Published in final edited form as:

Pain. 2018 December; 159(12): 2442–2460. doi:10.1097/j.pain.000000000001342.

Catastrophizing, pain, and functional outcomes for children with chronic pain: a meta-analytic review

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Abstract

Pediatric chronic pain is associated with numerous negative outcomes including increased physical disability, increased rates of depression and anxiety, and decreased quality of life. Pain catastrophizing - broadly conceptualized as including rumination, magnification, and helplessness cognitions surrounding one's pain - has been linked with poor functional outcomes in children with chronic pain. Pain catastrophizing in pediatric chronic pain is often considered a key factor on which to focus treatment efforts. However, absent a systematic review that integrates the relevant literature, this call for routine assessment and targeted treatment may be premature. The present study aimed to: 1) meta-analytically quantify the relationship between catastrophizing and pain and functional/psychosocial outcomes (functional disability/physical functioning, anxiety, depression, and quality of life) in children with chronic pain, and 2) examine potential moderators of these relationships. Using a random effects model, a total of 111 effect sizes from 38 studies were analyzed. Effect sizes ranged from medium to large, with anxiety, depression, and quality of life demonstrating a strong association with catastrophizing. Pain intensity and physical disability had a moderate association with catastrophizing. These relationships were robust, minimizing potential publication bias. None of the examined moderators were significant. The strong relationships found between catastrophizing and anxiety, depression, and quality of life suggest that successfully intervening on catastrophizing could have far reaching implications in improving pain outcomes in pediatric chronic pain.

Keywords

meta-analy	ysis; catastrophizing; child; adolescent; chronic pain; pediatric; functioning	
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Disclosures:

This research was partially supported by the National Institutes of Health under award number T32 AR055885. We confirm that there have been no closely related manuscripts that have been submitted for simultaneous consideration to this or another journal.

There are no conflicts of interest that might be seen as influencing or prejudicing the research.

Introduction

Chronic pain is associated with negative outcomes in children and adolescents, including worse physical and emotional functioning [5,9,33,50,53,54,66,75,81,83,100]. Many factors have been explored to determine why chronic pain leads to such poor outcomes. Pain catastrophizing is one such factor [19,104]. As detailed by Turner and Aaron (2001), catastrophizing is historically rooted in the broader psychological literature and is most commonly linked with anxiety disorders [12,102] and depression [32]. Nearly two decades after what many consider to be the first use of the term by Albert Ellis [24], scholars began characterizing and examining the impact of catastrophizing about pain specifically [13,14,89,101]. Several of the early self-report measures of pain-related coping, such as the Cognitive Error Questionnaire (CEQ) [63] and the Coping Strategies Questionnaire (CSQ) [89], included items assessing catastrophic thinking about pain. However, the content of these items differ across measures. Many of the items from the CEQ assess pessimistic painrelated thoughts about the future (e.g., "If I don't get some time to relax during the day, I'm going to be bedridden and unable to work."), whereas items from the CSQ focus solely on helplessness cognitions (e.g., "I feel like I can't go on."). Over ten years later, in an effort to integrate the various theories and measurements of pain catastrophizing, Sullivan and colleagues (1995) created the Pain Catastrophizing Scale [104], which includes three separate but related domains: rumination, magnification, and helplessness. Although the work of Sullivan and colleagues mark a turning point in our understanding of pain catastrophizing (at least as indicated by the exponential growth in the literature and the predominance of the PCS as the gold standard measure), Turner and Aaron (2001) argue that none of the currently available measures, including the PCS, fully capture the construct of catastrophizing as defined in the broader field of psychology. Specifically, they contend that none tap into thoughts about the worse possible outcomes of pain (e.g., paralysis, complete disability), although they do acknowledge that the CEQ comes closest. Given these historical developments, and the fact that the three-factor model is privileged in most of the contemporary work in this area, we adopted Sullivan and colleagues' [104] conceptualization of pain catastrophizing for the current systematic review.

The aforementioned developments focused exclusively on adults with pain. Nearly a decade later, the pediatric literature began examining the impact of catastrophizing in pediatric pain experience. Self-report measures assessing pain catastrophizing in children and adolescents were created de novo (e.g., Pain Response Inventory [121] & Pain Coping Questionnaire [88]) or adapted from existing adult versions (e.g., CSQ – Child version [35], & PCS – Child version [19]). Research using these measures has found that catastrophizing in children with chronic pain is linked to greater pain and disability (cross-sectionally and longitudinally) [8,19], and to increased pain behaviors [74] and analgesic use [7]. In addition, previous research found that high catastrophizing children had significantly worse psychological outcomes than low catastrophizing children [21], with high catastrophizers scoring 4 times higher on a measure of depressive symptoms than their counterparts [74].

On account of these advancements in the theory and measurement of pain catastrophizing, along with the seemingly consistent pattern of findings suggesting that catastrophizing contributes to negative pain outcomes for children, several scholars have called for routine

assessment of pain catastrophizing in pediatric chronic pain and consider it a key factor on which to focus treatment efforts [2,55,127]. However, this call for routine assessment and targeted treatment may be premature. To date, no study has systematically integrated the previous results, quantified the magnitude of the relationship between catastrophizing and poor pain outcomes in children, or explored possible moderating factors. Such information is critical for healthcare providers and administrators to best allocate finite clinical and financial resources to the patients that are most in need.

Given the rapid developmental changes – physical and psychosocial – that take place during childhood and adolescence, several moderators should be examined in the context of catastrophizing and its relation to poor outcomes in pediatric chronic pain. Age is one such moderator. Research suggests that increased age is associated with increased levels of catastrophizing [7]; thus, the strength of the relationships between catastrophizing and pain outcomes may vary with age. Gender may also moderate the relationship between catastrophizing and pain outcomes. Socialization in relation to pain differs for girls and boys. In both healthy and chronic pain samples, pain catastrophizing is associated with higher levels of pain expression [116,117]. This is potentially important in the context of gender differences given that girls display more pain behaviors than boys, which may result in girls receiving more comfort from others when in pain [28]. This differential reinforcement may, in turn, contribute to gender differences in catastrophizing and pain outcomes.

In addition to developmental factors that may affect the relationship between catastrophizing and pain outcomes, there are several other factors to consider. Pain severity may influence the relationship between catastrophizing and pain outcomes, given that increasing levels of pain are related to higher levels of catastrophizing [94,108]. Pain duration may also influence this relationship. A longer pain duration may exacerbate catastrophic and helpless perceptions (i.e., "It's never going to get any better"), or it may attenuate them (i.e., "I need to accept this pain"). Another clinical factor to consider is type of chronic pain. Research suggests that levels of catastrophizing differ by pain diagnosis. For example, several studies have found that sickle cell patients have higher levels of pain catastrophizing than those with rheumatoid arthritis or musculoskeletal pain [16,47]. In addition to developmental and clinical factors, the specific measure used to assess catastrophizing may affect its relationship to pain. Measures of catastrophizing vary across clinical and research settings. These measures have different conceptualizations (unidimensional versus multidimensional) of catastrophizing and consequently, different psychometric properties. These differences between measures may influence the association between catastrophizing and pain outcomes.

In summary, catastrophizing is commonly touted as a key clinical construct to measure and focus treatment efforts on in the pediatric pain context. However, no systematic review has been conducted to integrate the relevant literature and guide these clinical efforts. Filling this knowledge gap would enhance treatment and guide allocation of resources to the patients who would benefit from treatment most. Thus, the aims of this study were to: 1) meta-analytically quantify the relationship between catastrophizing and pain and functional/psychosocial outcomes (physical functioning, anxiety, depression, and quality of life) in

children with chronic pain, and 2) examine age, gender, pain intensity, pain duration, pain diagnosis, and measure type as moderators of these relationships.

Methods

Literature Search

Empirical studies were identified using PsychInfo, Medline, PubMed, and Embase databases. Relevant articles published through March 23, 2017 were selected based on searches defined by all possible keyword combinations of terms for 1) catastroph*, 2) child*, youth, adoles*, and 3) pain, chronic pain, nociception. Electronic mail alerts were created using these terms to identify articles published after the initial search. Reference sections of identified empirical studies were reviewed for additional relevant studies. In addition, forward searches were conducted using identified articles to find additional relevant articles. Study authors were contacted for any necessary information needed for analyses from studies that reported insufficient information (see Table 1).

Inclusion and Exclusion Criteria

Studies were included if they 1) had measures of child-reported catastrophizing and one or more of the following: pain intensity, physical functioning, depression, anxiety, or quality of life (QOL), 2) were based on an independent child or adolescent sample (between the ages of 8 and 21) with chronic pain (i.e., persistent pain for 3 or more months), and 3) were available in English. Also, the studies needed to provide an effect size or contain information that allowed calculation of an effect size representing the relationship between catastrophizing and one or more of the pain outcomes of interest. Only effect sizes representing the relationship between baseline, or pre-intervention, levels of catastrophizing and functional/psychosocial outcomes were included.

