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PSYCHOLOGY, PSYCHIATRY, IMAGING & BRAIN NEUROSCIENCE SECTION

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 vs pain-specific PF in contributing to child outcomes. **Methods.** Children with JIA (age eight to 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 59 children and 48 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 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. **Conclusions.** Our findings indicate that child and parental PF are resilience factors and that pain acceptance buffers the negative impact of pain intensity. Implications for psychosocial interventions that target (pain-specific) PF in children and parents are discussed.

Key Words: 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 toward, 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 [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 has 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 and Gauntlett-Gilbert [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 buffer 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 the negative effects of child pain intensity (as a risk factor). We also expected parental general and 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 between eight and 18 years old 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, $M_{\text{age}} = 13.76$ years, $SD = 2.67$ year) and 48 parents (33 mothers, $M_{\text{age}} = 43.78$ years, $SD = 4.8$ years) (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 and the Declaration of Helsinki for the protection of people who participate in clinical studies. All families who participated in this study received a multimedia gift card (15 euros).

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 six months were measured via the Dutch version of the Graded Chronic Pain Scale (GCPS [27]), adapted for children [7]. The adapted GCPS contains three items that assess current, worst, and average experienced pain intensity during the past six months, rated on a numerical rating scale (NRS; range = 0 = no pain to 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 to 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 et al. [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

Table 1. Sample descriptive characteristics

	N (%)	M (SD)	Range
Child characteristics			
Age, y	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			
I	20 (33.9)		
II	18 (30.5)		
III	9 (15.3)		
IV	12 (20.3)		
Parent characteristics			
Age, y	45*	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)		

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.

*No data available on age of three parents (6%).

into five pain grades, which were used to describe the sample (Table 1) [7]. 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, the Cronbach's alphas were 0.85 for the mean pain intensity subscale and 0.76 for the disability subscale.

Child Functioning

Pediatric Quality of Life Inventory

The Generic Core version of the Pediatric Quality of Life Inventory (PedsQL; Dutch version) [31] is a 23-item self-report measure that assesses health-related quality of life (HRQOL) in children during the past six months. In this study, a child version (age eight to 12 years) and a teen version (age 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, with higher scores reflecting better health-related functioning. Four generic core scale scores can be calculated measuring physical functioning (eight items), emotional functioning (five items), social functioning (five items), and school functioning (five 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, the Cronbach's alphas were 0.92 for the child version and 0.89 for the teen version of the psychosocial functioning subscale.

Positive and Negative Affect Scale for Children

In this study, the Dutch version of the 27-item Positive and Negative Affect Scale for Children (PANAS-C) [33] was used. This self-report measure consists of two subscales: one assessing positive affect (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 were 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 and nonclinical samples [33,34]. In the current study, Cronbach's alpha was 0.88 for both the positive and negative affect scales.

Child Resilience Factors

Avoidance and Fusion Questionnaire for Youth

The 17-item Dutch version of the Avoidance and Fusion Questionnaire for Youth (AFQ-Y) [35] was used to assess PF in children and adolescents. This questionnaire was completed by children age 10 years 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 were rated on a scale ranging from 0 (not at all true) to 4 (very true). For this study, items were reverse-scored, with higher scores indicating 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 0.87.

Chronic Pain Acceptance Questionnaire–Adolescent version

The Dutch Chronic Pain Acceptance Questionnaire–Adolescent version (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 age 10 or older. Items were 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 (nine 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 0.89.

Parental Resilience Factors

Acceptance and Action Questionnaire

The 10-item Dutch version of the Acceptance and Action Questionnaire (AAQ-II) [38,39] was used to assess general levels of PF in the parent. Items were 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 nonclinical adult samples [38]. In the current study, Cronbach's alpha for the 10-item AAQ-II scale was 0.82.

