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**Parent cognitive, affective, and behavioral responses in the context of their child's chronic pain: A systematic review and meta-analysis of measures**

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### Abstract

**Objective.** Parent responses can have a major impact on their child's pain. The purpose of this systematic review is to (1) identify and describe measures assessing pain-related cognitive, affective, and behavioral responses in parents of children with chronic pain; and (2) meta-analyze reported correlations between parent constructs and child outcomes (i.e., pain intensity, functional disability, school functioning). Prospero protocol registration ID: CRD42019125496. **Methods.** We conducted a systematic search of studies including a measure of parent/caregiver responses to their child's chronic pain. Study characteristics and correlations between parent measures and child outcomes were extracted. Data were summarized and meta-analyzed. **Results.** Seventy nine met inclusion criteria using 18 different measures of cognitive/affective (n=3), behavioral (n=5), and multidimensional responses (n=10). Measures were used a median of 3 times (range 1-48), predominantly completed by mothers (88%), and primarily in mixed pain samples. Psychometrics of measures were generally adequate. Meta-analyses were based on 42 papers across 5 measures. Results showed that each of the cognitive, affective, and behavioral parent constructs we examined was significantly associated with pain-related functional disability. A small number of measures assessing parent cognitions and affective functioning were associated with higher child pain intensity, however the majority were not. **Conclusion.** Findings demonstrate that there is a wealth of measures available, with adequate reliability overall but a lack of psychometrics on temporal stability. Synthesizing data across studies revealed small effects between parent responses and child functioning, and even smaller and/or absent effects on child pain intensity.

**Key words:** parents, chronic pain and recurrent pain, family functioning, meta-analysis

### **Introduction**

Chronic pain (i.e., pain lasting more than three months; Merskey, 1994) is a common experience during childhood (King et al., 2011), negatively impacting academic, social, emotional, and behavioral functioning (Fales, Essner, Harris, & Palermo, 2014; Gaughan, Logan, Sethna, & Mott, 2014; Goubert, Eccleston, Vervoort, Jordan, & Crombez, 2006; Jordan, Eccleston, McCracken, Connell, & Clinch, 2008; Palermo, Valrie, & Karlson, 2014; Simons, Smith, Kaczynski, & Basch, 2015). Children with chronic pain also experience significant functional impact, which extends to family members (Palermo & Chambers, 2005). Over the last several years, there has been increased attention toward parent cognitive, affective, and behavioral responses in the context of their child's pain, and how this contributes to the child's pain experience and associated disability.

Historically, two frameworks, the biopsychosocial model and the ecological model, have been used to better understand the experience of chronic pain and disability in children. Developed in part from a social learning theory perspective, the biopsychosocial model (Fordyce et al., 1973) suggests that biological, psychological, and social factors are all potential contributors to the child's experience of pain and related disability. The ecological model (Bronfenbrenner, 1979), influenced by family systems theory, emphasizes the individual's behavior within the context of family situations. Although components of these theories have been applied to pediatric chronic pain populations, these models generally lack an examination of specific pathways by which family factors may influence children's pain and associated disability.

In an attempt to overcome these limitations, intergenerational and interpersonal models have been developed by integrating the aforementioned frameworks and focusing on the bidirectional relationships between the child's pain experiences as well as the influence that parent

and family variables have on chronic pain and related disability (Goubert & Simons, 2013; Palermo & Chambers, 2005; Palermo et al., 2014). Palermo and Chambers (2005) proposed an integrative model of parent and family factors in pediatric chronic pain. Within this model, individual parenting variables (e.g., parenting style, parental reinforcement) are situated within a broader context of dyadic variables (e.g., quality of parent-child interaction), which is further embedded within the more global familial environment. This model highlights the shared influence of pain and related disability, as well as child and parent variables, interacting with individual, dyadic, and family variables as they relate to pediatric chronic pain. More recently, the Interpersonal Fear Avoidance Model of Pain (IFAM; Goubert & Simons, 2013; Simons et al., 2015) was put forth. This model further emphasizes parent cognitive-affective and behavioral variables and their impact on child functioning. Within the IFAM, parents interpret their child's pain expression through the lens of their own catastrophic appraisals and pain-related fears, leading some to engage in parenting behaviors that may result in negative consequences for the child (e.g., functional decline).