Coding of Studies

Each article was read and coded independently by two study authors (M.M.M. and S.M.M.) using a standardized coding form.

Basic study information and study variables.—Sample-level information included publication year and type of publication. Sample characteristics included total sample size, race (percent White), gender (percent female), mean age, and whether or not the sample included participants over 18 years of age.

Moderator coding.—Gender (percent female), mean age of sample, mean pain intensity rating, and mean pain duration (in months) were coded as continuous moderators. Type of chronic pain and catastrophizing measure were coded as categorical moderators.

Quality Assessment

Studies were assessed for common sources of bias in observational studies [41,91]. Many items on the typical quality rubrics used for meta-analyses are not relevant for correlational designs [44,79]. Thus, for the current study, we used a modified version of the rubric from Salyers and colleagues' [90] meta-analysis, such that study quality ratings ranged from 0 to

10. The initial rating system was tested and refined on several studies before applying it to the full sample of studies. Interrater reliability of the initial codes was strong (t=.81, p<.01), and disagreements were resolved through discussion.

Meta-Analytic Method

Mean Effect Size.—Pearson's *r* was used as the effect size statistic for the relationship between catastrophizing and the pain outcomes of interest. Effect sizes were coded such that higher values reflect higher levels of catastrophizing and greater pain intensity, poorer physical functioning, higher levels of anxiety symptoms, higher levels of depressive symptoms, and worse QOL. All effect sizes were corrected using Fisher's *r*-to-Z transformation, which mitigates the problematic standard error formulation inherent in using *r* in its raw form. Effect sizes at the study level were weighted by sample size in order to account for the standard error in effect size estimates [11]. When a study provided multiple values for an association, an average effect size was calculated to reduce bias [11].

A random effects model was used, when appropriate (k 6), due to effects of both withinstudy and between-study variability [65]; when k < 6, the fixed effects model is reported. Effect sizes, mean effect sizes, and moderation models were calculated using IBM SPSS Statistics 24 and macros provided by Wilson (2017) [126]. Effect sizes were transformed back to r for ease of interpretation using the inverse of the Fisher's r-to-z transformation. Correlation coefficients of less than 0.10 were considered small, correlations of 0.25 were considered medium, and correlations greater than or equal to 0.40 were considered large [65].

Heterogeneity for each overall effect size was examined using the Q-statistic [11], with significant results (p < .10) suggesting moderation [45]. The I^2 index was calculated to examine the extent of heterogeneity [45]. When I^2 values were greater than or equal to 25%, moderator analyses were conducted [49], as this suggests that between-study variability in effect sizes exceeds levels that would be expected by chance [49].

A fail-safe Nanalysis was conducted to estimate how many studies with null findings would be necessary to reduce the effect sizes to non-significance [80]. Q-test effect size comparisons (with follow-up z tests) were conducted to determine the largest effect size for each outcome. The influence of publication bias was evaluated using Egger's regression approach and funnel plots [23].

Moderation Analyses—Categorical moderators were tested using Q statistics and \mathcal{P} indices (polarized effect sizes, decreased \mathcal{P} , and small confidence interval ranges indicating moderation), and continuous moderators were assessed for significant beta weights and decreased \mathcal{P} using meta-regressions [49]. Because meta-regressions use list-wise deletion, each moderator was examined independently in order to maximize the number of studies included in the analysis. A minimum of two studies per comparison group for categorical moderators was considered necessary for conducting planned meta-analyses.

For the first aim, the mean effect sizes between catastrophizing and pain, physical functioning, anxiety, depression, and QOL were calculated. For the second aim, categorical

and continuous moderators were explored when heterogeneity indexes indicated potential moderation.

Subgroup Analyses—In an effort to be inclusive, studies that included participants over the age of 18 or a small subset of participants reporting pain for < 3 months were included. Differences in mean effect size based on inclusion or exclusion of these samples were explored in subgroup analyses.

Results

Study Sample

One thousand two hundred and eighty-two records were identified through the initial database search. Sixty five studies met inclusion criteria. Of these 65 studies, 36 did not report sufficient effect size information. All corresponding authors were contacted via email to obtain required information. Twelve authors provided the necessary information and these data were included in the final sample, resulting in 38 unique samples of children with chronic pain (see Figure 1 for PRISMA flow diagram). Two studies were included that overlapped with other study samples but reported a unique catastrophizing – pain outcome relationship and so were retained for analysis. Descriptions of these 2 studies were omitted in the study summary (Table 1) and study quality (Table 2) tables to avoid duplication.

A total of 111 effect sizes from 38 studies were included in the final sample. Each study provided an average of 2.5 effect sizes. Studies used the following self-report measures of pain catastrophizing: Pain Catastrophizing Scale – Child (PCS-C) and adult report (PCS), Pain Coping Questionnaire (PCQ), Coping Strategies Questionnaire – Child (CSQ-C), Pain Response Inventory (PRI), and Pain-related Cognitions Questionnaire (PRCQ). The most commonly used measures were PCS-C (k=23), PCQ (k=8), and PCS (k=5).

The overall sample size contained 6202 participants, with the mean sample size for included studies equal to 77 participants. Study samples were, on average, predominately female (M=72%) and White (M=80%). The mean age for the samples was 14.4 years (range: 11.2–18.3). Three studies provided multiple effect sizes for one of the catastrophizing-pain outcome relationships; these effect sizes were averaged within each study for subsequent analyses. Additional study summary characteristics are reported in Table 1.

Relationship between Catastrophizing and Pain Outcomes

Table 3 presents the original and corrected (Fisher's *r*-to-Z transformed) effect sizes and sample level information for studies included in the final meta-analysis.

Pain intensity—The meta-analysis of the relationship between catastrophizing and pain intensity included 35 independent samples and resulted in a positive medium effect size, with r=0.29 (95% CI: 0.24 – 0.34, Table 4). The overall effect was significantly different from zero (z=11.37, p<0.01). Orwin's fail safe N analysis indicated an additional 172 studies with null effects would be needed to reduce the overall mean effect to non-significance. Egger's regression test of asymmetry resulted in a precision value = -0.17 (90% CI: -1.37 to 1.04, p=0.78), indicating no significant amount of asymmetry and suggesting no

significant amount of publication bias [23]. The funnel plot indicates a roughly symmetric distribution of effect sizes, with most of the effects around the mean intercept, further suggesting that publication bias is unlikely (Figure 2). The Q-statistic of the overall effect was 85.12, with a substantial amount of heterogeneity (P=60.06%) warranting additional moderation analyses.

Physical functioning—The meta-analysis of the relationship between catastrophizing and physical functioning included 30 independent samples and resulted in a positive medium effect size, with r=0.39 (95% CI: 0.35 – 0.43, Table 4). The overall effect was significantly different from zero (z=15.26, p<0.01), and an additional 210 studies with null effects would be needed to reduce this to non-significance. Egger's regression test of asymmetry resulted in a precision value = -0.17 (90% CI: -1.44 to 1.09, p=0.78), indicating no significant amount of asymmetry and minimal publication bias (Egger et al., 1997). The funnel plot was roughly symmetrical, further suggesting that publication bias is unlikely (Figure 3). The results of heterogeneity analyses (Q=72.33, P=59.91%) supported additional moderation analyses.

Anxiety—The meta-analysis of the relationship between catastrophizing and anxiety included 15 independent samples and resulted in a positive large effect size, with r=0.55 (95% CI: 0.50 – 0.59, Table 4). The overall effect was significantly different from zero (z=18.96, p<0.01). Orwin's fail safe N analysis indicated an additional 165 studies with null effects would be needed to reduce the overall mean effect to non-significance. Egger's regression test of asymmetry resulted in a precision value = 0.37 (90% CI: -1.00 to 1.73, p=0.57), indicating no significant amount of asymmetry and minimal publication bias [23]. The funnel plot was roughly symmetrical, further suggesting that publication bias is unlikely (Figure 4). The Q-statistic of the overall effect was 28.04, with a moderate amount of heterogeneity (P=45.91%).

Depression—The meta-analysis of the relationship between catastrophizing and depression included 19 independent samples and resulted in a positive large effect size, with r=0.49 (95% CI: 0.43 – 0.55, Table 4). The overall effect was significantly different from zero (z=12.58, p<0.01) and an additional 180 studies with null effects would be needed to reduce this to non-significance. Egger's regression test of asymmetry resulted in a precision value = 0.49 (90% CI: -1.48 to 2.46, p=0.61), indicating no significant amount of asymmetry, suggesting no significant amount of publication bias [23]. The funnel plot indicates a roughly symmetric distribution of effect sizes (Figure 5). The results of heterogeneity analyses (Q=66.96, P=73.12%) supported additional moderation analyses.

