Quality of Life in Youth with Chronic Pain: An Examination of Youth and Parent Resilience and Risk Factors

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Abstract

Objectives: Pediatric chronic pain has often been examined from a risk perspective, and relatively less is known about the individual and family-level resilience factors that help youth with chronic pain maintain their quality of life. This cross-sectional study: a) examined the relations among purported youth and parent resilience (youth pain acceptance and pain self-efficacy, parent psychological flexibility) and risk (youth pain intensity, parent protectiveness) factors with youth quality of life, and b) tested exploratory statistical mechanisms that may explain relations between parent and youth variables. **Methods:** Participants included 122 youth (10 to 17 years; $M=14.26$, $SD=2.19$) seen in an interdisciplinary pediatric chronic pain program and a parent. Youth completed measures of their average pain, quality of life, pain acceptance, and pain self-efficacy. Parents completed measures of their pain-related psychological flexibility and behavioral responses to pain (i.e., protectiveness, distraction, monitoring, minimizing). **Results:** Youth pain acceptance, pain self-efficacy, and parent psychological flexibility were highly positively correlated with each other, and with overall youth quality of life. Evidence for a buffering effect of pain acceptance and pain self-efficacy on the association between pain intensity and quality of life was not found. Protectiveness was found to be a significant mediator of the relation between parental psychological flexibility and youth quality of life. **Discussion:** The results are discussed in the context of the resilience-risk framework and current understandings of the role of parental factors for pediatric chronic pain.

Key words: pediatric chronic pain, resilience, parents, quality of life.

Introduction

Pediatric chronic pain is a prevalent health concern that is commonly associated with negative effects on daily functioning in the social, academic, physical, and emotional domains, which is reflected in an overall decreased quality of life (QOL) [1]. Research on pediatric chronic pain has largely focused on risk factors and adverse consequences, such as factors that negatively impact QOL [1,2]. However, individual variation in pain adaptation and functioning has been observed [3,4], suggesting that some factors may be protective for youth. Within pediatric chronic pain interventions, helping the youth return to school, maintain social interactions, and engage in physical activity have been identified as primary goals of treatment, rather than focusing on reducing pain levels [5,6]. Thus, it is important to understand the youth and parent factors contributing to QOL in youth with chronic pain, in order to further optimize family-based interventions.

The resilience-risk model of pediatric chronic pain identifies resilience and risk factors that promote pain adaptation or exacerbate pain at the individual and familial level [7]. Further, it has been proposed that resilience factors may both be relatively stable/broad traits, such as optimism or mindfulness [7,8], as well as pain-specific factors such as pain acceptance (living with pain without reaction, disapproval, or attempts to reduce/avoid it)[19,20] and pain self-efficacy (confidence that one can function despite pain)[11]. Pain acceptance and pain self-efficacy may be important factors to consider, as helping youth to live and function with their pain is often a goal of chronic pain interventions. In youth, pain acceptance and pain self-efficacy have been associated with reduced depressive symptoms, pain catastrophizing, pain interference, and mobility difficulties, as well as improved school functioning and QOL [12–16]. In adults with chronic pain, pain acceptance moderated the relation between pain intensity and

QOL [17], as well as ‘buffered’ expected increases in negative affect during intense pain [18]. Additionally, a study of children with juvenile arthritis found evidence for a buffering effect of pain acceptance on the relation between youth pain intensity and functional disability [19]. However, the potentially moderating role of both pain acceptance and pain self-efficacy on QOL outcomes in youth with a wide range of chronic pain conditions has not yet been investigated.

The associations between pain intensity and youth resilience factors have been inconsistent in the literature. Small yet significant associations between pain self-efficacy and pain intensity have been reported [13], as well as no associations at all [11]. Similarly, some studies have found significant associations (with ranging effect sizes) between pain acceptance and pain intensity [13,16,19,20], whereas others have reported no significant associations, suggesting that higher pain acceptance is not simply a function of lower pain [14]. This inconsistency demonstrates the importance of, and need for, continued replication of research findings in the field, in order to draw more confident conclusions from the literature [21,22].

Parent factors may also shape the development and maintenance of youth pain from both a resilience and risk perspective [7]. For example, how parents respond to the pain is important; four parental response styles to youth pain have been identified via the Adult Responses to Children’s Symptoms scale (ARCS) [23]. Protectiveness – characterized by a restriction of the child’s activities – is the most widely studied response style and is related to greater youth pain, functional disability, pain interference, and parental distress [24–28], indicating a potential parental risk factor for pediatric chronic pain outcomes [7]. Though relatively less studied, one study found that greater monitoring (checking in on the pain) and minimizing (dismissing/downplaying the pain), associated with increased youth functional disability, particularly if youth were experiencing higher levels of depression and anxiety [29]. The role of

a fourth response style, distraction (taking attention away from the pain), has not yet been investigated within a pediatric chronic pain context in relation to youth outcomes. In acute pain contexts, it has been demonstrated that parental distraction may be a helpful response style in reducing both youth pain and distress [30]. However, it could be argued that distraction for chronic, long-standing pain may not be helpful because it may encourage pain avoidance rather than acceptance, which has generally been associated with poorer youth outcomes [23,31,32]. To date, few studies have examined or reported on associations among the recently identified parent responses aside from protectiveness. Further, the studies which have reported on the other parent response styles have either not included associations with youth outcomes [33,34], or have not considered youth resilience factors such as pain self-efficacy [19,35].