There is building evidence to support interpersonal models. For example, parents report significant distress as a result of parenting a child with chronic pain (Fales et al., 2014; Gaughan et al., 2014; Goubert et al., 2006; Jordan et al., 2008; Palermo et al., 2014; Simons et al., 2015). Moreover, certain parent cognitions and affective responses (e.g., catastrophizing, fears related to their child's pain) are associated with poorer pain-related outcomes in children (Chow, Otis, & Simons, 2016; Fales et al., 2014; Logan, Simons, & Carpino, 2012; Palermo et al., 2014; Simons et al., 2015). Parent behavioral responses, including protective behaviors, minimizing, and encouragement/reinforcement of symptoms, have also been associated with poorer child outcomes, including increased pain intensity and functional disability (Noel et al., 2015; Peterson

& Palermo, 2004; Sieberg, Williams, & Simons, 2011). While these parental responses are understandable, normative, and care-promoting in the context of acute pain, prolonged and high levels of distress and protective behaviors within the context of chronic pain is maladaptive. On the other hand, other cognitive and behavioral responses (e.g., psychological flexibility, pain acceptance) might serve as resilience factors by promoting child functioning through effects on parent behaviors as well as on child coping (Gauntlett-Gilbert, Alamire, & Duggan, 2018; Kanstrup et al., 2016; McCracken & Gauntlett-Gilbert, 2011; Timmers, Simons, Hernandez, McCracken, & Wallace, 2019; Wallace, Woodford, & Connelly, 2016).

Taken together, there is accumulating evidence on the instrumental influence of parent pain-related cognitive, affective, and behavioral responses on their child's pain experience and associated functioning. Given this, sound assessment of parent responses in the context of their child's pain is important for building upon current research, as well as identifying treatment targets within this population. Thus, it is timely to systematically review measures assessing these parent variables. This systematic review has two aims: (1) identify measures used to assess parent pain-related responses to their child's pain, summarizing their use and psychometric properties; (2) meta-analyze correlations between parent constructs and child outcomes (i.e., pain intensity, functional disability, school impairment). In relation to the second aim, we hypothesize that higher parent cognitive and affective distress is associated with higher child pain intensity, functional disability, and school impairment. We also hypothesize that higher reports of parents' protective, solicitous, monitoring, and minimizing behavioral responses are associated with higher child pain intensity, functional disability, and school impairment. Additionally, we hypothesize that higher parental psychological flexibility and pain acceptance will be associated with lower child pain intensity, functional disability, and school impairment.

## Method

### Inclusion Criteria

We included studies with samples of children and adolescents (<18 years in age) with chronic pain and a parent, guardian, or caregiver. All chronic pain conditions, including but not limited to headache, abdominal pain, neuropathic pain, and musculoskeletal pain were included. We excluded studies that exclusively recruited disease-related pain conditions (e.g., cancer, sickle-cell, juvenile idiopathic arthritis) or reported acute pain or post-operative pain samples. While we believe that parent responses to child's pain are also relevant for children with disease-related pain, there are likely a number of additional variables that drive parent responses and emotional functioning in the context of their child's disease-related pain that are not captured by the more traditional interpersonal models. We included cross-sectional, longitudinal, and treatment outcome studies. For longitudinal and treatment outcome studies, only baseline data were extracted. We did not include qualitative studies, expert opinion articles, reviews, case studies/reports, or meta-analyses. We only included studies from peer-reviewed journals, published in English.

Studies had to include parent or guardian self-report measures of cognitive, affective, and/or behavioral responses in the context of their child's chronic pain. For the purpose of the current review, we conceptualized parent cognitive and affective factors as pain-related anxiety, depression, self-blame/helplessness, catastrophizing, fear of pain, pain acceptance, and psychological flexibility. Parent behavioral responses were conceptualized as protective, solicitous, helpless, minimizing, and/or avoidant behaviors. We did not include parent-proxy measures. While many studies include general measures of psychological functioning for parents (e.g., Beck Depression Inventory; Beck, Ward, Mendelson, Mock, & Erbaugh, 1961), we only included measures assessing parent distress and behavioral responses in the context of parenting a

child with chronic pain, as opposed to overall parent psychological functioning. A protocol for this systematic review was registered on PROSPERO (ID: CRD42019125496).

