The interplay of parent and child coping responses in understanding child functioning in the context of living with a parent with or without chronic pain

Elke Van Lierde[a,b*], MSc, Liesbet Goubert[a], PhD, Tim Lammens[c,d], PhD, Lamia Ben Brahim[d], Eva Van den Bussche[e], PhD, Tine Vervoort[a], PhD

a Department of Experimental-Clinical and Health Psychology, Ghent University, Ghent, Belgium

b Department of Experimental and applied psychology, Vrije Universiteit Brussel, Brussels, Belgium

c Department of Pediatric Hematology-Oncology and Stem Cell Transplantation, Ghent University Hospital, Ghent, Belgium

d Vzw FibroRes, Ghent, Belgium

e Brain & Cognition, KU Leuven, Leuven, Belgium

*Corresponding author: Elke Van Lierde, Department of Experimental-Clinical and Health Psychology, Ghent University, Henri Dunantlaan 2, B-9000 Gent, Belgium. Tel: +32(0)2-629 14 26, elke.vanlierde@ugent.be.

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Abstract (250 words)

Objectives: Pain problems tend to run in families and children of individuals with chronic pain (ICPs) have been found to report lower functioning. Drawing upon a social learning perspective, the current study examined how diverse maternal pain coping responses (i.e., pain catastrophizing and distraction) may, via corresponding child pain coping responses, act as a vulnerability or protective factor for child functioning in the context of parental chronic pain (CP).

Methods: A cross-sectional study was conducted in mothers with CP and their pain-free child (N = 100) and mothers without CP and their pain-free child (N = 74). Moderated mediation analyses were performed to test whether associations between maternal coping responses and child functioning (i.e., somatic symptoms, physical functioning and psychosocial health) were mediated by corresponding child coping responses and whether these associations were moderated by the presence or absence of maternal CP.

Results: Maternal pain catastrophizing was indirectly related to more somatic symptoms, lower physical functioning and lower psychosocial health in their child via child pain catastrophizing. Relationships were moderated by the presence or absence of maternal CP, such that mediated relationships were only found in mothers without CP and their child. No (in)direct relationships between maternal distraction, child distraction and child functioning were observed.

Discussion: The current findings demonstrated that child functioning was associated with maternal and child pain catastrophizing, but only in children of mothers without CP. No evidence was found in support of maternal pain coping responses as vulnerability or protective factors in the context of parental CP.

Key words

Parental chronic pain, coping, social learning theory, children

Introduction

Children of individuals with chronic pain (ICPs) are at increased risk of experiencing pain and other negative outcomes, including illness, internalizing and externalizing symptoms and pain catastrophizing [1–5]. According to Social Learning Theory (SLT), pain-related behaviours and beliefs can be learned directly by receiving parental reinforcing responses and indirectly through observing parental behaviour [6–8]. Since more observable pain behaviours are present in ICPs compared to parents without chronic pain (CP) [9], observational learning is considered particularly relevant in the intergenerational transmission of chronic pain. Research that aims to understand child functioning in a context of parental CP is still in its infancy, with most studies focusing on children who already developed CP themselves. Indeed, studies in children with CP have shown that child functioning and pain outcomes are related to parental CP status (i.e., absence or presence of CP) and parental pain behaviour [9–12]. Recently Stone and Wilson [1] developed an integrative conceptual model to help understand and study why and how parental CP heightens a child's risk for adverse outcomes and, potentially, CP. Crucially, the model states that child vulnerabilities (e.g., altered pain processing, pain-related cognitions and affect, pain coping behaviours) may be affected by having a parent with CP. These child vulnerabilities are known to be related to and affected by children's own experience of pain [13]. However, it remains largely unexplored whether child vulnerabilities are, as proposed by the conceptual model, more pronounced in pain-free children of ICPs compared to pain-free children of parents without CP. Importantly, a heightened presence of child vulnerability factors may predispose currently pain-free children to develop CP or other adverse outcomes later in life. To our knowledge, only one previous study examined child vulnerability factors (i.e., pain-related cognitive biases) in pain-free children of parents with and without CP, but failed to find support for hypothesized differences between both groups of children [14]. We aim to extend these first results by examining another proposed critical child vulnerability factor (i.e., child pain coping behaviours) in pain-free children of parents with and without CP.

How children and parents cope with pain might be particularly important, as both child and parental coping with personal pain have been found to shape child functioning. In accordance with previous studies investigating parental responses to child pain (e.g., [15–19]), we focused on pain catastrophizing and distraction. Child pain catastrophizing, a coping style characterized by excessive negative beliefs about pain [20], is associated with poorer adjustment, while children who distract themselves from pain usually report better outcomes [21–25]. Importantly, parental pain catastrophizing was also found to be associated with negative child outcomes [26,27]. Conversely, there is currently no evidence on the potential advantages of parental ability to distract from pain for child functioning. Based on SLT, it is likely that parental coping responses, child coping responses and child outcomes are interrelated and that parental coping responses contribute to child functioning indirectly via child coping responses. Specifically, children who observe parental pain coping responses and associated behaviours (e.g., parents who catastrophize about pain also tend to express more pain [28,29]) may engage in similar coping strategies because of observational learning processes [6,7]. In turn, child coping affects child functioning. An accordance in parental and child pain coping strategies may arise especially in children of ICPs, as they observe more parental pain behaviours [9].

To date, research examining this indirect relationship between parental coping responses and child functioning through child coping responses in ICPs and their children is lacking. Moreover, evidence on the relationship between parental and child coping responses is limited and remains inconclusive. Indeed, while some studies in parents without CP and their children found that parental and child pain catastrophizing were related [26,27], another study in ICPs and their children failed to find substantial associations in coping responses [30]. In addition, research has focused predominantly on pain catastrophizing, neglecting more adaptive pain coping responses such as distraction [22–24].

The aim of the current study was to examine how diverse parental pain coping responses (i.e., pain catastrophizing and distraction) may, through their association with corresponding child pain coping responses, either act as a vulnerability or protective factor in a context of parental CP. Relationships were examined in a sample of ICPs together with their pain-free child and parents without CP together with their pain-free child, allowing us to explore whether the expected associations were particularly pronounced amongst children of ICPs. Accordingly, we evaluated a

moderated-mediation model (Figure 1) whereby we expected that higher levels of parental pain catastrophizing would be associated with poorer child functioning (i.e., more somatic complaints, lower physical functioning and lower psychosocial health) and that higher levels of parental distraction would be associated with better child functioning. We expected these relationships to be mediated by corresponding child coping responses. Finally, based on recent theoretical accounts[1], a moderation by parental pain status was included in the model. It was explored whether the (in)direct relationships between parental coping responses and child functioning are strongest in ICPs and their child compared to parents without CP and their child.