Parent responses to pain are likely at least partially informed by their own cognitions and beliefs regarding their youth's pain [32]. For example, pain-related psychological flexibility is defined as the capacity of a parent to accept their own distress related to their youth's pain, and continue to engage the youth in valued activities; pain-related psychological flexibility is related to better youth social and emotional functioning, lower functional disability, and higher youth pain acceptance [19,35,36]. However, what is less clear is *how* parents' own attitudes/cognitions may be contributing to positive youth functioning. One study has demonstrated that parent psychological flexibility may indirectly influence youth outcomes via youth pain acceptance [19]. However, it is also possible that how parents think about their youth's pain may potentially impact how they are likely to respond to their youth, which in turn may lead to differential outcomes in youth QOL. Recently, it has been found that parent protectiveness appears to statistically mediate the relation between parent psychological flexibility and youth functional disability [35], although the role of other parental responses is unknown. Therefore, it is

important to extend this single study finding to examine youth QOL as an outcome, as well as consider other parental response styles.

A resilience-risk approach to pediatric chronic pain considering both youth and parent factors is necessary to better understand how they contribute to differential QOL in youth with chronic pain. Youth pain intensity and parent protectiveness are identified risk factors, and youth pain acceptance and pain self-efficacy, as well as parental psychological flexibility are identified resilience factors [7]. The role of other parental response styles, such as monitoring, minimizing, and distraction within the resilience-risk framework is less studied and understood, especially in relation to youth outcomes. Thus, the aims of this cross-sectional study were to investigate the relations among youth pain acceptance and pain self-efficacy, and parent psychological flexibility and responses to pain, in relation to youth pain intensity and QOL. We expected positive associations among the resilience factors of youth pain acceptance, pain self-efficacy, parent psychological flexibility, and youth quality of life. We also expected negative associations between the youth and parent resilience factors and youth pain intensity. Parent protectiveness, monitoring, and minimizing were expected to share negative associations with youth pain acceptance, pain self-efficacy, quality of life, and parent psychological flexibility. In line with frameworks that suggest distraction may be associated with an avoidant response style, it was hypothesized that parental distraction would be negatively associated with youth pain acceptance, pain self-efficacy, and quality of life.

Additionally, this study sought to add to the sparse literature showing that youth pain acceptance buffers the effect of pain intensity on disability [19], and that parent protectiveness statistically mediates the relation between parent psychological flexibility and youth functional disability [37]. Specifically, this study sought to extend these findings by examining the: (1)

moderating role of youth pain acceptance and pain self-efficacy on QOL outcomes, and (2) exploratory mediating effects of parental responses on the association between parent psychological flexibility and youth QOL. Informed by previous findings [19], it was hypothesized that both youth pain acceptance *and* pain self-efficacy would moderate the relation between pain intensity and QOL. It was also hypothesized that parental protectiveness, monitoring, minimizing, and distraction would statistically partially mediate the relation between parent psychological flexibility and youth quality of life.

Method

Participants and Procedure

Participants for this study were recruited as part of an ongoing research database that is being collected at a publicly funded, outpatient pediatric chronic pain clinic in Canada. Youth and parents provided informed consent and/or assent to participate in an ongoing research database which tracks patients throughout the program. Inclusion criteria were youth with chronic pain (pain for >3 months) between 10 to 17 years and the participation of a parent. Both youth and parent consent/assent were necessary in order to participate. Parents were considered to be any primary caregiver of the youth. Exclusion criteria included inability of the youth and/or parent to complete the questionnaires (e.g., inability to read or understand English or significant developmental delays) or youth aged younger than 10 years. Data for this study were drawn from questionnaires completed prior to or at the intake appointment only (i.e., prior to receiving any treatment). Institutional ethics approvals from the hospital and the university were granted.

Data were collected between January 2017 and March 2018. One hundred and fifty families were approached at this time. Eleven patients did not meet inclusion/exclusion criteria, four declined participation (e.g., did not want to or were not interested in participating in

research), and five were lost to follow-up (e.g., did not return after intake). In total, data were drawn from 130 youth and parent dyads. Of these dyads, a further 8 were excluded from the analyses because they did not meet inclusion criteria due to missing measures ($n=7$), or pain for <3 months ($n=1$; Complex Regional Pain Syndrome in its early stages). Thus, in the final sample, participants were 122 youth between the ages of 10 to 17 years ($M=14.26$, $SD=2.19$) and their parent/caregiver. In total, 93 girls (76.2%) and 29 boys (23.8%) participated in the study, and participating parents were predominantly mothers (82.8%), with a smaller subset of fathers (10.7%), grandparents (1.6%), and other (i.e., parents completed questionnaires together; 4.9%). Demographic information is presented in Table 1.

Measures

Youth Questionnaires

Pain Intensity. The Faces-Pain Scale Revised (FPS-R) was used to capture youth self-report of average pain intensity over the past one week [38]. The FPS-R presents six faces depicting increasing levels of pain that correspond to a numeric rating score from 0-10, in two-point intervals, from 0 (“No Pain”) to 10 (“Very Much Pain”). Higher scores demonstrate greater pain intensity. The FPS-R has been recommended for use in clinical trials [39], and has demonstrated high content validity and convergent validity for use with children older than 4 years [38].

Quality of Life. The Pediatric QOL Inventory Generic Core Scales Short Form (PedsQL SF15) is a 15-item self-report scale used to assess for QOL in youth, which has been adapted from the original 23-item version [40]. Items are rated on a 5-point scale ranging from 0 (“Never”) to 4 (“Almost Always”). The PedsQL SF15 includes four subscales of physical functioning, emotional functioning, social functioning, and school functioning. Subscales can be

combined to derive a total score, which ranges from 0 to 100 with higher scores reflecting higher QOL. The total score was used in the analyses for this study. The PedsQL SF15 has demonstrated adequate internal reliability in a sample of youth 12-18 years [41]. Overall, the PedsQL SF15 demonstrated adequate internal consistency in the present study ($\alpha=.85$) consistent with previous studies [41].