### **Search methods**

Searches were conducted through Pubmed, EMBASE, PsycINFO, and CINAHL databases from inception to February 13, 2019. The search terms were generated in collaboration between the research team and a biomedical librarian and systematic review expert from the Lane Medical Library at Stanford University School of Medicine. Search terms included terms relating to chronic pain, parents/caregivers, family, and questionnaire/assessment. Specifically, we searched for papers that included any of the questionnaires most commonly used to assess the constructs of interest, in addition to papers that included terms relating to chronic pain, children, parent/caregivers, and validation/assessment (see Supplemental Materials 1 for full search strategy). Following the initial search, we inspected reference lists of included studies for additional papers and measures. Results of the database searches were uploaded to Covidence (<http://covidence.org>), a web-based application that facilitates screening and analyzing studies for systematic reviews. Three independent reviewers screened all titles and abstracts (LEH, LCH, and IT), with the first reviewer screening all abstracts and the other two reviewers screening half of the abstracts each. The three reviewers also discussed papers for inclusion and read full texts. Disagreements were resolved between the authors.

### **Data Extraction**

**Study and measure characteristics.** We extracted study characteristics including sample size, child age, child sex, chronic pain condition, caregiver age, caregiver sex, caregiver relationship to the child, author group, and country of study origin. For each measure that assessed one of our target constructs, we extracted the name, number of items, and subscales names.



Measures were categorized into “Parent pain-related cognitive and affective responses”, “Parent behavioral responses” or “Multidimensional measures of parent pain-related responses” (i.e., having components of pain-related cognitive, affective, and/or behavioral responses). Psychometric properties (i.e., alpha coefficients, test-retest reliability correlations) of the measures were also extracted, where reported. We identified overlapping samples by identifying multiple papers from the same research lab and/or author groups, noting similar recruitment dates, and assessing whether or not the sample size and descriptive statistics were matching. We clarified overlapping samples with the author(s) where possible. When overlapping samples were identified, we classified one paper as primary based on the largest sample size. Secondary studies were included if they reported on measures and correlations not included in the primary paper.

**Outcomes.** Meta-analyses were conducted by extracting reported bivariate correlations between the measures of parent constructs and child outcomes (i.e., child pain intensity, functional disability, school functioning). Functional disability was assessed by the Functional Disability Inventory (Walker & Greene, 1991) and the Child Activity Limitations Interview (Palermo et al., 2004); school functioning was assessed by the Pediatric Quality of Life Inventory-School Functioning Subscale (Peds QL School Functioning; Varni et al., 2001). All outcomes were child-report. Parent-proxy reports of child outcomes were not included. For studies that included both clinical and community samples, data were extracted from the chronic pain sample (when reported separately).

### **Data Analysis**

First, we summarized all identified measures that were used to assess parent cognitive, affective, and behavioral responses in the context of their child’s chronic pain and described their use and psychometric properties. Second, we pooled all reported correlations between parent

measures and child outcomes. When data were not reported in the paper, we contacted authors to request correlational data and allowed 6 weeks for them to reply data. Data of samples < 20 were not included in the meta-analysis. We performed meta-analyses where we were able to pool together data from two studies or more (Deeks et al., 2011).

We conducted meta-analyses in STATA, using the DerSimonian–Laird random effects method (DerSimonian & Laird, 1986). This method assumes that studies are heterogeneous. We interpreted heterogeneity ( $I^2$ ) following Cochrane Review methodology (Higgins & Green, 2011): 0–40% might not be important; 30–60% moderate heterogeneity; 50–90% substantial heterogeneity, and 75–100% considerable heterogeneity. Trim and fill analyses (Duval & Tweedie, 2000) and Rosenthal’s failsafe N (Rosenthal, 1979) were conducted to detect potential publication bias in analyses where more than 10 studies were included.

## Results

### Search Results

Search of the databases yielded 5,805 papers. An additional 239 papers were identified through searching reference lists (see Figure 1 for PRISMA flow chart). After duplicates were removed, we screened 4,188 titles and abstracts, we read 320 full papers, and we found 79 papers that met full inclusion criteria. There were 36 papers reporting correlation data in the manuscript. We attempted to contact the authors of the remaining (n=44) papers to obtain correlational data, resulting in 16 additional papers. In total, 52 papers (65% of those eligible) were included in the meta-analyses from 33 separate studies, with unique correlational data extracted from all 52 papers. Twenty-two of these papers were identified as having overlapping samples, and only data

reporting on non-overlapping measures were retained for meta-analyses (i.e., all studies reported on at least one correlation that was not overlapping<sup>1</sup>, thus data were extracted from all papers).