Parental Psychological Flexibility Questionnaire

The Parental Psychological Flexibility Questionnaire (PPFQ) [24,25] was developed to assess the PF of parents in the context of their child's (chronic) pain. Items were 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 (five items), emotional acceptance (five items), pain acceptance (four items), and pain willingness (three items). Items of the latter three subscales were 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 17-item [25] versions 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 0.91.

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 (age eight to 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 age 12 years 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.

All analyses were performed in SPSS (version .23; IBM Statistics). Cronbach's alpha reliability statistics (see the *Methods* section) 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

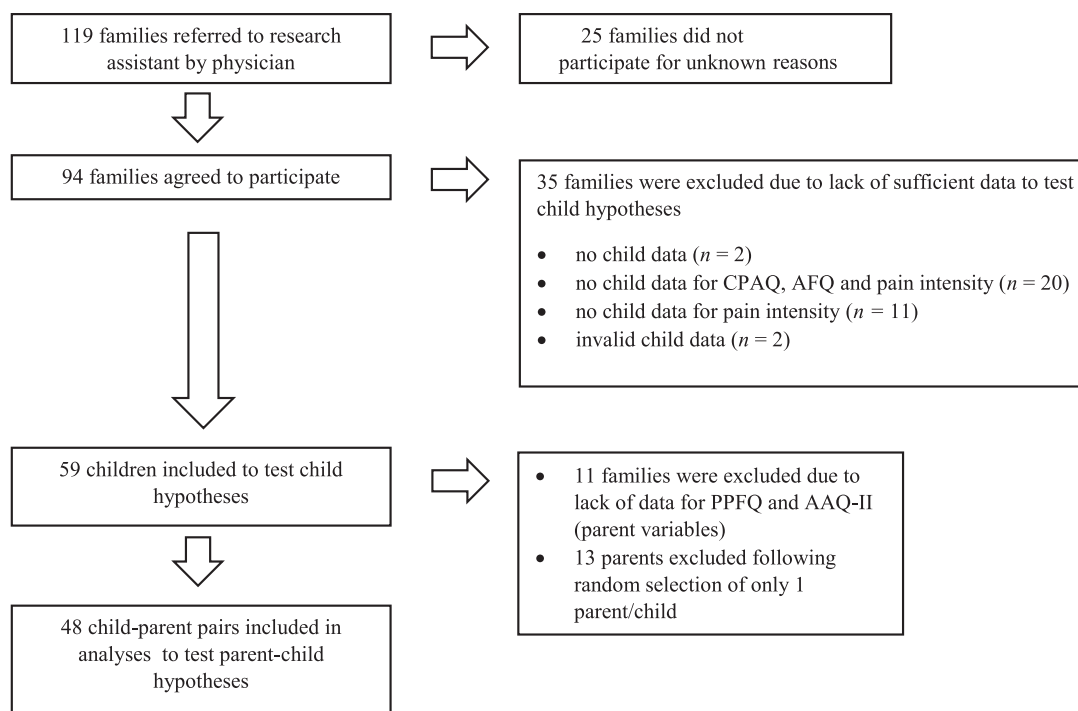


Figure 1. Flow chart of recruitment and data cleaning. AAQ-II = Acceptance and Action Questionnaire; AFQ = Avoidance and Fusion Questionnaire for Youth; CPAQ = Chronic Pain Acceptance Questionnaire-Adolescent version; PPFQ = Parental Psychological Flexibility Questionnaire.

pain-related PF and child outcomes (Figure 2), and 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 CIs) as they have been argued to be the most accurate with a high level of statistical power, especially with smaller sample sizes ($N < 80$) (see Mallinckrodt et al. [43] for more info). All analyses were executed at the 5% significance level.

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 ($r = -0.46, P < 0.01$) and between mean pain intensity and positive affect ($r = -0.40, P < 0.01$); there was a positive association between pain intensity and pain-related disability ($r = 0.64, P < 0.01$). Child mean pain intensity was not related to negative affect ($r = 0.15, ns$). Consistent with our expectations, child “resilience” factors (i.e., general PF and pain acceptance) were related to better scores on almost all outcome variables (Table 2).