Pain Acceptance. Youth completed the Chronic Pain Acceptance Questionnaire for Adolescents (CPAQ-A) to assess for pain acceptance [14]. On the CPAQ-A, pain acceptance is measured by two factors: activity engagement (e.g., “I can do activities well even if I do not control my pain”), and pain willingness (e.g., “It’s O.K. to experience pain”). Items are rated on a scale from 0 (“Never True”) to 4 (“Always True”), with higher scores indicating higher pain acceptance. The CPAQ-A has shown high internal consistency and good construct validity for use with youth aged 10-19 years [14,42]. Internal consistency in this study was adequate and comparable to prior research ($\alpha=.87$)[42].

Pain Self-Efficacy. The Child Self-Efficacy Scale (CSE) was used to assess for youth pain self-efficacy in chronic pain [11]. The CSE is a 7-item scale that has been validated for use with youth aged 9-18 years. It measures the strength of a youth’s belief that they are able to engage in activities, rated on a 5-point scale ranging from 1 (“Very Sure”) to 5 (“Very Unsure”). Examples of activities on the CSE include the youth’s belief in their ability to complete homework, do chores, and see their friends. In this study, the CSE was reverse scored and mean scale scores were computed, such that higher scores indicated higher pain self-efficacy. Internal consistency in this study was adequate and consistent with prior research ($\alpha=.89$)[11].

Parent Questionnaires

Responses to Youth's Pain. The Adult Responses to Children's Symptoms (ARCS) was used to measure parental responses to their child's pain. The ARCS was scored and interpreted via the most recently reported four-factor structure for youth aged 7-18 years: protectiveness, distraction, monitoring, and minimizing [23]. Parents were asked to rate how often they engaged in a behavior on a 5-point scale ranging from 0 ("Never") to 4 ("Always"). Mean scale scores were computed for each response style, with higher scores indicating greater use of that response. Internal consistency for the four-factor structure of the ARCS was variable in this study ($\alpha = .83$ for protectiveness, $\alpha = .61$ for minimizing, $\alpha = .78$ for monitoring, and $\alpha = .58$ for distraction), though consistent with previously published research [23].

Pain-related Psychological Flexibility. Parents also completed the Parent Psychological Flexibility Questionnaire (PPFQ), which is a 17-item measure designed to assess parents' level of psychological flexibility regarding their child's pain [36]. Items are rated on a 7-point scale ranging from 0 ("Never True") to 6 ("Always True"). A mean score was calculated for parents' reported level of psychological flexibility, with higher scores indicating greater psychological flexibility. The PPFQ has demonstrated high internal consistency ($\alpha = .89$) and good convergent and concurrent validity for youth 10-18 years [43,44]. Internal consistency in the present study ($\alpha = .87$) was consistent with previous findings [43].

Analytic Plan

All statistical analyses were conducted using the Statistical Package for Social Sciences software program (SPSS; Version 24.0). Minimum sample size estimates were derived from statistical software (i.e., R) and previously published guidelines [45,46]. Thus, in order to detect medium sized effects with a power of 0.8 and significance level of .05, a minimum sample of 85, 71, and 76 were required to conduct correlations, mediation, and moderation respectively. The

current sample size exceeds the minimum sample size requirements. Age and gender were controlled for in all analyses.

Less than 5% of the data were missing for each outcome (pain intensity [1.6%], quality of life [0.8%], pain self-efficacy [4.1%], pain acceptance [3.2%], parental responses [3.2%], and parent psychological flexibility [2.5%]). Additionally, Little's MCAR test revealed that data were missing completely at random. Thus, list-wise deletion was used to account for missing data in all analyses [47]. Pearson and Spearman correlations (for non-normal data) were conducted with all variables. Given that many variables are considered in the preliminary correlation, it is possible that multiple comparisons may increase the Type I error rate, however a conventional statistical correction (i.e., Bonferroni correction) was not applied due to its stringent and conservative nature and likelihood for Type II error [48]. Discussions of the results will focus on interpreting effect sizes and CI, rather than p -values.

Moderation and cross-sectional mediation analyses were conducted using the PROCESS macro for SPSS. Two separate moderation analyses were conducted: the relation of pain intensity (predictor) and QOL (outcomes) moderated by (a) pain acceptance, and (b) pain self-efficacy. Predictors and moderators were mean-centered prior to analyses, and bias-corrected bootstrapping was performed with 5000 samples.

Given the low reliability of the distraction and minimizing subscales, these were excluded from the planned mediation analysis. Instead, a parallel mediation was conducted as follows: the relation between parental psychological flexibility (predictor) and youth QOL (outcome) mediated by (a) protectiveness and (b) monitoring. A regression-based path analytic framework and bias-corrected bootstrapping was used to test for indirect effects with 5000 samples.

Results

Associations Among Youth and Parental Factors, Pain, and QOL

Pearson's and Spearman's correlations, means, standard deviations, and 95% bootstrapped confidence intervals (95% CI) are reported in Table 2. Pain intensity was significantly and negatively associated with QOL and pain acceptance, and positively correlated with parental distraction. As hypothesized, youth and parent factors of pain acceptance, pain self-efficacy, and parental psychological flexibility were highly and positively correlated with each other and with youth QOL. Parent psychological flexibility was negatively associated with the parental response styles of protectiveness, distraction, and monitoring. Only protectiveness, distraction, and minimizing were found to be significantly and negatively correlated with youth QOL (i.e., not parental monitoring). Significant positive correlations were found among the parental response styles, except between parental minimization and distraction as well as between parental minimization and monitoring.