Quality of life—The meta-analysis of the relationship between catastrophizing and QOL included 10 independent samples and resulted in a positive large effect size, with r= -0.48 (95% CI: -0.56 - -0.39, Table 4). The overall effect was significantly different from zero (z=9.32, p<.01) with fail safe N analysis indicated 91 additional studies with null effects would be needed to reduce the overall mean effect to non-significance. Egger's regression test of asymmetry resulted in a precision value = 0.37 (90% CI: -2.10 to 2.84, p=0.74), indicating no significant amount of

publication bias [23]. The funnel plot indicates a roughly symmetric distribution of effect sizes, further suggesting that publication bias is unlikely (Figure 6). The Q-statistic of the overall effect was 30.31, with a substantial amount of heterogeneity (P=70.31%), indicating the need for additional moderation analyses.

Moderation

Moderator analyses for the relationship between catastrophizing, pain, and pain outcomes are shown in Tables 5 – 7. Among the continuous moderators and contrary to hypotheses, gender, age, pain intensity, and pain duration did not significantly moderate the relationship between catastrophizing and any of the pain outcomes. Similarly, among the categorical moderators, neither type of chronic pain nor catastrophizing measure significantly moderated (p>.05 for Q(b) values) any of the relationships between catastrophizing and pain outcomes. We could not conduct categorical moderation analyses of the relationship between catastrophizing and QOL due to having less than 2 studies for pain diagnosis or catastrophizing measure subgroups.

Study Quality

Study quality is reported in Table 2. The mean QR for studies was 5.97 (Median=6, Mode=5). In line with previous studies [17,27,48,90], quality rating (QR) was examined as a moderator of subgroup differences in effect sizes. Studies were grouped using sample mode (5) as a cut point [90], with studies 6 or above coded as "higher quality" and studies scoring at or below 5 considered "lower quality". Effect sizes between these two groups were compared. Study quality was not a significant moderator (p>0.05 for Q(b) values) for any of the relationships between catastrophizing and pain outcomes. Detailed results are presented in Table 8.

Sub-group Analyses

Two study characteristics were explored to assess their impact on the mean effect size: (1) whether or not samples included any participants older than 18 years of age, and (2) whether or not samples contained any participants reporting pain duration less than 3 months. Mean ESs were estimated including and excluding studies with these characteristics (i.e., participants older than 18 and participants with pain <3 months). As seen in Table 9, minimal differences in mean effect sizes were observed between the total study sample and the subgroups, indicating that these study characteristics did not substantially influence the results.

Discussion

The current meta-analysis assessed the magnitude of the relationship between catastrophizing, pain, and functional outcomes in children with chronic pain. Effect sizes ranged from medium to large, with anxiety, depression, and QOL demonstrating strong associations with catastrophizing. Pain intensity and physical functioning had moderate associations with catastrophizing. These relationships were robust, minimizing the potential influence of publication bias and study quality. Gender, age, pain intensity rating, pain

duration, pain diagnosis, and type of catastrophizing measure did not significantly moderate the relationship between catastrophizing and pain or functional/psychosocial outcomes.

Of the outcomes examined in the current meta-analysis, anxiety and depression had the strongest absolute relationships with catastrophizing. This strong relationship may be partially due to individuals having a general maladaptive thinking style that transcends painrelated stressors. Both in a broader psychological context and specifically within the chronic pain literature, catastrophizing has been characterized as a maladaptive thinking style in response to stress [6,24,104], frequently observed among individuals with anxiety [12,102] and depressive disorders [32], as well as those with chronic pain [87]. Thus, children who catastrophize in response to pain – and/or who appraise pain in catastrophic ways – may do similarly for non-pain stressors. This general cognitive-emotional style may explain the strong relationships between catastrophizing and anxiety and depression observed herein. Worth noting, a handful of studies point to conceptual overlap as a reason for the strong association between pain catastrophizing and negative mood (e.g., depression, anxiety) [1,46,105]. Although we acknowledge these blurry demarcations, cross-sectional [34,58,106] and longitudinal [57] evidence supports the conceptual distinctiveness of pain catastrophizing. These conceptual issues are paralleled by concerns about measurement overlap and common-method variance. Subscale or item level analysis between measures of catastrophizing, anxiety, and depression may clarify the relevance of these methodological issues. Unfortunately, because the majority of studies included in the current meta-analysis did not report subscale or item level scores, we could not undertake such analyses ourselves.

The strong relationship between catastrophizing and emotional outcomes suggests that intervention efforts aimed at decreasing catastrophic thinking may reduce anxiety and depressive symptoms among children with pain. Only a few studies have examined this issue. Kashikar-Zuck and colleagues [56] evaluated an 8-week CBT program for children with Juvenile Fibromyalgia and found that while catastrophizing decreased over the course of treatment, it did not mediate improvement in depressive symptoms. Similarly, Wicksell and colleagues [124] found that catastrophizing did not mediate changes in depression in response to an Acceptance and Commitment Therapy program (ranging from 7 to 20 therapy sessions) for pediatric chronic pain. Although these studies suggest that cognitive-behavioral and acceptance-based approaches can effectively reduce pain catastrophizing in children, much remains to be known about whether and how these changes translate into improvement in psychological functioning. Of note, these studies used less common measures of catastrophizing (PRI and PCQ), and neither assessed anxiety symptoms, thus constraining our understanding of these relationships.

Catastrophizing and physical functioning were moderately related in our analysis. The fear-avoidance model (FAM) of chronic pain provides one lens through which to interpret this relationship [3,62,85,97]. The FAM posits that poor physical functioning results from pain-related catastrophizing and threat appraisals. Thinking about pain in this way increases fear of future pain, leading to avoidance of activities and ultimately to disuse and disability [97]. This model has been applied to pediatric populations, lending support to the idea that intervening on pain catastrophizing could lead to improved physical functioning in children and adolescents with pain through decreases in their fear of pain and avoidance of activities.

The biopsychomotor model offers another perspective on the connection between catastrophizing and physical functioning in children and adolescents. This model posits that pain behavior is an integral part of the pain system [103], serving communicative, protective, and social-relational purposes. Pain expression is a form of communication, transmitting information that has survival value while also soliciting attention and support for the pain sufferer [36,40,125]. Although catastrophizing has been linked to increased communicative pain behaviors, specifically facial expressions of pain [72,117,118], the protective and social-relational aspects of the biopsychomotor model seem particularly relevant to the current findings regarding the link between catastrophizing and physical functioning. Children who endorse high levels of catastrophizing about their pain may, consequently, engage in protective behaviors (e.g., guarding, bracing) that have been linked to increased functional disability [107]. These behaviors may also trigger the social response system of proximal others, leading to solicitous behaviors from parents, teachers, and peers. Such solicitousness may reinforce the child's pain and avoidance behaviors thereby leading to continued physical disuse and worse functional disability.

QOL was strongly associated with catastrophizing. In child and adolescent chronic pain samples, QOL is often measured with the PedsQL [18,52,64,71,72,74,76,112], which assesses physical, emotional, social, and school domains [114]. Thus, the strong association between catastrophizing and QOL may be driven, in part, by the physical and emotional domains, which overlap with our other outcomes of interest (i.e., anxiety, depression, physical functioning) that demonstrated moderate-strong associations with catastrophizing. However, the PedsQL also assesses social and school functioning, domains of the pediatric pain experience that have received less attention in this literature. Children/adolescents with chronic pain frequently struggle with peer relations and academic achievement [31,82], and there is some research suggesting that catastrophizing contributes to poorer functioning in these domains [78]. Pain expressions and behaviors might tie these intra- and inter-personal factors together. As discussed above, although such behaviors may elicit emotional and functional support from others, they can also occasion more punitive responses, such as ignoring or expressions of anger or irritation [10], and are associated with interpersonal problems [61]. Although the current findings support the notion that pain catastrophizing and general QOL are strongly related in children with pain, future research should take a more nuanced approach, separating the construct into individual domains to elucidate their unique associations with pain catastrophizing.

