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.

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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_{age} = 13.76; SD = 2.67) and 48 parents (33 mothers; M_{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™ **(PedsQL™ 4.0).** The Generic Core version of the

PedsQL™ (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 *in*flexibility: 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 (β = -.38, p < .01), disability (β = -.60, p < .001), and

positive affect (β = -.38, p < .01), but not negative affect (β = .00, ns). Furthermore, the child's

level of general PF did significantly contribute to the explanation of psychosocial health-related

functioning (β = .34, p < .01) but not of disability (β = -.17, ns). Moreover, the child's general PF

had a significant contribution in explaining negative affect (β = -.60, p < .01), but did not

contribute to the explanation of positive affect (β = .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 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 to 0.41; $ab_{negative}$ = -.27, SE =

.14, 95% BC CI: -0.62 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 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 (β = .31, $p < .05$), disability (β = -.45, $p < .001$) and negative affect (β = -.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 (β = .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 (β = .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 (β = .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

(β = -.73, p < .0001) as compared to children who experienced low levels of pain (β = -.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'

$_{psychosocial}$ = .05, p = .75; c' $_{disability}$ = -.02, p = .10; c' $_{positive}$ = -.10, p = .32; c' $_{negative}$ = .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*

$_{psychosocial}$ = .22, *SE* = .10, 95% BC CI: 0.09 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* $_{disability}$ = -.02, *SE* = .01, 95% BC

CI: -0.04 to -0.01; *ab* $_{negative}$= -.08, *SE* = .04, 95% BC CI: -0.18 to -0.02). This was not the case

for positive affect (*ab* $_{positive}$ = .03, *SE* = .03, 95% BC CI: -0.05 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 live. 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.

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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%)		
Educational level			
Primary school	13 (22%)		
Secondary school	31 (52.5%)		
Higher education	1 (1.7 %)		
Missing	14 (23.7 %)		
Pain grades			
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%)		
Marital Status			
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
Psychosocial functioning	1	Pain intensity	-.38**			
		Psychological flexibility	.34**	.32***		
	2	Pain x Psychological flexibility	-.02	.02	.32***	50
Disability	1	Pain intensity	.60***			
		Psychological flexibility	-.17	.44***		
	2	Pain x Psychological flexibility	.01	.00	.44***	56
Positive affect	1	Age (control)	-.34*	.12*		
	2	Pain intensity	-.38**			
		Psychological flexibility	.14	.19**		
	3	Pain x Psychological flexibility	.23	.05	.36***	50
Negative affect	1	Pain intensity	.00			
		Psychological Flexibility	-.60***	.37***		
	2	Pain x Psychological Flexibility	.06	.00	.37***	50