### **Included Studies**

The 69 studies from 79 papers that met inclusion criteria comprised 16,642 children and adolescents with chronic pain (11,657 female; two studies did not report sex of the child) and at least one parent or caregiver ( $n = 16,515$ ). Parents were mostly mothers (on average 66%, ranging from 50-100%), although there were 5 studies that included grandparents or other caregiver (18 studies did not report sex of the parents). The mean age of children and adolescents was 13.71 ( $SD = 2.08$ ; 1 study did not include data on child age). The mean age of parent-responders was 48.32 ( $SD = 6.20$ ; 54 studies did not include data on parent age). The majority of studies included mixed chronic pain samples, including youth with musculoskeletal pain, neuropathic pain, headache, migraine, and abdominal pain. Eleven studies included samples of only abdominal pain, two studies included only headache, and two studies included only musculoskeletal pain. See Supplemental Materials (S2) for a full overview of studies and their characteristics.

### **Aim 1: Summary of Measures Assessing Parent Cognitive, Affective and Behavioral Responses**

Across the 70 studies reported in 79 papers that met inclusion criteria, we found 18 separate measures assessing parent cognitive, affective, and behavioral responses within the context of

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<sup>1</sup> For example, Chow et al. (2016) described (partly) the same sample as Simons et al. (2015). As the sample in Simons et al. was larger, data reported by both Simons et al. and Chow et al. were only extracted from Simons et al. (e.g., correlation between PCS-P and child pain intensity). Data reported by Chow et al. that were not included in Simons et al. (e.g., correlation of PCS-P Magnification subscale with Peds QL School Functioning) were extracted and included in the analyses.

parenting a child with chronic pain. Of these, 3 measures assessed parent cognitive and affective responses, 5 assessed parent behaviors, and 10 measures were mixed (assessing cognitive, affective responses and behaviors). The following sections describe those measures that are most commonly used (i.e., reported in > 5 papers). See Table 1 for a description of all included measures, and Table 2 for an overview of the use and reported psychometrics.

**Parent Pain-Related Cognitive and Affective Responses.** Three measures assessed parent cognitive and affective responses (and not behavioral responses). These included the constructs pain catastrophizing, pain-related self-efficacy, pain beliefs, and injustice experiences. One measure in this category (Parental Pain-Related Self-Efficacy) focused on children with abdominal pain, while the other two (Pain Catastrophizing Scale, Parent Version, and Injustice Experience Questionnaire, Parents in Reference to Child) included mixed pain samples. The most commonly used measure was the Pain Catastrophizing Scale, Parent version (PCS-P; Goubert et al., 2006), used in 35 studies. The PCS-P assesses parent catastrophizing about their child's pain (e.g., *“when my child is in pain, I worry all the time about whether the pain will end”* *“when my child is in pain, it's awful and I feel that it overwhelms me”*). This measure comprises three subscales: Rumination, Magnification, and Helplessness. A total or subscale score is derived by summing items; higher scores indicate higher levels of catastrophic thinking. Across studies, Cronbach's Alpha for the total score of the PCS-P was Good to Excellent ( $\alpha = .87-.96$ , numbers of studies  $k = 22$ ;  $\alpha = .91$  in the measure development paper).

**Parent Pain-Related Behavioral Responses.** Five measures were identified as exclusively assessing parent behavioral responses (and not cognitive and/or affective responses). The measures included constructs such as protective behaviors, solicitous responding, distraction from pain, and monitoring of pain symptom complaints and behaviors. All measures assessed

mixed pain samples, except for the Parent Response Scale, which was used in children with headache. The most widely used measure in this category was the Adult Responses to Children's Symptoms (ARCS; Claar, Guite, Kaczynski, & Logan, 2010; Noel et al., 2015; Van Slyke & Walker, 2006; Walker, Levy, Walker, Levy, & Whitehead, 2006), used in 48 studies. The second most widely used measure was the Helping for Health Inventory (HHI; Fales et al., 2014), used in seven studies.