Catastrophizing and pain intensity were moderately associated. This relationship was the most modest and varied (r= -.25 to .58) among those examined in our meta-analysis. Several physiological and neural explanations have been proposed to explain the link between catastrophizing and pain intensity; these include exaggerated muscle responses at the site of injury [86], altered hypothalamic-pituitary-adrenal axis activity [22,51], and lack of activation in brain regions responsible for top-down inhibitory control [92]. Although these studies suggest that pain catastrophizing is related to greater activity in brain regions involved in affective processing of pain, attention to pain, and pain behaviors, all were conducted in samples of adults with chronic pain. Whether and how such explanations apply to children remains an open question. Erpelding and colleagues [26] found that, among children with complex regional pain syndrome, pain catastrophizing was correlated with

increased gray matter and activation in brain regions involved in motor function, sensorimotor integration, and anxiety-driven exacerbations of pain – these findings suggest that pain catastrophizing may predispose children and adolescents to develop chronic pain, experience greater pain intensity when they do have pain, and engage in particular behavioral and emotional responses to that pain. Additional studies are needed to better understand the extent to which catastrophizing is a cause and/or consequence of increased pain in children, as well as the neurophysiological underpinnings of these relationships.

The varying effect sizes – magnitude and direction – observed in the current meta-analysis suggest a complex relationship between catastrophizing and pain in children that is moderated by other factors. We examined several candidate moderators that were suggested by theory and prior studies, however, none significantly moderated the relationships examined herein. Though evidence suggests that the pain experience differs for boys and girls of varying ages [7,28], the samples included in our analyses were comprised mostly of adolescent girls. Additionally, because the majority (83%) of included samples reported moderate (NRS=4) to severe (NRS=7) average pain intensity, our moderation analyses of gender, age, and pain intensity may have lacked adequate power. Future, high-powered studies are needed to better understand whether and how these factors impact the relationship between catastrophizing and pain outcomes in children.

Pain duration and pain diagnosis were also not supported as moderators, which suggests that the catastrophizing-pain nexus is not altered by the length of time or the specific pain diagnosis a child/adolescent has been experiencing. However, it bears noting that the number of studies included in specific pain diagnoses subgroups was low (all Ns < 6), thus reducing confidence in the reliability of the null finding. Lastly, pain catastrophizing measure did not explain significant heterogeneity in the observed relationships. Nevertheless, both Q and I^2 values were substantially reduced within categorical moderation subgroups for catastrophizing measure and pain diagnosis, indicating a reduction of variability in effect size heterogeneity between studies [49].

Findings from this meta-analysis have implications for treatment. Evidence supports the efficacy of several psychological interventions for reducing pain catastrophizing in pediatric samples – these include CBT for chronic pain [29,70], relaxation training [29], and Acceptance and Commitment Therapy [29,123]. These therapies often educate patients about pain catastrophizing, and some target self-reported catastrophic thoughts directly (i.e., through cognitive restructuring) or indirectly (i.e., through experiential exercises). Despite their wide use, much remains to be known about the effectiveness of such approaches – individually and/or collectively – as well their durability and mechanisms of action. Answers to these questions would provide insights for the further refinement of existing treatments and the formulation of new treatments to target catastrophic thinking and improve the functioning of children with pain. Results of the current meta-analysis suggest that such improvement may be especially achieved in the domains of anxiety and depression, as well as overall QOL.

Several limitations should be acknowledged. The majority of studies used clinical data, which introduces several forms of error, including selection bias and unsystematic

administration of measures [93]. Additionally, clinical data sets are often used repeatedly for various publications, a detail that is not specifically and consistently disclosed, making it difficult to guarantee samples and corresponding effect sizes are unique. Furthermore, the variability in outcome measures used to assess a particular outcome (e.g. physical functioning) may influence effect sizes. Another limitation, common in meta-analyses, is the file drawer problem [80]. Though the results of fail-safe analyses instill confidence in the meta-analytic findings for all of our primary outcome variables, we cannot rule out the existence of unpublished studies that would have changed these findings had they been included. This meta-analysis only included a selection of outcomes that may be associated with catastrophizing. Unfortunately, other potentially important outcomes, such as school performance, are not commonly reported in the pediatric pain literature and thus were not included herein. Lastly, all measures were self-report and used similar item-response formats, thus, increasing the possibility that common method bias contributed to the observed associations.

Future studies should aim to elucidate possible moderators of the relationship between catastrophizing and pain outcomes, as the current results indicate considerable between-study variability in the nature of this relationship. Future research may also investigate the effectiveness of current interventions in reducing catastrophic thinking and the magnitude of these effects over time. These findings would serve to enhance the individualization of treatments for chronic pain in children. The role of parents is important to examine as well. Many studies have reported significant relationships between parental catastrophizing and child pain outcomes [37,67,74], but these relationships have yet to be meta-analytically quantified.

Acknowledgments:

This research was partially supported by the National Institutes of Health under award number T32 AR055885. There are no conflicts of interest to disclose.

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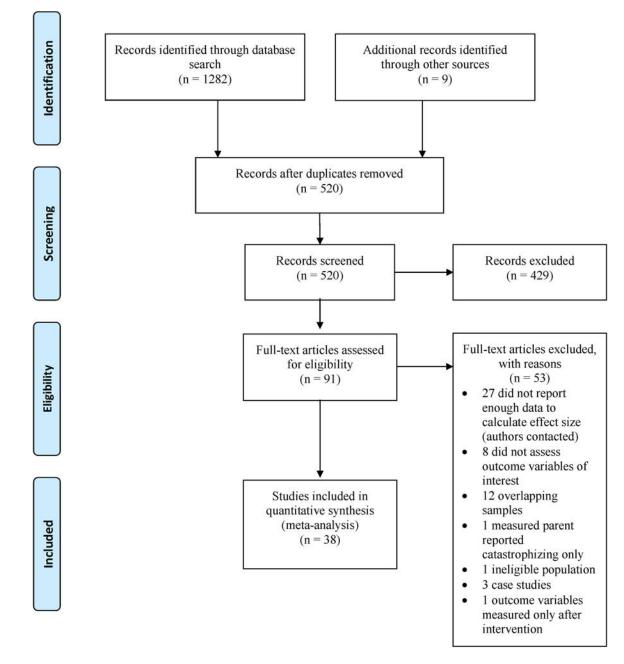