- Insert Figure 1 about here -

Method

Participants and procedure

To obtain data of parents with and without CP, together with their pain-free child (only one child participated in families with more than one child), we recruited participants through schools and the Flemish League for Fibromyalgia Patients (FLFP). The FLFP is a support group for patients whose main goal is to bring patients in contact with each other, to organize meetings in which patients can get in touch with other people who have similar experiences and to provide information to patients (e.g., about diagnosis, treatment options, returning to work). The period of recruitment and testing of participants through schools was January 2014-April 2014 and the period of recruitment and testing of FLFP-members was March 2015-December 2015. This led to two participant pools (see Figure 2).

Twenty-eight schools (grades 4 to 12) were first contacted by letter and subsequently by telephone. Sixteen schools agreed to participate. Reasons for non-participation were already participating in other studies or the lack of fit of the study in their planned school activities. In these schools, a total of 4978 schoolchildren and their parents received an invitation letter. In this letter the purpose of the study was explained (i.e., it was mentioned that the goal was to study how children and parents experience pain and how they cope with it), potential participants were told that it would take maximum one hour to complete the survey and that children who participated together with at least one parent would be included in a lottery and could win a prize (i.e., movie ticket, IPod or an IPad mini). Moreover, a link to the online survey, which participants were requested to complete at home, and three personal codes (one for the child, one for the mother and one for the father) were provided. At the start of the survey, participants received information about the study and were asked to provide written informed consent if they agreed to participate. For 536 families, at least one member of the family (child, mother or father) opened the link to the online survey and consented to participate. In total, 457 children and adolescents (from now on referred to as “children”), 204 mothers and 79 fathers consented to participate. In order to be eligible for further analyses, data of both a parent and

child needed to be available and the child needed to be pain-free. Because the presence of chronic or recurrent pain in the child was not directly assessed in the child, the pain status of the child was determined based on maternal report. Mothers were asked whether the child that participated in the study had chronic or recurrent pain (item: "Is your child experiencing chronic or recurrent pain (with chronic or recurrent pain we mean pain that was continuously or intermittent present during the last 3 months)? If yes, indicate how long the pain was present."). Children were considered pain-free if the mother responded 'no' on this item[a]. Based on these eligibility criteria, 160 parent-child dyads (134 mother-child dyads and 26 father-child dyads) were retained from this participant pool.

Furthermore, all FLFP-members (N = 1395) received a letter with general information concerning studies of the Ghent Health Psychology Lab. A total of 563 persons responded to the invitation letter and 481 persons agreed to be contacted for participation in studies of the Ghent Health Psychology Lab. All parents who reported having one or more children aged between 8 and 18 years (N = 133) were contacted by telephone. Parents and children received information about the study and 105 parents were interested to participate and consented to receive the necessary documents. These parents were asked whether they had a computer with internet access to complete the online survey. If so, they received a letter with more information about the study (purpose, duration and compensation, see above), a link to the online survey and three personal codes. If not, paper and pencil questionnaires were sent and were returned by mail in a prepaid envelope. In 62 families at least one member of the family (child, mother or father) consented to participate. Of these 62 families, in total 53 children, 61 mothers and 31 fathers consented to participate. Again, only families for which we had data of both a parent and child, and for which the child was pain-free, were eligible for further analyses. Based on this criterion, 41 parent-child dyads (40 mother-child dyads and 21 father-child dyads) were retained from this participant pool. Given the unequal sample sizes of mother-child and father-child dyads and the small number of eligible father-child dyads, we decided to include only mother-child dyads in our analyses. Post hoc power analysis (G*power [31]) indicated that, given a total of 47 father-child dyads, the power to detect small effects (f^2 = .10) at α = .05 for the

[a] Note: 12 mothers recruited through schools and 13 mothers recruited through the FLFP reported that their child has chronic or recurrent pain

moderated paths, was only .69. With a total of 174 mother-child dyads, on the other hand, power to detect small sized moderated effects was .99.

The level of pain severity for all 174 mothers was examined by means of the Graded Chronic Pain Scale (GCPS; [32]). This is a reliable and valid instrument to assess pain severity [32,33]. Based on the results of the GCPS, mothers were classified in one of five pain grades, by combining characteristic pain intensity with self-reported pain-related disability. Pain intensity is indexed by the average of current pain, worst pain in the past six months and average pain in the past six months (all measured on a scale from 0 = 'no pain' to 10 = 'pain as bad as could be'). Pain-related disability is a combination of the number of disability days in the past six months (i.e., number of days that the person has been kept from his/her usual activities because of pain) and the degree of interference with daily activities, social activities/family and work (all measured on a scale from 0 = 'no interference' to 10 = 'unable to carry out any activity'). Based on this, mothers' pain severity was then classified in one of the following five grades: grade 0, no pain problem; grade I, low pain intensity and low disability; grade II, high pain intensity, low disability; grade III, moderate disability and grade IV, high disability [32]. In Figure 2, an overview of the number of mothers classified in each grade is given per recruitment source. It can be concluded that the mother-child dyads recruited through school do not generally represent a control group of mothers without CP. Therefore, participants from both participant pools were reclassified into our study groups. Classification of mother-child dyads in the ICP group (i.e., ICPs and their child) versus control group (i.e., mother without CP and their child) was based on mothers' level of pain severity (GCPS) in combination with the duration of the pain. In line with the recently outlined 'IASP Classification of Chronic Pain for the International Classification of Diseases (ICD-11)'[34], pain was defined to be chronic whenever the duration was longer than three months. Mothers that reported no pain (grade 0) and mothers in grades I-IV who reported a pain duration of less than three months, were assigned to the control group, together with their child. Although some mothers in this group do report pain with low or moderate disability (i.e., five mothers were classified in grades I-II), according to the guidelines presented in the ICD-11 their pain is not considered to be chronic as the duration is less than three months[34]. Since we aimed at classifying mothers based on their chronic pain status, these five mothers were therefore retained in

the control group. Mothers in grades I-IV who reported a pain duration of at least three months were categorized as ICPs and were assigned to the ICP group, together with their child. In total, 46.3% of mother-child dyads recruited through schools were included in the ICP group. The final control group consisted of 74 dyads in the control group, of which 72 were recruited through schools. The final ICP group consisted of 100 dyads, of which 62 were recruited through schools. For each study group, an overview of the number of mothers per grade is given in Figure 2.

