

Running Title: CHILD AND PARENT PSYCHOLOGICAL FLEXIBILITY

Title: Resilience factors in children with Juvenile Idiopathic Arthritis and their parents: the role of child and parent psychological flexibility.

Melanie Beeckman, MSc^a, Sean Hughes, PhD^a, Dimitri Van Ryckeghem, PhD^{a,b} Eline Van Hoecke, PhD^c, Joke Dehoorne, MD, PhD^d, Rik Joos, MD^e, & Liesbet Goubert, PhD^a

Author Affiliations: ^a Department of Experimental-Clinical and Health Psychology, Ghent University, H. Dunantlaan 2, 9000 Ghent, Belgium. Mebeeckm.Beeckman@ugent.be, Sean.Hughes@ugent.be, Liesbet.Goubert@ugent.be ^b Institute for Health and Behaviour, INSIDE, University of Luxembourg, Maison des Sciences Humaines, Porte des Sciences 11, L-4366 Esch-sur-Alzette, Luxembourg. Dimitri.VanRyckeghem@uni.lu ^c Department of Pediatric Psychology, Ghent University Hospital, De Pintelaan 185, 9000 Ghent, Belgium.

Eline.VanHoecke@uzgent.be ^d Department of Pediatric Rheumatology, Ghent University Hospital, De Pintelaan 185, 9000 Ghent, Belgium. Joke.Dehoorne@uzgent.be

^e Department of Pediatric Rheumatology, Ghent University Hospital, De Pintelaan 185, 9000 Ghent, Belgium. Rik.Joos@uzgent.be

Corresponding Author: All correspondence concerning this article should be addressed to Melanie Beeckman, Department of Experimental, Clinical and Health Psychology, Ghent University, H. Dunantlaan 2, 9000 Ghent E-mail: Mebeeckm.Beeckman@UGent.be.

The authors have no conflicts of interest to declare.

This manuscript was supported by a grant from the Special Research Fund of Ghent University, grant number BOF15/24j/017, awarded to Liesbet Goubert and Sean Hughes.

Dimitri Van Ryckeghem is supported by the European Union's Horizon 2020 research and innovation program under the Marie Skłodowska-Curie grant No. 706475.

Abstract

Objective: Chronic pain is central to Juvenile Idiopathic Arthritis (JIA) and is predictive of impaired functioning. Whereas most work has focused on identifying psychosocial risk factors for maladaptive outcomes, we explored the idea that child and parental psychological flexibility (PF) represent resilience factors for adaptive functioning of the child. We also explored differences between general versus pain-specific PF in contributing to child outcomes.

Methods: Children with JIA (8-18 years) and (one of) their parents were recruited at the department of pediatric rheumatology at the Ghent University Hospital in Belgium. They completed questionnaires assessing child and parent general and pain-specific PF and child psychosocial and emotional functioning, and disability.

Results: The final sample consisted of fifty-nine children and forty-eight parents. Multiple regression analyses revealed that child PF contributed to better psychosocial functioning and less negative affect. Child pain acceptance contributed to better psychosocial functioning, lower levels of disability and lower negative affect, and also buffered against the negative influence of pain intensity on disability. Bootstrap mediation analyses demonstrated that parental (general) PF indirectly contributed to child psychosocial functioning and affect via the child's (general) PF. Parent pain-specific PF was indirectly linked to child psychosocial functioning, disability, and negative affect via child pain acceptance.

Conclusion: Our findings indicate that child and parental PF are resilience factors and show that pain acceptance buffers against the negative impact of pain intensity. Implications for psychosocial interventions that target (pain-specific) PF in children as well as in parents are discussed.

Keywords: chronic pain; children; parents; psychological flexibility; pain acceptance; functioning

Introduction

Juvenile Idiopathic Arthritis (JIA) is a common chronic inflammatory disease of childhood with pain as a central complaint (1). Chronic pain has been frequently associated with impairments in physical, school, social, and emotional functioning (2,3). To better understand and target these relations, most research has focused on factors that increase the risk of maladaptive outcomes, such as pain intensity, pain-related fear, and catastrophizing (4–6). However, a significant portion of children do function well despite the continued presence of pain (7). Therefore a number of models have recently argued that attention to both risk *and* resilience factors is crucial if we are to understand how, why, and in which contexts, some individuals function well despite pain whereas others do not (8–10).

Psychological flexibility (PF), described as one's openness and acceptance towards, and disengagement from, unwanted experiences such as pain and (fearful) emotions and thoughts while still being able to engage in valued activities, is a resilience factor that has been consistently linked to adaptation to chronic pain (11–13). Until now, research addressing PF in chronic pain has mainly been restricted to adult populations (e.g., 11,13,14). Recently, however, research has begun to focus on the role of psychological (in)flexibility and one of its subcomponents (i.e., pain acceptance) in children with chronic pain as well (15–17).

Parents play a crucial role in their child's pain-related functioning given the dependence of children upon their parents for care (18). Previous research predominantly focused on parents as a risk factor for child maladjustment (19–21). For instance, parental catastrophizing about their child's pain and protective parenting behaviors have been related to impaired child functioning (e.g., disability and depression (22,23)). Yet, parents may also function as a resilience factor, with

some recent studies showing positive associations between parental PF and child functioning (24,25). Earlier studies suggest a possible transfer of PF from parent to child. For example, McCracken et al. (24) found that parents' PF about the child's pain was related to the child's acceptance of the pain in a pediatric chronic pain sample. Further, research in a healthy child sample showed that this relation is mediated by certain parenting styles (i.e., high warmth, low control) (26).

Although a risk-resilience perspective has recently been forwarded in the context of pediatric pain (9), most studies to date have only focused on risk (e.g., catastrophizing) *or* resilience variables (e.g., PF, pain acceptance (15–17)) for child functioning. A risk-resilience perspective however (8,9) requires that both risk and resilience factors *as well as* their interaction in influencing pain-related functioning are investigated. A resilience factor may contribute to adaptive functioning, but may also be a buffer against the adverse impact of risk factors. The present study is one of the first to investigate the link between risk *and* resilience in the context of pediatric pain. Pain intensity was examined as a risk, and PF as a resilience factor for child outcomes (i.e., disability, psychosocial and emotional functioning). Specifically, we expected child PF and pain acceptance to be positively related with child outcomes, and to buffer against the negative effects of child pain intensity (as a risk factor). We also expected parental general as well as pain-specific PF to contribute positively to their child's functioning. Based on findings in previous studies (24,26), we expected this relationship to be mediated by the child's own (general) PF and pain acceptance. Finally, we explored whether general or pain-specific PF (in both parent and child) would exert more of an impact on the child's daily functioning.

Methods

Participants

Patients with juvenile idiopathic arthritis (JIA) between 8 and 18 years and (one of) their parents who attended the department of pediatric rheumatology of Ghent University hospital between January 2014 and August 2016, and who met eligibility criteria (i.e., currently experiencing pain and sufficient Dutch language skills) were asked to participate in this study. The final sample consisted of 59 children (36 females; $M_{\text{age}} = 13.76$; $SD = 2.67$) and 48 parents (33 mothers; $M_{\text{age}} = 43.78$; $SD = 4.8$) (see Table 1). The study was approved by an independent Commission for Medical Ethics related to the hospital (number of ethical approval: 2013/544), and was carried out according to the guidelines for Good Clinical Practice (ICH/GCP) and the declaration of Helsinki for the protection of people who participate in clinical studies. All families who participated in this study received a multi-media gift card (15 euro).

