447)	.520 (.426)	.447 (.382)	.358 (.423)	.514 (.423)	.564 (.478)
AU6 (cheek raiser)	.834 (.290)	.519 (.491)	.668 (.385)	.901 (.234)	.641 (.450)	.887 (.200)	.834 (.331)	.841 (.300)	.750 (.337)	.799 (.336)	.536 (.470)	.724 (.421)

	Exaggerate				Minimize				Control			
	Mother-daughter (*n*=13)	Mother-son (*n*=8)	Father-daughter (*n*=8)	Father-son (*n*=9)	Mother-daughter (*n*=7)	Mother-son (*n*=10)	Father-daughter (*n*=9)	Father-son (*n*=8)	Mother-daughter (*n*=9)	Mother-son (*n*=9)	Father-daughter (*n*=11)	Father-son (*n*=9)
AU7 (lid tightener)	.338 (.354)	.408 (.397)	.528 (.380)	.518 (.381)	.511 (.476)	.727 (.405)	.347 (.416)	.735 (.456)	.839 (.299)	.682 (.479)	.541 (.442)	.491 (.490)
AU9 (nose wrinkle)	.122 (.281)	.047 (.133)	.205 (.392)	.025 (.076)	.143 (.378)	.197 (.311)	.092 (.180)	.047 (.124)	.219 (.286)	.382 (.373)	.088 (.107)	.235 (.387)
AU10 (upper lid raiser)	.036 (.071)	.094 (.186)	.092 (.259)	.109 (.326)	.143 (.378)	.051 (.101)	.156 (.329)	.125 (.354)	.176 (.337)	.219 (.384)	.085 (.216)	.231 (.437)
AU12 (lip corner puller)	.978 (.069)	.978 (.045)	.804 (.381)	.971 (.065)	.992 (.016)	.996 (.008)	.886 (.305)	.988 (.035)	.868 (.328)	.830 (.346)	.865 (.215)	.886 (.311)
AU17 (chin raiser)	.219 (.341)	.221 (.359)	.231 (.354)	.224 (.380)	.168 (.373)	.167 (.315)	.156 (.330)	.327 (.385)	.314 (.352)	.378 (.428)	.219 (.282)	.268 (.390)

	Exaggerate				Minimize				Control			
	Mother-daughter (n=13)	Mother-son (n=8)	Father-daughter (n=8)	Father-son (n=9)	Mother-daughter (n=7)	Mother-son (n=10)	Father-daughter (n=9)	Father-son (n=8)	Mother-daughter (n=9)	Mother-son (n=9)	Father-daughter (n=11)	Father-son (n=9)
AU23 (lip tightener)	.049 (.104)	.250 (.463)	.022 (.049)	.000 (.000)	.000 (.000)	.032 (.101)	.000 (.000)	.000 (.000)	.001 (.003)	.000 (.000)	.038 (.116)	.000 (.000)
AU24 (lip pressor)	.230 (.237)	.023 (.054)	.183 (.346)	.259 (.362)	.084 (.123)	.175 (.241)	.043 (.094)	.000 (.000)	.080 (.116)	.141 (.214)	.165 (.338)	.122 (.257)
AU25 (lips part)	.529 (.357)	.413 (.384)	.590 (.424)	.401 (.386)	.497 (.433)	.465 (.326)	.422 (.455)	.406 (.351)	.823 (.246)	.633 (.393)	.712 (.361)	.639 (.450)
AU27 (mouth stretch)	.002 (.007)	.000 (.000)	.022 (.062)	.109 (.326)	.143 (.378)	.025 (.079)	.000 (.000)	.000 (.000)	.100 (.164)	.120 (.301)	.117 (.305)	.222 (.441)
AU43 (eyes closed)	.230 (.299)	.310 (.378)	.386 (.361)	.330 (.442)	.184 (.366)	.435 (.427)	.171 (.333)	.598 (.375)	.471 (.335)	.466 (.408)	.343 (.414)	.514 (.430)

Note. Only AUs included in the final analyses (i.e., that had a mean frequency of 5% or greater) are present in this table. As a mean score for each AU was calculated for each participant for the first 10 seconds of the cold pressor, and frequency was coded as 0 (not present) or 1 (present), the above means are represented on a scale of 0 to 1, with 0 meaning the AU was never present for any participant during the first 10 seconds of the CPT, and 1 meaning that the AU was present 100% of the time for all participants during the first 10 seconds of the CPT. Note that due to missing data (e.g., computer modeling failed to detect the face of the participant in the video), number of participants per group is not equivalent to the overall sample size.

Table 3.7.6.

Mean (SD) of the intensity of children's facial action units during the first 10 seconds of the cold pressor task, by experimental condition and sex-paired dyads.

	Exaggerate				Minimize				Control			
	Mother-daughter (*n*=13)	Mother-son (*n*=8)	Father-daughter (*n*=8)	Father-son (*n*=9)	Mother-daughter (*n*=7)	Mother-son (*n*=10)	Father-daughter (*n*=9)	Father-son (*n*=8)	Mother-daughter (*n*=9)	Mother-son (*n*=9)	Father-daughter (*n*=11)	Father-son (*n*=9)
AU1 (inner brown raiser)	.145 (.239)	.223 (.483)	.112 (.306)	.191 (.363)	.862 (1.302)	.270 (.594)	.281 (.656)	.000 (.000)	.008 (.012)	.002 (.006)	.625 (1.158)	.466 (.775)
AU2 (outer brow raiser)	.557 (.815)	.297 (.629)	.055 (.156)	.677 (.950)	.470 (1.244)	.337 (.763)	.467 (.981)	.024 (.062)	.645 (1.247)	.407 (.668)	.741 (1.210)	.847 (1.592)
AU4 (brow lowerer)	1.228 (1.701)	2.218 (1.935)	.768 (1.030)	.866 (1.094)	1.598 (1.469)	1.802 (1.529)	1.343 (1.460)	1.511 (1.710)	1.349 (1.601)	.818 (.972)	1.415 (1.387)	1.699 (1.649)
AU6 (cheek raiser)	1.426 (.759)	1.046 (1.067)	1.284 (.949)	1.610 (.611)	1.273 (1.019)	2.002 (1.089)	1.510 (.877)	1.799 (.693)	1.388 (.792)	1.449 (.939)	1.041 (1.053)	1.608 (1.110)

	Exaggerate				Minimize				Control			
	Mother-daughter (*n*=13)	Mother-son (*n*=8)	Father-daughter (*n*=8)	Father-son (*n*=9)	Mother-daughter (*n*=7)	Mother-son (*n*=10)	Father-daughter (*n*=9)	Father-son (*n*=8)	Mother-daughter (*n*=9)	Mother-son (*n*=9)	Father-daughter (*n*=11)	Father-son (*n*=9)
AU7 (lid tightener)	.380 (.398)	.670 (.713)	.890 (.944)	.553 (.370)	.828 (1.061)	1.737 (1.165)	.613 (.730)	1.515 (1.100)	1.612 (.694)	1.504 (1.147)	.991 (.938)	1.076 (1.207)
AU9 (nose wrinkle)	.287 (.670)	.047 (.133)	.499 (.964)	.025 (.076)	.429 (1.134)	.446 (.774)	.112 (.210)	.076 (.207)	.607 (.808)	.925 (1.011)	.156 (.217)	.648 (1.131)
AU10 (upper lid raiser)	.076 (.176)	.094 (.186)	.145 (.411)	.306 (.917)	.429 (1.134)	.062 (.131)	.373 (.875)	.375 (1.061)	.186 (.336)	.413 (.761)	.206 (.507)	.496 (.941)
AU12 (lip corner puller)	3.681 (.663)	3.517 (.568)	3.019 (1.651)	3.741 (.647)	3.640 (.443)	3.832 (.232)	3.462 (1.287)	3.789 (.425)	3.403 (1.308)	3.241 (1.353)	3.098 (1.212)	3.421 (1.239)
AU17 (chin raiser)	.611 (.962)	.622 (1.094)	.918 (1.735)	.621 (1.115)	.485 (1.119)	.454 (.798)	.511 (1.317)	.800 (.888)	.912 (1.190)	1.257 (1.792)	.516 (.663)	.975 (1.534)
AU23 (lip tightener)	.158 (.383)	.757 (1.402)	.041 (.076)	.000 (.000)	.000 (.000)	.097 (.306)	.000 (.000)	.000 (.000)	.003 (.009)	.000 (.000)	.089 (.288)	.000 (.000)

	Exaggerate				Minimize				Control			
	Mother-daughter (*n*=13)	Mother-son (*n*=8)	Father-daughter (*n*=8)	Father-son (*n*=9)	Mother-daughter (*n*=7)	Mother-son (*n*=10)	Father-daughter (*n*=9)	Father-son (*n*=8)	Mother-daughter (*n*=9)	Mother-son (*n*=9)	Father-daughter (*n*=11)	Father-son (*n*=9)
AU24 (lip pressor)	.462 (.520)	.025 (.059)	.443 (.828)	.714 (1.327)	.091 (.132)	.260 (.432)	.052 (.115)	.000 (.000)	.097 (.137)	.274 (.508)	.382 (.913)	.191 (.439)
AU25 (lips part)	1.256 (1.242)	1.206 (1.604)	1.784 (1.493)	1.052 (1.273)	1.563 (1.744)	1.139 (1.186)	1.241 (1.676)	1.256 (1.175)	2.765 (1.322)	1.855 (1.604)	2.575 (1.604)	2.272 (1.708)
AU27 (mouth stretch)	.007 (.026)	.000 (.000)	.025 (.070)	.295 (.886)	.571 (1.512)	.036 (.115)	.000 (.000)	.000 (.000)	.280 (.589)	.393 (1.117)	.227 (.590)	.882 (1.751)
AU43 (eyes closed)	.619 (.822)	.901 (1.374)	.874 (.798)	.591 (.883)	.476 (1.115)	1.354 (1.476)	.578 (1.323)	1.781 (1.365)	1.627 (1.593)	1.660 (1.822)	1.202 (1.554)	1.812 (1.851)

Note. Only AUs included in the final analyses (i.e., that had a mean frequency of 5% or greater) are present in this table. Intensity for the Facial Action Coding System is coded on a 6-point scale, ranging from 0 (action unit not present) to 5 (action unit present at maximum intensity). Note that due to missing data (e.g., computer modeling failed to detect the face of the participant in the video), number of participants per group is not equivalent to the overall sample size.

3.8 Footnotes

1 Though children and parents were asked not to touch the water until instructed, and not to place their hand back in the water once they had removed it, some participants did not follow these instructions. In such situations, pain tolerance time was defined as commencing at the first full immersion of the hand after the research assistant's instruction to lower their hand into the water, and ending at the point that the hand was fully removed from the water.