Moderating Roles of Pain Acceptance and Pain Self-Efficacy on the Relation between Pain Intensity and QOL

Pain Acceptance. Pain acceptance was tested as a moderator of the relation between youth pain intensity and QOL. The main effects of pain intensity and pain acceptance, as well as the interaction effect, accounted for 25% of the variance in QOL ($F=7.07, p<.001$; Table 3). However, the interaction term was not significant ($b=.46, SE=1.10, 95\%CI [-1.82, 2.74]$), R^2 change=.00), demonstrating no evidence for a moderating role of pain acceptance.

Pain Self-Efficacy. Pain self-efficacy was also tested as a moderator of the relation between youth pain intensity and QOL. The main effects of pain intensity and pain self-efficacy, as well as the interaction effect accounted for 26% of the variance in QOL ($F=7.39 p<.001$;

Table 4). However, the interaction term for pain self-efficacy and pain intensity was not significant ($b=.79$, $SE=.62$, 95%CI [-.69, 2.27], R^2 change=.01), demonstrating no evidence of a moderating role of pain self-efficacy.

Mediating Roles of Parental Responses on Youth QOL

In the total effect model, the positive relation between parent psychological flexibility and youth quality of life was significant (path c; $b=5.45$, $p<.01$, 95%CI [1.91, 9.00]; Figure 1). Results of the parallel mediation (Figure 2) revealed that only protectiveness significantly partially mediated the effect of parent psychological flexibility on youth QOL (95% CI [.49, 5.00]; ratio of the indirect effect to the total effect was 44%), statistically suggesting that parents with low psychological flexibility were more likely to have youth who reported low QOL via a greater use of parental protectiveness. In contrast, monitoring did not significantly mediate the effect of parental psychological flexibility on youth QOL (95% CI [-2.55, .68]). The effect of parent psychological flexibility on youth quality of life was not significant after taking into account parents' reported protectiveness and monitoring responses.

Discussion

This study sought to apply a family-based, resilience-risk framework to understand quality of life outcomes for pediatric chronic pain. Results from this study build upon existing research by addressing gaps in the current literature, such as the paucity of research on parent responses other than protectiveness, as well as novel associations among parent resilience factors and youth outcomes (e.g., parent psychological flexibility, youth pain self-efficacy). Moreover, this study is the first to report on the parent response style of distraction in relation to youth outcomes in a chronic pain context.

Results indicated that youth pain acceptance, pain self-efficacy, and parental psychological flexibility are resilience factors moderately to strongly related to higher youth QOL. Additionally, strong positive associations and large effect sizes were found among these variables, consistent with a positive feedback loop within the family in thinking about pain from a resilience perspective (e.g., emphasizing acceptance over avoidance, and believing that one can function despite pain).

Consistent with previous research, results revealed that high parental psychological flexibility cannot be fully explained by low youth pain, as the two appear weakly related [36,43]. Indeed, all parent variables examined were weakly related with pain intensity, although all parent variables except monitoring showed small to moderate associations with youth QOL. In our study, pain self-efficacy was not associated with pain intensity, and pain acceptance shared a significant but small correlation with pain intensity. Thus, it appears that for youth, pain intensity plays a small and potentially variable role with their levels of pain acceptance and pain self-efficacy. Although directionality is unknown in this cross-sectional study, the importance of the family context in promoting resilience despite pain levels is clear, and family-based interventions may be important to optimize QOL.

In contrast to a previous study that found that youth pain acceptance moderated the association between youth pain intensity and functional disability in youth with juvenile arthritis [19], the moderation models in our study with QOL as an outcome did not support similar findings. Of note, pain intensity in the study by Beeckman et al. (2018) was calculated via averaging youth's scores on their current, worst, and average pain ratings, whereas this study examined the youth's single average pain intensity rating. Within the resilience-risk model, pain acceptance and pain self-efficacy are conceptualized as processes or 'states' that are utilized

when a youth is confronted with pain. Therefore, it is possible that pain acceptance and pain self-efficacy may have a stronger effect when youth are confronted with more severe pain suggesting that examining ‘worst’ pain ratings is an avenue for future research. Moreover, our results are in contrast to a previous study of adults with chronic pain that utilized an accelerometer to examine momentary associations between pain intensity, pain acceptance, and pain interference found that pain acceptance moderated the association between pain intensity and pain interference [49]. Therefore, it is possible that the method of retroactive assessment was also not sensitive enough to detect a moderating effect. This study also reported on a Canadian sample from an outpatient chronic pain clinic within a children’s hospital. Given that moderation analyses may be particularly vulnerable to the effects of situational factors [22], another possibility is that cultural or contextual factors influenced the ability to detect a moderating effect. Continued replication, as well as the use of more rigorous, momentary data collection methods is important in order to draw firm conclusions.

Parents’ responses to their youth’s pain play an important role in the youth’s pain experience [26,32]. One previous study examined the role of parental distraction in youth with chronic abdominal pain during an acute pain task [50]. Ours is the first study to examine the association of parental distraction with youth outcomes in a pediatric chronic pain context. Importantly, the low internal consistency of the distraction subscale makes the results difficult to interpret and we were unable to conduct the planned mediation including this variable. Tentatively, based on the present results, associations between parent distraction and youth pain, QOL, and pain acceptance suggest that distraction may be more consistent with an avoidant than an acceptance-based response. It is understood that acceptance rather than avoidance of pain is necessary for pain adaptation and maintained functioning [51]. However, the role of parental

distraction is unclear because some types may be helpful if they encourage the youth to engage in valued activities that they enjoy (e.g., playing a game with friends), or may be unhelpful if interpreted as parental dismissiveness or encouraging of more cognitive distraction (e.g., ARCS item: “talk to your child about something else to take your child’s mind off it [pain]”). In part, this variability may explain the low reliability of the distraction subscale used in this study.