- Insert Table 1 about here –

Measures

Sociodemographic Measures. Information regarding child and parent age, ethnicity, child gender, the parent's relation to the child, family situation, and parental education was collected from parents at the beginning of the survey.

Child Pain Intensity and Disability. Child pain experiences during the past 6 months were measured via the Dutch version of the Graded Chronic Pain Scale (GCPS (27)), adapted for children (7). The adapted GCPS contains 3 items that assess current, worst, and average experienced pain intensity during the past 6 months, which are rated on a numerical rating scale

(NRS; range: 0 = *no pain*, 10 = *a lot of pain*). Pain disability was assessed via two items: one item assessing the total number of days the child was prevented from carrying out his/her usual activities during the past six months (i.e., disability days; 0-180) and one item assessing the degree to which pain interfered with those activities in that same period (i.e., ‘pain interference’; NRS, range: 0 = *no interference*; 10 = *unable to carry on any activities*). Scores for mean pain intensity and disability were calculated based on the recommended strategy described by Vervoort et al. (7) and Von Korff (27). A ‘mean pain intensity’ score was calculated by averaging the scores on the items assessing current, worst, and average pain intensity. A ‘disability’ score (0-6) was calculated based upon a combined score of points related to the number of disability days (0: < 7 days; 1: ≥ 7 and < 15 days; 2: ≥ 15 and < 31 days; 3: ≥ 31 days) and points related to the degree of pain interference (0: < 3; 1: ≥ 3 and < 5; 2: ≥ 5 and < 7; 3: ≥ 7). Higher values indicate greater disability. This score was treated as a continuous variable in all subsequent analyses. Based on the scores for pain intensity and disability children were classified into 5 pain grades which were used to describe the sample (7) (see Table 1). The original version of the GCPS has been shown to be a valid measure of pain severity in primary care, chronic pain, and the general population (28–30). The adapted child version that was used here has been validated in a large sample of children and adolescents in the general population (7). In the current study, Cronbach’s alpha was .85 for the mean pain intensity subscale and .76 for the disability subscale.

Child Functioning.

***Pediatric Quality of Life Inventory*TM (PedsQLTM 4.0).** The Generic Core version of the PedsQLTM (31) (Dutch version) is a 23-item self-report measure that assesses health-related quality of life (HRQOL) in children during the past 6 months. In this study a child version (8-12 years) and a teen version (13-18 years) were used. Children rated items on a scale ranging from 0

(*never a problem*) to 4 (*almost always a problem*) and scores were transformed to a 0-100 scale and reverse-scored so that higher scores reflect better health-related functioning. Four generic core scale scores can be calculated measuring physical functioning (8 items), emotional functioning (5 items), social functioning (5 items) and school functioning (5 items). In this study, the psychosocial health summary score was used, which was calculated by taking the mean of the items on the emotional, social, and school functioning scales. Higher scores indicate better psychosocial health-related functioning. The Dutch PedsQL™ 4.0 has been proven to be reliable and valid to assess pediatric health-related functioning in both healthy children and those with a chronic condition(32). In the current study, Cronbach's alpha's were .92 for the child version and .89 for the teen version of the psychosocial functioning subscale.

Positive and Negative Affect Scale for Children (PANAS-C). In this study the Dutch version of the 27-item PANAS-C(33) was used. This self-report measure consists of two subscales: one assessing positive (12 items; e.g. '*happy*') and the other assessing negative affect (15 items; e.g., '*sad*') as experienced by the child during the past two weeks. Items are rated on a scale ranging from 1 (*totally not*) to 5 (*a lot*). The 27-item PANAS-C has been demonstrated as a valid and reliable measure in clinical as well as non-clinical samples (33,34). In the current study, Cronbach's alpha was .88 for both the positive and negative affect scale.

Child Resilience Factors.

Avoidance and Fusion Questionnaire for Youth (AFQ-Y). The 17-item Dutch version of the AFQ-Y (35) was used to assess PF in children and adolescents. This questionnaire was completed by children aged 10 or older. The scale is designed to assess two core concepts that reflect psychological *inflexibility*: experiential avoidance (i.e., avoidance of negative or unwanted

experiences) and cognitive fusion (i.e., being entangled with the content of one's thoughts or feelings). Items are rated on a scale ranging from 0 (*not at all true*) to 4 (*very true*). For this study, items were reverse scored so that higher scores indicate a greater level of PF (for a similar strategy see Williams et al. (26)). The AFQ-Y shows good reliability and validity in children and adolescents (35,36). In the current study Cronbach's alpha was .87.

Chronic Pain Acceptance Questionnaire – Adolescent version (CPAQ-A). The Dutch CPAQ-A(37) is a 20-item self-report measure designed to assess acceptance of chronic pain in children/adolescents. This questionnaire was completed by children aged 10 or older. Items are rated on a scale ranging from 0 (*never true*) to 4 (*always true*). The overall acceptance score consists of two subscales: activity engagement (11 items) and pain willingness (9 items). 'Activity Engagement' items assess the extent to which children attempt to participate in daily life activities while they are in pain whereas 'Pain Willingness' items assess the extent to which the child rates the goal to reduce or control pain as being less important than other life goals. Higher total scores (0-80) indicate greater levels of (chronic) pain acceptance. The CPAQ-A has demonstrated good reliability and validity in samples of adolescents with chronic pain (15,37). In the current study, Cronbach's alpha for the total scale was .89.

Parental Resilience Factors.

Acceptance and Action Questionnaire (AAQ-II). The 10-item Dutch version of the AAQ-II (38,39) was used to assess general levels of PF in the parent. Items are rated on a scale ranging from 1 (*never true*) to 7 (*always true*). In this study, negatively worded items were reverse scored so that a higher total score (0-70) reflected greater acceptance and less avoidance of unwanted (private) experiences. The 10-item AAQ-II has proven reliable and valid across various clinical

and non-clinical adult samples (38). In the current study Cronbach's alpha for the 10-item AAQ-II scale was .82.

Parental Psychological Flexibility Questionnaire (PPFQ). The PPFQ (24,25) was developed to assess PF of parents in the context of their child's (chronic) pain. Items are rated on a scale ranging from 0 (*never true*) to 6 (*always true*). The PPFQ scale incorporates items in four subscales: parents' values-based action (5 items), emotional acceptance (5 items), pain acceptance (4 items), and pain willingness (3 items). Items of the latter three subscales are reverse-scored. Higher scores (0-102) indicate a greater level of parental PF in the context of their child's pain. Both the original 24-item (24) and the 17-item (25) version of the PPFQ demonstrate reliability and validity in samples of parents of children with chronic pain. For the current study a Dutch translation (following a back-translation procedure) of the 17-item version of the PPFQ was used and Cronbach's alpha was .91.

Procedure

This study was part of a larger survey-based study, the Multi-informant Pediatric Quality of Life Study (i.e., MiPedQoL), which included all children (8-18 years) who came to see their physician at the department of pediatrics of Ghent University hospital (2014-2016). Children diagnosed with JIA and (one of) their parents were approached and asked to complete an additional set of questionnaires for this specific study. Their pediatric rheumatologist first evaluated if they met eligibility criteria (i.e., age, currently in pain, and sufficient Dutch language skills) and referred the family to a research assistant if they showed interest. If they agreed to participate after receiving more information, informed consent was obtained at the hospital by both parent and child. Out of 119 children who were referred to us by their physician, 94 consented to participate

(78 %). Children younger than 12 were required to obtain the consent of their parent before taking part in the study whereas children older than 12 were asked to give their own consent. Surveys were completed via LimeSurvey 2.00, a web-based survey platform. We asked children younger than 12 to complete the first section of the survey on a tablet or computer at the hospital so that the research assistant could provide guidance and clarification if necessary. Thereafter they completed the survey at home. Children older than 12 were given a similar option or told that they could complete all questionnaires at home. Parents completed all surveys at home. A paper version of the survey was made available in case participants did not have access to a computer or internet (nobody availed of this option). If participants did not complete all measures within two weeks, a reminder e-mail was sent.