2 The participating parents included 82 mothers, 83 fathers, 1 stepmother, 1 stepfather, and 1 grandmother.

3 Simple contrasts with the Control group as the reference category indicated that action units 2 (outer brow raiser) and 6 (cheek raiser) were more frequent in the Control compared to Minimize condition, and action units 10 (upper lip raiser), 14 (dimpler), 15 (lip corner depressor), and 24 (lip pressor) were more frequent in the Minimize condition compared to the Control condition. Action unit 18 (lip puckerer) was more frequent in the Exaggerate condition compared to the Control condition.

4 Simple contrasts with the Control group as the reference category indicated that action units 1 (inner brow raiser) and 2 (outer brow raiser), were more intense in the Control condition compared to the Minimize condition, action units 10 (upper lip raiser), 14 (dimpler), and 24 (lip pressor) were more intense in the Minimize condition than the Control condition, and that action units 4 (brow lowerer), 17

(chin raiser), and 18 (lip puckerer) were more intense in the Exaggerate condition compared to the Control condition.

[5] As there was a significant difference between experimental groups in child age, analyses for all child pain responses (anxiety, pain intensity, pain tolerance, and facial expression) were conducted including child age as a covariate. However, as this did not change the significance of any of the results, the simplest analysis (i.e., without the covariate) is presented here.

[6] Note that due to missing data (e.g., computer modeling failed to detect the face of the participant in the video), number of participants (n = 147) is not equivalent to the overall sample size, or the sample size used for the analysis of only the first 10 seconds.

[7] The results for all of the pain models reported by children are presented here, and as several children reported on more than one pain model, the total number of pain models (n= 100) is greater than the total number of children who reported the presence of a pain model.

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CHAPTER 4. PARENT AND CHILD GENDER ROLES AND THEIR ASSOCIATION WITH PAIN RESPONSES

The manuscript prepared for this study is presented below. Readers are advised that Katelynn Boerner, under the supervision of Dr. Christine Chambers, was responsible for all aspects of the project, including securing funding, developing the research questions and protocol, obtaining ethical approval, training and supervising study staff on participant recruitment and data collection, as well as data analysis and manuscript preparation. The manuscript was submitted for publication on April 18, 2016. The current reference for this manuscript is:

Boerner, K.E. & Chambers, C.T. (submitted). Parent and child gender roles and their association with pain responses.

4.1 Abstract

Objective: Gender (the characteristics and behaviours considered to be typically masculine or feminine) is related to health outcomes in adults; few studies have considered gender in the context of child health. The present study examined gender in pediatric pain.

Methods: Participants were 168 parent-child dyads (50% fathers; 50% sons), who completed self-reported measures of gender. Parents completed the cold pressor task (CPT) while their child watched, then children completed the CPT themselves.

Results: Higher levels of masculinity in fathers was related to higher pain tolerance and lower pain intensity during the CPT. Gender was not related to self-reported pain in children or mothers. In children, lower anxiety after watching their parent complete the CPT was predicted by higher femininity in girls, but not in boys.

Conclusions: Gender influences aspects of the pain experience in children and parents. Additional research is needed to explore the impact of gender on pain.

4.2 Introduction

Gender is defined by the World Health Organization (2012) as *"the socially constructed roles, behaviours, activities, and attributes that a given society considers appropriate for men and women."* Gender has been of interest in the study of children with gender dysphoria or disorders of sex development (Jürgensen et al., 2014; Reiner, 2005; Stout, Litvak, Robbins, & Sandberg, 2010; Sung, Han, Chung, Lee, & Cho, 2014; Thyen, Richter-Appelt, Wiesemann, Holterhus, & Hiort, 2005). However, gender is a construct that influences the health and behaviour of *all* individuals, and may offer a means to help understand the significant variability that is observed within the sexes. As described by Walter Mischel, a pioneer in research on the learning of gendered behaviour: *"(...) the abundant individual differences found within each sex, and the fact that the behaviours of the sexes overlap to a great degree, suggest that there are many ways to be a boy or girl (...)"* (Mischel (1966), p. 62). Investigating the role of gender in health is more complicated than simply comparing males and females, as it involves measuring constructs that are continuously changing on the basis of the sociocultural environment and development. Further, gender both influences and is influenced by an individual's biological sex (Hausmann, Schoofs, Rosenthal, & Jordan, 2009; Mitchell, Baker, & Jacklin, 1989; van Anders, Steiger, & Goldey, 2015).

There has been a substantial movement in recent years towards examining sex differences in pain, with studies showing higher rates of and increased pain in women (Fillingim et al., 2009; Mogil, 2012; M. Racine et al., 2012a). However, little research has considered the contributions of gender in understanding how sex differences develop and are maintained. In adults, masculinity has been shown to predict increased pain tolerance and threshold, but is also a barrier to accessing health services, while femininity is associated with decreased tolerance and increased sensitivity to pain (Alabas et al., 2012; Keogh, 2015).

Only two studies to date have examined the impact of child gender on pain responses. Myers and colleagues (2006) reported stronger relationships between masculinity and pain in male than female 8-18 year olds. In contrast, Vierhaus and colleagues (2011) found that masculinity and femininity were not mediators between sex and pain in children and adolescents aged 10-17 years. No studies to date have examined the impact of parent's own gender on children's pain.

The present exploratory study aimed to describe the role of parent's and children's gender in pain responses. As parental adherence to gender-typed beliefs have been shown to significantly impact their children's behaviour and development of gender schemas (Fagot & Leinbach, 1989; Fagot & Hagan, 1991; Lytton & Romney, 1991; Williams et al., 1985), the present study also aimed to describe parent's gender role expectations as they relate to children's pain. It was expected that higher levels of parent's and children's self-reported masculinity would be associated with decreased pain intensity and increased pain tolerance, particularly in boys and fathers (Alabas et al., 2012; Myers et al., 2006). As femininity has not emerged as a significant predictor of pain or anxiety in children (Ginsburg & Silverman, 2000; Myers et al., 2006), it was hypothesized that femininity would not predict children's responses to experimental pain. It was hypothesized that parents would endorse similar gender role expectations of pain for their children as adults endorse for themselves, reporting that females are more sensitive to and express/report more pain, and males have higher pain tolerance (Robinson et al., 2001).

4.3 Methods

The present study was conducted using data collected as part of a larger study. The first paper (Boerner, Chambers, McGrath, LoLordo, & Uher, submitted) reports on the role of parent and child sex in understanding the impact of parental modeling of pain behaviours on their children. As the larger study involved an experimental manipulation of parental behaviour, analyses that were conducted on variables measured after the experimental manipulation were conducted controlling for experimental group. Readers are directed to the first paper by Boerner and colleagues (submitted) for more detailed description of the larger study methods.

4.3.1 Participants

One hundred and sixty-eight parent-child dyads were recruited from the community. Both parents and children had to live together at least 50% of the time, have no uncorrected hearing/vision impairments, have no conditions that contraindicated participation in the cold pressor task, and had to be able to read, write, and speak English at a level proficient enough to complete questionnaires (research assistants were available to assist children when necessary). Children were between the ages of 6 and 8 years, generally healthy and free of any chronic or recurrent pains, and could not have participated in a cold pressor task before. An equal number of sex-matched (42 father-sons; 42 mother-daughters) and sex-unmatched (42 fathers-daughters; 42 mother-sons) dyads were recruited.

4.3.2 Measures

Parent gender. Parents completed the Bem Sex Role Inventory (Bem, 1974) as a measure of self-reported endorsement of behaviours and attributes that represent stereotypically masculine and feminine behaviours. The Bem Sex Role Inventory (BSRI) provides participants with 60 items, each of which are an adjective or phrase that describes a trait or behaviour (20 items considered to be characteristically masculine, 20 items considered to be characteristically feminine, and 20 neutral items). Participants were asked to rate how true each item is of themselves on a scale of 1 (“never or almost never true”) to 7 (“always or almost always true”). Mean scores were calculated for the masculinity and femininity subscales. The BSRI has been previously used to predict pain responses (Fillingim, Edwards, & Powell, 1999; Myers, Robinson, Riley, & Sheffield, 2001; Otto & Dougher, 1985). Cronbach’s alpha for the present sample was excellent at $\alpha = .840$ for the masculinity subscale and $\alpha = .811$ for the femininity subscale.

Child gender. Children completed the Children’s Sex Role Inventory (Boldizar, 1991) as a measure of self-reported femininity and masculinity. The Children’s Sex Role Inventory (CSRI) is a children’s version of the BSRI (Boldizar, 1991). The CSRI presents children with 60 statements (20 items that are considered to be characteristically masculine, 20 items that are characteristically feminine, and 20 neutral items), which they are asked to rate on a scale of 1 (“not at all true of me”) to 4 (“very true of me”). Each of the statements corresponds to an adjective on the BSRI, though are presented in a different order. Mean scores were calculated for the masculinity and femininity subscales. The CSRI has been used in research with children as young as 6 years (Conti, Collins, & Picariello, 2001; Ginsburg & Silverman, 2000). Reliability was excellent for the present sample; Cronbach’s alpha of $\alpha = .825$ for masculinity and $\alpha = .808$ for femininity.

Gender role expectations of pain. The Gender Role Expectations of Pain Questionnaire – Parent version (GREP-P) is an adaptation of the GREP (Robinson et al., 2001) to assess parent’s perceptions of gender roles with regards to pain for their children. The GREP-P is adapted such that questions refer to the participant’s child (rather than the participant themselves) and typical “boys” and “girls” (rather than typical men and women). Four categories of the pain experience are assessed: sensitivity to pain, endurance of pain, willingness to report pain, and pain expression, the latter of which was an addition to this adapted version of the GREP. For each area, participants are asked to compare their child to the typical boy and typical girl, and compare the typical boy to the typical girl. For each question, participants provided their rating on an 11-point scale, where 0 indicated “far less” and 10 indicated “far greater”, with 5 denoting “the same.”

4.3.3 Procedure

This study was approved by the institutional research ethics board. After obtaining informed consent from parents and assent from participating children, parents and children completed questionnaire measures that included self-reports of gender and a measure of parent’s gender role expectations of pain as they relate to their children. Parents were then randomly assigned to either exaggerate their expression of pain, minimize their expression of pain, or act naturally during the cold pressor pain induction task (CPT). Children were unaware that their parent may have been asked to alter their behaviour. Then parents and children were reunited, during which time parents completed the CPT while their child watched. After observing their parent complete the CPT, children rated their own anxiety on a 10-cm visual analogue scale (VAS). The child then completed the CPT themselves, and children’s pain responses were measured by timing the children’s pain tolerance and with children’s self-report of pain intensity using the *Faces Pain Scale – Revised* (Hicks et al., 2001). Upon completion, children were fully debriefed as to the deception involved in the study, and families were compensated for their time.