This study was approved by the ethical committee of the faculty of Psychology and Educational Sciences at Ghent University; participants gave written informed consent at the start of the (online) survey.

- Insert Figure 2 about here -

Measures

Child report measures

Sociodemographic measures. Information on the children's gender, age, family situation and socioeconomic status was collected. Socioeconomic status of the child's family was assessed by means of an adapted Dutch version of the Family affluence scale (FAS; [35,36]). The FAS consists of four items reflecting the child's family affluence: 'Does your family own a car, van or truck?' (0 = 'no'; 1 = 'yes, one'; 2 = 'yes, two or more'); 'Do you have your own bedroom for yourself?' (0 = 'no'; 1 = 'yes'); 'During the past 12 months, how many times did you travel away on holiday with your family?' (0 = 'not at all'; 1 = 'once'; 2 = 'twice', 3 = 'more than twice'); 'How many computers does your family own?' (0 = 'none'; 1 = 'one'; 2 = 'two', 3 = 'more than two'). Items were summed in a total score ranging between 0 and 9, with higher scores indicating greater affluence. Following Vervoort and colleagues [36], family affluence was categorized based on the total score into low (0-3), medium (4-6) and high (7-9) family affluence.

Catastrophizing thoughts about pain. Children's pain catastrophizing was assessed with the Pain Catastrophizing Scale for children (PCS-C; [37]). This scale is based on the Pain Catastrophizing Scale for adults (PCS; [38]) and consists of 13 items measuring thoughts and feelings that children might experience when they are in pain. Children were asked to rate the frequency of each of these thoughts

and feelings on a 5-point scale (0 = 'not at all'; 4 = 'extremely'). The total score (ranging between 0-52) was used in the current study. The PCS-C has been shown to be a reliable and valid instrument for measuring pain catastrophizing in community samples of children [37]. In the current study, the internal consistency (Cronbach's alpha) was 0.93.

Child distraction. Children's use of distraction was assessed with the Dutch version of the Pain Coping Questionnaire (PCQ; [22,39]). Children were asked to rate how often they use each of the coping responses when facing pain on a 5-point scale (1 = 'never'; 5 = 'very often'). As described by Reid and colleagues [22], the 10 items of the subscales 'behavioral distraction' and 'cognitive distraction' were averaged to get a score (ranging between 1-5) for the higher-order factor problem-focused avoidance (in this study, we refer to this factor as "child distraction"). The PCQ has been shown to be a reliable and valid instrument for measuring pain coping responses in community samples of children [22,24]. Cronbach's alpha in the current study was 0.90.

Somatic symptoms. The Dutch version of the Children's Somatization Inventory (CSI; [40,41]) was used to measure severity of 35 nonspecific somatic symptoms (e.g., headaches, dizziness, nausea). Children rated the extent to which they experienced the symptoms in the past two weeks on a scale from 0 (not at all) to 4 (a whole lot). A total score (0-140) was computed by summing all item ratings and was used as an outcome variable in the current study. The CSI has been shown to be a reliable and valid measure for assessing somatic symptoms in community samples of children [41,42]. Cronbach's alpha in the current study was 0.90.

Health-related Quality of Life. Children's perception of their health-related quality of life (HRQOL) was assessed with the 23-item Dutch version of the PedsQL 4.0 [43,44]. A version for 8-12 and 13-18-year-old children was used, both consisting of four generic core scales: physical (8 items), emotional (5 items), social (5 items) and school functioning (5 items). Each item was rated on a 5-point scale ranging from 0 = 'never a problem' to 4 = 'almost always a problem' (Scores were reverse-scored and transformed to a range between 0-100 (0 = 100, 1 = 75, 2 = 50, 3 = 25, 4 = 0); with higher scores indicating a better HRQoL). Next to the four subscales, a psychosocial health summary score (summing of scores on the emotional, social and school functioning subscales) and a total score were computed. In the current study the psychosocial health summary score and the scale

score of physical functioning were used as outcome variables. The Dutch version of the PedsQL 4.0 has been shown to be a reliable and valid instrument in community samples of children [44,45]. For the child version (8-12 yrs), Cronbach's alpha in the current study was 0.90 for the total score, 0.90 for psychosocial health and 0.83 for physical functioning for the child-version. For the adolescent version (13-18 yrs), Cronbach's alpha was 0.88 for the total score, 0.86 for psychosocial health and 0.81 for physical functioning.

Parent report measures

Sociodemographic measures. Information on mothers' age, educational level, occupation, and family status was gathered. Furthermore, mothers were asked whether the child that participated in the study had chronic or recurrent pain (item: "Is your child experiencing chronic or recurrent pain (with chronic or recurrent pain we mean pain that was continuously or intermittent present during the last 3 months)? If yes, indicate how long the pain was present.").

Catastrophizing thoughts about pain. The Dutch version of the PCS [38,46] was used to measure the level of catastrophizing thoughts and feelings about personal pain in the mothers. Mothers were asked to think about previous pain experiences and to rate the frequency of 13 thoughts and feelings that can be present when feeling pain. Each item was rated on a scale ranging from 0 ('not at all') to 4 ('always'); summing the item scores yielded a total score (ranging between 0-52), which was used in the current study. The Dutch version of the PCS has proven to be a reliable and valid instrument in healthy adults and clinical populations [46]. Cronbach's alpha in the current study was 0.95.

Maternal distraction. A Dutch version of the PCQ for adults [22,24] was used to measure the level of maternal distraction when confronted with personal pain. Mothers were asked to rate how often they use each of the coping responses when facing pain on a 5-point scale (1 = 'never'; 5 = 'very often'). In line with the child report, the 10 items of the subscales 'behavioural distraction' and 'cognitive distraction' were averaged to obtain a score (ranging between 1-5) for maternal distraction. The PCQ has been shown to be a reliable and valid instrument for measuring pain coping responses in adults [24]. Cronbach's alpha in the current study was 0.90.