Statistical Analyses

Analyses were performed on a sample of 59 children and 48 parents. For 13 children both parents completed questionnaires; given this small number we decided to randomly select the data of one parent for each of these children. For 11 children no (sufficient) parent data were available.

Additional information on our data preparation strategy can be found in Figure 1.

- Insert Figure 1 about here –

All analyses were performed in SPSS (v.23; IBM Statistics). Cronbach's Alpha reliability statistics (see 'Methods') were used to examine internal consistency for all scales. Normality was checked through visual inspection of the QQ-plots of the residuals. These checks revealed that all outcome variables were normally distributed. Bivariate Pearson correlations between child predictors, parent variables, and child outcomes were carried out to test hypothesized correlational patterns and to detect potential variables to control for (e.g., child age, gender, or

mean pain intensity [in mediation analyses]). If a relation between these variables and child outcomes was significant at the 5% level, they were entered as control variables in subsequent analyses. First, a series of hierarchical multiple regression analyses was performed for each of the child outcomes (i.e., psychosocial health-related functioning, disability, positive affect, and negative affect) in order to investigate their association with pain intensity (risk factor) and PF and/or pain acceptance (resilience factors). Furthermore, interaction terms were created to examine whether child PF and/or pain acceptance moderated the association between the child's self-reported pain intensity and outcomes (i.e., risk-resilience association). All continuous independent and moderator variables were (grand-mean) centered. Separate analyses were performed for general PF and pain acceptance as moderators of the association between pain intensity and each outcome. Significant moderation effects were plotted to facilitate interpretation of the effects using methods by Dawson (40) and Holmbeck (41). Second, separate analyses were performed to examine whether general PF or pain acceptance in the child mediated the relationship between parental general or pain-related PF and child outcomes (see Figure 2), mediation analyses were carried out, using a bootstrapping resampling method described by Hayes(42) by means of the PROCESS plug-in in SPSS. Instead of inspecting *p*-values to conclude for significance of the effects, bootstrapping methods focus on confidence intervals and effect sizes. According to this analytic method, a significant indirect effect (ab) or mediation is observed if the 95% bootstrap confidence interval does not include 0. We examine bias-corrected bootstrap confidence intervals (BC CI) since they have been argued to be the most accurate with a high level of statistical power, especially with smaller samples sizes ($N < 80$) (see Mallinckrodt et al. (43) for more info). All analyses were executed at the 5% significance level.

- Insert Figure 2 about here -

Results

Pearson correlations between all child and parent variables of interest are presented in Table 2. Results indicated significant negative associations between child mean pain intensity and psychosocial health-related functioning ($r = -.46, p < .01$), and between mean pain intensity and positive affect ($r = -.40, p < .01$), as well as a positive association between pain intensity and pain-related disability ($r = .64, p < .01$). Child mean pain intensity was not related with negative affect ($r = .15, ns$). Consistent with our expectations, child ‘resilience’ factors (i.e., general PF and pain acceptance) were related with better scores on almost all outcome variables (see Table 2). That said, no significant association was found between pain acceptance and positive affect ($r = .15, ns$). Results also showed that parental general and pain-specific PF were not directly related to any of the child outcome variables. These correlational patterns, however, showed that general PF in parents did relate to general PF in their child ($r = .35, p < .05$) as well as child acceptance of pain ($r = .38, p < .05$). Likewise, parents’ PF in the context of their child’s pain was positively related to the child’s own pain acceptance ($r = .44, p < .01$), but not to the child’s general PF ($r = -.01, ns$).

- Insert Table 2 about here –

General Psychological Flexibility

Does Child General Psychological Flexibility Moderate the Relation Between Child Pain

Intensity and Child Outcomes? Hierarchical multiple regression analyses (see Table 3) revealed that the child’s mean pain intensity had a significant contribution in explaining children’s

psychosocial health-related functioning ($\beta = -.38, p < .01$), disability ($\beta = -.60, p < .001$), and positive affect ($\beta = -.38, p < .01$), but not negative affect ($\beta = .00, ns$). Furthermore, the child's level of general PF did significantly contribute to the explanation of psychosocial health-related functioning ($\beta = .34, p < .01$) but not of disability ($\beta = -.17, ns$). Moreover, the child's general PF had a significant contribution in explaining negative affect ($\beta = -.60, p < .01$), but did not contribute to the explanation of positive affect ($\beta = .14, ns$). Inspection of the interaction between child general PF and mean pain intensity showed that this interaction did not significantly contribute to the explanation of any of the outcomes. In other words, and in contrast with expectations, the level of general PF in the child did not moderate any of the above associations between pain intensity and child outcomes (see Table 3).

- Insert Table 3 about here -

Does Parental General Psychological Flexibility Contribute, through Child Psychological Flexibility, to Child Outcomes? Bootstrap analyses (with 5000 resamples and controlling for child pain intensity [all outcomes] and child age [only positive affect]) showed no significant *direct* contributions of parental general PF on any of the child outcomes ($c'_{psychosocial} = .00, p = .99$; $c'_{disability} = -.02, p = .47$; $c'_{positive} = -.29, p = .10$; $c'_{negative} = -.01, p = .97$). Although we found no direct contribution, we did find a significant *indirect* contribution of parent's general PF via child general PF in explaining the child's psychosocial health-related functioning ($ab_{psychosocial} = .25, SE = .15, 95\% BC CI_{psychosocial}: 0.04 \text{ to } 0.69$). Analyses revealed a similar significant indirect contribution of parental general PF via child general PF on the child's positive and negative affect ($ab_{positive} = .16, SE = .09, 95\% BC CI: 0.03 \text{ to } 0.41$; $ab_{negative} = -.27, SE = .14, 95\% BC CI: -0.62 \text{ to } -0.10$). However, we found no significant indirect link with the child's pain-related disability ($ab_{disability} = .00, SE = .00, 95\% BC CI: -0.02 \text{ to } 0.04$). According to

typology offered by Zhao et al.(44) the above-described significant indirect effects in combination with the absence of direct effects might lead us to conclude that these reflect indirect-only mediations of child's general PF in the relationship between parent's general flexibility and their child's psychosocial health-related functioning, positive and negative affect.