That said, no significant association was found between pain acceptance and positive affect ($r = 0.15, ns$). 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 ($r = 0.35, P < 0.05$), as well as child acceptance of pain ($r = 0.38, P < 0.05$). Likewise, parents’ PF in the context of their child’s pain was positively related to the child’s own pain acceptance ($r = 0.44, P < 0.01$) but not to the child’s general PF ($r = -0.01, ns$).

General Psychological Flexibility

Does Child General Psychological Flexibility Moderate the Relation Between Child Pain Intensity and Child Outcomes?

Hierarchical multiple regression analyses (Table 3) revealed that the child’s mean pain intensity had a significant contribution in explaining children’s psychosocial health-related functioning ($\beta = -0.38, P < 0.01$), disability ($\beta = -0.60, P < 0.001$), and positive affect ($\beta = -0.38, P < 0.01$) but not negative affect ($\beta = 0.00, ns$). Furthermore, the child’s level of general PF did significantly contribute to the explanation of psychosocial health-related functioning ($\beta = 0.34, P < 0.01$) but not of disability ($\beta = -0.17, ns$). Moreover, the child’s general PF had a significant contribution in explaining negative affect ($\beta = -0.60, P < 0.01$) but did not contribute to the explanation of positive affect ($\beta = 0.14, ns$). Inspection of the interaction between child general PF

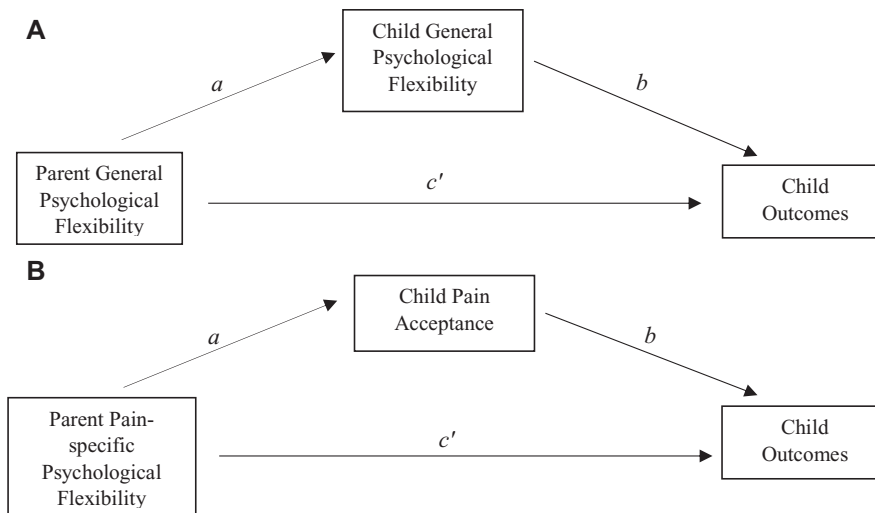


Figure 2. Three-variable mediation models that were examined for each of the child outcomes.

Note 1. 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. *Note 2.* 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]).

Table 2. Numbers, Means, SDs, 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)	0.14	0.05	-0.18	0.08	0.15	-0.11	-0.12	0.14	-0.36**	0.07
2. Child gender	59	N/A	—	0.04	-0.10	-0.05	0.09	0.02	-0.02	-0.05	0.09	0.10
Child variables												
3. Pain intensity	59	5.20 (2.13)	—	-0.25	-0.42**	-0.11	0.18	-0.46**	0.64**	-0.40**	0.15	
4. Psychological flexibility (general)	56	46.43 (11.51)	—	0.35**	0.35*	-0.01	0.43**	-0.32*	0.29*	-0.60**		
5. Pain acceptance	56	47.93 (12.88)	—	0.38*	0.44**	0.45**	-0.64**	0.14	-0.33*			
Parent variables												
6. Psychological flexibility (general)	43	54.09 (7.39)	—	0.27	0.19	-0.18	-0.07	-0.24				
7. Psychological flexibility (pain-specific)	44	57.68 (16.40)	—	0.19	-0.21	-0.13	-0.08					
Child Outcomes												
8. Psychosocial functioning	52	73.17 (16.63)	—	-0.62***	0.48**	-0.45**						
9. Disability	59	2.22 (1.98)	—	-0.47**	0.13							
10. Positive affect	52	40.69 (9.14)	—	-0.08								
11. Negative affect	52	28.90 (9.10)	—									