Figure 1. PRISMA Flow Diagram

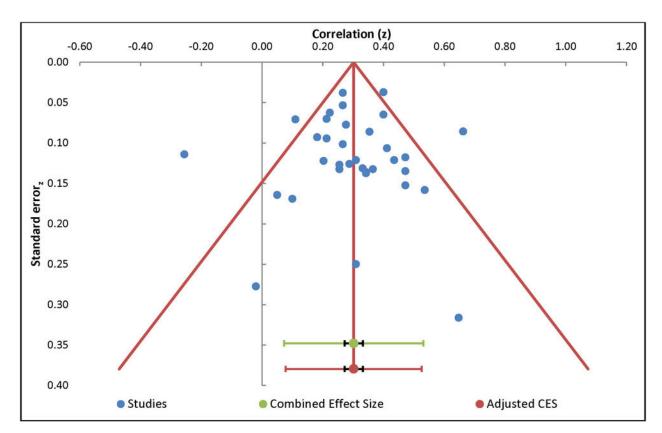


Figure 2. Funnel Plot – Pain Intensity

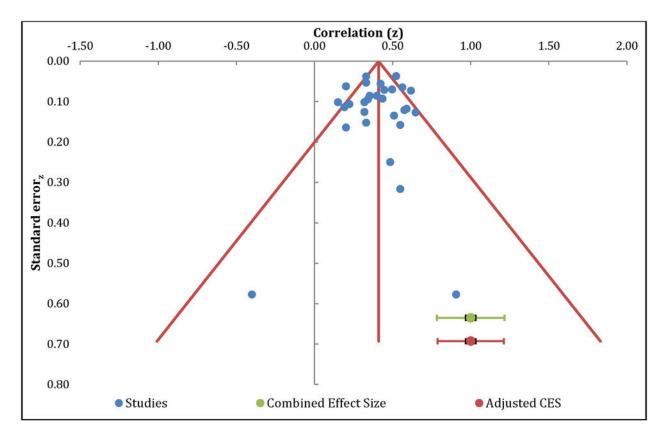


Figure 3. Funnel Plot – Physical Disability

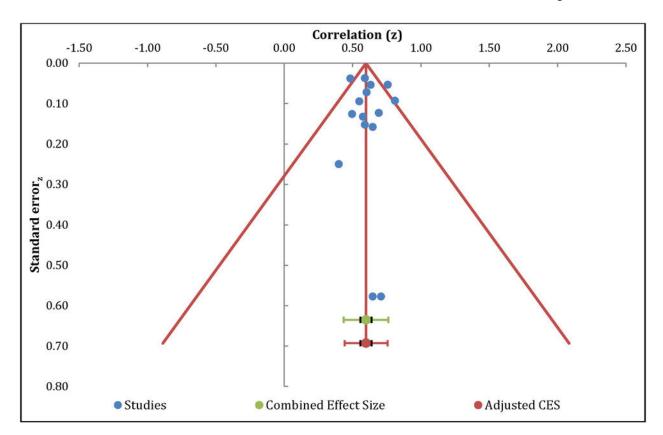


Figure 4. Funnel Plot – Anxiety

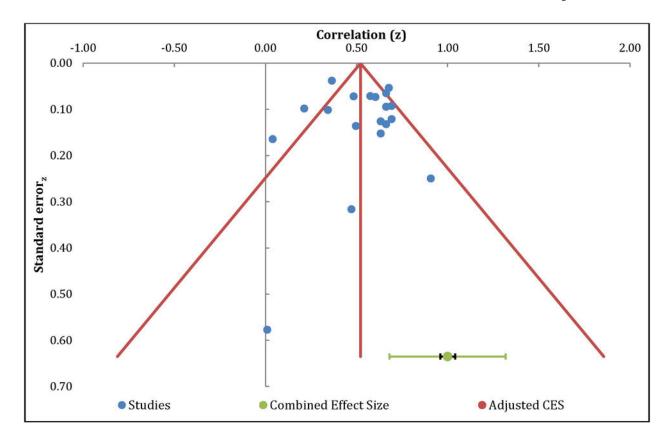


Figure 5. Funnel Plot – Depression

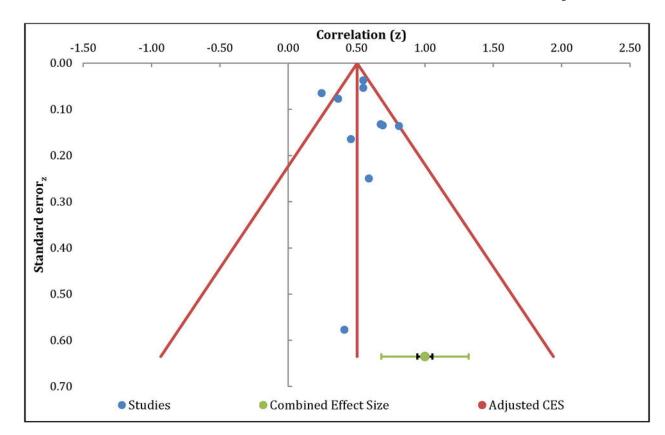


Figure 6. Funnel Plot – Quality of Life

Table 1.

Summary of Study Characteristics Across Independent Samples (k=38)

Sample Characteristics	k/N	%
Median year (range)	2013 (1998 – 2017)	
Mean sample size (SD)	145 (159.4)	
Median sample size (Range)	77 (6 – 725)	
Mean Age (SD)	14.4 (1.50)	
Gender (% Female)		72
Race (% White)		80
Country of Study		
United States	24	63.2
Belgium	2	5.3
Germany	3	7.9
Denmark	4	10.5
Netherlands	1	2.6
United Kingdom	3	7.9
Sweden	1	2.6
Catastrophizing Measure Used		
PCS-C	23	60.5
PCS (adult)	5	13.2
PRI	1	2.6
CSQ-C	1	2.6
PCQ	8	21.1
PRCQ	1	2.6
Outcomes Measured		
Pain Intensity	35	92.1
Physical Functioning	30	78.9
Anxiety	15	39.5
Depression	19	50
Quality of Life	2	5.3
Setting		
Inpatient	2	5.3
Outpatient	36	94.7
Pain Diagnosis		
Mixed Chronic Pain	21	55.3
Sickle Cell	2	5.3
Abdominal Pain	4	10.5
Headache	2	5.3
Fibromyalgia	3	7.9
Arthritis	4	10.5
Lupus	1	2.6
Chronic Low Back Pain	1	2.6

Sample Characteristics	k/N	%

Abbreviations: PCS = Pain Catastrophizing Scale (adult version), PCS-C = Pain Catastrophizing Scale for Children, PRCQ-C = Pain-Related Cognitions Questionnaire for Children, PCQ = Pain Coping Questionnaire, CSQ-C = Coping Strategies Questionnaire for Children, PRI = Pain Response Inventory

Table 2.

Quality
f Study
Ratings of

Quality Score	e	е	∞	7	∞	9	w	9	w	7	4	9	9	3	w	∞	∞	7	w	w	w	w
Power analysis?	No	No	Yes	No	No	No	No	No	No	No	No	No	No	No	No	Yes	No	No	No	No	No	No
Single pain diagnosis pain sample?	No	No	No	No	Yes	No	No	No	No	No	No	Yes	Yes	No	Yes	Yes	Yes	Yes	No	Yes	No	No
Was the participation rate of eligible individuals at least 50%	No	NR	Yes	Yes	Yes	NR	Yes	NR	NR	Yes	NR	NR	No	NR	NR	No	Yes	No	NR	NR	NR	NR
Was the data collected as part of a research study	No	No	Yes	Yes	Yes	Yes	Yes	Yes	No	Yes	No	No	Yes	No	Yes	Yes	Yes	Yes	No	No	No	No
Were both variables continuous?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Was the study part of a RCT?	No	No	No	No	No	No	No	No	No	No	No	No	Yes	No	No	No	No	No	No	No	oN	No
Was reliability information for the dependent variable reported and a=.70 for the current sample?	No	No	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	No	Yes	No	No	°N	Yes	Yes	Yes	Yes	No	Yes	Yes
Was the dependent variable clearly defined, using a measure that had been validated before?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Was reliability information for the independent variable reported and above α=.70 for the current sample?	No	No	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes	Yes	No	No	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Was the independent variable assessed with a validated measure?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Study	Benore et al. (2015)	Bhandari et al. (2016)	Cousins et al. (2015)	Crombez et al. (2003)	Cunningham et al. (2014)	Engel et al. (2013)	Flink et al. (2016)	Guite et al (2011b)	Guite et al (2011a)	Heathcote et al. (epub)	Hermann et al. (2007)	Jones et al. (2016)	Kashikar-Zuck et al. (2013)	Keogh & Eccleston (2006)	Kroner-Herwig & Maas (2013)	Libby & Glenwick (2010)	Lomholt et al. (2013)	Lomholt et al. (2015)	Lynch et al. (2007)	Lynch et al. (2006)	Lynch-Jordan et al. (2013)	Mano et al. (2012)

Study	Was the independent variable assessed with a validated measure?	Was reliability information for the independent variable reported and above α=.70 for the current sample?	Was the dependent variable clearly defined, using a measure that had been validated before?	Was reliability information for the dependent variable reported and $\alpha=70$ for the current sample?	Was the study part of a RCT?	Were both variables continuous?	Was the data collected as part of a research study	Was the participation rate of eligible individuals at least 50%	Single pain diagnosis pain sample?	Power analysis?	Quality Score
Mano et al. (2013)	Yes	No	Yes	No	Yes	Yes	Yes	No	$_{\rm o}^{\rm N}$	No	w
Miller et al. (2016)	Yes	Yes	Yes	Yes	No	Yes	No	NR	No	No	w
Pielech et al. (2014)	Yes	Yes	Yes	Yes	No	Yes	No	NR	No	No	w
Sil et al. (2016a)	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes	No	œ
Sil et al. (2016b)	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes	Yes	6
Simons et al. (2015a)	Yes	No	Yes	No	No	Yes	Yes	Yes	Yes	No	9
Simons et al. (2015b)	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	No	No	7
Thastum et al. (2005)	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes	No	œ
Thastum et al. (1999)	Yes	Yes	Yes	No	No	Yes	Yes	NR	Yes	Yes	7
Tran et al. (2017)	Yes	No	Yes	No	No	Yes	Yes	Z	Yes	No	ß
Tran et al. (2015)	Yes	Yes	Yes	Yes	No	Yes	No	NR	No	No	ß
van Tilburg et al. (2015)	Yes	No	Yes	No	Yes	Yes	Yes	Yes	Yes	No	7
Verbunt et al. (2015)	Yes	No	Yes	No	No	Yes	Yes	z	No	No	4
Vervoort et al. (2009)	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	N_{0}	No	7
Vervoort et al. (2008)	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	No	No	7
Warschburger et al (2014)	Yes	Yes	Yes	Yes	No	Yes	Yes	No	Yes	No	7
Chow et al. (2016)				Same san	Same sample as Simons, 2015	ns, 2015					
Vervoort et al. (2006)				Same san	Same sample as Crombez 2003	bez 2003					

NR=Not reported

Description of Included Studies

Table 3.