The ARCS assesses multiple behavioral responses to the child's pain. The original factor structure of the ARCS supports a three-factor measure consisting of Protect, Encourage/Monitor, and Minimize subscales (Claar et al., 2010; Walker et al., 2006). Items are averaged for total subscale scores, with higher scores indicating higher engagement in that behavioral response. More recently, Noel and colleagues (2015) reexamined the factor structure for child and adolescent samples separately and found evidence to support two new four-factor models, one in a child-only sample (ages 7-11) and one in a combined child-adolescent sample (ages 7-18), as well as a five-factor model for an adolescent-only sample (ages 12-18; Noel et al., 2015). The number of items in subscales across different factor structures differ (see Table 1). Across the 48 studies, there is substantial variation in how the measure is used and scored. Only one study calculated a total ARCS score and found the internal consistency to be Good ( $\alpha = .84$ ) (Walker et al., 2006); this study did not report on descriptive statistics for the total ARCS score. Some studies reported summed subscale scores, while others report average scores (as recommended). Further, some studies use the original factor structure (also after the publication of the new structure), while others use a more recent one (e.g., Noel et al., 2015). Also, most studies use only one or two subscales, while others use all subscales (see S2 for more details). The most utilized subscale is the Protect subscale, used in 43 studies (of which  $k = 30$  or 70% of studies reported an average

score as recommended). Internal consistencies for the Protect subscale ranged from fair to excellent ( $\alpha = .70-.92$ ,  $k = 32$ ;  $\alpha = .84$  in measure development paper).

The Helping for Health Inventory (HHI; Fales et al., 2014)), used in seven studies, assesses miscarried helping (i.e., parental behaviors intended to be helpful, but inadvertently reinforcing pain behaviors and functional disability). Items are summed to create a total score; higher scores indicating greater miscarried helping. Reported internal consistencies were considered Good ( $\alpha = .82-.88$ ,  $k = 5$ ).

**Multidimensional Measures of Parent Pain-Related Responses.** We identified 10 measures that assessed both parent cognitive/affective and behavioral responses across the different subscales. Of these measures, most have been utilized within mixed pain populations; however, the Parental Pain-Related Behavior measure has only assessed abdominal pain samples. The most commonly used measures were the Bath Adolescent Pain-Parent Impact Questionnaire (BAP-PIQ; Jordan et al., 2008; used in 11 studies), and the Parent Fear of Pain Questionnaire, the Parent Psychological Flexibility Questionnaire (PPFQ; McCracken & Gauntlett-Gilbert, 2011; Wallace, McCracken, Weiss, & Harbeck-Weber, 2015; used in 7 studies).

The BAP-PIQ assesses parent pain-related distress and behavioral responses across eight domains: Depression, Anxiety, Child-Related Catastrophizing, Self-Blame and Helplessness, Partner Relationships, Leisure Functioning, Parental Behaviors, and Parental Strain. The authors recommend using subscale scores only (Jordan et al., 2008). Total scores for each domain are calculated by summing all items, with higher scores indicating higher level of distress or behavioral response for each domain. Internal consistencies across subscales were questionable to excellent (ranging from  $.62-.92$ ,  $k = 11$ ). The measure development paper also reported data on test-retest reliability, ranging from  $.70-.80$  (see Table 2).

The Parent Fear of Pain Questionnaire (P-FOPQ; Simons et al., 2015) assesses a parents' fear and avoidance of activities in relation to their child's pain (e.g., "*My child's feelings of pain are scary for me,*" "*I avoid making plans because of my child's pain*"). The P-FOPQ contains 4 subscales: Avoidance, Fear of Pain, Fear of School, and Fear of Movement, of which the first two were most often used in the included studies. Items are summed to create a total score, with higher scores indicating greater parent fear of their child's pain. Cronbach's alphas for the total score were Good to Excellent across studies ( $\alpha = .84-.97$ ,  $k = 3$ ;  $\alpha = .91$  in the measure validation paper), with alphas for subscales ranging from acceptable to good ( $\alpha = .69-.88$ ,  $k = 2$ ; see Table 2).