Pain-specific Psychological Flexibility

Does Child Pain Acceptance Moderate the Relation Between Child Pain Intensity and Child

Outcomes? Hierarchical multiple regression analyses (Table 4) indicated that child pain acceptance had a significant contribution in explaining psychosocial health-related functioning ($\beta = .31, p < .05$), disability ($\beta = -.45, p < .001$) and negative affect ($\beta = -.32, p < .05$), indicating better psychosocial functioning, and lower levels of disability and negative affect in children who reported higher levels of pain acceptance. Child pain acceptance did not contribute to the explanation of positive affect ($\beta = .02, ns$). Regression analyses examining the interaction between child pain acceptance and mean pain intensity on child outcomes showed that the child's acceptance of pain significantly moderated the positive association between pain intensity and pain-related disability, $\Delta R^2 = .05, F(1,52) = 6.89, p < .05$. To explore the nature of this moderation effect, regression lines were plotted for children with high (1 SD above the mean) versus low (1 SD below the mean) pain acceptance scores (see Figure 3). Significance tests with disability as an outcome showed that the simple slope was only significant for low pain acceptance ($\beta = .67, p < .0001$), suggesting that higher levels of pain were associated with higher levels of disability when pain acceptance was low. However, when pain acceptance was high the significant association between pain and disability disappeared ($\beta = .18, ns$), suggesting that high pain acceptance completely buffered for the negative effect of pain on disability. Additional

analyses (see also Figure 3) revealed that higher pain acceptance was related to lower levels of disability for children who reported low as well as for children who reported high levels of pain intensity. However, this association was stronger in children who experienced high levels of pain ($\beta = -.73, p < .0001$) as compared to children who experienced low levels of pain ($\beta = -.25, p < .05$). Finally, in contrast to our hypotheses, pain acceptance did not moderate the associations between mean pain intensity and psychosocial health-related functioning, positive affect, or negative affect.

- Insert Table 4 about here -

- Insert Figure 3 about here -

Does Parental Pain-specific Psychological Flexibility Contribute, through their Child's Acceptance of Pain, to Child Outcomes? Bootstrap analyses (with 5000 resamples and controlling for child pain intensity [all outcomes] and child age [positive affect]) showed no significant *direct* contributions of parent pain-specific PF on any of the child outcomes ($c'_{\text{psychosocial}} = .05, p = .75$; $c'_{\text{disability}} = -.02, p = .10$; $c'_{\text{positive}} = -.10, p = .32$; $c'_{\text{negative}} = .01, p = .91$). Yet, analyses did reveal that the *indirect* contribution of parent pain-specific PF through child pain acceptance helped explaining child's psychosocial health-related functioning ($ab_{\text{psychosocial}} = .22, SE = .10, 95\% \text{ BC CI: } 0.09 \text{ to } 0.46$). Analyses also revealed that parent pain-specific PF added significantly to the explanation of both pain-related disability and negative affect in an indirect way, via their child's pain acceptance ($ab_{\text{disability}} = -.02, SE = .01, 95\% \text{ BC CI: } -0.04 \text{ to } -0.01$; $ab_{\text{negative}} = -.08, SE = .04, 95\% \text{ BC CI: } -0.18 \text{ to } -0.02$). This was not the case for positive affect ($ab_{\text{positive}} = .03, SE = .03, 95\% \text{ BC CI: } -0.05 \text{ to } 0.14$). The significant indirect contributions again suggest an indirect-only mediation of child pain acceptance in the relation between parent pain-specific PF and the child's psychosocial health-related functioning,

disability, and negative affect.

Discussion

Chronic pain is known to impact daily functioning in children (3). Although most research has focused on identifying factors that increase the risk for maladjustment (4–6), risk-resilience models have directed attention towards resilience factors that enhance adaptive functioning in the face of chronic pain (8,9). In this study we investigated whether: (a) child and parental psychological flexibility (PF) are resilience factors that promote adaptive functioning in children with Juvenile Idiopathic Arthritis (JIA); (b) child PF and pain acceptance buffer against the negative influence of pain intensity (risk factor) on impaired functioning; (c) parental (general and pain-specific) PF represent an interpersonal source of resilience; and (d) general and pain-specific PF differ in their contribution to child functioning.

In this study higher levels of pain intensity were associated with poorer psychosocial functioning, higher levels of disability, and lower levels of positive affect, which is consistent with previous work showing that pain intensity is a risk factor for maladaptive functioning in children with JIA (17,45,46). That said, pain intensity was not related to (higher) negative affect. This discrepancy with previous research (47,48) could be due to the measures used: we employed the negative affect scale of the PANAS-C, which asks children to report their experience of some general negative emotions (e.g., ‘upset’ or ‘sad’) during the past two weeks, whereas previous studies typically assessed symptoms of emotional disorders (e.g., anxiety, depression) (17,47,48).

As hypothesized, we found that child general PF and pain acceptance were both related to better

psychosocial functioning and less negative affect, a finding that is consistent with previous literature on PF (15,17,37). Our results also indicated that child pain acceptance buffered against the adverse impact of pain intensity on disability. Children who showed high levels of pain acceptance reported less disability. This effect was strongest when pain intensity was high. This is in line with an adult study in which pain acceptance also buffered against the negative effects of pain on pain interference (49). In short, PF and pain acceptance both seem to represent resilience factors for child adaptive functioning and confirm why it is important to focus on risk *and* resilience factors to increase our knowledge on pain-related functioning in children (8). Notably, the current study is one of the first to show how pain acceptance not only contributes to adaptive functioning but also buffers the negative effects of risk factors on maladaptive child functioning.

Unlike previous studies (24,26), we did not observe a significant *direct* link between parental (general or pain-specific) PF and child functioning. However, we did find that a parent's general PF *indirectly* contributed to their child's psychosocial and emotional functioning via the child's own general PF. Parental pain-specific PF was also indirectly linked with child psychosocial functioning, negative affect, and disability, via the child's pain acceptance. Thus it seems that parents who show an open and accepting attitude towards their child's pain, and who promote engagement in valued activities despite associated pain, might promote PF in their child. Earlier studies in the general population (26) showed that the transfer of PF from parent to child could be mediated by parenting-specific psychological flexibility (50) or parenting style (i.e. high warmth, low control) (26). Parents may also promote PF in their child by overtly modeling openness to pain or other unwanted experiences (e.g., fear) and/or engagement in valued activities despite pain (24). Future work could test this idea and examine whether other social agents in the child's

environment (e.g., peers, teachers) might promote PF in the child as well.

Finally, we intended to explore (different) contributions of general versus pain-specific PF to child outcomes. First, small (child) to no (parent) relations were found between general and pain-specific PF. These types of flexibility in children were also related to outcomes in different ways. Notably, only pain acceptance was related to, and buffered for the impact of pain on disability. A possible explanation for this might be that pain acceptance takes the specific pain situation into account whereas general PF does not. JIA is characterized by pain in the joints which is known to mainly interfere with the performance of (physical) activities (1,45,46). Thus, it may be that a child who accepts this specific pain, even when pain intensity is high, will engage more in (physical) activities as compared to a child who has difficulties in accepting the pain. Future research could further examine different contributions of general versus pain-specific PF to pain-related functioning. If consistently shown, these differences could inform future interventions to target pain-specific and/or general PF depending on the type of chronic pain and domain of functioning mainly affected by pain.

Taking a step back, our findings are consistent with the theoretical idea that (pain-specific) PF can be characterized by (a) the ability to flexibly adapt or change behavior in the presence of painful or other negative experiences in order to serve long-term goals and values while (b) not rigidly behaving in the service of a single (pain-reduction) goal that undermines contact with other goals and values (51). This is true for children in pain as well as for their parents. Being able to focus on and engage in valued activities instead of persistently resisting any contact with pain may help the child to engage in and sustain a valued life. Our results showed that (a) child PF is indeed related to better psychosocial and emotional functioning and that (b) parents can

contribute to this by modelling or shaping PF in their child.