Study	Year	Sample Size	Percent Female	Percent White	Mean Age	Chronic Pain Diagnosis	Catastrophizing Questionnaire	Outcome Association	Measure Used to Assess Outcome	ES	ES
Benore et al. [8]	2015	119	77%	95%	15.1	Mixed Pain Sample	PCS-C	Pain Intensity	NRS (0–10)	0.18	0.18
								Physical Functioning	BAPQ-PFss	0.41	0.37
								Physical Functioning	PedsQL-PFss	0.46	0.50
								Anxiety	BAPQ - GAss	0.57	0.65
								Anxiety	BAPQ - PSAss	0.77	1.02
								Depression	BAPQ - Dss	09.0	69.0
Bhandari et al. [4]	2016	352	75%	%89	13.9	Mixed Pain Sample	PCS-C	Pain Intensity	NRS (0-10)	0.26	0.26
								Physical Functioning	PROMIS - mobility	0.32	0.33
								Anxiety	PROMIS - anxiety	0.64	0.76
								Depression	PROMIS - depression	0.59	0.68
Chow et al. [15]	2016	195	%9L	93%	13.8	Mixed Pain Sample	PCS-C	Anxiety	RCMAS	0.54	09.0
								Depression	CDI	0.45	0.48
Cousins et al. [18]	2015	58	%6L	%09	14.6	Mixed Pain Sample	PCS-C	Pain Intensity	VAS	0.44	0.47
								Physical Functioning	FDI	0.47	0.51
								Quality of Life	PedsQL	-0.60	-0.69
Crombez et al. [19]	2003	43	54%	100%	11.83	Mixed Pain Sample	PCS-C	Pain Intensity	VAS	0.49	0.54
								Physical Functioning	FDI	0.50	0.55
Cunningham et al. [20]	2014	75	77%	83%	13.84	Abdominal Pain	PCS-C	Pain Intensity	NRS (0-10)	0.44	0.47
								Physical Functioning	FDI	0.53	0.59

	rear	Size	Female	Percent White	Mean Age	Chronic Pain Diagnosis	Catastrophizing Questionnaire	Outcome Association	Measure Used to Assess Outcome	ES original	ES corrected
Engel et al. [25]	2013	80	43%	77%	14.35	Mixed Pain Sample	PCS	Pain Intensity	NRS (0–10)	-0.25	-0.26
								Physical Functioning	FDI	0.19	0.19
Flink et al. [30]	2016	9	83%	100%	18.33	Mixed Pain Sample	PCS-C	Physical Functioning	FDI	0.72	0.91
								Anxiety	HADS-A	0.57	0.65
								Depression	HADS-D	0.01	0.01
Guite et al.(a) [39]	2011	138	84%	87%	15.6	Mixed Pain Sample	PCS	Pain Intensity	VAS	0.34	0.35
								Physical Functioning	FDI	0.38	0.40
Guite et al.(b) [38]	2011	259	78%	93%	15.1	Mixed Pain Sample	PCS-C	Pain Intensity	NRS (0-10)	0.22	0.22
								Physical Functioning	FDI	0.20	0.20
Heathcote et al. [42]	2017	99	83%	ı	13.97	Mixed Pain Sample	PCS-C	Pain Intensity	NRS (0-10)	0.28	0.29
								Physical Functioning	FDI	0.31	0.32
								Anxiety	RCADS-GADss	0.46	0.49
								Depression	RCADS-MDDss	0.56	0.63
Hermann et al. * [43]	2007	71	ı	1	1	Mixed Pain Sample	PRCQ	Pain Intensity	CSI	0.30	0.31
		69	ı	ı	ı			Anxiety	CAT-II	09.0	69.0
		106	,	1	ı			Depression	DTC-DMss	0.30	0.31
								Depression	DTC-ABss	0.13	0.13
								Depression	DTC-Ess	0.21	0.21
Jones et al. [52]	2016	09	%88	20%	16.1	Lupus	PCS	Pain Intensity	VAS	0.35	0.37
								Anxiety	SCARED	0.52	0.58
								Depression	CDI	0.58	99.0
								Quality of Life	PedsQL	-0.59	-0.68
Kashikar-Zuck et al. [56]	2013	100	93%	%06	15.02	Juvenile Fibromyalgia	PCQ	Physical Functioning	FDI	0.15	0.15
								Depression	CDI	0.33	0.34

Study	Year	Sample Size	Percent Female	Percent White	Mean Age	Chronic Pain Diagnosis	Catastrophizing Questionnaire	Outcome Association	Measure Used to Assess Outcome	ES original	ES
Keogh & Eccleston ** [59]	2006	46 (male)	%0		i	Mixed Pain Sample	PCQ	Pain Intensity	NRS (0–10)	0.44	0.47
								Physical Functioning	FDI	0.32	0.33
								Anxiety	SCAS	0.53	0.59
								Depression	CDI	0.56	0.63
		115 (female)	100%		ı			Pain Intensity	NRS (0-10)	0.21	0.22
								Physical Functioning	FDI	0.33	0.34
								Anxiety	SCAS	0.50	0.55
								Depression	CDI	0.58	99.0
Kröner-Herwig & Maas [60]	2013	09	%55	N. R.	12.6	Headache	PCS-C	Pain Intensity	NRS (0-10)	0.25	0.26
Libby & Glenwick [64]	2010	57	93%	2%	15.5	Juvenile Fibromyalgia	CSQ-C	Pain Intensity	VAS	0.33	0.34
								Depression	CDI	0.46	0.50
								Quality of Life	PedsQL	-0.67	-0.81
Lomholt et al. (a) [69]	2013	91	77%	Z.	12.7	Arthritis	PCQ	Pain Intensity	FPS-R	0.39	0.41
								Physical Functioning	СНАО	0.22	0.22
Lomholt et al. (b) [68]	2015	19	%62	N. R.	11.72	Arthritis	PCQ	Pain Intensity	FPS-R	0.30	0.31
								Physical Functioning	FDI	0.45	0.48
								Anxiety	BAI	0.38	0.40
								Depression	BDI	0.72	0.91
								Quality of Life	PedsQL	-0.53	-0.59
Lynch et al. ** [72]	2007	70	%0			Mixed Pain Sample	PCQ	Pain Intensity	VAS	0.20	0.20
		202	100%		,			Pain Intensity	VAS	0.11	0.11
Lynch et al. [73]	2006	65	%08	91%	14.9	Chronic Back Pain	PCQ	Pain Intensity	VAS	0.25	0.26
								Physical Functioning	FDI	0.57	0.65

Study	Year	Sample Size	Percent Female	Percent White	Mean Age	Chronic Pain Diagnosis	Catastrophizing Questionnaire	Outcome Association	Measure Used to Assess Outcome	ES original	ES corrected
Lynch-Jordan et al. [74]	2013	240	77%	%68	14.76	Mixed Pain Sample	PCS-C	Pain Intensity	NRS (0–10)	0.38	0.40
								Physical Functioning	FDI	0.51	0.56
								Depression	CDI	0.58	99.0
								Quality of Life	PedsQL	-0.24	-0.24
Mano et al. [76]	2012	349	%69	%11%	14.2	Mixed Pain Sample	PCS-C	Anxiety	SCARED	0.56	0.63
								Quality of Life	PedsQL	-0.50	-0.55
Mano et al. [77]	2013	9	83%	%19	13.75	Mixed Pain Sample	PCS	Physical Functioning	CALQ	-0.38	-0.40
								Anxiety	STAI-C Trait Scale	0.54	09.0
								Anxiety	STAI-C State Scale	0.68	0.83
Miller et al. [78]	2016	139	72%	92%	15	Mixed Pain Sample	PCS-C	Pain Intensity	NRS (0–10)	0.58	0.66
								Physical Functioning	FDI	0.34	0.35
Pielech et al. [84]	2014	269	77%	92%	13.9	Mixed Pain Sample	PCS-C	Pain Intensity	NRS (0–10)	0.26	0.27
								Physical Functioning	FDI	0.32	0.33
								Anxiety	RCMAS	0.45	0.48
								Depression	CDI	0.35	0.37
Sil et al. (a) [95]	2016	40	NR	NR R	NR	Sickle Cell	PCS-C	Pain Intensity	NRS (0-10)	0.05	0.05
								Physical Functioning	FDI	0.20	0.20
								Depression	CDI	0.04	0.04
								Quality of Life	PedsQL	-0.43	-0.46
Sil et al. (b) [96]	2016	100	61%	%0	13.54	Sickle Cell	PCS-C	Pain Intensity	NRS (0-10)	0.26	0.27
								Physical Functioning	FDI	0.31	0.32
Simons et al. (a) [98]	2015	206	73%	%06	13.6	Headache	PCS-C	Pain Intensity	NRS (0-10)	0.21	0.22
								Physical Emetioning	FDI	0.46	0.49