The Parent Psychological Flexibility Questionnaire (PPFQ; McCracken & Gauntlett-Gilbert, 2011) assesses parents' choice of valued-based behavior even in the presence of their own distress associated with their child's pain (e.g., "*I suffer terribly from my child's pain and need to make this suffering stop*" "*it is possible to live a normal life while my child suffers with pain*"). Select items are reversed scored and a total score indicates greater flexibility. The original 24-item version of the PPFQ was used in one study (McCracken & Gauntlett-Gilbert, 2011), internal consistency for the total score was Excellent ( $\alpha = .91$ ). While the original list had no subscales, short forms have identified a four-factor structure (PPFQ-17; Values-Based Action, Pain Acceptance, Emotional Acceptance, and Pain Willingness; Wallace et al., 2015) and a three-factor structure (PPFQ-10; Values-Based Action, Pain Acceptance, Emotional Acceptance; Timmers et al., 2019; Wiwe Lipsker, Kanstrup, Holmstrom, Kemani, & Wicksell, 2016). Four studies utilized the PPFQ-17, all reporting data for a total score, while one study reported additional descriptive statistics for each subscale. Two studies utilized the 10-item version of the PPFQ, with both studies reporting a total score and one also reporting descriptive statistics for the subscales. Across versions, the internal consistencies reported for the total score ranged from Good to Excellent ( $\alpha$

= .82-.91,  $k = 5$ ), with alphas for the subscales ranging from questionable to good ( $\alpha = .60-.88$ ,  $k = 2$ ; see Table 2).

### **Aim 2: Correlations Among Parent Pain-Related Responses and Child Outcomes**

Of the 79 papers included, correlational data between a parent measure and at least one child outcome variable was extracted from 42 papers ( $n=30$  reported in paper,  $n=12$  obtained directly from authors). Reported data were sufficient to allow meta-analyses for correlations between the PCS-P (total score and Helplessness and Magnification subscales), the ARCS (Protect, Minimize, Distract, and Encourage/Monitor subscales), and the PPFQ total score with child outcomes (i.e., pain intensity, functional disability, school impairment; in various combinations). Results will be discussed per measure below. A visual overview of results, as well as of all conducted analyses, is presented in Figure 2. Table 3 also presents an overview as well as all details of meta-analyses results. Note that for measures not included in these tables (but included in Table 1 and 2), there was insufficient data to conduct analyses. See Supplementary Materials (S3) for all individual meta-analysis tables and forest plots and (S4) for trim and fill plots.

#### **Measures of parent pain-related cognitive and affective responses.**

*Pain Catastrophizing Scale for Children-Parent Report.* We found a very small, but significant, positive effect between parent pain catastrophizing (PCS-P total score) and child pain intensity (effect size (ES) = 0.15; 95% confidence interval (CI) 0.10 to 0.19;  $k = 16$ ) and a small effect between PCS-P total and child functional disability (ES = 0.27; 95% CI 0.20 to 0.32;  $k = 19$ ).

Publication bias analyses were conducted for the analysis of PCS-P and pain intensity. Analyses indicated that 3 studies may have been missing from the pain intensity analysis (see S4).



Despite this, overall effects were found to be similar to effect sizes noted in the meta-analyses (Table 3; S4). The fail-safe  $N$  in the analysis examining associations between PCS-P and pain intensity was  $n = 29$ .

Regarding the PCS-P subscales, we found very small but significant positive effects between PCS-P Helplessness and child pain intensity (ES = 0.18; CI = 0.10 to 0.27;  $k = 2$ ) and between PCS-P Magnification and child pain intensity (ES = 0.15; CI = 0.03 to 0.27;  $k = 2$ ). There were insufficient data to conduct additional analyses.

***Bath Adolescent Pain – Parental Impact Questionnaire: cognitive/affective subscales***

*Depression subscale.* We found a very small, significant, positive effect between the Depression subscale of the BAP-PIQ and child pain intensity (ES = 0.11; 95% CI = 0.02 to 0.20;  $k = 3$ ). *Anxiety subscale.* We observed a very small, significant, positive effect between the Anxiety subscale and child pain intensity (ES = 0.13; 95% CI = 0.04 to 0.22;  $k = 3$ ). *Self-Blame and Helplessness subscale.* A small significant, positive effect was found for the Self-blame and helplessness subscale and child functional disability (ES = 0.25; 95% CI = 0.15 to 0.34;  $k = 3$ ). There was insufficient data to conduct additional.

**Measures of parent pain-related behavioral responses.**

*Adult Responses to Children's Symptoms.* A small, significant, positive effect was found between the ARCS Protect and child functional disability (ES = 0.30; 95% CI = 0.21 to 0.38;  $k = 6$ ). No significant effect between the ARCS Protect and child pain intensity (ES = 0.04; 95% CI = -0.01 to 0.09;  $k = 20$ ) was observed. No significant effect was observed between ARCS Protect and child activity limitations (ES = 0.07; 95% CI = -0.10 to 0.24;  $k = 4$ ).