Limitations and Future Directions

The current study sets the stage for new research directions in pediatric chronic pain. For instance, although it is one of the first to adopt a risk-resilience perspective to chronic pediatric pain it is certainly not conclusive. As the risk-resilience literature grows (8–10,52,53) it will be important to explore other risk factors (e.g., pain-related fear (4,6)) as well. Doing so will expand our understanding of how psychosocial risk factors and PF interact in predicting children's pain-related functioning. Likewise, we showed that positive and negative affect were unrelated and are thus unique outcomes (54,55). This highlights the importance of including positive outcomes in research on pain-related functioning. Future research could include other positive outcomes such as long-term psychological well-being or perceptions of a fulfilling life (8).

We employed a cross-sectional design and therefore cannot draw any causal conclusions. Future studies should adopt longitudinal or experimental designs to determine the direction of the effects. Given that PF involves actions, feelings, and thoughts that interact with each other within a specific (interpersonal) context (51,56), future research could adopt a (functional) study design to accommodate such factors. For instance, diary methodology may be useful to explore PF in relation to daily variations in pain and contexts.

It might also be that the current study did not completely capture pain-specific PF in children, because there is no well-validated child instrument to do so. We used the child version of the Chronic Pain Acceptance Questionnaire(57), which measures only one subcomponent of PF. Future research should aim at constructing such a measure. Furthermore, our sample consisted of children with chronic pain related to JIA. Before any general conclusions about the role of child

and parental PF can be made, our hypotheses should also be tested in children with other chronic pain types (e.g., not disease-related).

Finally, it should be noted that our findings speak to the potential of interventions that focus on fostering adaptive functioning in children with chronic pain by enhancing PF and acceptance, such as Acceptance and Commitment Therapy(58–62). Our results additionally support the importance of including parents in (ACT-based) child treatment programs (63–65). Targeting PF in parents might help to boost PF and, as a result, adaptive functioning in the child.

Conclusion

The current study demonstrates that child PF and pain acceptance represent resilience factors that foster adaptive psychosocial and emotional functioning in children with JIA. Pain acceptance additionally buffered against the negative influence of pain on disability. Several indirect associations between parental PF and child functioning (via the child's own PF) emerged, suggesting that parents may be a source of resilience for the child as well. Our findings further highlight the importance of discriminating between general and pain-specific PF. Future (longitudinal) research should examine both risk and resilience factors in relation to (the absence of) negative as well as positive outcomes. Doing so could contribute to the improvement of pain treatment programs, highlighting the importance of enhancing resilience factors in both children and parents.

Acknowledgements

We thank Sara Van der Mispel and Anja Kloosterman for their help with data collection.

References

1. Weiss JE, Ilowite NT. Juvenile Idiopathic Arthritis. *Rheum Dis Clin North Am.* 2007;33(3):441–70.
2. Gauntlett-Gilbert J, Eccleston C. Disability in adolescents with chronic pain: Patterns and predictors across different domains of functioning. *Pain.* 2007;131(1–2):132–41.
3. Palermo TM. Impact of recurrent and chronic pain on child and family daily functioning: a critical review of the literature. *J Dev Behav Pediatr.* 2000;21(1):58–69.
4. Asmundson GJG, Noel M, Petter M, Parkerson HA. Pediatric fear-avoidance model of chronic pain: foundation, application and future directions. *Pain Res Manag.* 2012;17(6):397–405.
5. Simons LE, Smith A, Ibagon C, Coakley R, Logan DE, Schechter N, et al. Pediatric Pain Screening Tool: Rapid identification of risk in youth with pain complaints. *Pain.* 2015;156(8):1511–8.
6. Simons LE, Kaczynski KJ. The Fear Avoidance model of chronic pain: examination for pediatric application. *J Pain.* 2012;13(9):827–35.
7. Vervoort T, Logan D, Goubert L, De Clercq B, Hublet A. Severity of pediatric pain in relation to school-related functioning and teacher support: An epidemiological study among school-aged children and adolescents. *Pain.* 2014;155(6):1118–27.
8. Goubert L, Trompetter H. Towards a science and practice of resilience in the face of pain. *Eur J Pain (United Kingdom).* 2017;21(8):1301–15.

9. Cousins LA, Kalapurakkal S, Cohen LL, Simons LE. Topical review: Resilience resources and mechanisms in pediatric chronic pain. *J Pediatr Psychol.* 2015;40(9):840–5.
10. Sturgeon JA, Zautra AJ. Resilience: A New Paradigm for Adaptation to Chronic Pain. *Curr Pain Headache Rep.* 2010;14(2):105–12.
11. McCracken LM, Morley S. The psychological flexibility model: a basis for integration and progress in psychological approaches to chronic pain management. *J Pain.* 2014;15(3):221–34.
12. Scott W, McCracken LM, Norton S. A Confirmatory Factor Analysis of Facets of Psychological Flexibility in a Sample of People Seeking Treatment for Chronic Pain. *Ann Behav Med.* 2016;50(2):285–96.
13. Vowles KE, McCracken LM. Comparing the role of psychological flexibility and traditional pain management coping strategies in chronic pain treatment outcomes. *Behav Res Ther.* 2010;48(2):141–6.
14. Scott W, Hann KEJ, McCracken LM. A Comprehensive Examination of Changes in Psychological Flexibility Following Acceptance and Commitment Therapy for Chronic Pain. *J Contemp Psychother.* 2016;46(3):139–48.
15. Wallace DP, Harbeck-Weber C, Whiteside SPH, Harrison TE. Adolescent Acceptance of Pain: Confirmatory Factor Analysis and Further Validation of the Chronic Pain Acceptance Questionnaire, Adolescent Version. *J Pain.* 2011;12(5):591–9.
16. Weiss KE, Hahn A, Wallace DP, Biggs B, Bruce BK, Harrison TE. Acceptance of pain: Associations with depression, catastrophizing, and functional disability among children and adolescents in an interdisciplinary chronic pain rehabilitation program. *J Pediatr*

- Psychol. 2013;38(7):756–65.
17. Feinstein AB, Forman EM, Masuda A, Cohen LL, Herbert JD, Nandini Moorthy L, et al. Pain Intensity, Psychological Inflexibility, and Acceptance of Pain as Predictors of Functioning in Adolescents with Juvenile Idiopathic Arthritis: A Preliminary Investigation. *J Clin Psychol Med Settings*. 2011;18(3):291–8.
 18. Palermo TM, Eccleston C. Parents of children and adolescents with chronic pain. *Pain*. 2009;146(1):15–7.
 19. Chow ET, Otis JD, Simons LE. The Longitudinal Impact of Parent Distress and Behavior on Functional Outcomes Among Youth With Chronic Pain. *J Pain*. 2016;17(6):729–38.
 20. Goubert L, Simons L. Cognitive styles and processes in paediatric pain. In: McGrath P, Stevens B, Walker S, Zempsky W, editors. *Oxford textbook of paediatric pain*. New York: Oxford University Press; 2013. p. 95–101.
 21. Logan DE, Scharff L. Relationships Between Family and Parent Characteristics and Functional Abilities in Children with Recurrent Pain Syndromes: An Investigation of Moderating Effects on the Pathway from Pain to Disability. *J Pediatr Psychol*. 2005;30(8):698–707.
 22. Caes L, Vervoort T, Eccleston C, Vandenhende M, Goubert L. Parental catastrophizing about child's pain and its relationship with activity restriction: the mediating role of parental distress. *Pain*. 2011;152(1):212–22.
 23. Caes L, Vervoort T, Eccleston C, Goubert L. Parents who catastrophize about their child's pain prioritize attempts to control pain. *Pain*. 2012;153(8):1695–701.