Parent minimizing was also characterized by low internal consistency. Minimizing was the least frequently utilized parental response style, potentially indicating a social desirability bias. In this study, minimizing was generally negatively associated with youth outcomes. A challenge in the current research base around parental responses to pediatric chronic pain has been that most of the research has focused on protectiveness, and very few report data including the other identified response styles. Parental distraction and minimizing may be important response styles to consider; it is therefore crucial that further development of psychometrically sound assessments is considered.

Consistent with previous findings, protectiveness was only weakly related with youth pain intensity [27,52,53]. However, a medium-sized relation was found between protectiveness and psychological flexibility, suggesting that parents’ own cognitions share a stronger association with protectiveness than the youth’s reported pain. Previously, parents’ own catastrophizing and confidence about their youth’s ability to function were found to be more strongly related to their protectiveness than their youth’s actual functioning [33]. Taken together, these findings highlight some important considerations. First, parents’ own attitudes and beliefs can contribute to their protectiveness more than child-specific factors, and thus are important to consider in pediatric pain assessment and treatment. Second, parental responses stem from a desire to help their child’s pain, and thus, protectiveness may be a reflection of parents

inadvertently engaging in unhelpful responses in an effort to protect their child from pain [54]. Taken together, interventions targeting parent behaviors, attitudes, and beliefs about pain should be considered an important part of pediatric chronic pain management.

Protectiveness showed a significant but small-sized association with youth pain acceptance. In contrast, a strong relation was found between protectiveness and youth pain self-efficacy. Although specifying directionality is not possible given the cross-sectional design, one interpretation of the latter finding is that the more parents try to restrict their youth's activities, the less confident the youth may feel in his/her ability to engage in tasks of daily living, such as homework and chores. Alternatively, youth who have lower confidence in their ability to complete daily tasks may be more likely to elicit protective responding from their parents. The mechanisms underlying these relations are unclear. For example, it is possible that parental protectiveness negatively impacts youth QOL via its negative impact on the youth's pain self-efficacy. Investigating mechanisms would provide important information regarding the role of parental responses on youth QOL, and factors to target in treatment.

Surprisingly, parental monitoring was not significantly related with any youth outcomes (effect sizes nearly zero), although it was moderately correlated with higher protectiveness and distraction. Additionally, mediation models that examined protectiveness and monitoring together highlighted the importance of protectiveness (and not monitoring), such that parents with low psychological flexibility had youth who reported significantly lower QOL via higher parent protectiveness. Previously, parental catastrophizing has been correlated with higher monitoring [33], suggesting that monitoring is closely associated with factors that have been found to underlie protectiveness and predict youth functional disability [55]. However, monitoring in isolation does not appear to relate strongly to outcomes. Current understandings of

parental solicitous behaviors are that they tend to reinforce pain versus function and are associated with negative youth outcomes [26,56]. Notably, solicitousness is characterized by behaviors associated with both “monitoring” and “protectiveness” (e.g., attention to the pain and restriction/avoidance of activities). Thus, it may be possible that parental attention exerts a negative influence when combined with protectiveness. Indeed, the two are moderately correlated, and parents who check-in more with their youth may be more likely to act on that information with protectiveness. Outcomes may also differ depending on when the monitoring occurs. Parents who check-in more frequently while their child is exhibiting pain behaviors may reinforce those behaviors [57]. However, if monitoring is non-contingent on pain (i.e., parents check-in at consistent intervals rather than in response to pain in the moment), it may be less reinforcing. Further research, which includes live, observational data of parental responses to their child’s pain is warranted in order to better understand the role of these behaviors in-the-moment, and optimally inform parent and family-based interventions for pediatric chronic pain.

Strengths, Limitations, and Future Directions

This study included a comprehensive examination of youth and parent resilience and risk factors, including youth pain self-efficacy, and the recently identified parent response styles of distraction, minimizing, and monitoring. The results from this study add to a growing literature of the individual and family factors that help youth with chronic pain maintain their functioning. Replication and logical extensions are important and necessary to verify the likelihood that the effects and mechanisms are reliable across a wide range of contexts [22], as well as clarify inconsistencies in the literature. Our results were consistent with some previous findings, such as the cross-sectional mediating role of protectiveness in the relation between parent psychological flexibility and youth outcomes [37], and inconsistent with other studies, such as the potential

buffering role of youth pain acceptance on pain intensity [19]. Therefore, results from this study contribute to current research findings, as well as demonstrate the need for further, rigorous examination.

The results of this study must be considered within the context of some limitations. One limitation that has been briefly discussed is the internal reliability of the ARCS. Although the internal reliabilities are consistent with previously published research [23,58], the distraction and minimizing subscales demonstrated low internal consistency, making it difficult to draw conclusions from the results. Further instrument development and psychometric testing are necessary to contribute more meaningful research that examines a full complement of parent responses beyond protectiveness. Moreover, youth perceptions of their parents' typical responses to pain, and how this in turn relates to youth outcomes is an area for future research. Secondly, this study recruited a clinical sample of youth from a hospital outpatient chronic pain clinic; thus, it is unknown whether the results generalize to youth who do not seek or have access to pain management services, such as youth who may be seen in community-based clinics. Efforts to include community-based samples may add to our increasing understanding of resilience and risk mechanisms in pediatric chronic pain, and potential implications for healthcare utilization. This study also utilized a cross-sectional design (and thus inferences about causality, especially in mediation models, cannot be made) and parent respondents were predominantly mothers. Longitudinal research is needed to confirm exploratory results found from the mediation models, and further studies examining the role of both mothers and fathers in the youth's pain and functioning outcomes could elucidate the complex relations among chronic pain and its psychosocial correlates, throughout development and within the family context. This study also focused solely on parent factors, although other social influences such as friends/peers are also

important to consider. Given that some evidence has been found that the quality of peer relationships may negatively impact functioning in youth with chronic pain regardless of parents' cognitions and behaviours [59], further widening our conceptualization of social risk and resilience is a crucial direction for future research. Additionally, although the analyses in the current study controlled for age and gender effects, it is possible that parent cognitions and responses could differ by youth age, and future research into age and gender based differences may be important. Finally, further development and examination of the resilience-risk model including other youth and/or parent risk factors of pain catastrophizing, pain interference, fear of pain, anxiety, or depression may be helpful to providing greater information on the factors that interact to impact quality of life in youth with chronic pain.