* $P < 0.05$; ** $P \leq 0.01$; *** $P \leq 0.001$.

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 (Table 3).

Does Parental General Psychological Flexibility Contribute, Through Child Psychological Flexibility, to Child Outcomes?

Bootstrap analyses (with 5,000 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 ($c'_{psychosocial} = 0.00, P = 0.99$; $c'_{disability} = -0.02, P = 0.47$; $c'_{positive} = -0.29, P = 0.10$; $c'_{negative} = -0.01, P = 0.97$). Although we found no direct contribution, we did find a significant indirect contribution of parents’ general PF via child general PF in explaining the child’s psychosocial health-related functioning ($ab_{psychosocial} = 0.25, SE = 0.15, 95\% BC CI_{psychosocial} = 0.04 \text{ to } 0.69$). Analyses revealed a similar significant indirect contribution of parental general PF via child general PF on the child’s positive and negative affect ($ab_{positive} = 0.16, SE = 0.09, 95\% BC CI = 0.03 \text{ to } 0.41$; $ab_{negative} = -0.27,$

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	-0.38**			
	2	Psychological flexibility	0.34**	0.32***		
Disability	1	Pain × psychological flexibility	-0.02	0.02	0.32***	50
	2	Pain intensity	0.60***			
Positive affect	1	Psychological flexibility	-0.17	0.44***		
	2	Pain × psychological flexibility	0.01	0.00	0.44***	56
Negative affect	1	Age (control)	-0.34*	0.12*		
	2	Pain intensity	-0.38**			
Disability	3	Psychological flexibility	0.14	0.19**		
	3	Pain × psychological flexibility	0.23	0.05	0.36***	50
Positive affect	1	Pain intensity	0.00			
	2	Psychological flexibility	-0.60***	0.37***		
Negative affect	2	Pain × psychological flexibility	0.06	0.00	0.37***	50

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 the analyses are shown. N = number of children included in analyses.

* $P < 0.05$; ** $P \leq 0.01$; *** $P \leq 0.001$.

SE = 0.14, 95% BC CI = -0.62 to -0.10). However, we found no significant indirect link with the child's pain-related disability ($ab_{disability} = 0.00$, SE = 0.00, 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 parents' general flexibility and their child's psychosocial health-related functioning and 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 ($\beta = 0.31$, $P < 0.05$), disability ($\beta = -0.45$, $P < 0.001$), and negative affect ($\beta = -0.32$, $P < 0.05$), 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 ($\beta = 0.02$, 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 ($\Delta R^2 = 0.05$, $F(1, 52) = 6.89$, $P < .05$). To explore the nature of this moderation effect, regression lines were plotted for children with high (1 SD above the mean) vs low (1 SD below the mean) pain acceptance scores (Figure 3). Significance tests with disability as an outcome showed that the simple slope was only significant for low pain acceptance ($\beta = 0.67$, $P < 0.0001$), 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 ($\beta = 0.18$, ns), suggesting that high pain acceptance completely buffered the negative effect of pain on disability. Additional analyses (Figure 3) revealed that higher pain acceptance was related to lower levels of disability in children who reported low as well as high levels of pain intensity. However, this association was stronger in children who experienced high levels of pain ($\beta = -0.73$, $P < 0.0001$) as compared with children who experienced low levels of pain ($\beta = -0.25$, $P < 0.05$). 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.