Statistical analyses

Before conducting our main analyses, total scores were computed for all self-report measures. Total scores were only computed if no more than 25% of the items was missing. Using IBM SPSS Statistics (Version 25.0) [47] descriptive statistics and correlation analyses on the variables of interest were conducted. Little's MCAR test indicated that missing data were missing completely at random ($\chi^2(11) = 18.31$, $p = .075$). In this case, all imputation methods are possible [48] and we have opted to adopt a pairwise deletion method for descriptive statistics and correlations. Pearson's chi-square test was used to examine relationships between maternal pain status and other categorical variables. When cell frequencies were smaller than five, the likelihood ratio statistic was reported. To test for group differences *t*-tests were used for continuous variables and Mann-Whitney *U* tests for categorical variables.

The primary aim of the current study is to examine how parental coping responses (predictor: maternal pain catastrophizing and distraction) are related to corresponding child coping responses (mediator: child pain catastrophizing and distraction) and how these child coping responses in turn affect child outcomes (somatic symptoms, physical functioning and psychosocial health). Moreover, we aim to examine the impact of maternal pain status (moderator: presence or absence of chronic pain) on these relationships. We first evaluated the level of independence between the scores of the mothers and the children. As proposed by Kenny, Cashy and Cook [49], we calculated a canonical correlation between the coping of the mother (i.e., maternal pain catastrophizing and maternal distraction) and the coping of the child (i.e., child pain catastrophizing and child distraction) to evaluate the multivariate shared relationship between these two sets of variables. Given that the canonical correlation between the two sets of variables was not significant ($r_c = .20$, Wilks $\lambda = 0.96$, $F(4,312) = 1.75$, $p = .14$), a single-level model was used in our analyses. In line with other research examining (in)direct associations between parental and child variables (e.g., [50–52]), the PROCESS macro for SPSS (3rd version; [53]) was used. To examine the hypothesized model (Figure 1), moderated mediation analyses were performed. In these analyses, mothers in the control group were coded as "0" and ICPs as "1" and continuous predictors and mediators were mean-centered. Cases with missing

data on any of the variables in the model were excluded from the analyses (i.e., listwise deletion). A series of six models was estimated, one for each child outcome (i.e., somatic symptoms, physical functioning score, psychosocial health score) and each coping response (i.e., pain catastrophizing and distraction). The models with pain catastrophizing as coping response were run on 161 mother-child dyads; the models with distraction as coping response on 160 mother-child dyads. As depicted in Figure 1, in each of these models an a-path, b-path and c'-path is estimated using an ordinary least squares (OLS) regression procedure [53]. We estimated the effect of maternal coping responses on child coping responses (a-path), the effect of child coping responses on child outcomes (b-path) and the effect of maternal coping responses on child outcomes (c'-path[b]). Significance of these coefficients was tested against an α-level of 0.05. To test our main hypothesis, we examined whether the indirect effect of maternal coping responses on child outcomes through child coping responses differed significantly between the ICP group and control group (i.e., mediation effect was moderated by maternal pain status). The indirect effect consists of the product of the coefficients of the a-path and b-path, therefore moderation of the a-path (Figure 1) also implies moderation of the indirect effect [54]. As proposed by Hayes [54], we used the Index of Moderated Mediation (IMM) as a quantification of the association between a moderator and the indirect effect. The indirect effect is moderated if this index is significantly different from zero [54]. For the current study, a significant IMM indicates that the conditional indirect effect (i.e., the indirect effect for different values of maternal pain status) differs significantly between the ICP group and the control group. To further elaborate on these group differences, the conditional indirect effects were estimated in both groups and their significance was tested. Significance of the IMM and the conditional effects was tested using bootstrapping. Bootstrapping is a non-parametric resampling procedure [53]. We constructed the 95% bootstrap confidence intervals (BCI) using 5000 bootstrap samples and an effect was deemed significant if the 95% BCI did not include zero. To make the bootstrap results reproducible, we used 271216 as a seed for the random number generator.

[b] In line with Hayes [53] we use the term c'-path, to clearly differentiate this effect from the direct effect of maternal coping responses on child outcomes when a mediator is not added in the model (c-path, total effect). This total effect is not the focus of the current study.

Results

Descriptive group statistics and correlations

Descriptive statistics of sociodemographic and pain-related variables of mothers and children in the control group and ICP group are presented in Table 1. In our sample, mothers' and children's age did not differ significantly between groups and the distribution of boys and girls was similar in both groups (Table 1). In the ICP group 61% of the mothers had higher education (beyond the age of 18 years); in the control group this was the case for 71.6% of the mothers. The level of education did not differ between groups ($\chi^2(1) = 2.12$, $p = .15$). In the ICP group (76.0%) significantly less mothers were employed at the time of the study compared to the control group (93.2%; $\chi^2(1) = 9.10$, $p = .003$). In both groups, the number of children in the family ranged from 1 to 6 and 58.0% of the families in the ICP group and 47.3% of the families in control group consisted of two children. Compared to children in the control group, children of ICPs more often grew up in other family compositions (e.g., stepparent or one parent family) and less often in a classic family (i.e., two parent nuclear family) (Table 1). Furthermore, family affluence was on average higher in the control group (Mean Rank = 95.54) compared to the ICP group (Mean Rank = 81.55; $U = 3105.00$, $p = .038$). Since group classification was based on the GCPS, pain grades of the mothers were significantly higher in the ICP group (Mean Rank = 121.98) compared to the control group (Mean Rank = 40.91*;* $U = 252.50$, $p < .001$). Moreover, average pain duration in months was higher in the ICP group compared to the control group (Table 1). Frequently reported pain complaints in ICPs were back pain (70.0%), neck pain (59.0%) and headaches (56.0%). About one third (32.0%) of ICPs reported being diagnosed with fibromyalgia.

- Insert Table 1 about here -

Next, we examined group differences for the variables which are included in the moderated mediation models (Table 2). Results indicated that children in the ICP group reported lower psychosocial health than children in the control group, somatic symptoms and physical functioning did not differ between both groups (Table 2). The use of coping responses (pain catastrophizing and distraction from pain) did not differ between both groups on the child level. However, on the maternal level, ICPs reported more catastrophizing thoughts about pain and lower use of distraction than mothers in the control group (Table 2).

- Insert Table 2 about here -

Pearson correlation analyses indicated that all child variables were significantly correlated (Table 3). Furthermore, child pain catastrophizing and child distraction were negatively correlated. Child pain catastrophizing was negatively related to physical functioning and psychosocial health and positively to somatic symptoms. Child distraction, on the other hand, was positively related to physical functioning and psychosocial health and negatively to somatic symptoms. Maternal pain catastrophizing and maternal distraction were negatively correlated. Importantly, regarding mother-child correlations, higher levels of maternal pain catastrophizing were associated with higher levels of child pain catastrophizing (Table 3).