Notes. * $p < .05$. ** $p \le .01$. *** $p \le .001$ 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
Psychosocial functioning	1	Pain intensity	-.34*			
		Pain acceptance	.31*	.30***		
	2	Pain x Pain acceptance	.02	.00	.30**	50
Disability	1	Pain intensity	.46***			
		Pain acceptance	-.45***	.58***		
	2	Pain x Pain acceptance	-.23*	.05*	.63***	56
Positive Affect	1	Age (control)	-.34*	.12*		
	2	Pain intensity	-.41**			
		Pain acceptance	.02	.17**		
	3	Pain x Pain acceptance	.10	.01	.30**	50
Negative Affect	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.
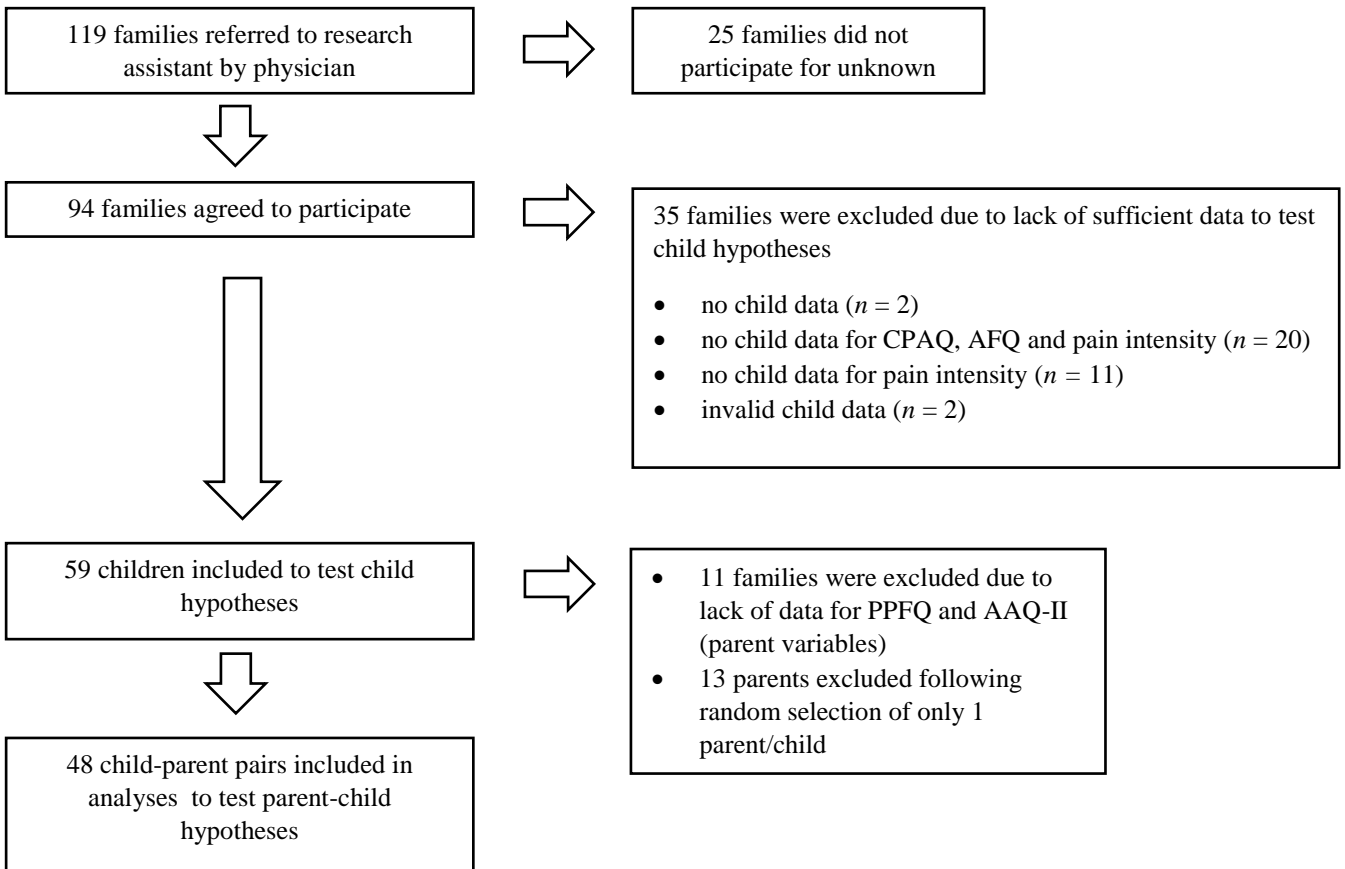
Figure 2. Three-variable mediation models that were examined for each of the child outcomes.
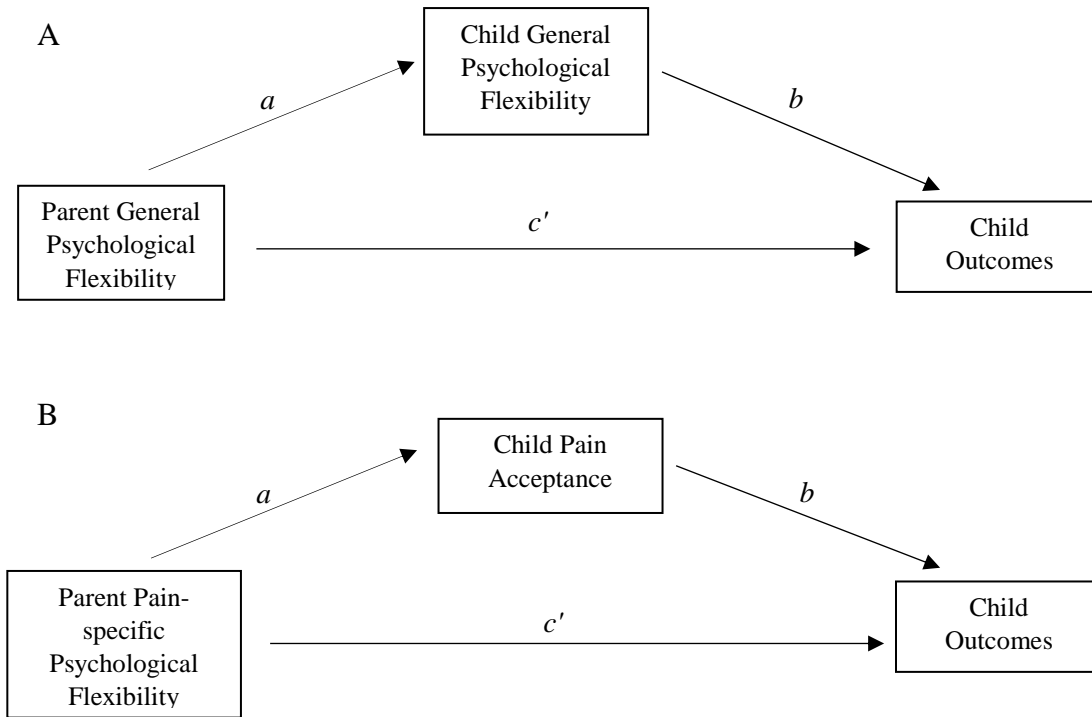
Note₁. **A**: Mediation model with child general psychological flexibility as a mediator between parent general psychological flexibility and child outcomes. **B**: Mediation model with child pain acceptance as a mediator between parental pain-specific psychological flexibility and child outcomes.

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 x 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 x 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.