Study	Year	Sample Size	Percent Female	Percent White	Mean Age	Chronic Pain Diagnosis	Catastrophizing Questionnaire	Outcome Association	Measure Used to Assess Outcome	ES original	ES corrected
Simons et al. (b) [99]	2015	321	75%	%06	13.73	Mixed Pain Sample	PCS-C	Physical Functioning	FDI	0.40	0.42
Thastum et al. [109]	2005	56	%08		11.4	Arthritis	PCQ	Pain Intensity	VAS	0.33	0.34
Thastum et al. [110]	1999	16	1			Arthritis	PCQ	Pain Intensity	VAS	-0.02	-0.02
Tran et al. [112]	2015	725	%69	75%		Mixed Pain Sample	PCS-C	Pain Intensity	NRS (0–10)	0.38	0.40
								Physical Functioning	CALQ	0.48	0.52
								Anxiety	SCARED	0.53	0.59
								Quality of Life	PedsQL	-0.50	-0.55
Tran et al. [111]	2017	13	100%	95%	15.94	Juvenile Fibromyalgia	PCS-C	Pain Intensity	VAS	0.57	0.65
								Physical Functioning	FDI	0.50	0.55
								Depression	CDI	0.44	0.47
van Tilburg et al. ^t [113]	2015	189	49%	%88	13.76	IBD	PRI	Physical Functioning	FDI	0.55	0.62
								Depression	CDI	0.54	0.61
		200	73%	%96	11.2	Abdominal Pain		Physical Functioning	FDI	0.42	0.45
								Depression	CDI	0.52	0.58
Verbunt et al. [115]	2015	71	91%		17	Mixed Pain Sample	PCS-C	Pain Intensity	VAS	0.41	0.44
								Physical Functioning	FDI	0.52	0.58
								Depression	CDI	09.0	0.69
Vervoort et al. [119]	2006	43	54%	100%	11.8	Mixed Pain Sample	PCS-C	Anxiety	STAI-C Trait Scale	0.57	0.65
Vervoort et al. [120]	2009	38	76%	100%	15.74	Mixed Pain Sample	PCS-C	Pain Intensity	NRS (0–10)	0.10	0.10
Vervoort et al. [117]	2008	19	57%	%96	13.33	Mixed Pain Sample	PCS	Pain Intensity	VAS	0.32	0.33
Warschburger et	2014	170	64%	NR	11.7	Abdominal Pain	PRCQ-R	Pain Intensity	FPS-R	0.27	0.28

-0.37

ES corrected

ES original

Author Manuscript

Measure Used to Assess Outcome	KINDL-R
Outcome Association	Quality of Life
Catastrophizing Questionnaire	
Chronic Pain Diagnosis	
Mean Age	
Percent White	
Percent Female	
Sample Size	
Year	
ıdy	

^{*}Study reported different Ns for each measure;

Abbreviations: IBD = Inflammatory Bowel Disease, PCS = Pain Catastrophizing Scale (adult version), PCS-C = Pain Catastrophizing Scale for Children, PRCQ-C = Pain-Related Cognitions Questionnaire Revised Child Anxiety and Depression Scale - Major Depressive Disorder subscale, RCMAS = Revised Children's Manifest Anxiety Scale, SCARED = Screen for Child Anxiety-Related Disorders, SCAS Anxiety and Depression Scale - Anxiety subscale, HADS-D = Hospital Anxiety and Depression Scale - Depression subscale, NRS = Numeric Rating Scale, FPS-R = Faces of Pain Scale - Revised, CHAQ = Spence Children's Anxiety Scale, STAL-C State Scale = State-Trait Anxiety Inventory for Children-State scale, STAI-C Trait Scale = State-Trait Anxiety Inventory for Children-Trait scale, VAS = Visual for Children, PCQ = Pain Coping Questionnaire, CSQ-C = Coping Strategies Questionnaire for Children, PRI = Pain Response Inventory, BAPQ - Dss = Bath Adolescent Pain Questionnaire - Depression Depression Test for Children - Dysphoric Mood subscale, DTC-Ess = Depression Test for Children - Exhaustion/somatic complaints subscale, FDI = Functional Disability Inventory, HADS-A = Hospital Childhood Health Assessment Questionnaire, PedsQL = Pediatric Quality of Life, PedsQL - EFss = Pediatric Quality of Life - Emotional Functioning subscale, PedsQL-PFss = Pediatric Quality of Life Adolescent Pain Questionnaire - Physical functioning subscale, CALQ = Child Activity Limitations Questionnaire, CAT.II = Children's Anxiety Test - II, CDI = Children's Depression Inventory, CHQ-Physical Functioning subscale, BAI = Beck Anxiety Inventory, BDI = Beck Depression Inventory, PROMIS - anxiety = PROMIS Anxiety Question Bank, PROMIS - depression = PROMIS Depression CF87 - MHss = Child Health Questionnaire - Mental Health subscale, CS1 = Children's Somatization Inventory, DTC-ABss = Depression Test for Children - Agitated Behavior subscale, DTC-DMss = Question Bank, PROMIS - mobility = PROMIS Mobility Question Bank, RCADS-GADss = Revised Child Anxiety and Depression Scale - Generalized Anxiety Disorder subscale, RCADS-MDDss = subscale, BAPQ - GAss = Bath Adolescent Pain Questionnaire - General anxiety subscale, BAPQ - PSAss = Bath Adolescent Pain Questionnaire - Pain-specific anxiety subscale, BAPQ-PFss = Bath Analog Scale, PRCQ-R = Pain Related Coping Questionnaire - Revised

Study reported separate correlations for boys and girls;

fstudy reported separate correlations by diagnosis subsample

Table 4.

Mean Effect Sizes

Association	K	N	ES (r)	SE	95% C.I.	Z	Q	I ²
Random-effects model								
Pain Intensity	35	4661	0.29**	0.03	[0.24, 0.34]	11.37	85.12**	60.06
Physical Disability	30	4622	0.39**	0.03	[0.35, 0.43]	15.26	72.33**	59.91
Anxiety	15	2867	0.55**	0.03	[0.50, 0.59]	18.96	28.04**	50.07
Depression	19	2691	0.49**	0.04	[0.43, 0.55]	12.58	66.96**	73.12
Quality of Life	10	1724	-0.48**	0.06	[-0.56, -0.39]	-9.32	30.31**	70.31

^{*}p<.05

^{**} p<.01

Continuous Moderator Analyses

Table 5.

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Association	K (N)	β (SE)	95% CI	\mathbf{Z}
Pain Intensity				
Gender	32 (4534)	0.04 (0.14)	[-0.23, 0.31]	0.29
Age	27 (3376)	-0.002 (0.02)	[-0.05, 0.04]	-0.10
Pain Intensity	30 (4246)	-0.0002 (0.02)	[-0.05, 0.04]	-0.01
Pain Duration	20 (3082)	0.001 (0.002)	[-0.002. 0.005]	0.73
Physical Disability				
Gender	29 (4582)	-0.07 (0.16)	[-0.39, 0.25]	-0.42
Age	26 (3696)	-0.004 (0.02)	[-0.05, 0.04]	-0.16
Pain Intensity	25 (3900)	0.03 (0.02)	[-0.01, 0.08]	1.47
Pain Duration	17 (3228)	0.001(0.003)	[-0.005, 0.006]	0.28
Anxiety				
Gender	14 (2798)	-0.06 (0.19)	[-0.43, 0.32]	-0.29
Age	11 (1912)	0.03 (0.04)	[-0.05, 0.12]	0.74
Pain Intensity	11 (2591)	0.02 (0.04)	[-0.05, 0.10]	0.65
Pain Duration	11 (2434)	-0.002 (0.003)	[-0.008, 0.003]	-0.84
Depression				
Gender	17 (2545)	-0.08 (0.18)	[-0.43, 0.27]	-0.46
Age	15 (2384)	-0.001 (0.03)	[-0.05, 0.05]	-0.02
Pain Intensity	14 (1995)	01 (0.04)	[-0.10, 0.07]	-0.33
Pain Duration	11 (1728)	.01 (0.003)	[-0.0003, 0.01]	1.87
Quality of Life				
Gender	9 (1684)	-1.07 (0.62)	[-2.29, 0.14]	-1.73
Age	8 (959)	-0.06 (0.15)	[-0.15, 0.04]	-1.20
Pain Intensity	9 (1718)	0.04 (0.04)	[-0.04, 0.13]	0.96

-0.002 (0.006)

[-0.01, 0.01]

-0.27

Pain Duration

6 (1451)

^{*} p<.05

^{**} p<.01

Table 6.