- Insert Table 3 about here -

Moderated mediation analyses

Using moderated mediation analyses, we examined whether maternal coping responses were indirectly related to child outcomes through corresponding child coping responses. Furthermore, we examined whether this hypothesized indirect effect was moderated by the presence or absence of maternal CP. The results of the moderated mediation analyses are presented in Table 4 (see Supplementary Table for all path values). Running the models while controlling for the employment

status of the mother and family affluence scores, yielded the same conclusions as the results presented below.

The analyses indicated that the modelled indirect effects of maternal pain catastrophizing on child outcomes through child pain catastrophizing were significantly moderated by maternal pain status for all child outcomes (none of the bootstrap confidence intervals (BCI) for the index of moderated mediation included zero, see Table 4). Counter to expectations, the conditional indirect effects of maternal pain catastrophizing on child outcomes (i.e., child somatic symptoms, child physical functioning and child psychosocial health) through child pain catastrophizing were only significant in the control group (BCIs for the conditional indirect effects in the control group did not include zero, see Table 4). Findings demonstrated that, in the control group, higher maternal pain catastrophizing was related to higher child pain catastrophizing (a-path: B = 0.49, $t(157) = 3.90$, $p < .001$). Higher child pain catastrophizing, in turn, was related to more somatic symptoms (b-path: B = 0.50, $t(156) = 5.86$, $p < .001$), lower physical functioning (b-path: B = -0.39, $t(156) = -4.19$, $p < .001$) and lower psychosocial health (b-path: B = -0.65, $t(156) = -6.91$, $p < .001$) in children. The conditional indirect effects in the ICP group were not significant[c] (all BCIs included zero, see Table 4).

- Insert Table 4 about here -

No evidence was found for a moderated mediation effect of maternal distraction on child outcomes through child distraction (all BCIs for the index of moderated mediation included zero, see Table 4). We only identified an association between child distraction and child outcomes, with higher levels of child distraction related to less somatic symptoms (B = -4.78, $t(155) = -4.04$, $p < .001$), higher physical functioning (B = 4.92, $t(155) = 3.98$, $p < .001$) and higher psychosocial health (B = 5.44, $t(155) = 4.04$, $p < .001$).

[c] Separate mediation models with maternal pain catastrophizing as predictor, child pain catastrophizing as mediator and child functioning as outcomes were run for ICPs recruited through schools and ICPs recruited via the FLFP. Indirect effects were not significant in both groups.

Discussion

The aim of this study was to increase our understanding of child functioning in a context of parental chronic pain (CP). More specifically, we examined whether maternal and child pain coping responses may contribute to the vulnerability of children to develop adverse outcomes in the context of maternal CP. Based on Social Learning Theory (SLT) [6], we hypothesized that parent and child coping responses are interrelated and that parent coping responses are associated with child functioning through child coping responses. Moreover, we expected that these associations would be especially present in children of individuals with chronic pain (ICPs) as they tend to observe parental pain behaviours more frequently [9]. To the best of our knowledge, this is the first study to examine these interrelationships in a group of pain-free children of ICPs and pain-free children of mothers without CP. Moreover, only one previous study examined whether child vulnerabilities (i.e., pain-related cognitive biases) are affected by parental CP, but failed to find differences between children of parents with and without CP [14]. In contrast, moderated mediation analyses in the current study indicated that maternal pain catastrophizing was indirectly related with child functioning through child pain catastrophizing. Unexpectedly, this indirect effect was only present among children of mothers without CP (control group). Maternal distraction was not found to be indirectly related with child functioning through child distraction.

The current findings partially support a social learning perspective, but also raise some questions that require further research. Specifically, for mothers without CP, maternal pain catastrophizing was positively related to child pain catastrophizing which, in turn, was related to more somatic symptoms, lower physical functioning and lower psychosocial health in their child. Notably, this lowered functioning might potentially indicate a heightened risk for later development of CP (see also [55]). The observed concordance between parent and child pain catastrophizing may arise because children adapt their coping responses from observing parental coping responses and associated behaviours [6,7]. These results confirm previous studies [26,27] and theoretical models about paediatric pain [13,56]. Given the cross-sectional nature of the data, conclusions cannot be drawn on the direction of the observed associations. Longitudinal research is needed to confirm and extend the current results by

indicating how parent and child pain catastrophizing may impact child functioning in a context of parental CP over time. Unexpectedly, indirect relationships between maternal pain catastrophizing and child functioning through child pain catastrophizing were not present in children of ICPs. Because of repeated observations of pain behaviours associated with both parental pain catastrophizing [28,29] and parental CP status [9], we expected that children of ICPs would be more likely to report catastrophic beliefs about pain and consequently more negative outcomes [50,57]. Our findings indicated higher levels of maternal pain catastrophizing in the ICP group compared to the control group. However, while children of ICPs reported lower psychosocial health compared to children in the control group, findings demonstrated that child functioning was not (indirectly) related with maternal pain catastrophizing amongst children of ICPs. This suggests that high levels of parental catastrophizing about personal pain may not act as a vulnerability factor in the context of parental CP. Likewise, low levels of parental pain catastrophizing in ICPs do not seem to protect against the development of adverse outcomes.

Some possible explanations can be put forward for this unexpected pattern of results. First, the concordance between CP status of mother and child may have an impact on observational learning processes. SLT posits that observational learning is enhanced when model and observer share characteristics [6] and our results seem to support this statement. Children without CP may identify more with their mother's episodic pain experiences compared to chronic pain experiences. Hence, observing maternal pain catastrophizing and associated behaviours in everyday pain situations may lead to similar child coping responses when confronted with acute pain. Similarly, and supported by previous results (e.g., [9–12]), children with CP may identify more with maternal CP experiences and therefore learn more from observing maternal coping with CP. To fully understand vulnerability and protective factors in the context of parental CP, future research should therefore continue to examine children without CP next to children who already experience CP. Second, it is possible that direct parental reinforcing responses are more important in children of ICPs. Indeed, parental solicitous responding (e.g., granting the child special privileges, comforting the child) is related to adverse child outcomes (e.g., increased disability, somatic symptoms) [15,16,18,19] and solicitous responses are more common in high catastrophizing parents [58–60] and parents with CP [9]. Although research comparing