4.3.4 Data analysis

All analyses were conducted using IBM SPSS Statistics version 21. If more than 10% of the data was missing from a participant's BSRI or CSRI, the questionnaire was deemed not to be meaningfully interpretable, and the entire questionnaire was considered missing data for that participant. If less than 10% of the questionnaire was missing, the available data was included in the analyses. As the BSRI and CSRI relies on mean scores rather than sum scores, a mean score was calculated using only the items that the participant responded to. Participants who had data missing for a complete questionnaire were excluded from any analyses involving that particular questionnaire, but were included in all other analyses. As the Gender Role Expectations of Pain questionnaire was analyzed on an individual-item basis, all available responses were included.

4.4 Results

Demographics

Children were 84 boys and 84 girls with a mean age of 7.14 years (SD = 0.83, range = 6 years 0 months to 8 years 11 months), who were primarily "White" (78.6%, n = 132). Parents were 84 fathers and 84 mothers, who had a mean age of 39.59 years (SD = 5.77). Parents were primarily "White" (85.1%, n = 143), married (75%, n = 126), highly educated (62.9%, n = 105 had university education or graduate/professional training), and had high income (62.6%, n = 102 had an annual household income of ≥$75,000)[1].

Parent and child self-reported gender

Parents were categorized on gender using the median-split approach described in the manual for the BSRI (Bem, 1981). There was nearly equal numbers of parents who were classified as characteristically masculine (*n* = 39, 23%), feminine (*n* = 41, 25%;), androgynous (*n* = 42, 25%; indicating high scores on both masculine and feminine scales), and undifferentiated (*n* = 44, 27%; indicating low scores on both masculine and feminine scales). Mothers (*M* = 4.99, *SD* = 0.54) reported significantly higher femininity scores than fathers (*M* = 4.68, *SD* = 0.56), $t(164) = -3.534$, $p = .001$. While fathers (*M* = 5.03, *SD* = 0.61) reported higher masculinity than mothers (*M* = 4.83, *SD* = 0.71), the difference was not statistically significant, $t(164) = 1.909$, $p = .058$.

When children were categorized by gender using the median-split approach suggested by the scale author (Boldizar, 1991), the majority of children were classified as androgynous (*n* = 61, 37%) or undifferentiated (*n* = 58, 35%). Only a small sample of children identified as being characteristically masculine (*n* = 24, 15%) or feminine (*n* = 22, 13%). There were no significant differences in the proportion of boys and girls in each gender category, $\chi^2(3, N = 165) = 7.218$, $p = .065$. Criterion group validity was established for the CSRI: there were sex differences in the expected directions for masculinity ($t(163) = 2.366$, $p = .019$, with boys (*M* = 2.98, *SD* = 0.55) scoring significantly higher than girls (*M* = 2.80, *SD* = 0.43)) and femininity ($t(142.308) = -2.278$, $p = .024$, with girls (*M* = 3.34, *SD* = 0.34) scoring significantly higher than boys (*M* = 3.19, *SD* = 0.50)). Paired-sample t-tests demonstrated that both girls ($t(82) = -12.959$, $p < .001$) and boys ($t(81) = -4.130$, $p < .001$) reported higher levels of femininity than masculinity.

Correlations between parent and child gender and cold pressor responses[2]

Correlations between participants' self-reported gender and their pain responses are presented split by sex in Table 4.7.1. Fathers who reported higher levels of masculinity tolerated the pain task for longer and reported lower pain intensity. Girls who reported higher levels of femininity were rated by their parents as having greater average pain during the cold pressor task. No significant correlations were present between measures of gender and pain responses in mothers or boys.

Gender as a predictor of child pain responses

A series of hierarchical regression analyses were conducted with scores of child gender as predictors for the children's anxiety and pain responses (Table 4.7.2). Experimental condition (one variable for each of Exaggerate and Minimize conditions, dummy-coded) and child age were entered in Step 1 of the regression analysis, and the child's masculinity and femininity scores were entered in Step 2. As previous research has suggested that masculinity and femininity may impact pain responses in men and women in different ways (Applegate et al., 2005; Otto & Dougher, 1985), the regression analyses were conducted separately for boys and girls. For children's self-reported pain intensity, the model (for Step 1 or Step 2) was not significant for children's self-reported pain intensity for either boys or girls, and is therefore not presented here.

With regards to children's self-reported anxiety after having watched their parent complete the cold pressor task, Step 1 (age and experimental condition) accounted for 13% of the variance in anxiety for boys ($R^2 = .133$, $p = .010$) and 10% of the variance in anxiety for girls ($R^2 = .098$, $p = .042$). Entering self-reported gender in Step 2 explained an additional 0.3% of the variance in boy's anxiety ($\Delta R^2 = .003$, $p = .884$) and an additional 6% of the variance in girl's anxiety ($\Delta R^2 = .057$, $p = .081$); in neither case did the addition of gender provide statistically significant variance to the model. The final model significantly predicted anxiety in boys ($F(5,76)=2.398$, $p = .045$) and girls ($F(5,76)=2.823$, $p = .021$). Participating in the Exaggerate condition was associated with increased levels of anxiety for boys (β = .274, *p* = .038), while participating in the Minimize experimental condition (β = -.275, *p* = .030) was associated with decreased anxiety in girls. Increased self-reported femininity among girls was associated with decreased anxiety after watching their parent complete the cold pressor task (β = -.285, *p* = .026).

With regards to children's pain tolerance, Step 1 (age and experimental condition) accounted for 13% of the variance in pain tolerance for boys (R^2 = .130, *p* = .012), and 8% of the variance in pain tolerance for girls (R^2 = .084, *p* = .072). Entering self-reported gender in Step 2 explained an additional 2% of the variance in boy's anxiety (ΔR^2= .018, *p* = .459) and an additional 0.02% of the variance in girl's pain tolerance (ΔR^2 = .002, *p* = .908); in neither case did the addition of gender provide statistically significant variance to the model. The final model significantly predicted pain tolerance in boys (F(5,76)=2.624, *p* = .03) but not girls (F(5,77)=1.456, *p* = .214). The only significant predictor in the model for boys was child age, indicating that boys who were older kept their hand in the water for longer (β = .337, *p* = .003).

Parent gender role expectations of pain in typical children

Visual inspection of the mean scores on the items of the GREP-P that asked about the typical differences between boys and girls indicates that parents generally believe that the typical boy and the typical girl are "the same" on most of the pain indicators (i.e., mean was approximately = 5). In cases where the mean was slightly above or below 5, parents endorsed beliefs that the typical girl is more sensitive to pain, expresses more pain, and is more willing to report pain; while the typical boy has greater endurance of pain. T-tests were conducted comparing responses between mothers and fathers (means and standard deviations provided in Table 4.7.3). When indicating sex differences in willingness to report pain, there was a significant difference between mothers and fathers when they were asked to compare the typical girl to the typical boy, $t(148.988) = -2.563$, $p = .011$, though interestingly when parents were asked to compare the typical boy to the typical girl (i.e., the opposite wording), the difference between mothers and fathers was not significant, $t(146.332) = 1.750$, $p = .081$. In both cases, the mean score of fathers indicated that they generally believed that girls and boys were the same in willingness to report pain, while the mean scores of mothers indicated that they believed that girls were slightly more willing to report pain than boys. No significant differences were observed between mother's and father's beliefs about sex differences in typical children's pain sensitivity, endurance, or expression.

Parent gender role expectations of pain in their own children

With regards to the items on the GREP-P that asked parents to compare their child to the typical boy or the typical girl, analyses of variance (ANOVAs) were conducted comparing responses based on parent and child sex (means and standard deviations provided in Table 4.7.4). When parents were asked to compare their child's pain sensitivity, endurance, expression, and willingness to report to the typical child of the same sex, there were no significant differences based on parent or child sex.

However, when comparing their child's pain sensitivity to pain and endurance of pain to the typical child of the opposite sex, significant differences emerged. There was a main effect of child sex for pain sensitivity, whereby parents reported higher scores for girls compared to boys for pain sensitivity, $F(1,164) = 5.950$, $p = .016$. This indicates that parents of daughters generally believed that their child had slightly higher sensitivity to pain compared to typical boys, and that parents of sons generally believed that their child had similar levels of sensitivity to pain compared to typical girls. There was also a significant main effect of child sex for pain endurance, whereby parents reported higher scores for boys compared to girls for pain endurance, $F(1,163) = 4.841$, $p = .029$. This indicates that parents of daughters reported that their child had about the same level of endurance of pain compared to typical boys, and that parents of sons reported that their child had greater endurance of pain compared to typical girls. There was a significant main effect of parent sex on willingness to report pain, whereby mothers reported higher scores for their children (i.e., that their children had greater expression of pain compared to the typical child of the opposite sex), compared to fathers, $F(1,164) = 5.619$, $p = .019$. No differences were observed for pain expression.

4.5 Discussion

The objective of this exploratory study was to conduct a preliminary investigation of the relationship between gender and pain responses in 6-8 year old children and their parents. As expected, gender was related to experimental pain responses differently in males and females. In parents, increased masculinity was associated with decreased pain intensity and increased pain tolerance in fathers, but not in mothers. These results mirror previous findings in the adult literature related to the differential impact of gender on men and women's pain responses (Alabas et al., 2012). Contrary to the hypotheses, child gender was not related to any of the children's own self-reported pain responses. Self-reported femininity was a significant predictor of child anxiety, but only in girls. Interestingly, the relationship between femininity and anxiety in girls was the opposite of what might have been expected: increased self-reported femininity was associated with decreased anxiety. The femininity subscale of the Children's Sex Role Inventory (and the BSRI) is comprised primarily of items that described other-oriented traits (i.e., caring about other people), as contrasted with the self-oriented traits on the masculinity scale (i.e., self-reliance and self-sufficiency). It is possible that the present finding relates to girls who were not high in femininity per se, but rather were high in empathy. As such, their concern and empathic feelings experienced while observing their parent in pain may have been more salient than their own anxious feelings.

Masculinity did not predict any of the pain responses in children. It is possible that in this age group, masculinity is a less important predictor of behaviour, or is less variable between children; while boys in the present sample reported higher masculinity than girls, they also reported higher femininity than masculinity scores. The general lack of gender-related findings may be due to the fact that children's adherence to gender-stereotyped behaviour may not be rigid as is seen in adolescent or adult populations (Boldizar, 1991; Galambos, Almeida, & Petersen, 1990). Indeed, the majority children in the present study were categorized as either androgynous or undifferentiated (i.e., not stereotypically masculine or feminine). Additionally, previous research has shown that measures of gender specifically related to pain are more highly associated with the sex differences seen in adult pain responses than general measures of masculinity and femininity (Alabas et al., 2012). As the present study only measured pain-related gender behaviour from the perspective of the parent such analyses were not possible for children; future studies would be prudent to include a pain-specific measure of child gender.