24. McCracken LM, Gauntlett-Gilbert J. Role of psychological flexibility in parents of adolescents with chronic pain: Development of a measure and preliminary correlation analyses. *Pain*. 2011;152(4):780–5.
25. Wallace DP, Mccracken LM, Weiss KE, Harbeck-Weber C. The Role of Parent Psychological Flexibility in Relation to Adolescent Chronic Pain: Further Instrument Development. *J Pain*. 2015;16:235–46.
26. Williams KE, Ciarrochi J, Heaven PCL. Inflexible Parents, Inflexible Kids: A 6-Year Longitudinal Study of Parenting Style and the Development of Psychological Flexibility in Adolescents. *J Youth Adolesc*. 2012;41(8):1053–66.
27. Von Korff M, Ormel J, Keefe FJ, Dworkin SF. Grading the severity of chronic pain. *Pain*. 1992;50(2):133–49.
28. Elliott AM, Smith BH, Smith CW, Chambers AW. Changes in chronic pain severity over time: the Chronic Pain Grade as a valid measure. *Pain*. 2000;88(3):303–8.
29. Smith BH, Penny KI, Purves AM, Munro C, Wilson B, Grimshaw J, et al. The Chronic Pain Grade questionnaire: validation and reliability in postal research. *Pain*. 1997;71(2):141–7.
30. Von Korff M, Deyo RA, Cherkin D, Barlow W. Back pain in primary care. Outcomes at 1 year. Vol. 18, *Spine*. 1993. p. 855–62.
31. Varni J, Seid M, Kurtin P. PedsQL 4.0: reliability and validity of the Pediatric Quality of Life Inventory version 4.0 generic core scales in healthy and patient populations. *Med Care*. 2001;39(8):800–12.

32. Engelen V, Haentjens MM, Detmar SB, Koopman HM, Grootenhuis MA. Health related quality of life of Dutch children: psychometric properties of the PedsQL in the Netherlands. *BMC Pediatr.* 2009;9:68.
33. Laurent J, Catanzaro JS, Joiner, T. E. J, Rudolph KD, Potter KI, Lambert S, et al. A Measure of Positive and Negative Affect for Children: Scale Development and Preliminary Validation. *Psychol Assess.* 1999;11(3):326–38.
34. Hughes AA, Kendall PC. Psychometric Properties of the Positive and Negative Affect Scale for Children (PANAS-C) in Children with Anxiety Disorders. *Child Psychiatry Hum Dev.* 2009;40(3):343–52.
35. Greco L, Lambert W, Baer R. Psychological inflexibility in childhood and adolescence: Development and evaluation of the Avoidance and Fusion Questionnaire for Youth. *Psychol Assess.* 2008;20(2):93–102.
36. Livheim F, Tengström A, Bond FW, Andersson G, Dahl J, Rosendahl I. Psychometric properties of the Avoidance and Fusion Questionnaire for Youth: A psychological measure of psychological inflexibility in youth. *J Context Behav Sci.* 2016;5(2):103–10.
37. McCracken LM, Gauntlett-Gilbert J, Eccleston C. Acceptance of pain in adolescents with chronic pain: Validation of an adapted assessment instrument and preliminary correlation analyses. *Eur J Pain.* 2010;14(3):316–20.
38. Bond FW, Hayes SC, Baer R a., Carpenter KM, Guenole N, Orcutt HK, et al. Preliminary Psychometric Properties of the Acceptance and Action Questionnaire–II: A Revised Measure of Psychological Inflexibility and Experiential Avoidance. *Behav Ther.* 2011;42(4):676–88.

39. Jacobs N, Kleen M, De Groot F, A-Tjak J. The measurement of experiential avoidance. [The Dutch language version of the Acceptance and Action Questionnaire-II (AAQ-II)]. *Gedragstherapie*. 2008;41(4):349–346.
40. Dawson JF. Moderation in Management Research: What, Why, When, and How. *J Bus Psychol*. 2014;29(1):1–19.
41. Holmbeck GN. Post-hoc probing of significant moderational and mediational effects in studies of pediatric populations. *J Pediatr Psychol*. 2002;27(1):87–96.
42. Hayes AF. Introduction to mediation, moderation, and conditional process analysis : a regression-based approach. The Guilford Press; 2013.
43. Mallinckrodt B, Abraham WT, Wei M, Russell DW, Frazier PA, Tix AP, et al. Advances in Testing the Statistical Significance of Mediation Effects. *J Couns Psychol*. 2006;53(3):372–8.
44. Zhao X, Lynch Jr. JG, Chen Q. Reconsidering Baron and Kenny: Myths and Truths about Mediation Analysis. *J Consum Res Inc*. 2010;37(2):197–206.
45. Anthony KK, Schanberg LE. Pain in children with arthritis: A review of the current literature. *Arthritis Rheum*. 2003;49(2):272–9.
46. Varni J, Wicox K, Hanson V, Brik R. Chronic musculoskeletal pain and functional status in juvenile rheumatoid arthritis: an empirical model. *Pain*. 1988;32(1):1–7.
47. Schanberg LE, Anthony KK, Gil KM, Maurin EC. Daily pain and symptoms in children with polyarticular arthritis. *Arthritis Rheum*. 2003;48(5):1390–7.
48. Margetic B, Aukst-Margetic B, Bilic E, Jelusic M, Tambic Bukovac L. Depression,

- anxiety and pain in children with juvenile idiopathic arthritis (JIA). *Eur Psychiatry*. 2005;20(3):274–6.
49. Kratz AL, Ehde DM, Bombardier CH, Kalpakjian CZ, Hanks RA. Pain acceptance decouples the momentary associations between pain, pain interference, and physical activity in the daily lives of people with chronic pain and spinal cord injury. *J Pain*. 2017;18(3):319–31.
 50. Brassell AA, Rosenberg E, Parent J, Rough JN, Fondacaro K, Seehuus M. Parent's psychological flexibility: Associations with parenting and child psychosocial well-being. *J Context Behav Sci*. 2016;5(2):111–20.
 51. Hayes SC, Luoma JB, Bond FW, Masuda A, Lillis J. Acceptance and commitment therapy: model, processes and outcomes. *Behav Res Ther*. 2006;44(1):1–25.
 52. Cousins LA, Cohen LL, Venable C. Risk and resilience in pediatric chronic pain: Exploring the protective role of optimism. *J Pediatr Psychol*. 2015;40(9):934–42.
 53. Sturgeon JA, Zautra AJ. Psychological resilience, pain catastrophizing, and positive emotions: Perspectives on comprehensive modeling of individual pain adaptation topical collection on psychiatric management of pain. *Curr Pain Headache Rep*. 2013;17(3).
 54. Kratz AL, Mary MA, Davis C, Zautra AJ. Pain Acceptance Moderates the Relation Between Pain and Negative Affect in Female Osteoarthritis and Fibromyalgia Patients. *Ann Behav Med*. 2007;33(3):291–301.
 55. Diener E, Emmons RA. The independence of positive and negative affect. *J Pers Soc Psychol*. 1984;47(5):1105–17.