both SLT processes (i.e., reinforcing responses and observational learning) is scarce, one recent study in paediatric CP patients showed that parental solicitousness was not related with child pain severity or functional impairment in a model that also incorporated observational learning [9]. To our knowledge, reinforcing processes are not yet examined in a sample of pain-free children growing up in a context of parental CP and studies jointly examining both SLT processes are highly needed. Third, the lack of an indirect relationship between maternal pain catastrophizing and child functioning through child pain catastrophizing might be due to heterogeneity in the group of children of ICPs. For example, some children may have been exposed to their mother experiencing pain from an early age on, while for other children, maternal CP may have developed at a later point in time. Consequently, learning time may vary significantly. In future studies, these and other child and family characteristics need to be further explored in children of ICPs. Finally, it should be noted that maternal pain catastrophizing was higher in ICPs compared to controls in the current study, making it difficult to disentangle the effects of maternal pain catastrophizing and pain status. Further research is needed to clarify their relative contribution on child functioning by, for example, exploring SLT processes in parents with and without CP but with similar levels of pain catastrophizing.

To fully understand child functioning in a context of parental CP, consideration of mechanisms that might protect children from developing adverse outcomes is also needed [1]. Therefore, we added distraction as a supposedly adaptive and observable coping response and, as expected, the results indicated that children's use of distraction was associated with less somatic symptoms, higher physical functioning and higher psychosocial health. Furthermore, ICPs reported lower use of distraction compared to mothers without CP. However, maternal distraction was not related with child functioning via child distraction. Thus, our results did not support the idea that maternal distraction could act as a protective factor in the context of parental CP. Possibly, other constructs are more suited to capture adaptive coping with CP and to examine the impact of maternal adaptive coping on child coping and functioning. For example, measures of pain acceptance (i.e., a concept derived from Acceptance and Commitment Therapy, ACT [61]) have been shown to be good predictors of adjustment to CP [62]. Further research is needed to examine whether observing parental pain acceptance and associated behaviours, contribute to child pain acceptance and functioning. If

research would indicate that children start to use adaptive coping responses (e.g., pain acceptance) by observing parental adaptive coping responses, social learning principles may be used in interventions.

Our results extend limited and inconclusive previous findings and are clinically relevant by increasing our knowledge on the vulnerabilities of pain-free children growing up with a parent with CP. Still, clinical implications remain rather limited given that indirect effects were only observed in mothers without CP who generally report low levels of pain catastrophizing. Additionally, some limitations should be mentioned. First, data are cross-sectional, so we cannot infer causality; longitudinal and experimental designs are necessary to address causality. Second, although the aims of this study can be framed within SLT, we did not explicitly measure observable parental (pain) behaviours or test observational learning processes. Experimental studies should be carried out to test whether child functioning differs after observation of different coping responses used by a parent experiencing (laboratory-induced) pain. Furthermore, the recently developed Parent Pain Behavior-Proxy form [63] can be used in future studies to let parents and children report on daily parental pain behaviours. This might increase our understanding of which parental behaviours are actually observed by children and enables the examination of parental (pain) behaviours as an important mediator in the relationship between parental coping responses on the one hand and child coping responses and functioning on the other. Third, we only included mother-child dyads in our final analyses. This is a common problem in this field of research but can be especially problematic given previously observed sex differences in (parental) pain catastrophizing [38,64]. Therefore, our results might not be generalizable to father-child dyads. Moreover, when examining social learning processes, it would be valuable to include all caregivers and examine caregiver-child associations on the one hand and interactions between caregivers on the other. Fourth, our sample was potentially impacted by a self-selection bias. Indeed, the proportion of mothers recruited through schools who met the criteria for chronic pain, was larger than one would expect based on European prevalence studies [65,66]. Based on the information given in the invitation letters, mothers who experience chronic or recurrent pain may have been more inclined and motivated to participate. Fifth, we assessed the presence of chronic or recurrent pain in the children via maternal report. Future studies should asses this directly in the child

as well. Finally, it remains to be examined whether our results extend to children with CP and whether the results can be replicated in a larger sample.

Despite these limitations, the current findings suggest that maternal pain catastrophizing is indirectly related to her child's somatic symptoms, physical functioning and psychosocial health through child's pain catastrophizing. Yet, this effect was only observed amongst pain-free children of mothers without CP. Finally, we did not observe any parent-child associations regarding the use of distraction. Our results highlight the need for a continued search for vulnerability and protective factors in the context of parental CP, both in children with and without CP. Combining knowledge about these vulnerability and protective factors will be crucial in defining which parental (coping) responses should be targeted in interventions aimed at improving child functioning.

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Figures and Tables

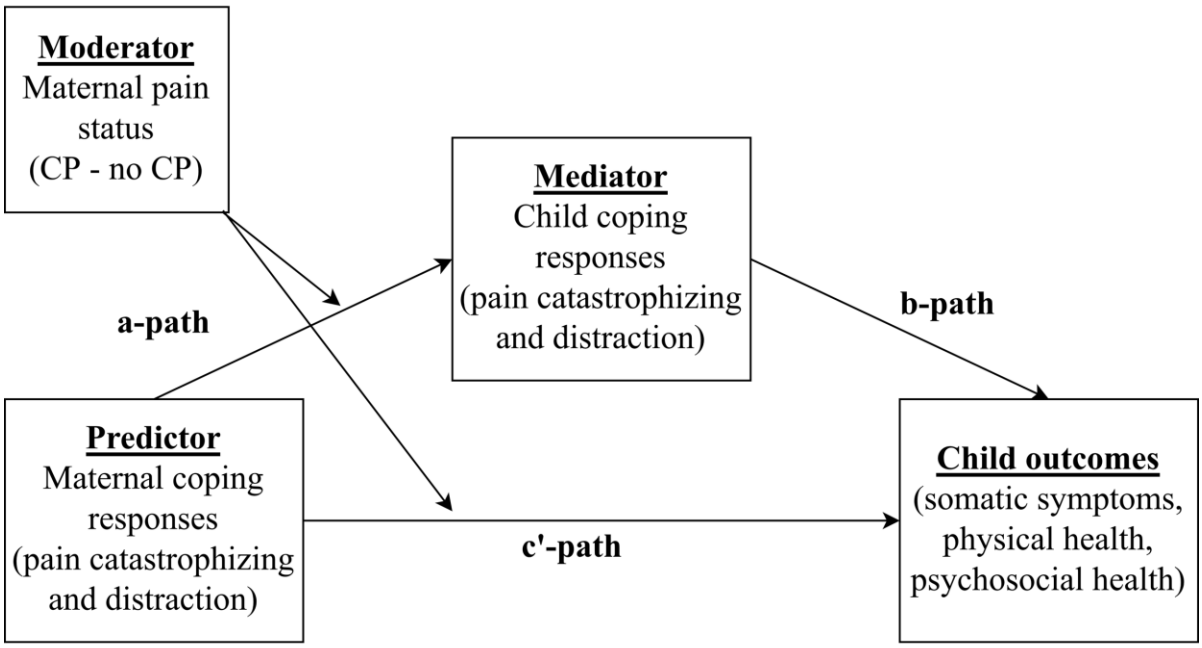

Figure 1. Hypothesized moderated-mediation model. a-path = effect of predictor on mediator; b-path = effect of mediator on outcomes; c'-path = effect of predictor on outcomes, controlling for the mediator; CP = Chronic Pain