When reporting on their gender role expectations of pain in children, parents mean scores indicted that parents were generally endorsing the belief that typical boys and girls experience and express pain similarly, and only reported slight differences between boys and girls (if any). When differences were present they described perceived differences that were in line with the sex differences that are observed in the adult literature

(Fillingim et al., 2009), despite the fact that generally no sex differences are observed in these variables between boys and girls at this age (Boerner, Birnie, Caes, Schinkel, & Chambers, 2014). When asked to compare their child's responses to pain to the responses of a typical child of the same sex, parents of girls did not differ significantly in their responses compared to parents of boys. However, when comparing their child to a typical child of the opposite sex, parents participating with their daughter reported that their child would have slightly higher sensitivity and lower endurance of pain than the typical boy, with the opposite finding observed for parents participating with their son. These findings are important to consider, as they speak to the expectations that parents hold regarding their children's pain. Such beliefs are likely to influence the way that parents interact with and make decisions about their children during pain, as well as potentially influence differences between boys and girls that may occur as a result of gendered expectations (i.e., self-fulfilling prophecies). For example, a parent who adheres strongly to gender-typical beliefs may subtly reinforce or punish their child for acting or failing to act in accordance with the behaviours that would be considered acceptable for their gender.

The present research illustrates the challenges of exploring gender within a health context. Just as it is essentially impossible to tease apart influences of biological and social influences on the pain experience, it is difficult to tease apart the relative contributions of sex and gender. The methodological issues related to measuring gender, such as the outdated measures and confusion regarding their theoretical basis, further compound such complexity (Hoffman & Borders, 2001).

This exploratory study is the first in the field of pediatric pain to incorporate a measure of parent and child gender in understanding children's pain responses in children younger than 8 years old. The findings suggest that gender may play a role in understanding the experience of pediatric pain and related anxiety beyond sex alone, but that this may look different from the research on gender and pain in adults. Future studies should focus on developing a more current measure of child gender, and focuses on the specific aspects of gender that are related to health outcomes.

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4.7 Tables

Table 4.7.1.

Correlations between self-reported gender and pain responses.

	Parent pain intensity (most)	Parent pain intensity (average)	Parent pain tolerance
Fathers *(n* = 82)			
Parent masculinity	- .245*	- .121	.229*
Parent femininity	.073	- .018	- .060
Mothers (*n* = 84)			
Parent masculinity	- .095	- .070	.082
Parent femininity	- .031	- .020	.089

	Child pre-CPT anxiety	Child pain intensity (most)	Parent proxy rating of child's pain intensity (most)	Child pain intensity (average)	Parent proxy rating of child's pain intensity (average)	Child pain tolerance
Boys (*n* = 82)						
Child masculinity	- .023	- .073	.012	- .118	.136	- .116
Child femininity	- .068	- .089	.025	- .102	.122	.024
Parent masculinity [a]	.010	.019	.102	- .035	.134	.053
Parent femininity [a]	- .158	.047	.060	.005	.117	- .080
Girls (*n* = 83)						
Child masculinity	- .027	.112	.013	.144	.028	- .027
Child femininity	- .199	.128	.090	.095	.219*	- .008
Parent masculinity	- .078	- .026	- .203	.113	- .061	.132
Parent femininity	.002	- .003	.085	.020	.038	- .037

Note. The following measures were used in the present correlations: Masculinity and Femininity subscales of the CSRI for children and BSRI for parents, VAS (pre-CPT anxiety), FPS-R (pain intensity and parent proxy rating of pain intensity – most and average).

[a] $N = 83$ for the correlations between parent gender and boys' outcomes.

*$p < .05$

Table 4.7.2.

Summary of hierarchical multiple regression analyses predicting children's pre-CPT anxiety and pain tolerance during the CPT.

	Children's pre-CPT anxiety				Children's pain tolerance			
	β	Adj. R^2	ΔR^2	F	β	Adj. R^2	ΔR^2	F
Boys (n = 81)								
Step 1		.100	.133	4.005*		.096	.130	3.870*
Age	.102				.327**			
Exaggerate condition[a]	.290*				- .066			
Minimize condition[a]	-.110				- .138			
Step 2		.079	.003	2.398*		.091	.018	2.624*
Age	.095				.337**			
Exaggerate condition[a]	.274*				- .022			
Minimize condition[a]	- .128				- .083			
Masculinity[b]	.042				- .155			
Femininity[b]	- .071				.167			

	Children's pre-CPT anxiety				Children's pain tolerance			
	β	Adj. R^2	ΔR^2	F	β	Adj. R^2	ΔR^2	F
Girls (n = 82)								
Step 1		.064	.098	2.860*		.049	.084	2.418
Age	- .007				.245*			
Exaggerate condition[a]	.103				.226			
Minimize condition[a]	- .250				.055			
Step 2		.100	.057	2.823*		.027	.002	1.456
Age	- .048				.257*			
Exaggerate condition[a]	.088				.231			
Minimize condition[a]	- .275*				.059			
Masculinity[b]	.139				.000			
Femininity[b]	- .285*				.049			

[a]Experimental condition includes two variables: Exaggerate condition and Minimize condition (dummy-coded).
[b]Masculinity and Femininity was measured with the CSRI.
*p < .05; **p < .01; ***p < .001

Table 4.7.3.

Mean (SD) of parent responses to individual items on the Gender Role Expectations of Pain questionnaire – Parent Version comparing typical boys and girls, separated by parent sex.

	Sex of parent	
	Male (*n*= 84)	Female (*n*= 83)
Pain sensitivity		
Compared to the typical boy, the typical girl is…	5.38 (1.13)	5.54 (1.43)
Compared to the typical girl, the typical boy is…	4.86 (1.00)	4.58 (1.35)
Pain endurance		
Compared to the typical boy, the typical girl is…	4.87 (1.08)	4.98 (1.36)
Compared to the typical girl, the typical boy is…	5.29 (0.96)	5.42 (1.47)
Pain expression		
Compared to the typical boy, the typical girl is…	5.52 (1.10)	5.93 (1.68)
Compared to the typical girl, the typical boy is…	4.63 (1.12)	4.39 (1.51)
Willingness to report pain		
Compared to the typical boy, the typical girl is… [b]	5.63 (1.18)[a]	6.19 (1.63)
Compared to the typical girl, the typical boy is…	4.85 (1.01)	4.51 (1.45)

Note. Comparisons of the typical boy/girl to the typical girl/boy was scored by parents on an 11-point scale ranging from 0 (“far less”) to 10 (“far greater”), with the middle option (5) denoting “the same”. For all options, with the exception of pain endurance, higher scores denote greater sensitivity/response to pain.
[a]One response missing for this question (*n*=83).

[b]Significant difference between mothers and fathers at $p < .05$

Table 4.7.4.

Mean (SD) of parent responses to individual items on the Gender Role Expectations of Pain questionnaire – Parent Version comparing their child to the typical same- or opposite-sex child, separated by parent and child sex.

	Male parent		Female parent	
	Male child (*n* = 42)	Female child (*n* = 42)	Male child (*n* = 42)	Female child (*n* = 42)
Pain sensitivity				
Comparing the participating child to a typical child of the same sex	5.57 (1.81)	5.45 (1.77)	5.12 (2.46)	4.90 (1.94)
Comparing the participating child to a typical child of the opposite sex[a]	5.05 (1.90)	5.60 (1.93)	4.52 (2.54)	5.60 (2.18)
Pain endurance				
Comparing the participating child to a typical child of the same sex	5.05 (1.53)	5.38 (1.55)	5.86 (2.07)	5.40 (1.64)
Comparing the participating child to a typical child of the opposite sex[a]	5.46 (1.57)[c]	5.07 (1.76)	6.00 (2.31)	5.12 (1.76)
Pain expression				
Comparing the participating child to a typical child of the same sex	5.90 (1.78)	5.38 (1.45)	6.17 (2.23)	6.24 (2.39)
Comparing the participating child to a typical child of the opposite sex	5.29 (1.69)	5.67 (1.65)	5.60 (2.48)	6.43 (2.47)

	Male parent		Female parent	
	Male child (n = 42)	Female child (n = 42)	Male child (n = 42)	Female child (n = 42)
Willingness to report pain				
Comparing the participating child to a typical child of the same sex	6.38 (1.62)	5.62 (1.41)	6.40 (2.20)	6.40 (2.20)
Comparing the participating child to a typical child of the opposite sex[b]	5.86 (1.54)	5.74 (1.51)	6.07 (2.30)	6.88 (1.95)

Note. Comparisons of the participating child's response to pain to the typical child of the same or opposite sex were scored by parents on an 11-point scale ranging from 0 ("far less") to 10 ("far greater"), with the middle option (5) denoting "the same". For all options, with the exception of pain endurance, higher scores denote greater sensitivity/response to pain.

[a]Significant difference between boys and girls at $p < .05$

[b]Significant difference between mothers and fathers at $p < .05$

[c]One response missing for this question (n=41).

4.7 Footnotes

1 Fathers (*M* = 41.15 years, *SD* = 5.39) were significantly older than mothers (*M* = 38.03 years, *SD* = 5.73) in the present sample, $t(166) = 3.624$, $p < .001$. Additionally, mothers and fathers did differ with in marital status, *Fisher's Exact Test* = 10.342, $p = .021$.

2 Correlations should be interpreted in the context that cold pressor responses were measured in the context of the experimental manipulation, however, the correlations were repeated only with individuals assigned to the Control condition and the pattern of results remained the same for fathers, mothers, and boys. However, when correlations were conducted only for girls in the Control group (n = 28), several significant findings emerged: there was a significant correlation between parent's self-reported femininity and their daughter's self-reported most pain during the CPT (r = - .387, p = .042), a significant correlation between parent's self-reported femininity and their own proxy ratings of their daughter's most pain during the CPT (r = - .381, p = .045), a significant correlation between parent's self-reported masculinity and their own proxy ratings of their daughter's most pain during the CPT (r = - .389, p = .041), and a significant correlation between parent's self-reported masculinity and their daughter's ratings of anxiety after watching the parent CPT (r = - .564, p = .002). Additionally, the relationship between girl's self-reported femininity and their parent's proxy ratings of their average pain during the CPT was no longer significant when only girls in the Control group were examined (r = .159, p = .420).