Does Parental Pain-Specific Psychological Flexibility Contribute, Through Their Child's Acceptance of Pain, to Child Outcomes?

Bootstrap analyses (with 5,000 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 ($c'_{psychosocial} = 0.05$, $P = 0.75$; $c'_{disability} = -0.02$, $P = 0.10$; $c'_{positive} = -0.10$, $P = 0.32$; $c'_{negative} = 0.01$, $P = 0.91$). Yet analyses did reveal that the indirect contribution of parent pain-specific PF through child pain acceptance helped explain the child's psychosocial health-related functioning ($ab_{psychosocial} = 0.22$, SE = 0.10, 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 ($ab_{disability} = -0.02$, SE = 0.01, 95% BC CI = -0.04 to -0.01; $ab_{negative} = -0.08$, SE = 0.04, 95% BC CI = -0.18 to -0.02). This was not the case for positive affect ($ab_{positive} = 0.03$,

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	-0.34*			
	2	Pain acceptance	0.31*	0.30***		
Disability	1	Pain intensity	0.46***		0.30**	50
	2	Pain acceptance	-0.45***	0.58***		
Positive affect	1	Age (control)	-0.23*		0.05*	56
	2	Pain intensity	-0.41**		0.17**	
Negative affect	3	Pain \times pain acceptance	0.10	0.01	0.30**	50
	1	Pain intensity	0.02			
Disability	2	Pain acceptance	-0.32*	0.11		
	2	Pain \times pain acceptance	0.27	0.06	0.17*	50

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 the analyses are shown. N = number of children included in analyses.

* $P < 0.05$; ** $P \leq 0.01$; *** $P \leq 0.001$.

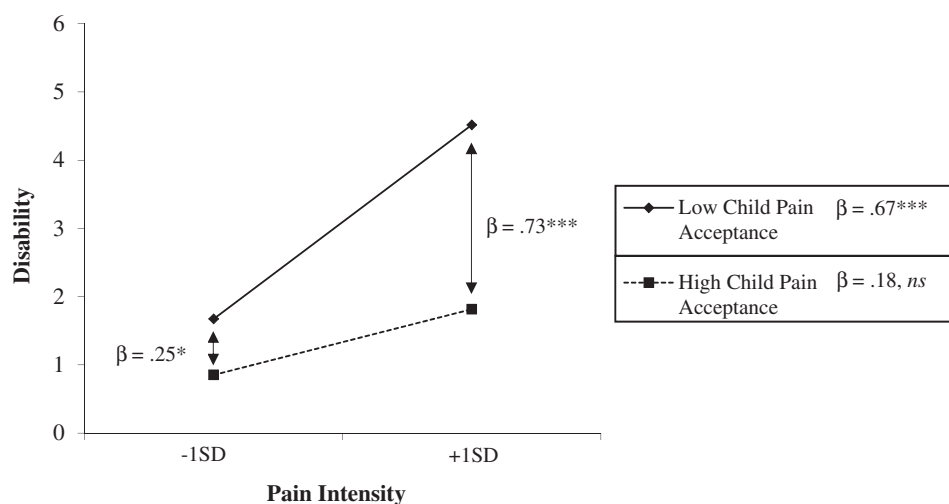


Figure 3. Regression lines for the associations between child pain intensity and disability as moderated by child pain acceptance. β = standardized regression coefficients (i.e., simple slopes).

SE = 0.03, 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 toward 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 are resilience factors that promote adaptive functioning in children with juvenile idiopathic

arthritis; b) child PF and pain acceptance buffer 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 have 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 the previous literature on PF [15,17,37]. Our results also indicated that child pain acceptance buffered 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 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 toward 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 vs 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, 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 with a child who has difficulty accepting the pain. Future research could further examine different contributions of general vs 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 and 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 modeling 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 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 (ACT) [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.

Conclusions

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 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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