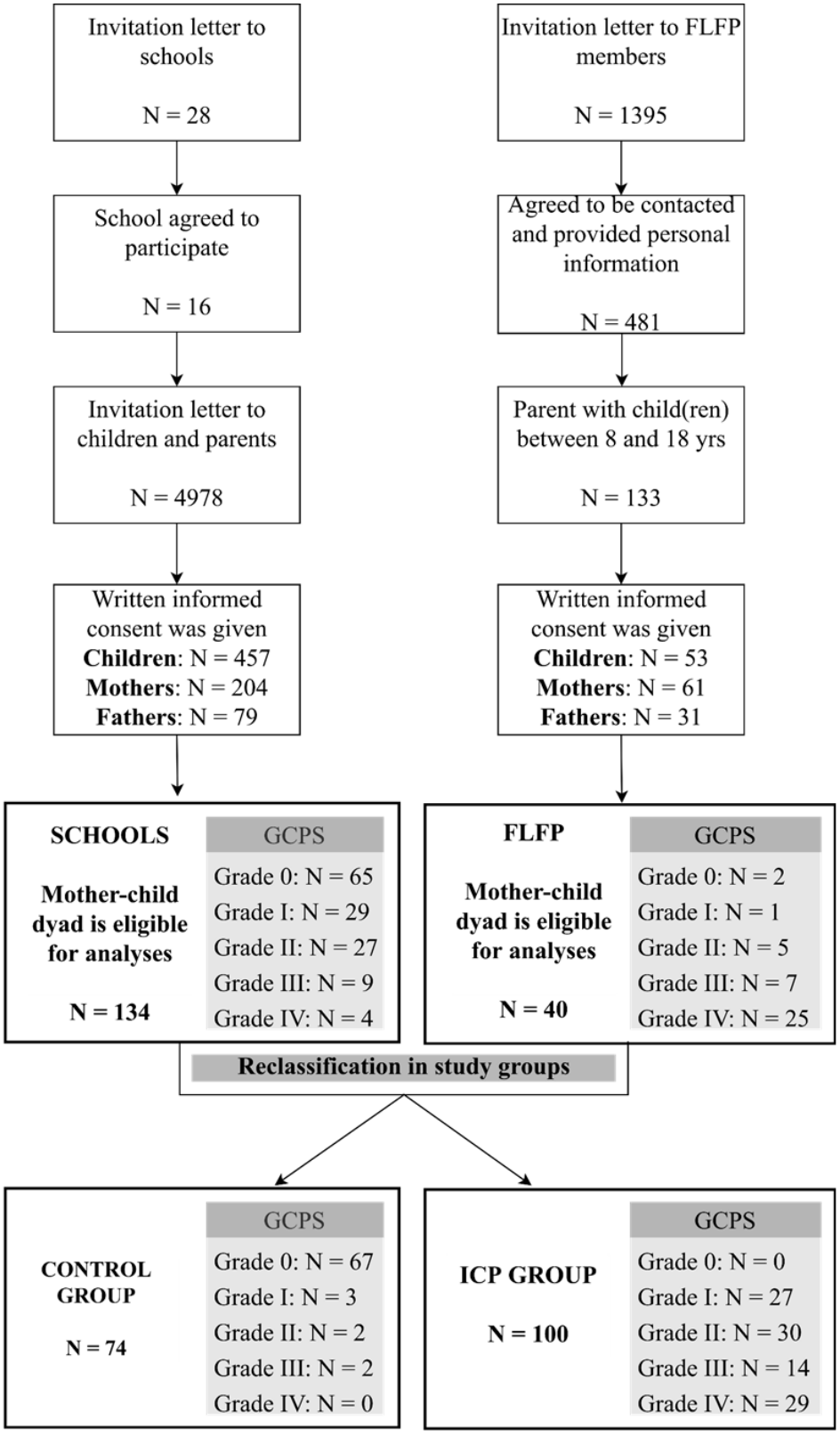

Figure 2. Flowchart of how the sample was obtained through schools and the Flemish League for Fibromyalgia Patients (FLFP); GCPS = Graded Chronic Pain Scale (GCPS [32]); ICP = Individual with Chronic Pain

Table 1. Participant characteristics and group differences.

		ICP				Control				Group differences
		n	%	*M*	*SD*	*n*	%	*M*	*SD*	Test statistic
Child variables										
Age				13.46	2.81			12.84	2.56	$t(172) = -1.50$
Gender	Girl	62	62.0			44	59.5			$\chi^2(1) = 0.12$
	Boy	38	38.0			30	40.5			
Mother variables										
Age				43.57	4.57			43.20	4.58	$t(172) = -0.52$
Pain grade	0	0	0			67	90.5			
	1	27	27.0			3	4.1			
	2	30	30.0			2	2.7			$U = 252.50$***
	3	14	14.0			2	2.7			
	4	29	29.0			0	0			
Pain duration				115.21	112.93			0.96	0.74	$t(99.12) = -10.11$***

ICP: mother with chronic pain; Control: mother without chronic pain; Child pain: the presence or absence of chronic or recurrent pain, as reported by the mother
*p <.05; ** p <.01; *** p <.001.

Table 1. (continued)

		ICP				Control				Group differences
		n	%	*M*	*SD*	*n*	%	*M*	*SD*	Test statistic
Family variables										
Composition	classic	69	69.0			66	89.2			
	one parent	11	11.0			3	4.1			$L_{\chi^2}(3) = 11.47$**
	step parent	16	16.0			3	4.1			
	other	4	4.0			2	2.7			
Affluence score	low	4	4.0			0	0			
	medium	51	51.0			30	40.5			$U = 3105.00$*
	high	45	45.0			44	59.5			

ICP: mother with chronic pain; Control: mother without chronic pain
*p <.05; ** p <.01; *** p <.001.