56. Hayes SC, Levin ME, Plumb-Villardaga J, Villatte JL, Pistorello J. Acceptance and commitment therapy and contextual behavioral science: examining the progress of a distinctive model of behavioral and cognitive therapy. *Behav Ther.* 2013;44(2):180–98.
57. McCracken LM, Vowles KE, Eccleston C. Acceptance of chronic pain: component analysis and a revised assessment method. *Pain.* 2004;107(1–2):159–66.
58. Coyne LW, McHugh L, Martinez ER. Acceptance and Commitment Therapy (ACT): Advances and applications with children, adolescents, and families. *Child Adolesc Psychiatr Clin N Am.* 2011;20(2):379–99.
59. Pielech M, Vowles K, Wicksell R. Acceptance and Commitment Therapy for Pediatric Chronic Pain: Theory and Application. *Children.* 2017;4(2):10.
60. Wicksell RK, Dahl J, Magnusson B, Olsson GL. Using acceptance and commitment therapy in the rehabilitation of an adolescent female with chronic pain: A case example. *Cogn Behav Pract.* 2005;12(4):415–23.
61. Wicksell RK. The role and function of acceptance and commitment therapy and behavioral flexibility in pain management. *Pain Manag.* 2015;46(editorial):319–22.
62. Wicksell RK, Melin L, Lekander M, Olsson GL. Evaluating the effectiveness of exposure and acceptance strategies to improve functioning and quality of life in longstanding pediatric pain – A randomized controlled trial. *Pain.* 2009;141(3):248–57.
63. Kanstrup M, Wicksell R, Kemani M, Wiwe Lipsker C, Lekander M, Holmström L. A Clinical Pilot Study of Individual and Group Treatment for Adolescents with Chronic Pain and Their Parents: Effects of Acceptance and Commitment Therapy on Functioning. *Children.* 2016;3(4):30.

64. Martin S, Wolters PL, Toledo-Tamula MA, Schmitt SN, Baldwin A, Starosta A, et al. Acceptance and commitment therapy in youth with neurofibromatosis type 1 (NF1) and chronic pain and their parents: A pilot study of feasibility and preliminary efficacy. *Am J Med Genet Part A*. 2016;170(6):1462–70.
65. Wallace DP, Woodford B, Connelly M. Promoting psychological flexibility in parents of adolescents with chronic pain: Pilot study of an 8-week group intervention. *Clin Pract Pediatr Psychol*. 2016;4(4):405–16.

Table 1. Sample descriptive characteristics. N, means (M), standard deviations (SD) and range.

	N (%)	M (SD)	Range
Child characteristics			
Age (in years)	59	13.76 (2.67)	8-18
Boys	23 (39%)		
<i>Educational level</i>			
Primary school	13 (22%)		
Secondary school	31 (52.5%)		
Higher education	1 (1.7 %)		
Missing	14 (23.7 %)		
<i>Pain grades</i>			
Grade I	20 (33.9 %)		
Grade II	18 (30.5%)		
Grade III	9 (15.3 %)		
Grade IV	12 (20.3 %)		
Parent characteristics			
Age (in years)	45 ^a	43.78 (4.78)	32-54
Mothers	33 (68,8%)		
<i>Marital Status</i>			
Married or living together	41 (85,4%)		
Divorced	3 (6,3 %)		
Single parent or not married	1 (2,1 %)		
Missing	3 (6,3 %)		

Notes. Grade 0 = no pain; Grade I = low intensity and low disability; Grade II = high intensity and low disability; Grade III = moderate disability, regardless of pain intensity; Grade IV = high disability, regardless of pain intensity. ^a no data available on age of 3 parents (6%).

Table 2. N, Means (M), standard deviations (SD), and Pearson correlations of child age, gender, pain intensity, general psychological flexibility and pain acceptance, parent general and pain-specific psychological flexibility, and child outcomes.

	N	M (SD)	2	3	4	5	6	7	8	9	10	11
Child Demographics												
1. Child age	59	13.76 (2.67)	.14	.05	-.18	.08	.15	-.11	-.12	.14	-.36**	.07
2. Child gender	59	n/a	--	.04	-.10	-.05	.09	.02	-.02	-.05	.09	.10
Child variables												
3. Pain intensity	59	5.20 (2.13)	--	-.25	-.42**	-.11	.18	-.46**	.64**	-.40**	.15	
4. Psychological flexibility (general)	56	46.43 (11.51)		--	.35**	.35*	-.01	.43**	-.32*	.29*	-.60**	
5. Pain acceptance	56	47.93 (12.88)			--	.38*	.44**	.45**	-.64**	.14	-.33*	
Parent variables												
6. Psychological flexibility (general)	43	54.09 (7.39)				--	.27	.19	-.18	-.07	-.24	
7. Psychological flexibility (pain-specific)	44	57.68 (16.40)					--	.19	-.21	-.13	-.08	
Child Outcomes												
8. Psychosocial functioning	52	73.17 (16.63)						--	-.62***	.48**	-.45**	
9. Disability	59	2.22 (1.98)							--	-.47**	.13	
10. Positive affect	52	40.69 (9.14)								--	-.08	
11. Negative affect	52	28.90 (9.10)									--	

Note. * $p < .05$. ** $p \leq .01$. *** $p \leq .001$

Table 3. Results of hierarchical multiple regression analyses examining the moderating role of child general psychological flexibility in the relationship between child pain intensity and child outcomes.

Criterion variable	Block	Predictor	β	ΔR^2	Total R^2	N
<i>Psychosocial functioning</i>	1	Pain intensity	-.38**			
		Psychological flexibility	.34**	.32***		
	2	Pain x Psychological flexibility	-.02	.02	.32***	50
<i>Disability</i>	1	Pain intensity	.60***			
		Psychological flexibility	-.17	.44***		
	2	Pain x Psychological flexibility	.01	.00	.44***	56
<i>Positive affect</i>	1	Age (control)	-.34*	.12*		
	2	Pain intensity	-.38**			
		Psychological flexibility	.14	.19**		
	3	Pain x Psychological flexibility	.23	.05	.36***	50
<i>Negative affect</i>	1	Pain intensity	.00			
		Psychological Flexibility	-.60***	.37***		
	2	Pain x Psychological Flexibility	.06	.00	.37***	50

Notes. * $p < .05$. ** $p \leq .01$. *** $p \leq .001$ Control variables were entered in block 1 (if applicable), predictors in block 2 and interaction terms in block 3. Standardized regression coefficients (β) of the last step in analyses are shown. N = number of children included in analyses.

Table 4. Results of hierarchical multiple regression analyses to examine the moderating role of child pain acceptance in relation between child pain intensity and child outcomes.

Criterion variable	Block	Predictor	β	ΔR^2	Total R^2	N
<i>Psychosocial functioning</i>	1	Pain intensity	-.34*			
		Pain acceptance	.31*	.30***		
	2	Pain x Pain acceptance	.02	.00	.30**	50
<i>Disability</i>	1	Pain intensity	.46***			
		Pain acceptance	-.45***	.58***		
	2	Pain x Pain acceptance	-.23*	.05*	.63***	56
<i>Positive Affect</i>	1	Age (control)	-.34*	.12*		
	2	Pain intensity	-.41**			
		Pain acceptance	.02	.17**		
	3	Pain x Pain acceptance	.10	.01	.30**	50
<i>Negative Affect</i>	1	Pain intensity	.02			
		Pain acceptance	-.32*	.11		
	2	Pain x Pain acceptance	.27	.06	.17*	50

Notes. * $p < .05$. ** $p \leq .01$. *** $p \leq .001$ Control variables were entered in block 1 (if applicable), predictors in block 2 and interaction terms in block 3. Standardized regression coefficients (β) of the last step in analyses are shown. N = number of children included in analyses.