This study is part of a growing literature focused on exploring factors that help youth maintain functioning despite chronic pain. Findings from this study support the resilience-risk model of pediatric chronic pain [7] and highlight important youth and parent treatment targets to optimize QOL in youth with chronic pain.

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References

- 1 Hunfeld JA, Perquin CW, Duivenvoorden HJ, *et al.* Chronic pain and its impact on quality of life in adolescents and their families. *J Pediatr Psychol* 2001;**26**:145–53.<http://www.ncbi.nlm.nih.gov/pubmed/11259516> (accessed 8 May2016).
- 2 Gold JI, Yetwin AK, Mahrer NE, *et al.* Pediatric chronic pain and health-related quality of life. *J Pediatr Nurs* 2009;**24**:141–50. doi:10.1016/j.pedn.2008.07.003
- 3 Edwards RR, Dworkin RH, Sullivan MD, *et al.* The Role of Psychosocial Processes in the Development and Maintenance of Chronic Pain. *J Pain* 2016;**17**:T70–92. doi:10.1016/j.jpain.2016.01.001
- 4 Turkel S, Pao M. Late consequences of chronic pediatric illness. *Psychiatr Clin North Am* 2007;**30**:819–35. doi:10.1016/j.psc.2007.07.009
- 5 Odell S, Logan DE. Pediatric pain management: the multidisciplinary approach. *J Pain Res* 2013;**6**:785–90. doi:10.2147/JPR.S37434
- 6 Friedrichsdorf SJ, Giordano J, Desai Dakoji K, *et al.* Chronic Pain in Children and Adolescents: Diagnosis and Treatment of Primary Pain Disorders in Head, Abdomen, Muscles and Joints. *Child (Basel, Switzerland)* 2016;**3**. doi:10.3390/children3040042
- 7 Cousins LA, Kalapurakkel S, Cohen LL, *et al.* Topical Review: Resilience Resources and Mechanisms in Pediatric Chronic Pain. *J Pediatr Psychol* 2015;**40**:840–5. doi:10.1093/jpepsy/jsv037
- 8 Cousins LA, Cohen LL, Venable C. Risk and Resilience in Pediatric Chronic Pain: Exploring the Protective Role of Optimism. *J Pediatr Psychol* 2015;**40**:934–42. doi:10.1093/jpepsy/jsu094
- 9 McCracken LM. Learning to live with the pain: acceptance of pain predicts adjustment in

- persons with chronic pain. *Pain* 1998;**74**:21–7.
<http://www.ncbi.nlm.nih.gov/pubmed/9514556> (accessed 26 Jun2016).
- 10 McCracken LM. Learning to live with the pain: acceptance of pain predicts adjustment in persons with chronic pain. *Pain* 1998;**74**:21–7. doi:10.1016/S0304-3959(97)00146-2
 - 11 Bursch B, Tsao JCI, Meldrum M, *et al.* Preliminary validation of a self-efficacy scale for child functioning despite chronic pain (child and parent versions). *Pain* 2006;**125**:35–42. doi:10.1016/j.pain.2006.04.026
 - 12 Cramm JM, Strating MMH, Nieboer AP. The Importance of General Self-Efficacy for the Quality of Life of Adolescents with Diabetes or Juvenile Rheumatoid Arthritis Over Time: A Longitudinal Study among Adolescents and Parents. *Front Pediatr* 2013;**1**:40. doi:10.3389/fped.2013.00040
 - 13 Kalapurakkel S, A Carpino E, Lebel A, *et al.* ‘Pain Can’t Stop Me’: Examining Pain Self-Efficacy and Acceptance as Resilience Processes Among Youth With Chronic Headache. *J Pediatr Psychol* 2015;**40**:926–33. doi:10.1093/jpepsy/jsu091
 - 14 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**:316–20. doi:10.1016/j.ejpain.2009.05.002
 - 15 Tomlinson RM, Cousins LA, McMurtry CM, *et al.* The power of pain self-efficacy: applying a positive psychology framework to pediatric pain. *Pediatr Pain Lett* 2017;**19**.http://childpain.org/ppl/issues/v19n1_2017/v19n1_tomlinson.shtml
 - 16 Feinstein AB, Sturgeon JA, Bhandari RP, *et al.* Risk and Resilience in Pediatric Pain: The Roles of Parent and Adolescent Catastrophizing and Acceptance. *Clin J Pain* 2018;**34**:1096–105. doi:10.1097/AJP.0000000000000639