Table 2. Descriptive statistics of variables included in moderated-mediation models (for each group) and group differences.

	ICP				Control				Group differences	
	N	*M*	*SD*	Range	*N*	*M*	*SD*	Range	Test statistic	Effect size
Coping responses										
Child pain catastrophizing	93	16.71	10.54	0-43	68	16.34	11.30	0-52	$t(159) = -0.21$	0.03
Child distraction	92	2.81	0.80	1-5	68	2.80	0.78	1-5	$t(158) = -0.07$	0.01
Maternal pain catastrophizing	100	19.55	12.12	0-52	74	13.41	10.00	0-42	$t(172) = -3.55$***	0.54
Maternal distraction	100	2.65	0.72	1.1-3.9	74	3.01	0.66	1-4.5	$t(172) = 3.37$**	0.52
Child outcomes										
Somatic symptoms	100	14.5	11.30	0-48	74	11.46	12.85	0-81	$t(172) = -1.66$	0.25
Physical health	100	83.84	13.02	46.9-100	74	87.54	12.74	46.9-100	$t(172) = 1.87$	0.29
Psychosocial health	100	76.82	14.82	35-100	74	82.57	12.48	36.7-100	$t(172) = 2.70$**	0.41

ICP: mother with chronic pain; Control: mother without chronic pain
*p <.05; ** p <.01; *** p <.001.

Table 3. Pearson correlation coefficients between child and maternal variables of interest.

		1	2	3	4	5	6	7
Child variables								
1. Child pain catastrophizing	ICP	-	-.23*	.47***	-.44***	-.58***	.03	.001
	Control	-	-.29*	.37**	-.07	-.31**	.44***	-.05
2. Child distraction	ICP		-	-.31**	.33**	.30**	-.19	.12
	Control		-	-.29*	.26*	.32**	-.02	-.02
3. Somatic symptoms	ICP			-	-.57***	-.68***	.06	.11
	Control			-	-.41***	-.80***	.05	-.12
4. Physical health	ICP				-	.44***	.02	-.07
	Control				-	.43***	.14	-.03
5. Psychosocial health	ICP					-	-.11	-.05
	Control					-	-.06	.06
Maternal variables								
6. Maternal pain catastrophizing	ICP						-	-.55***
	Control						-	.18
7. Maternal distraction	ICP							-
	Control							-

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

Table 4. Index of moderated mediation and conditional indirect effects for both groups.

	Index of Moderated Mediation			Conditional indirect effects	
	IMM	Boot SE	95% Boot CI	ICP (95% BCI)	Control (95% BCI)
Indirect effect of maternal pain catastrophizing through child pain catastrophizing on…					
Somatic symptoms	**-0.23**	0.10	-0.47 to -0.06	0.01 (-0.07 to 0.11)	**0.25** (0.09 to 0.47)
Physical health score	**0.18**	0.07	0.05 to 0.33	-0.01 (-0.09 to 0.06)	**-0.19** (-0.34 to -0.07)
Psychosocial health score	**0.30**	0.12	0.10 to 0.55	-0.02 (-0.14 to 0.09)	**-0.32** (-0.54 to -0.14)
Indirect effect of maternal distraction through child distraction on…					
Somatic symptoms	-0.57	1.06	-2.75 to 1.66	-0.66 (-2.18 to 0.77)	-0.09 (-1.81 to 1.53)
Physical health score	0.59	1.08	-1.50 to 2.80	0.68 (-0.76 to 2.15)	0.09 (-1.64 to 1.69)
Psychosocial health score	0.65	1.18	-1.84 to 3.00	0.75 (-0.93 to 2.27)	0.10 (-1.78 to 1.94)

Values in bold are significant (i.e., 95% bootstrap confidence interval did not include zero)
IMM: Index of moderated mediation; ICP: mother with chronic pain; Control: mother without chronic pain

Supplementary Table. Estimated unstandardized coefficients of the moderated mediation models.

		Pain Catastrophizing	Distraction
		B (SE)	B (SE)
X → M, moderated by maternal pain status (a-path)			
Moderation of the a-path		-0.47 (0.15)**	0.12 (0.19)
Effect of maternal pain status on M		-1.20 (1.70)	0.04 (0.13)
Conditional effect of X on M	Control	0.49 (0.13)***	0.02 (0.15)
	ICP	0.03 (0.09)	0.14 (0.12)
M → Child outcomes (somatic symptoms, physical and psychosocial health) (b-path)			
Effect of M on somatic symptoms		0.50 (0.08)***	-4.78 (1.18)***
Effect of M on physical health		-0.39 (0.09)***	4.92 (1.24)***
Effect of M on psychosocial health		-0.65 (0.09)***	5.44 (1.35)***
X → Somatic symptoms, moderated by maternal pain status (c'-path)			
Moderation of the c'-path		0.23 (0.17)	4.76 (2.77)
Effect of maternal pain status on somatic symptoms		3.13 (1.85)	2.87 (1.94)
Conditional effect of X on somatic symptoms	Control	-0.18 (0.14)	-2.54 (2.17)
	ICP	0.05 (0.10)	2.23 (1.29)
X → Physical health, moderated by maternal pain status (c'-path)			
Moderation of the c'-path		-0.32 (0.18)	-0.82 (2.90)
Effect of maternal pain status on physical health		-4.52 (2.01)*	-4.08 (2.03)*
Conditional effect of X on physical health	Control	0.33 (0.15)*	-0.82 (2.27)
	ICP	0.01 (0.10)	-1.64 (1.80)
X → Psychosocial health, moderated by maternal pain status (c'-path)			
Moderation of the c'-path		-0.37 (0.19)	-2.91 (3.15)
Effect of maternal pain status on psychosocial health		-6.35 (2.04)**	-5.72 (2.21)*
Conditional effect of X on psychosocial health	Control	0.26 (0.16)	1.85 (2.47)
	ICP	-0.10 (0.10)	-1.06 (1.96)

Note: Column 'pain catastrophizing' displays the results for the models where maternal pain catastrophizing was used as predictor (X) and child pain catastrophizing as mediator (M); Column 'distraction' displays the results for the models where maternal distraction was used as predictor (X) and child distraction as mediator (M); ICP = Individual with chronic pain

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