Categorical Moderator Analyses

Diagnosi	is						
I^2	k(N)		r	95% CI	z(SE)	Q(w)	Q(b)
	30 (4213)		0.30	[0.24, 0.35]	9.90**(0.03)	30.19	1.97
27.59		Mixed Pain (<i>k</i> =20)	0.28	[0.22, 0.35]	7.99 **(0.04)	26.24	
58.33		Arthritis (k=2)	0.34	[0.15, 0.50]	3.48 **(0.10)	2.40	
0.00		Sickle Cell (k=2)	0.18	[-0.06, 0.41]	1.45(0.13)	0.68	
0.00		Abdominal Pain (k=2)	0.35	[0.14, 0.52]	3.22**(0.11)	0.75	
0.00		Musculoskeletal (k=2)	0.37	[0.16, 0.54]	3.40 **(0.11)	0.12	
	27 (4251)		0.39	[0.35, 0.42]	17.18**(0.02)	22.65	7.74
0.00		Mixed Pain (k=20)	0.37	[0.33, 0.42]	13.68**(0.03)	18.59	
0.00		Arthritis (k=5)	0.30	[0.09, 0.48]	2.81 **(0.11)	1.43	
0.00		Sickle Cell (<i>k</i> =2)	0.27	[0.08, 0.45]	2.75 **(0.10)	0.29	
28.06		Abdominal Pain (<i>k</i> =2)	0.50	[0.40, 0.58]	8.48 **(0.06)	1.39	
0.00		Musculoskeletal (k=2)	0.44	[0.29, 0.56]	5.31 **(0.09)	0.95	
	16 (2520)		0.50	[0.44, 0.55]	14.36**(0.04)	14.14	2.52
28.68		Mixed Pain (k=10)	0.50	[0.43, 0.56]		12.62	
0.00		Abdominal Pain (<i>k</i> =2)	0.53	[0.39, 0.65]	6.42 **(0.09)	0.03	
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^{*}p<.05

CAT = catastrophizing, PI = pain intensity, PD = physical functioning, DEP = depression, ANX = anxiety

^{**} p<.01

Table 7.

Categorical Moderator Analyses

Catastrophizi	ng Measu	ire						
Association								
	I^2	k(N)		Г	95% CI	z(SE)	Q(w)	Q(b)
CAT - PI		33 (4434)		0.29	[0.24, 0.35]	10.89*** (0.03)	32.79	3.93
	0.00		PCS-C (<i>k</i> =21)	0.33	[0.26, 0.38]	9.96**(0.03)	17.95	
	0.00		PCQ (<i>k</i> =9)	0.25	[0.15, 0.35]	4.48 **(0.06)	4.80	
	80.08		PCS (<i>k</i> =3)	0.16	[-0.01, 0.33]	1.80(0.09)	10.04	
CAT - PF		30 (4622)		0.39	[0.35, 0.43]	17.00**(0.02)	28.58	5.32
	0.47		PCS-C (<i>k</i> =20)	0.40	[0.35, 0.45]	14.39**(0.03)	19.09	
	7.41		PRI (<i>k</i> =2)	0.49	[0.36, 0.60]	6.81 **(0.08)	1.08	
	30.17		PCQ (<i>k</i> =6)	0.32	[0.21, 0.42]	5.41 **(0.06)	7.16	
	20.00		PCS (<i>k</i> =2)	0.30	[0.21, 0.45]	3.42 **(0.09)	1.25	
CAT - ANX		14 (2807)		0.55	[0.50, 0.59]	18.53 **(0.03)	9.52	0.8
	0.00		PCS-C (<i>k</i> =11)	0.56	[0.51, 0.60]	17.62**(0.04)	9.16	
	0.00		PCQ (<i>k</i> =2)	0.49	[0.42, 0.62]	5.82 **(0.09)	0.36	
CAT - DEP		17 (2574)		0.49	[0.42, 0.55]	9.28 **(0.05)	17.62	0.71
	29.28		PCS-C (<i>k</i> =11)	0.47	[0.38, 0.55]	9.28 **(0.05)	14.14	
	0.00		PRI (<i>k</i> =2)	0.53	[0.35, 0.67]	5.31 **(0.11)	0.02	
	13.29		PCQ (<i>k</i> =4)	0.52	[0.37, 0.65]	5.91**(0.10)	3.46	

^{*}p<.05

CAT = catastrophizing, PI = pain intensity, PF = physical functioning, DEP = depression, ANX = anxiety

^{**} p<.01

Table 8.

Study Quality Moderator Analysis

Association								
	I^2	k (N)		r	95% CI	z(SE)	Q(w)	Q(b)
CAT - PI		34 (4618)		0.29	[0.24, 0.34]	10.92**(0.03)	34.12	0.91
	0.00		QR 5 (k=16)	0.31	[0.26, 0.37]	8.64 **(0.04)	13.87	
	16.05		QR 6 (<i>k</i> =18)	0.26	[0.19, 0.34]	6.74**(0.04)	20.25	
CAT - PF		29 (4579)		0.39	[0.34, 0.43]	15.34**(0.03)	27.85	0.96
	0.00		QR 5 (k=14)	0.41	[0.36, 0.47]	11.36**(0.04)	10.32	
	20.14		QR 6 (k=15)	0.36	[0.31, 0.42]	10.35 **(0.04)	17.53	
CAT - ANX		15(2867)		0.55	[0.55, 0.65]	19.01**(0.03)	9.95	0.66
	0.00		QR 5 (k=10)	0.56	[0.56, 0.60]	17.09**(0.04)	8.96	
	0.00		QR 6 (<i>k</i> =5)	0.52	[0.44, 0.60]	8.58 **(0.07)	0.99	
CAT - DEP		19(2691)		0.49	[0.46, 0.55]	13.47 **(0.04)	19.84	0.30
	11.42		QR 5 (k=10)	0.51	[0.45, 0.58]	10.19**(0.06)	10.16	
	0.00		QR 6 (<i>k</i> =19)	0.48	[0.40, 0.56]	8.82 **(0.06)	9.68	
CAT - QOL		10 (1724)		-0.48	[-0.62, -0.39]	-9.50*(0.05)	7.84	1.37
	5.96		QR 5 (<i>k</i> =4)	-0.42	[-0.61, -0.29]	-5.82*(0.08)	3.19	
	0.00		QR 6 (<i>k</i> =6)	-0.52	[-0.73, -0.41]	-7.60**(0.08)	4.65	

^{*} p<.05

CAT = catastrophizing, PI = pain intensity, PF = physical functioning, DEP = depression, ANX = anxiety, QOL = quality of life

^{**} p<.01

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Table 9.

Comparison of Mean Effect Sizes

Association									
Random-effects model	K	N	ES (r)	SE	95% C.I.	Z	Q	I^2	
Full Sample									
Pain Intensity	35	4661	0.29**	0.03	[0.24, 0.34]	11.37	85.12**	60.06	
Physical Disability	30	4622	0.39**	0.03	[0.35, 0.43]	15.26	72.33**	59.91	
Anxiety	15	2867	0.55**	0.03	[0.50, 0.59]	18.96	28.04**	50.07	
Depression	19	2691	0.49**	0.04	[0.43, 0.55]	12.58	66.96**	73.12	
Quality of Life	10	1724	-0.48 **	0.06	[-0.56, -0.39]	-9.32	30.31**	70.31	
18 and under Sample									
Pain Intensity	30	4369	0.30**	0.02	[0.27, 0.33]	12.88	55.86**	48.08	
Physical Disability	26	4422	0.39**	0.03	[0.34, 0.43]	14.65	65.28**	61.70	
Anxiety	12	2606	0.55 **	0.04	[0.49, 0.60]	16.07	27.99**	60.70	
Depression	15	2359	0.49**	0.05	[0.41, 0.56]	10.62	62.77**	77.70	
Quality of Life	9	1664	-0.47**	0.06	[-0.55, -0.37]	-8.56	28.53**	71.96	
Exclusively Chronic Pain	n (3+ 1	nonths)							
Pain Intensity	27	2678	0.27**	0.03	[0.21, 0.34]	8.17	67.46**	61.46	
Physical Disability	22	2374	0.36**	0.04	[0.30, 0.42]	10.57	51.89**	59.53	
Anxiety	11	1207	0.58**	0.04	[0.53, 0.62]	18.29	12.44**	19.61	
Depression	17	1799	0.51 **	0.04	[0.45, 0.57]	13.11	41.67**	61.60	
Quality of Life	9	999	-0.48 **	0.07	[-0.58, -0.36]	-7.36	27.74**	71.16	

^{*}p<.05

^{**} p<.01