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CHAPTER 5. IT IS NOT AS SIMPLE AS BOYS VERSUS GIRLS: THE ROLE OF SEX DIFFERENCES IN PAIN ACROSS THE LIFESPAN

The manuscript for this editorial is presented below. Readers are advised that Katelynn Boerner was the lead on conceptualizing and writing this editorial, under the supervision of Dr. Christine Chambers and with the support of co-author Meghan Schinkel. The manuscript and was accepted for publication in *Pain Management* on November 26, 2014. The full reference for this manuscript is:

Boerner, K.E., Schinkel, M., & Chambers, C.T. (2015). It is not as simple as boys versus girls: The role of sex differences in pain across the lifespan. *Pain Management*. *5*(1), 1-4. doi: 10.2217/pmt.14.42

5.1 Abstract

Understanding sex differences is currently an area of great interest in the field of pain research. However, despite the recent proliferation of sex difference research in adult pain, the research in pediatrics is significantly less well developed. The present editorial aims to review the state of the literature on sex differences in children's pain, and highlight the areas in which pediatric researchers may learn from the challenges and successes demonstrated in the adult literature. Specific suggestions for directions of future research and study methodology are provided.

5.2 Editorial

Over the last decade, sex differences have emerged as a "hot topic" in the area of pain research, after being frequently ignored in early studies of pain mechanisms and experience. As described in a recent commentary by Mogil(2012), many studies of pain in both human and animal research only used male subjects, likely a result of convention in research practices and the unfounded belief that there was more inherent variability in female subjects. As anecdotal reports and research evidence began to accumulate showing notable differences in the pain experiences and health of men and women, the field has slowly shifted to accommodate this line of investigation. This shift is clearly reflected in current policies of granting agencies (e.g., the Canadian Institutes of Health Research) requiring researchers to define how they will incorporate sex/gender into their studies and analyses, and the recent decision of the National Institutes of Health to require males and female cells and animals be equally represented in preclinical research (Clayton & Collins, 2014). Such ventures have facilitated a rapid expansion in the adult pain literature of sex difference research. However, this body of research is fraught with methodological inconsistencies and controversial findings. Despite a vast body of literature the field is not ready to provide substantial clinically relevant directions for treatment, though early studies on "personalized pain management" provide promising avenues (Niesters et al., 2010). Further, confusion regarding the use of terminology has added additional complications in presenting and interpreting results. While the terminology "sex" and "gender" are related, and certainly both relevant to observed differences between men and women and boys and girls, the two terms are not interchangeable as one refers to biological differences (sex) while the other is a psychosocial construct (gender) (Fillingim et al., 2009).

In contrast to the expansive adult literature, the research on this topic in children and adolescents is limited. However, there are clearly numerous opportunities for researchers to explore the mechanisms and implications of sex differences in pediatric pain. Pediatric researchers would be prudent to attend to the trials and errors the adult research has endured to guide research design and implementation to ensure productive and meaningful exploration of sex and gender differences in pediatric research. Reviews of the adult literature have emphasized the importance of standardized methodology for assessing and examining pain, or mediating/moderating variables, when exploring sex differences in order to ease comparisons across studies (M. Racine et al., 2012a; M. Racine, Tousignant-Laflamme, Kloda, Dion, Dupuis, & Choinière, 2012b). However, in order to gain a clear and meaningful understanding of sex and gender differences in the field of pediatric pain, it is necessary to create well-designed studies aimed at exploring this valuable topic, rather than simply relying on reviews. This would involve researchers designing their studies to reflect this aim, in contrast to the current practice of simply exploring sex differences post-hoc. In order to have sufficient power to detect sex differences, future studies will likely need to include substantially larger sample sizes than what is standard in current pediatric studies (Boerner et al., 2014; M. Racine et al., 2012a). A recent study by Schmitz and colleagues (2013) provides an excellent example of the type of large-scale study required to detect the small effect sizes associated with sex differences in pain, which is of particular importance when comparisons are being made across developmental stages. Such attention to methodological detail allowed the researchers to demonstrate that sex differences in pain tolerance during the cold pressor task emerged over the course of puberty, and were driven by a decrease of pain threshold in girls after age 14.

Another valuable direction for future research would be to design studies that specifically explore biological, social, or psychological variables that may mediate or moderate the relationship between child sex and pain outcomes, such as pain catastrophizing or anxiety (see Racine and colleagues(2012b) for a review in the adult literature). Basic science research has suggested that the interaction of sex with other factors may explain more of the variance in pain than the additive effects of sex alone (Chesler, Wilson, Lariviere, Rodriguez-Zas, & Mogil, 2002). Exploring the replicability of this finding in clinical research, as well as which factors interact with sex in the most prominent way, may provide more powerful explanations than simply comparing across groups. This approach also reflects the inherent complexity of the pain experience as an interaction between multiple levels of biological and psychosocial factors. In addition to the within-person interactions involved in the pain experience, research in pediatric populations also offers a unique opportunity to examine important social components of the pain experience, such as the parent-child relationship, and how the sex and gender of each member of the dyad influences pain learning, experience, and outcomes. As the field of pediatric pain has historically focused primarily on the role of mothers in the pain experience, it is important for future experimental and clinical research to examine how fathers also contribute to the development and socialization of their children's pain experiences and development of gender-linked pain beliefs (Phares et al., 2005).

An additional opportunity unique to pediatric populations is the ability to examine the impact of changes in physical maturation and onset of puberty in the development of sex differences in pain. Reviews of experimental and chronic pain have identified sex differences in the pain experience that emerge around the time of puberty, however, these findings were limited by the fact that age was used as a proxy for pubertal status (Boerner et al., 2014; King et al., 2011). Given the potential importance of pubertal status as a contributing factor to sex differences, and the conflicting results to date regarding whether pubertal status explains more variability beyond chronological age (LeResche et al., 2005; Schmitz et al., 2013), designing studies that specifically measure and examine pubertal status and the presence of sex hormones will be an important next step in the field.

The suggestions outlined above involve large-scale programs to explicitly examine the mechanisms and outcomes involved in sex differences in pediatric pain. For researchers simply interested in incorporating a focus on sex and gender into their existing programs of research, an important first step is clarify our language use to be specific about which sex and gender variables are being considered. A recent systematic review by our group on experimental pain in healthy children found that less than half of the studies included used appropriate terminology when referring to “sex” or “gender”, with authors frequently using “gender” when referring to sex categories, or using the terms interchangeably (Boerner et al., 2014). The term "gender” has shown a steep rise in its usage in the scientific literature since the 1970’s, with researchers showing a preference for this term over “sex” (Haig, 2004). This has resulted in inconsistent use of terminology, and potential confusion in the interpretation of results. Pediatric pain researchers need to demonstrate better awareness of the important distinctions between sex and gender in their studies and papers, and scientific publications are encouraged to have guidelines about the reporting practices of sex and gender. Ideally, research would involve a combination of sex and gender questions. It is difficult to isolate these variables or look at one without the other. Even the most stringent of laboratory settings cannot eliminate all of the psychosocial factors that could contribute to a difference between boys and girls (e.g., sex of experimenter, gender-specific learning experiences in the past). Additionally, it is likely that while society has a great influence on gender and its impact on pain, that gender also interacts with biology (Hausmann et al., 2009).

Studying sex and gender will only continue to get more complicated as society increasingly continues to recognize the flexibility and fluidity of gender as a construct that is capable of defining and directing our behaviour. Researchers should also consider how their work will be disseminated and interpreted by the public, as health research examining sex differences is a challenging and controversial field in the current sociological climate. Modern movements for gender equality and gender-blind approaches are not necessarily agreeable to scientific findings that highlight the differences between men and women, and this issue can become particularly contentious in research that involves children. Making the intentions of such research clear (e.g., to provide more person-specific care options) may ease the delivery of this research to a lay audience.

Sex and gender differences are an exciting avenue of research that promises a whole new layer to our understanding of the pain experience in children. However, it is important to remember that sex differences are not as simple as boys versus girls. There is much more to sex differences than t-tests of group differences, or the addition of sex as a covariate to primary analyses. The nuances are subtle and there are a massive number of potential mechanisms to contribute to explanations of any one finding (Mogil, 2012). Developing programs of research specifically designed to elucidate the mechanisms and implications, taking into account issues such as measurement and sample size, will be far more valuable to the field than the current practice of controlling for sex in analyses or post-hoc examinations of sex as an afterthought.

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CHAPTER 6. GENERAL DISCUSSION

The present dissertation examined the role of sex and gender in the experience of pediatric pain, specifically examining the impact on experimental pain responses and the social learning of pain behaviours. Chapter 2 described a systematic review and meta-analysis of the literature on experimental pain responses in children to examine the results and reporting practices of sex and gender across the broader literature. The systematic review examined the results of studies that conducted statistical tests of sex differences. The results of the systematic review indicated that while sex differences in pain outcomes are occasionally observed, the majority of studies found no significant differences between boys and girls on a variety of pain outcomes across a variety of experimental pain tasks. The meta-analysis combined the data from all studies from which data was available separately for boys and girls (either from the published manuscript or from the author directly), regardless of whether the study actually examined sex differences. The results of the meta-analysis indicated that, with the exception of two outcomes, there were generally no sex differences observed in children's responses to experimental pain. When examining studies that used the cold pressor task, overall girls reported greater pain intensity than boys in response to the CPT, but the difference was not significant. However, when the analysis was repeated separately by participant age, the effect remained only in the adolescent sample, and not in the sample that was predominantly children. This finding suggests that sex differences in pain may emerge around the time of puberty for cold pain intensity. In the analysis of experimental heat pain tasks, boys were found to have a significantly higher pain threshold than girls. When split by age, the effect remained significant for children and was marginally significant for adolescents.

While studies included in the review generally enrolled relatively equal numbers of boys and girls, only half of the included studies reported that they had tested for sex differences, and a small proportion (approximately 10%) indicated that they included sex as a covariate. Adding to the existing confusion in the literature, studies were mixed in the use of terminology for describing sex differences, with many using the term "gender" instead of, or interchangeably with, "sex." Despite the frequent use of "gender" in the published articles, less than 3% of included studies actually measured child gender and included this in their analyses.

Chapter 3 investigated the impact of sex on children's responses to the parental modeling of pain behaviours. This study involved manipulating parent behaviour during the cold pressor task to examine whether parent behaviour had an impact on their child's own behaviour while completing the cold pressor task themselves. Analyses examined differences based on parent and child sex. Overall, the brief parent training appeared successful in manipulating children's perceptions of parent pain, with children who observed their parent exaggerate their expression of pain reporting that their parent experienced more than children in the control condition, followed by children whose parents minimized their expression of pain. The manipulation also appeared to impact children's responses to the CPT: children who observed their parent exaggerating their expression of pain during the CPT reported greater anxiety prior to completing their own pain task than those who observed their parent minimizing their pain display.