- 17 Elander J, Robinson G, Mitchell K, *et al.* An assessment of the relative influence of pain coping, negative thoughts about pain, and pain acceptance on health-related quality of life among people with hemophilia. *Pain* 2009;**145**:169–75. doi:10.1016/j.pain.2009.06.004
- 18 Kratz AL, Davis MC, Zautra AJ. Pain acceptance moderates the relation between pain and negative affect in female osteoarthritis and fibromyalgia patients. *Ann Behav Med* 2007;**33**:291–301. doi:10.1007/BF02879911
- 19 Beeckman M, Hughes S, Van Ryckeghem D, *et al.* Resilience Factors in Children with Juvenile Idiopathic Arthritis and Their Parents: The Role of Child and Parent Psychological Flexibility. *Pain Med* 2019;**20**:1120–31. doi:10.1093/pm/pny181
- 20 Feinstein AB, Forman EM, Masuda A, *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**:291–8. doi:10.1007/s10880-011-9243-6
- 21 Amir Y, Sharon I. Replication research: A “must” for the scientific advancement of psychology. *J Soc Behav Personal* 1990;**5**:51–69. <https://psycnet.apa.org/record/1991-01127-001> (accessed 1 Aug2019).
- 22 Simons DJ. The Value of Direct Replication. *Perspect Psychol Sci* 2014;**9**:76–80. doi:10.1177/1745691613514755
- 23 Noel M, Palermo TM, Essner B, *et al.* A developmental analysis of the factorial validity of the parent-report version of the Adult Responses to Children’s Symptoms in children versus adolescents with chronic pain or pain-related chronic illness. *J Pain* 2015;**16**:31–41. doi:10.1016/j.jpain.2014.10.005
- 24 Goubert L, Vervoort T, Sullivan MJL, *et al.* Parental emotional responses to their child’s

- pain: the role of dispositional empathy and catastrophizing about their child's pain. *J Pain* 2008;**9**:272–9. doi:10.1016/j.jpain.2007.11.006
- 25 Hechler T, Vervoort T, Hamann M, *et al.* Parental catastrophizing about their child's chronic pain: are mothers and fathers different? *Eur J Pain* 2011;**15**:515.e1-9. doi:10.1016/j.ejpain.2010.09.015
 - 26 Palermo TM, Valrie CR, Karlson CW. Family and parent influences on pediatric chronic pain: a developmental perspective. *Am Psychol* 2014;**69**:142–52. doi:10.1037/a0035216
 - 27 Simons LE, Claar RL, Logan DL. Chronic pain in adolescence: parental responses, adolescent coping, and their impact on adolescent's pain behaviors. *J Pediatr Psychol* 2008;**33**:894–904. doi:10.1093/jpepsy/jsn029
 - 28 Ross CK, Lavigne J V, Hayford JR, *et al.* Psychological factors affecting reported pain in juvenile rheumatoid arthritis. *J Pediatr Psychol* 1993;**18**:561–73. <http://www.ncbi.nlm.nih.gov/pubmed/8295080> (accessed 3 Jun2018).
 - 29 Claar RL, Simons LE, Logan DE. Parental response to children's pain: the moderating impact of children's emotional distress on symptoms and disability. *Pain* 2008;**138**:172–9. doi:10.1016/j.pain.2007.12.005
 - 30 Birnie KA, Noel M, Parker JA, *et al.* Systematic Review and Meta-Analysis of Distraction and Hypnosis for Needle-Related Pain and Distress in Children and Adolescents. *J Pediatr Psychol* 2014;**39**:783–808. doi:10.1093/jpepsy/jsu029
 - 31 Grewal S, Petter M, Feinstein AB. The use of distraction, acceptance, and mindfulness-based techniques in the treatment of pediatric pain. *Pediatr Pain Lett* 2012;**14**.
 - 32 Asmundson GJG, Noel M, Petter M, *et al.* Pediatric fear-avoidance model of chronic pain: foundation, application and future directions. *Pain Res Manag J Can Pain Soc = J la*

- société Can pour le Trait la douleur* 2012;**17**:397–405.<http://www.pubmedcentral.nih.gov/articlerender.fcgi?artid=3659014&tool=pmcentrez&rendertype=abstract> (accessed 22 Mar2016).
- 33 Jaaniste T, Jia N, Lang T, *et al.* The relationship between parental attitudes and behaviours in the context of paediatric chronic pain. *Child Care Health Dev* 2016;**42**:433–8. doi:10.1111/cch.12312
 - 34 Poppert Cordts KM, Stone AL, Beveridge JK, *et al.* The (Parental) Whole Is Greater Than the Sum of Its Parts: A Multifactorial Model of Parent Factors in Pediatric Chronic Pain. *J Pain* 2019;**20**:786–95. doi:10.1016/j.jpain.2019.01.004
 - 35 Timmers I, Simons LE, Hernandez JM, *et al.* Parent psychological flexibility in the context of pediatric pain: Brief assessment and associations with parent behaviour and child functioning. *Eur J Pain* Published Online First: 19 April 2019. doi:10.1002/ejp.1403
 - 36 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**:780–5. doi:10.1016/j.pain.2010.12.001
 - 37 Timmers I, Simons LE, Hernandez JM, *et al.* Parent psychological flexibility in the context of pediatric pain: Brief assessment and associations with parent behaviour and child functioning. *Eur J Pain* 2019;**23**:ejp.1403. doi:10.1002/ejp.1403
 - 38 Hicks CL, von Baeyer CL, Spafford PA, *et al.* The Faces Pain Scale-Revised: toward a common metric in pediatric pain measurement. *Pain* 2001;**93**:173–83.<http://www.ncbi.nlm.nih.gov/pubmed/11427329> (accessed 1 Nov2015).
 - 39 Stinson JN, Kavanagh T, Yamada J, *et al.* Systematic review of the psychometric properties, interpretability and feasibility of self-report pain intensity measures for use in