Publication bias analyses were conducted on one of the outcome variables (pain intensity). Analyses indicated that 1 study may have been missing from the pain intensity analysis (see S4).

Despite this, overall effects were found to be similar to effect sizes noted in the meta-analyses (Table 3; S4). The fail-safe  $N$  in the analysis examining associations between ARCS Protect and pain intensity was  $n = 25$ .

A very small, significant, positive effect was found between the ARCS Encourage/Monitor and child functional disability (ES = 0.14; 95% CI = 0.07 to 0.21;  $k = 4$ ). No significant effect was found between the ARCS Encourage/Monitor and child pain intensity (ES = -0.02; 95% CI = -0.12 to 0.09;  $k = 7$ ). No significant effects were found between ARCS Minimize and child pain intensity (ES = 0.01; 95% CI = -0.04 to .07;  $k = 6$ ) or child functional disability (ES = 0.03; 95% CI = -0.03 to 0.08;  $k = 6$ ). Also, no significant effect was found between ARCS Distract and child pain intensity (ES = -0.02; 95% CI = -0.17 to 0.13;  $k = 3$ ). There were insufficient data to conduct additional analyses.

***Helping for Health Inventory.*** A very small, significant, negative effect was found between HHI and child pain intensity (ES = -0.09; 95% CI = -0.18 to -0.01;  $k = 3$ ). Due to insufficient data, we were unable to conduct analyses between the HHI and other child outcomes.

#### **Multidimensional Measures of Parent Pain-Related Responses.**

***Parent Psychological Flexibility Questionnaire.*** No significant effects were found between the PPFQ Total score and child pain intensity (ES = -0.07; 95% CI = -0.18 to 0.04;  $k = 3$ ). No other analyses could be conducted due to insufficient data.

### **Discussion**

The purpose of this systematic review was to identify and summarize the different measures developed to assess parent cognitive, affective, and behavioral responses in the context of parenting a child with chronic pain. We provide a comprehensive list, by domain, of pain-related measures that are used in this context, their characteristics, and psychometric properties. As such,

our work can serve as a roadmap for researchers designing studies examining these constructs in the future, or for clinicians assessing these constructs in their practice. Over the last decade, there has been increased attention toward these parent variables with evidence suggesting a direct association with their child's pain-related functioning (e.g., Lewandowski, Palermo, Stinson, Handley, & Chambers, 2010; Palermo & Chambers, 2005; Palermo et al., 2014). Our second aim was therefore to synthesize data examining correlations between the parent measures and child outcomes.

### **What measures are out there?**

We identified 18 measures across 79 papers used to assess parent cognitive, affective and/or behavioral responses to their child's pain. While we identified several pain-related measures assessing strictly cognitive/affective (e.g., pain catastrophizing) or behavioral constructs (e.g., protective behavior), the majority of measures assess a combination of cognitive, affective and behavioral responses across different subscales (e.g., fear of pain and avoidance subscales of the P-FOPQ). Across all measures and subscales, a variety of constructs were covered, including fear of pain, pain acceptance, pain avoidance, psychological flexibility, and the impact of pain on the parent and family unit (e.g., partner relations, financial burden). There seem to be no obvious content gaps in available measures that assess parent distress about their child's chronic pain and behaviors. Only one measure, however, - the BAP-PIQ (Jordan et al., 2008) - assessed symptoms of parent mood (e.g., anxiety and depression) within the context of their child's pain. To date, most studies examining parent psychological functioning within this population have utilized general measures of psychological symptoms (e.g., Beck Depression Inventory; Beck et al., 1961), which are widely used and validated to assess psychological symptoms. Nevertheless, using a condition-specific measure like the BAP-PIQ will allow us to better understand how these psychological

symptoms present in relation to their child's pain specifically. This differentiation is important, as it might be useful in identifying which psychological symptoms seem to be most relevant to how they interact with their child, and thus may be useful for identifying treatment targets.