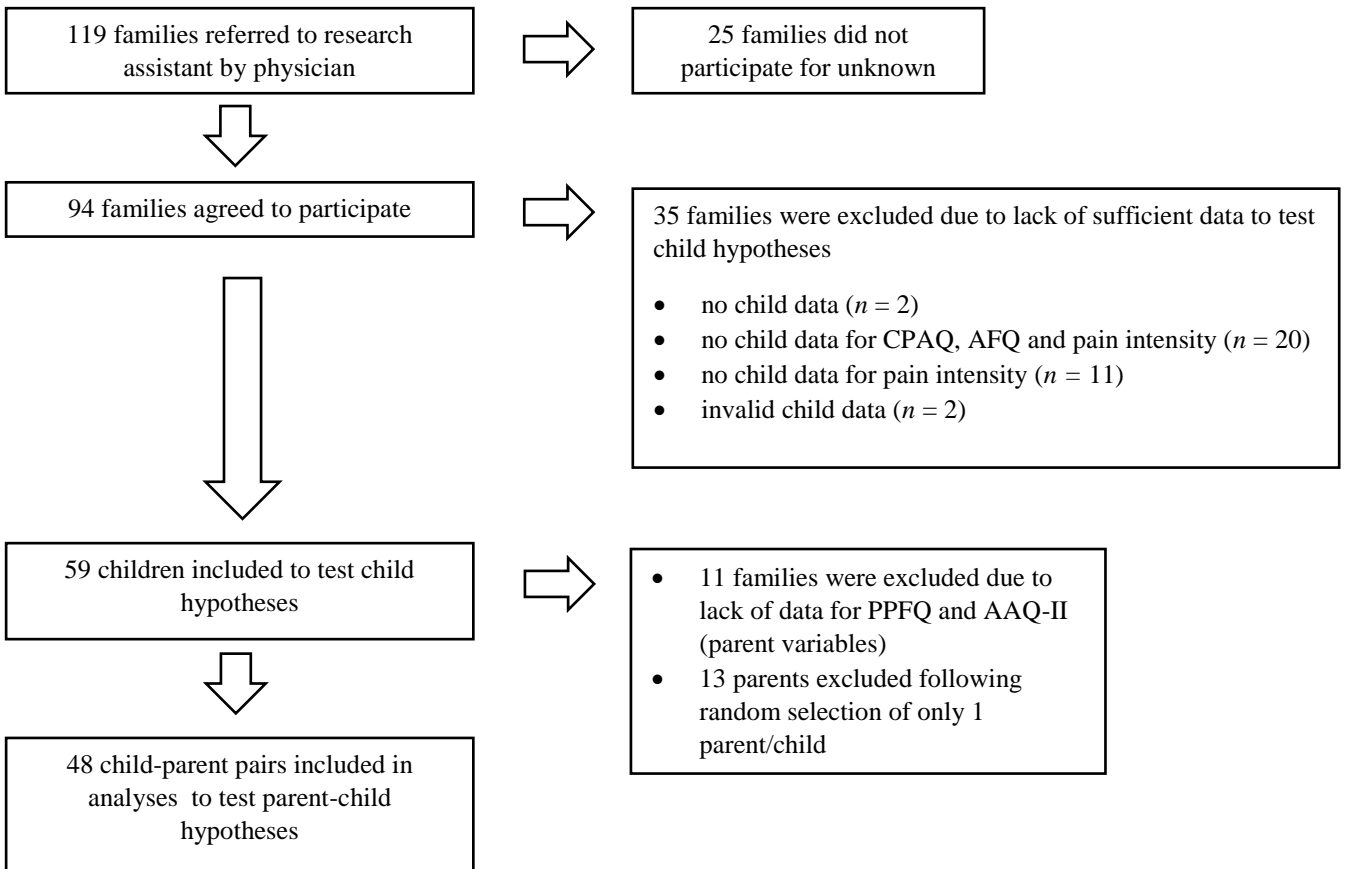
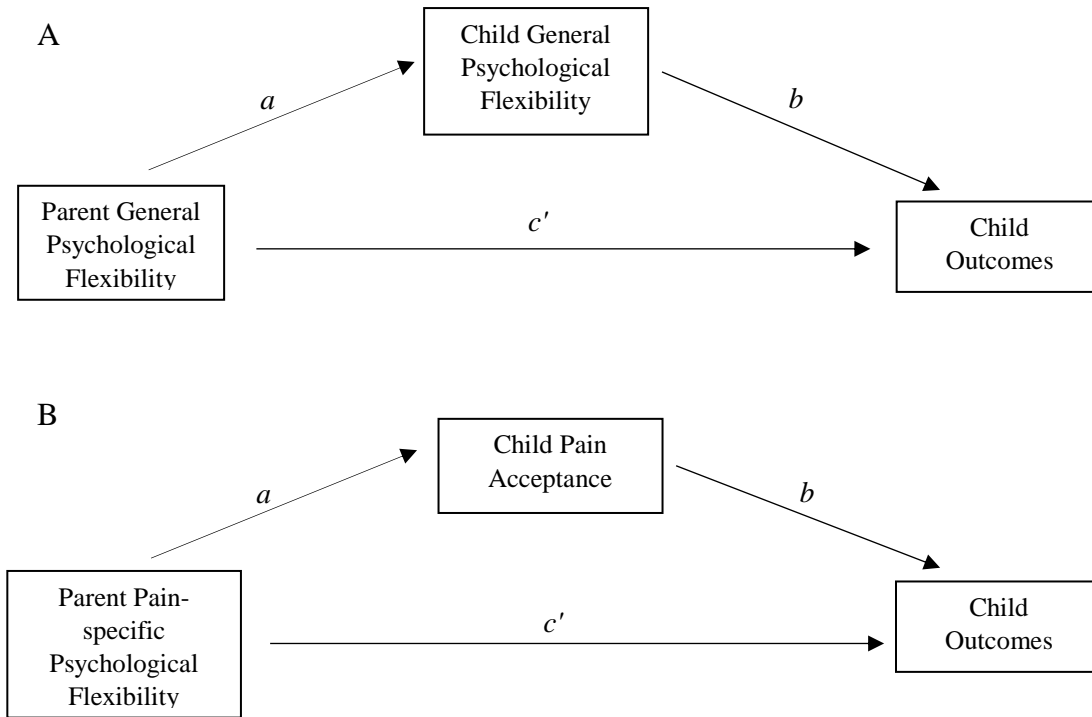
Figure 1. Flow chart of recruitment and data cleaning.

Figure 2. Three-variable mediation models that were examined for each of the child outcomes.

Note₁. **A:** Mediation model with child general psychological flexibility as a mediator between parent general psychological flexibility and child outcomes. **B:** Mediation model with child pain acceptance as a mediator between parental pain-specific psychological flexibility and child outcomes.

Note₂. These relations were tested using a bootstrapping method. This a modern approach to test statistical mediation which evolved as a response to several critiques towards the frequently used ‘normal theory’ or ‘causal steps’ methods based on a method by Baron and Kenny(63). One of the most common critiques is that this type of approach lacks power and has the risk for inflation of type I errors(64). A bootstrap approach focuses only on the indirect effect ($a \times b$), which is assumed to be the most relevant to conclude if mediation has occurred. There is no need of a direct effect (c') to indicate mediation. If there is a mediated effect ($a \times b$) in absence of a direct effect, this is classified as an ‘indirect-only’ mediation (typology offered by Zhao et al.(41)).

Figure 3. Regression lines for the associations between child pain intensity and disability as moderated by child pain acceptance. β = standardized regression coefficients (i.e., simple slopes); SD = standard deviation.