- clinical trials in children and adolescents. *Pain* 2006;**125**:143–57.
doi:10.1016/j.pain.2006.05.006
- 40 Varni JW, Seid M, Kurtin PS. 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**:800–12.<http://www.ncbi.nlm.nih.gov/pubmed/11468499> (accessed 6 Oct2015).
- 41 Chan KS, Mangione-Smith R, Burwinkle TM, *et al.* The PedsQL: reliability and validity of the short-form generic core scales and Asthma Module. *Med Care* 2005;**43**:256–65.<http://www.ncbi.nlm.nih.gov/pubmed/15725982> (accessed 11 May2018).
- 42 Wallace DP, Harbeck-Weber C, Whiteside SPH, *et al.* Adolescent acceptance of pain: confirmatory factor analysis and further validation of the chronic pain acceptance questionnaire, adolescent version. *J Pain* 2011;**12**:591–9. doi:10.1016/j.jpain.2010.11.004
- 43 Wallace DP, McCracken LM, Weiss KE, *et al.* The role of parent psychological flexibility in relation to adolescent chronic pain: further instrument development. *J Pain* 2015;**16**:235–46. doi:10.1016/j.jpain.2014.11.013
- 44 Wiwe Lipsker C, Kanstrup M, Holmström L, *et al.* The Parent Psychological Flexibility Questionnaire (PPFQ): Item Reduction and Validation in a Clinical Sample of Swedish Parents of Children with Chronic Pain. *Child (Basel, Switzerland)* 2016;**3**.
doi:10.3390/children3040032
- 45 Fritz MS, Mackinnon DP. Required sample size to detect the mediated effect. *Psychol Sci* 2007;**18**:233–9. doi:10.1111/j.1467-9280.2007.01882.x
- 46 Cohen J. A power primer. *Psychol Bull* 1992;**112**:155–9.<http://www.ncbi.nlm.nih.gov/pubmed/19565683> (accessed 4 Feb2015).

- 47 Kang H. The prevention and handling of the missing data. *Korean J Anesthesiol* 2013;**64**:402–6. doi:10.4097/kjae.2013.64.5.402
- 48 Perneger T V. What’s wrong with Bonferroni adjustments. *BMJ* 1998;**316**:1236–8. doi:10.1136/bmj.316.7139.1236
- 49 Kratz AL, Ehde DM, Bombardier CH, *et al.* 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**:319–31. doi:10.1016/j.jpain.2016.11.006
- 50 Walker LS, Williams SE, Smith CA, *et al.* Parent attention versus distraction: impact on symptom complaints by children with and without chronic functional abdominal pain. *Pain* 2006;**122**:43–52. doi:10.1016/j.pain.2005.12.020
- 51 Pielech M, Vowles K, Wicksell R. Acceptance and Commitment Therapy for Pediatric Chronic Pain: Theory and Application. *Children* 2017;**4**:10. doi:10.3390/children4020010
- 52 Sieberg CB, Williams S, Simons LE. Do Parent Protective Responses Mediate the Relation Between Parent Distress and Child Functional Disability Among Children With Chronic Pain? *J Pediatr Psychol* 2011;**36**:1043–51. doi:10.1093/jpepsy/jsr043
- 53 Wilson AC, Fales JL. Parenting in the context of chronic pain: a controlled study of parents with chronic pain. *Clin J Pain* 2015;**31**:689–98. doi:10.1097/AJP.0000000000000157
- 54 Fales JL, Essner BS, Harris MA, *et al.* When Helping Hurts: Miscarried Helping in Families of Youth With Chronic Pain. *J Pediatr Psychol* 2014;**39**:427–37. doi:10.1093/jpepsy/jsu003

- 55 Langer SL, Romano JM, Levy RL, *et al.* Catastrophizing and Parental Response to Child Symptom Complaints. *Child Health Care* 2009;**38**:169–84.
doi:10.1080/02739610903038750
- 56 Palermo TM, Eccleston C. Parents of children and adolescents with chronic pain. *Pain* 2009;**146**:15–7. doi:10.1016/j.pain.2009.05.009
- 57 Gatzounis R, Schrooten MGS, Crombez G, *et al.* Operant Learning Theory in Pain and Chronic Pain Rehabilitation. *Curr Pain Headache Rep* 2012;**16**:117–26.
doi:10.1007/s11916-012-0247-1
- 58 El-Behadli AF, Gansert P, Logan DE. Racial Differences in Parental Responses to Children’s Chronic Pain. *Clin J Pain* 2017;**33**:503–8.
doi:10.1097/AJP.0000000000000424
- 59 Ross AC, Simons LE, Feinstein AB, *et al.* Social Risk and Resilience Factors in Adolescent Chronic Pain: Examining the Role of Parents and Peers. *J Pediatr Psychol* 2018;**43**:303–13. doi:10.1093/jpepsy/jsx118

Figure 1. Total effect model for the relation between parent psychological flexibility and youth QOL (path c).

Figure 2. Standardized b -coefficients for the relation between parental psychological flexibility and youth QOL (path c') as mediated by parental protectiveness (path $a_1 \times b_1$) and monitoring (path $a_2 \times b_2$) ($n=107$). The indirect effect of protectiveness was found to be statistically significant, indicating that protectiveness partially mediated this relation. The ratio of the indirect effect to the total effect was 44% for protectiveness.

Table 3

Linear regression models examining the moderating role of youth pain acceptance on the relation between youth pain intensity and quality of life

	<i>b</i>	<i>SE b</i>	<i>p</i>
Pain Acceptance	11.43 [6.61, 16.26]	2.44	<.001
Pain Intensity	-.73 [-2.09, .63]	.69	.29
Pain Acceptance x Pain Intensity	.46 [-1.72, 2.64]	1.10	.68

*Note**: $n=114$. 95% confidence intervals are reported in square brackets.

Table 4

Linear regression models examining the moderating role of youth pain self-efficacy on the relation between youth pain intensity and quality of life

	<i>b</i>	<i>SE b</i>	<i>p</i>
Pain Self-Efficacy	7.17 [4.13, 10.21]	1.54	<.001
Pain Intensity	-.90 [-2.27, .47]	.69	.19
Pain Self-Efficacy x Pain Intensity	.79 [-.45, 2.02]	.62	.21

*Note**: $n=113$. 95% confidence intervals are reported in square brackets.