The manipulation also appeared to have a sex-specific impact on children's self-reports of pain during the cold pressor task, with girls who observed their parent exaggerating their expression of pain reporting greater overall pain intensity during the CPT than boys assigned to the same experimental condition. There was no impact of parent and child sex or experimental condition on child pain tolerance. When asked about the presence of pain models in their lives, children reported primarily similar-aged peers (e.g., friends, siblings) as pain models, and children whose participating parent reported having chronic pain did not identify their parent as a pain model.

Chapter 4 described an exploratory analysis of gender, examining whether child pain and anxiety responses were predicted by child self-reported endorsement of masculine or feminine behaviours. Child anxiety was significantly predicted by children's self-reported femininity, but this effect was only observed in girls. Masculinity did not predict child anxiety, and gender was not predictive of children's self-reports of pain intensity or their pain tolerance. Masculinity was associated with parent pain outcomes, but only in fathers. Parents did not report strong gender-related expectations regarding children's pain, but when gender stereotypes were endorsed they generally referred to girls having increased pain sensitivity and response compared to boys.

Finally, the editorial in Chapter 5 described the state of the literature on sex and gender in pain across the lifespan, highlighting areas for future research (including more studies of gender, and a developmental focus on sex differences), and suggestions for improving the methodology of such research. The following sections will discuss the findings of each study within the context of the existing literature and the theoretical application of the present findings, discuss the clinical implications, and outline the strengths and limitations of the present dissertation, concluding with an overview of areas that may guide future research.

6.1 Integration within the existing field of research and theoretical applications

6.1.1 Sex and gender differences in pain

Sex differences in children and adolescents, when present, appear to be in line with the findings of the adult literature, with girls reporting greater sensitivity to pain than boys (Fillingim et al., 2009). The present meta-analysis provides further evidence of the lack of sex differences in childhood, and that sex differences in the prevalence and experience of pain appear to emerge around the time of puberty (Finocchi & Strada, 2014; King et al., 2011; LeResche et al., 2005; MacGregor et al., 2011; N. M. Racine et al., 2016; Schmitz et al., 2013). There are several likely explanations for this phenomenon, including the emergence of sex hormones and brain-based changes that have shown to be implicated in the pain experience (Faria et al., 2015). Additionally, there are several likely contributing social factors that emerge or are consolidated around the time of puberty (e.g., identity development and stronger adherence to gender-normed behaviour that may result in more pain-promoting behaviours in adolescent girls or underreporting of pain in adolescent boys, the onset of menstruation providing more frequent pain experiences for adolescent girls that may impact or interact with other existing pain complaints (Unruh, 1996)). Sex differences that emerge around the time of puberty is a phenomenon seen in numerous areas (e.g., mathematics, self-esteem), and is not unique to pain, however researchers continue to speculate as to the exact mechanisms involved (Hyde, 2005). As described in Chapter 5, there is clearly a need for studies specifically designed to elucidate the mechanisms involved in the development and emergence of sex differences during the early adolescent years. However, the lack of sex differences in childhood should not be taken to indicate that sufficient work has been done in this area and that researchers can ignore sex differences prior to puberty. It is critical to continue to study the experience of pain in boys and girls prior to the onset of puberty, particularly in the social context, to understand how the experiences in these early years contribute to the sex differences observed in adolescence and adulthood.

In the study described in Chapter 3, as expected, sex differences were not generally observed in the child pain responses. However, there was a sex difference reported in children's pain intensity only during the Exaggerate condition. Given the overall findings of no child sex differences across experimental conditions, and the findings of the first study that suggest that children in this age group would not demonstrate observable sex differences, this suggests that the sex difference may reflect a differential impact of parental modeling of pain behaviours on girls and boys, rather than a sex difference in the experience of the pain task itself. Previous literature has suggested that females may be more impacted by the presence of a pain model than males (P. W. Edwards et al., 1985; Fillingim et al., 2000; Świder & Babel, 2013), which was also found in the present study, at least with regards to the presence of a threatening pain model (i.e., one that suggests that the pain experience will be intense).

There were sex differences in parent's own pain experience (as reported by both the parent and the child), and these differences were in the expected directions as per previous reviews on sex differences in adult pain, with mothers reporting greater pain intensity and tolerating the CPT for less time than fathers (Fillingim et al., 2009). Additionally, the sex difference in parent average pain intensity was present in the Minimize and Exaggerate conditions, but not in the Control condition.

The exploratory analysis of gender in Chapter 4 suggested that parents often do not report believing that sex differences are present in children's pain, but when they reported slight differences to be present they tended to endorse gender-role expectations that are in line with those observed in adult pain (Robinson et al., 2001). Masculinity was associated with pain responses for fathers, while femininity was associated with anxiety in girls. This study illustrates how a complete understanding of sex differences requires a concurrent study of gender influences to begin to appreciate the complex interplay of social, biological, and psychological factors involved in creating the pain experience of males and females. The conceptualization of “sex” and “gender” as “biological” and “social” influences, respectively, over-simplifies the distinction and ignores the overlap present between these constructs(Oliffe & Greaves, 2012). Research advances suggest that the bidirectional influence of sex and gender are perhaps even more intertwined than originally thought, such as recent research demonstrating an influence of gendered behaviour on sex hormones, which demonstrates a relationship in the opposite direction of what has been historically hypothesized (van Anders et al., 2015). Additionally, there is recognition now that gender traits such as masculinity and femininity have a genetic (in addition to cultural/environmental) influence (Mitchell et al., 1989).

In real life, sex is not the binary distinction that it is often referred to in the scientific literature, as not all individuals fall into male (XY) and female (XX) categories, and many of the differences observed between men and women exhibit greater differences within the sexes than between (Hyde, 2005). Researchers use sex categories for the ease of comparison, which mirrors the social phenomenon that individuals engage in every day: applying taxonomies for ease of using heuristics to understand and navigate the world, though assuming that the presence or absence of male and female sex organs provides the differentiation between the two categories ignores a host of other biological factors (e.g., hormones, metabolism, body chemistry, brain structure) that generally differ between the sexes, but in a more dimensional fashion (Oliffe & Greaves, 2012). There are multitudes of factors involved in the pain experience and therefore it is easier to conduct research looking at the broad categories that capture, generally, the majority of these differences by studying the typical male compared to the typical female. However, now that research has established these basic differences, there is a need for the field to shift towards understanding the mechanisms that drive these differences, both biologically and socially.

6.1.2 Social modeling as a mechanism for learning pain behaviours

Pain behaviours, i.e., the physical, verbal, and emotional responses that an individual has in response to a pain experience, influence how individuals cope with pain and communicate pain to others, and can have a significant impact on functioning. There are numerous ways that children learn how to behave during painful experiences; some pain behaviours are innate, some are learned through direct experience, and others are learned by observing the behaviour and consequences of other's pain. While a large body of experimental research exists on social modeling as a learning process for children, very little research has examined this process specifically as it relates to the experience of pain. There are numerous factors implicated with the pain experience, such as the associated physiological and emotional responses to pain, that may influence the social modeling experience differently than the experiences measured in the general social modeling literature.

This dissertation presents an exploratory analysis of how pain behaviours (specifically facial expressions) are learned via social modeling from parent to child. The primary analysis of the present study focuses on the direct impact of the parent's pain behaviour on the child's own experience of pain and anxiety, which has an impact on the modeling process. While modeling and reinforcement are often conceptualized in research as being separate processes, Bandura(1977) described reinforcement as being facilitative to the modeling process. In the case of the present study, the extent to which children choose to continue to model the behaviours they observed in their parent and integrate these into their behavioural repertoire is likely largely influenced by the consequences of doing so (i.e., how the behaviours impacted their own direct experience). The findings of the primary analyses will be discussed in the following sections with respect to their relevance to the social learning process.

The present study found that observing a parent exaggerating their expression of pain was associated in children's increased self-reported ratings of anxiety, compared to children who had observed their parent minimize an expression of pain. It cannot be determined from the present data exactly what about the situation increased the children's anxiety. The most obvious explanation would be that children were attending to a stimuli that was highly relevant to them (i.e., parental behaviour during an unfamiliar task that the child is about to complete), and that observing the parent expressing an intense expression of pain increased the child's anxiety about their imminent pain experience. Previous research has found that observing pictures of children expressing high facial expressions of pain was associated with increased anxiety in children about to complete the CPT, compared to children who were shown pictures of children expressing low facial expressions of pain, and that this appeared to have a greater impact on children's anxiety than high- or low-threat verbal instructions about the task (Boerner et al., E-pub ahead of print). The experience of observing an intense reaction to the CPT may have increased the threat value of the task that the child was about to experience, and therefore provoked an anxious response. Conversely, the opposite may have occurred in the case of the Minimize condition, where observing a parent display a relatively minimal or nonexistent reaction to the painful stimulus may have reduced the child's anxiety and increased their confidence in their ability to tolerate the task.

However, it is also possible that children reported increased anxiety due to the fact that their parent was acting in an unusual manner, as evidenced by children in the Exaggerate condition reporting that their parents were expressing more pain than usual. Rather than feeling anxious about the upcoming experience of pain itself, perhaps children's anxiety was driven by confusion as to why their parent was behaving in an unusual manner, or why the parent's exaggerated expression of pain did not match what the child had expected to be only a mildly painful experience. Regardless, it is notable that children's anxiety was significantly impacted by the brief manipulation in parent behaviour, suggesting that children experience an emotional response in relation to their parent's behaviour during a novel painful event that they are about to experience. It should be noted, however, that children's anxiety might have also been increased in the present situation as they were anticipating having to complete the cold pressor task themselves, and therefore may have been attending more to their parent's behaviour as it had a direct relevance to their immediate needs. As such, the results of the present study may offer an explanation as to what may occur in situations where children are observing their parent experiencing a pain that they themselves are about to experience (e.g., when families get their influenza vaccinations together), but perhaps not when the child witnesses their parent experiencing pain that is not imminent for themselves (e.g., when a parent has a chronic/recurrent pain problem).

Parent sex did not have a significant impact on child pain responses, despite the finding that there were differences between mothers and fathers in their own pain experience. As has been suggested in previous literature (Bussey & Perry, 1982; Perry &

Bussey, 1979), it is possible that a lack of effects based on parent sex in the present study was due to the fact that children were only presented with one sex of model. In daily life, children are presented with numerous models of both sexes, and thus have many more examples of behaviour to draw upon. As such, it is possible, for example, that boys in the present study attended to the behaviour of their mothers because she was the only parent present, but had they observed both their mother and father they may have been more inclined to select the behaviour of the same-sex model as the attend to. Future studies should employ an experimental paradigm that allows investigation of this issue.

However, if mothers and fathers were equally salient models to children because they were the only models presented to them at the time, then differences would have been expected based on parent sex because mothers and fathers were modeling different pain tolerances. Additionally, children were aware of the difference in pain intensity between mothers and fathers, even though the parent's own pain ratings were made privately, further confirming the fact that mothers and fathers were modeling different pain experiences. It is possible that differences in parent pain behaviour was enough for children to become aware of and increase their anxiety, but that this brief manipulation in facial expression alone was not enough to alter the children's true responses to the pain experience. An additional possibility is that the manipulation of behaviour (i.e., Exaggerate vs. Minimize vs. Control) was more salient to children than any parental sex differences. It is likely that repeated exposure to male and female pain models over time influences children's development of gender-appropriate pain behaviours that interact with the hormonal changes that occur during puberty to create the sex differences that are observed in adults, but the present experimental design was too brief and in children too young to capture the complexity and long-term nature of this hypothesis.

Additionally, it is possible that the assumption that children will model a same-sex parent is too simplistic, and a more detailed understanding of gender role orientation and the reciprocal nature of such interactions needs to be considered. Previous research has suggested that “tomboys” (i.e., girls with higher masculine traits) tend to model their fathers more than mothers, but also tend to have less traditionally feminine mothers (Williams et al., 1985). The early social modeling literature suggested that how “gendered” a behaviour was considered to be had an impact on how likely an individual was to model the behaviour of a same-sex individual (Bandura et al., 1961). Future research should take into consideration both the gender of the parent and child as well as their perceptions of how gender-typed particular pain behaviours are. This may help determine whether boys are more likely to attend to their parent’s behaviour if they display more masculine characteristics and the pain behaviour is considered to be more typical of a male, and vice versa for girls. Such research should also take into account the availability and characteristics of other models to understand how children choose which individual’s behaviour to model.

As expected, girls appeared to be more impacted by the Exaggerate condition than boys. One possibility is that girls are more attuned to the behaviour of others and attend more to social cues in their environment (Bayliss et al., 2005). An additional explanation is that the presence of a model displaying an overt expression of pain gave girls the “permission” to rate the task as being more painful. It is possible that even at this young age, the boys were already aware that it is considered socially unacceptable for males to express pain, and therefore boys may have found it uncomfortable to observe a pain model that was exaggerating their expression and chose not to attend to this behaviour (Perry & Bussey, 1979; Raskin & Israel, 1981). It is interesting to note that this difference was only observed for ratings of average, and not worst/most pain. Perhaps it is seen as being more socially acceptable for boys to express the intensity of pain at its worst, to demonstrate the extent to which they can tolerate intense pain, but less socially acceptable to express more longer-lasting “average” pain. Another possibility is that the effect of observing the parent exaggerate an expression caused an initial increase in the pain experience (worst pain), but that this effect dissipated more quickly for boys and did not impact their overall pain ratings as significantly.

The findings above generally describe differences between the Exaggerate condition compared to the Control and Minimize conditions. One potential explanation for the lack of differences between the Control and Minimize group is that high-functioning generally pain-free parents may tend to minimize their pain behaviour in front of their children anyway, so perhaps the Minimize condition was very similar to the Control condition. This hypothesis is supported by the children’s ratings of typicality of their parent’s behaviour during the pain task, where the Minimize and Control conditions were not rated as significantly different.

The present study provides a brief examination of the modeling process, and we can infer from this that the impact would be much greater if children experienced repeated exposures over time (i.e., in the context of chronic pain). This research builds on the results of Goodman and McGrath (2003) by examining the impact of parent and child sex on the modeling experience. The impact of sex and gender likely shifts over the developmental stages, which potentially explains why more sex-related effects were not observed in the present sample. It is possible that in this age group, modeling a gender-typed behaviour is less relevant to children as they are less inclined to adhere to rigid gender roles which appear to be more important in adolescence (Galambos et al., 1990).

There are a number of methodological differences from the original study by Goodman and McGrath (2003), including the age of the children in the present study being younger (6-8 years) than children in the original study (10-14 years). The choice of recruiting younger children in the present study was due to the hypothesis that younger children would look more to their parents in unfamiliar situations than older children. However, the results of the data children provided on the pain models in their lives provide preliminary evidence that same-age models may be more salient to younger children than parents. Similar to the literature on the development of masculine and feminine characteristics, this research suggests that it is not simply parental modeling that influences this process, but rather numerous other role models including other adults, siblings, peers, and the media that may all play a role to differing degrees in influencing the child's development and learning (McHale, Updegraff, Helms-Erikson, & Crouter, 2001; Schunk, 1987; Williams et al., 1985). It is possible that parents may tend to hide or minimize their experiences of pain from their children to protect them, therefore the pain exhibited by siblings and peers, which is likely more uninhibited, may be more salient to children. Alternatively, it is possible that children were less likely to report their parent as a pain model because the parent was participating in the study with them, or because they were not directly asked if their parent had chronic pain (as has been the case in many of the retrospective self-report studies). Regardless, the frequency at which same-age peers and siblings were reported as models is interesting and warrants further investigation.

The exploratory analysis of facial expression data found that the intensity of children's facial expressions was significantly different based on experimental condition, but not with regards to the same facial action units as the parents. The present findings suggest that children's facial expressions of pain are influenced by their parent's facial expressions of pain, but that it may not be a direct mimicing of facial actions that occurs.

6.2 Clinical implications

The present research provides increased support for the hypothesis that sex differences tend to emerge around the time of puberty. Knowing that girls are at higher risk than boys of developing increased pain sensitivity around the time of puberty can assist in the development of prevention efforts. A greater understanding of the processes and individual difference factors involved in the social learning experience may have treatment implications for chronic pain. Recent research has focused on developing and testing a form of cognitive-behavioural therapy that is based on Social Learning Theory, and targets the behaviours of parents that may be involved in maintaining or exacerbating their children's pain (Levy et al., 2010; Levy et al., 2014). The present study provides a preliminary investigation into how sex might be implicated in the social learning process, which may encourage future research examining how interventions based on the social learning theory may be differentially effective based on parent and child sex and gender.

The findings of the present research related to the impact of parental pain behaviours on children's pre-CPT anxiety have implications for understanding and potentially preventing the development of procedure-related fear. Procedural anxiety, particularly needle fear, has been shown to often originate from a negative modeling experience (Willemsen et al., 2002) and is associated with increased distress and pain during a procedure (Taddio et al., 2012). Therefore, the present dissertation has implications for the management of procedural pain and anxiety that may occur in a context that allows for modeling (e.g., family visits to their general practitioner for influenza vaccinations) as an intervention.

6.3 Strengths and Limitations

There are numerous strengths and limitations for each study that have been outlined within the manuscripts in Chapters 2, 3, and 4. The following will discuss in greater detail some of the broader issues with each of the studies in this dissertation.

6.3.1 Summary methods: The use of systematic review and meta-analysis

The use of systematic review and meta-analytic methodology in Chapter 2 has many of the strengths and weaknesses associated with these methods in other areas of research. We had a high response rate from authors with data that made it possible to conduct a meta-analysis, and low heterogeneity was observed in the meta-analysis. Care was taken to control for methodological variability as much as possible in choosing what data to extract and how to combine findings across studies, as well as in accounting for overlapping samples so that data from the same participants was not considered more than once. The use of summary methods allowed for a large sample size that made it possible to detect smaller effect sizes, as well as allowing for conclusions to be drawn across different experimental pain tasks in different laboratory settings with different protocols, participant samples, etc. However, the variability between studies was also a potential limitation, as was the lack of data from non-Western samples, as previous research has shown that both facial expressions of emotions (presumably including pain) and gender-stereotyped beliefs about pain expression differ across cultures (Hobara, 2005; Jack, Garrod, Yu, Caldara, & Schnys, 2012). Finally, due to the limitations of the data available, we were unable to compare sex differences in children and adolescents beyond splitting the data based on the mean age of the participant sample.

6.3.2 Self-report measures: Questionnaires and pain ratings

Several analyses in the second study relied on the use of children's self-reports of thoughts, feelings, and behaviours in the form of questionnaires. These questionnaires were administered to provide a richer understanding to the behaviours observed and measured in the study, and to help contextualize the findings. In particular, the use of a self-report questionnaire of gender allowed for a broader understanding of what may drive the sex differences that were observed by examining differences between boys and girls along a continuum, rather than merely as a binary outcome. While the questionnaires in the present study were selected for being the best available measure for a particular outcome, and the most appropriate for the age range of children in the study, there are several limitations to their use.

Previous research has questioned the validity of using questionnaires that use Likert response formats with children, as it requires an ability to think abstractly that most children have not developed (Mellor & Moore, 2014). However, the use of word-based (rather than number-based) response formats in the present study makes this slightly less problematic. Anecdotally, many of the children tended to respond to the questionnaires in a dichotomous way (i.e., only ever selecting the lowest or highest possible response option in the questionnaire and ignoring the middle options), which is in concordance with previous reports of children's use of Likert-type rating scales (Chambers & Johnston, 2002). In addition to the issues with scale use, there were also some issues related to language, as many children appeared to be confused by the pain models questions and often referred to recent accidents/injuries rather than chronic pain, and often also referred to emotional hurts rather than physical pain.

The self-report child measure of gender was validated on samples of children older than those in the present study (Boldizar, 1991), though it has been used before in children as young as 6 years (Conti et al., 2001; Ginsburg & Silverman, 2000). Previous research has asked parents to provide proxy reports for measures such as gender on behalf their children (Moon, 2010). However, it was deemed important for the present study to elicit the perspectives of the children themselves, and measures of validity (criterion group validity) and reliability (Cronbach's alpha) suggest that the use of these scales were appropriate in the present sample. As outlined in Chapter 1, there are still concerns related to the content validity of the measures of gender. In reviewing the items involved in each of the scales for the BSRI and CSRI, it is possible that the measures of femininity and masculinity may not be specific to these gender constructs per se, and rather may reflect constructs such as self- (masculinity) and other-oriented (femininity) traits, independence (masculinity) and empathy (femininity), assertiveness (masculinity) and passiveness (femininity), etc. Additionally, as the scales were created several decades ago, the relevance of these socially constructed gender norms may have shifted since then, and there are numerous critiques about the construction of these scales and the theories of gender they are based upon (Colley et al., 2009; Hoffman & Borders, 2001).

Inspection of Table 3.7.4 indicates that there may have been some issues with the children's comprehension of the pain intensity ratings, as one would expect that ratings of "most pain" would be at least equal to or greater than "average pain", but that this is not the case in all experimental groups. Parametric tests were used for continuous variables and pain intensity, as the Faces Pain Scale – Revised was designed to be an interval measurement (Hicks et al., 2001), though it has been suggested that younger children may not use the scale in this way and there is current debate about whether pain measures should be analyzed using parametric or nonparametric tests (von Baeyer, 2009).

Finally, while the present study aimed to measure and control for, if necessary, demographic variables that may have impacted the outcomes, to reduce participant burden there were some variables that may have impacted parent's engagement in the experimental manipulation or children's reaction to parental behaviour that were not measured in the present study (e.g., parental and child mental health status).

6.3.3 Computerized facial coding

The exploratory analyses of facial expression data in the present study are based on computerized coding of the parent and children's facial expressions in response to the painful experience. Initially, the data analytic plan for the present dissertation involved traditional FACS coding done by human coders. Two human coders were recruited to complete the FACS coding, both of whom had extensive prior experience with coding facial expressions of pain using FACS, were certified FACS coders (i.e., had passed the certification test), and had experience reliably coding with each other before. The first coder coded approximately 50% of the data, and 20% was reliability-coded by the second coder. Unfortunately, reliability between the two coders was lower than what would be considered acceptable for FACS coding. Despite discussions between the coders and dissertation author in an attempt to resolve systematic discrepancies, the coders were unable to achieve sufficient reliability. As such, the decision was made to instead use the computerized coding approach (Noldus FaceReader) for the present dissertation.

The FaceReader 6 Action Unit Module has been validated (Lewinski et al., 2014) by comparing the performance of the computer software to certified FACS coders on a dataset of over 200 images of facial expressions(van der Shalk, Hawk, Fischer, & Doosje, 2011). The agreement on individual action units is varied, with the majority of the AUs performing very well or "reasonably well" (den Uyl, 2015). Of note, validation was conducted with a dataset of young adults (ages 18-25), and therefore the validity of the computerized facial coding on children is unknown. However, the significant correlations of the facial coding data in the present study with children's self-reported pain intensity and pain tolerance provides preliminary support for the validity of this method.

As is often the case in facial coding research, there was a substantial amount of missing data. This generally was the result of the computer software failing to detect a face in the video frame, or failing to code a face in the frame due to poor image quality, which was usually due to the individual tilting their head down/to the side, or the face being obscured by hair, etc. This issue is not unique to computerized facial coding, and is influenced by a number of variables that are difficult to control, such as position of the camera and the extent to which the participant moves around during the task (an issue that is particularly salient when attempting to code the expressions of young children). The study described in Chapter 3 used security cameras placed in the upper corners of the testing room to capture facial expressions, meaning that the downward angle of the camera often made it difficult to see the bottom of the participant's face. As such, there is a potential bias introduced in that participants may tend to pair certain head movements (e.g., head down) with certain facial movements (e.g., increased expression of pain). While it would have been preferable, for facial coding purposes, to have the camera positioned directly in front of the participant's face, this was not done in the present study for both logistical reasons (it would be impossible to have a camera positioned directly in front of the participant completing the CPT without obscuring the view of the parent or child observing the CPT) and scientific reasons (e.g., having a camera directly in front of one's face may reduce the external validity of the experiment, and influence the facial expressions a participant may choose to portray). A research assistant who oversaw the filming of the videos from a separate room was able to unobtrusively manipulate the cameras to attempt to continue to capture the facial expression even if the participant moved their head or body. However, despite these efforts there was still a significant proportion of facial coding data that was missing, as described in Chapter 3. While this is a problem in any facial coding research, it should be acknowledged that the results of the present study are only a preliminary investigation and should be replicated as the technology and ability to conduct more sophisticated and reliable facial coding analysis advances. Computerized facial coding offers an exciting area for development and future research, and there are specific computerized programs currently being developed and validated for the use in pediatric pain populations (Sikka et al., 2015). That would reduce the time and logistical difficulties associated with human coders, as well as potentially reducing the bias introduced by human coders, who are not immune to implicit biases e.g., based on sex or gender (L. L. Cohen et al., 2014; Hall & Matsumoto, 2004; Schäfer, Prkachin, Kaseweter, & Williams, 2016).

6.4 Areas for future research

The findings of the present studies suggest numerous exciting opportunities for future research. While the present study solely examined sex differences in experimental pain in children, there is clearly a need to extend these investigations to clinical samples. Epidemiological studies have highlighted that sex differences tend to emerge around the time of puberty in rates of pediatric chronic pain (King et al., 2011), however longitudinal multi-method studies are needed to determine what processes differentially change for boys and girls around this time, with measures of sex hormones and closer monitoring of pubertal status being of clear importance. Current work is examining the impact of sex on treatment outcomes in clinical trials of psychological therapies for pediatric chronic pain (Boerner, Eccleston, Chambers, & Keogh, in preparation), and there is a need for studies that examine whether mechanisms of treatment differ between boys and girls, particularly as it relates to treatments with a basis in Social Learning Theory (Levy et al., 2010; Levy et al., 2014). Overall, within the field of sex differences in pediatric pain there is simply a need to increase the sample size of any studies that aim to investigate sex differences, as effect sizes are typically small (Fillingim et al., 2009). As such, there is a need for continued meta-analyses, to examine trends across larger, combined samples. Since the review described in Chapter 2 was conducted, a validated algorithm for conducting sex and gender searches in PubMed has been published, which may be of use to individuals conducting future reviews of sex and gender influences (Song, Simonsen, Wilson, & Jenkins, 2016). For example, while several studies have reported potential differences in boys' and girls' responses to and experience of painful procedures (Carr, Lemanek, & Armstrong, 1998; Fowler-Kerry & Lander, 1991; Goodenough et al., 1999), this literature has not been systematically reviewed, and there have been only limited investigations of sex differences in post-surgical pain or analgesia use (Logan & Rose, 2004).

There is a clear need for an increased appreciation of the differences between sex and gender, and how they interact to influence the experience of pain in men and women, boys and girls. Pelletier and colleagues (2015) recently developed a derived gender index specific to health that offers a more current and comprehensive alternative to the out-dated measures of gender that are currently used. Unfortunately, there are a number of variables included in this composite that are not relevant to children, such as income or primary earner status, therefore developing a comparable measure for children and adolescents would be a worthwhile venture for future research. There are numerous areas of sex difference research that would benefit from the addition of a gender perspective. For example, as the present meta-analysis and other studies have suggested, the impact of sex differs depending on the pain stimulus administered (Fillingim et al., 2009; Lautenbacher & Rollman, 1993). As the relative contribution of genetic and environmental influences also differs between types of pain stimuli, it would be interesting to also consider these differences from a framework that incorporates both sex and gender (Nielsen et al., 2008).

Following up on the sex-related findings of the present study, future research may experimentally manipulate the masculinity or femininity of the model's behaviour. While the present study gave instructions to parents to modify their facial expression of pain with no instructions with regards to verbal utterances, future studies may choose to instruct models in demonstrating adaptive or maladaptive coping behaviours, as well as positive or negative outcomes of the behaviour to observe the impact of this learning process on the observer. Additionally, employing more sophisticated measures such as eye-tracking may allow researchers to examine exactly what children are attending to in the presence of a pain model (e.g., facial expressions vs. body postures of pain), and which models children are attending to when presenting with more than one option of pain model (e.g., do they attend to same-sex models, or are there other characteristics that make a model more salient?). Finally, given the relationship between modeled behaviour and anxiety observed in the present study, future studies would be beneficial to further explore this relationship, particularly given the role of fear-induced hyperalgesia in increasing sensitivity to pain (Rhudy & Meagher, 2000). For example, future studies may examine whether children who are higher in trait anxiety pay more attention to models or are more likely to look to others for information in ambiguous pain-related situations.

6.5 Conclusions

Some sex differences in pediatric experimental pain responses are present, with some becoming apparent in adolescence, in same direction as seen in adults. Social modeling provides one avenue by which children may learn the pain behaviours that differentiate men and women. The present research has implications for our understanding of how sex impacts the social learning process. Additionally, gender was shown to provide an additional explanation for some of the social modeling outcomes, and parental expectations of gender roles related to pain may be involved in shaping their children's perceptions of gender-appropriate behaviours. More research needed to understand complex interplay of sex and gender in the pain experience, from both the perspective of the parent and child, as well as the developmental context in which sex differences begin to emerge. Parent and child sex and gender are important in understanding children's pain responses, and the way parents influence those experiences.

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APPENDIX B: CORRIGENDUM FOR CHAPTER 2

Corrigendum to "Sex differences in experimental pain among healthy children: A systematic review and meta-analysis" [PAIN® 155(5) (2014) 983-993]

The authors would like to draw your attention to two errors in the published article: Marginal means and standard errors, rather than means and standard deviations, had been extracted from Lu et al. (2005) and included in the meta-analysis of heat pain tolerance. Once corrected to include the means and standard deviations, the difference between boys and girls was no longer significant: SMD of -0.36 [-0.95, 0.23], $Z = 1.18$, $p = .24$. Further, in the systematic review portion of the article, the sex difference finding for pain tolerance in Zeltzer et al. (1989) was incorrectly reported. The correct categorization of this finding was "Girls < Boys" with regards to pain tolerance.

APPENDIX C: PARENT TRAINING SCRIPTS

Script for parent training 'Exaggerate' condition

"In a few minutes, you will be asked to do the cold pressor task. We would like you to exaggerate your facial expressions while doing the cold pressor task. This means you are being asked to show more pain on your face than you normally would. Please try to exaggerate the pain on your face while still maintaining a believable expression, that is not so extreme that *(child's name)* will be concerned about your level of distress or discomfort. Here are some examples of adults doing the cold pressor task who were given the same instructions as you, to exaggerate their expression of pain. Just so you know, it's alright to talk to your child while you are doing the task; the adults in this video did not have their child in the room with them, so they did not have anyone to talk to during the task." (*Show exaggerate condition video)*

"It's important that your child doesn't know that you may be changing the way you behave during the task."

"Do you have any questions?"

Script for parent training 'Minimize' condition

"In a few minutes, you will be asked to do the cold pressor task. We would like you to minimize your facial expressions while doing the cold pressor task. This means you are being asked to show less pain on your face than you normally would. Here are some examples of adults doing the cold pressor task who were given the same instructions as you, to minimize their expression of pain. Just so you know it's alright to talk to your child while you are doing the task; the adults in this video did not have their child in the room with them, so they did not have anyone to talk to during the task." (*Show minimize condition video)*

"It's important that your child doesn't know that you may be changing the way you behave during the task."

"Do you have any questions?"

Script for parent training 'Control' condition

"In a few minutes, you will be asked to do the cold pressor task. We would like you to act naturally and show your child how it really feels. We are **not** asking you to change your behaviour from what you would normally do in this situation. Just so you know, it's alright to talk to your child while you are doing the task. First I will show you a brief video about the cold pressor task and how it's cleaned after we use it." (*Show control condition video)*

"Do you have any questions?"