Most of the measures have been specifically developed in the context of pediatric chronic pain (as opposed to other chronic illness populations) or were modified from an existing adult or parent proxy measure, meaning several different, but related versions of measures exist, and this may not always be obvious to the clinician or researcher utilizing the measures. One example is the parent proxy versus the parent report of the Fear of Pain Questionnaire (FOPQ), which are referred to as the FOPQ-P (parent proxy) and P-FOPQ (parent report of their own fear about their child's pain). While one assesses the child's fear and avoidance behavior as reported by their parents, the other assesses the parent's own fear and avoidance in relation to their child's pain. It is important to be wary of such differences, and one of our intentions with the detailed summary of the measures (Table 1) is to increase awareness of the different measures and their related versions. With regard to psychometric properties, Cronbach alpha was the most often reported measure of internal consistency, with most measures showing adequate levels, indicating that the measures hold-up well across different pain samples and settings. We only found data on test-retest reliability for a few measures, indicating we still have little information on the temporal stability of the measures.

The ARCS was the mostly widely used measure across all domains. However, we found inconsistencies in how the measure is administered and scored. For example, there is variability in whether studies utilize the original three-factor structure (Protect, Encourage/Monitor, and Minimize), or the more recently developed four-factor or five-factor models (Noel et al., 2015). Moreover, it is often unclear which factor structure was used to create the subscale scores. The

original ARCS validation paper (Walker et al., 2006) suggests calculating average scores for each subscale, however, approximately one third of the studies reported sum scores.

The length of measures varied significantly, with half of the measures being short to moderate in length (number of items ranging from 9-21 items) and half of the measures being longer (25-62 items). Of note, however, the multidimensional 62-item measure (BAP-PIQ; Jordan et al., 2008) is unique in that it comprises individual domains (each with a maximum of 11 items) which are intended to be scored and interpreted independent of each other. Beyond this, it was also noticeable that effort has also been put forth to develop short forms for several measures, which also reduces burden for parents and family members completing the measures.

### **Meta-analyses of correlations between parent and child measures**

We found very small to small effects between all parent constructs examined and child functional disability, except for minimizing behaviors. These findings provide support for the association between parent cognitive, affective, and behavioral responses on pain-related disability and support the theoretical model put forth within the IFAM (Goubert & Simons, 2013; Simons et al., 2015). This is in accordance with the ample literature on the importance of parent factors in the context of their child's pain, as well as on the impact of parent-targeted interventions on their child's functioning. Our findings add important additional information, as they highlight that when synthesizing individual studies, effect sizes are relatively small, and seem to be greater for pain-related functioning compared to pain intensity. It is also clear that although there are ample data available, most studies utilize the same measures providing valuable information on those measures, while the available information on other measures is still very limited.

Based on our meta-analytic findings, it seems that parent effects on their child's functioning are greater than on their child's pain intensity. While virtually all parent measures

were significantly related to their child's functional disability, measures assessing cognitive/affective distress were significantly related to child pain intensity, although the effects were small. None of the subscales of the ARCS, nor parent psychological flexibility, were associated with child pain intensity. This suggests that the parent behavior associated with child functioning is stronger than the influence on pain intensity, which fits with current models of chronic pain, emphasizing that the child's functioning is not contingent on pain intensity (Simons et al., 2015; Walker et al., 2006; Wilson, Moss, Palermo, & Fales, 2014). On the other hand, it also suggests that cognitive and affective responding and emotional functioning is important to consider in relation to child pain expression. Even though our analyses did not consider changes over time, it is plausible that targeting these parent constructs in treatment will have positive effects on child functioning. Indeed, several studies have actively targeted parents in treatment and have shown positive effects, not only on parent variables, but also on child outcomes (Kanstrup et al., 2016; Wallace et al., 2016). The underlying mechanisms of change should be a topic of future studies.

**Limitations.**

Our findings should be interpreted in light of some limitations. First, we did not examine whether associations between parent variables and child outcomes were mediated by child distress and behaviors. We recognize that this approach may have missed other missing variables that may be more strongly associated with the examined parent variables (i.e., child distress and behavior). Second, we extracted reported indices of test-retest reliability but did not extract additional important psychometric indices, such as sensitivity to change. While extracting data on sensitivity to change was beyond the scope of this review, this is an important property for any measure used in a longitudinal or intervention study. Third, while our search strategy was developed by an

experienced science librarian and written to capture all necessary papers, we may have missed some papers that use less common measures, or where the measure was not included in the title, abstract, or key words. Lastly, because we restricted our search to “English only” we could not give a comprehensive overview of other languages in which the discussed measures might be available.

### **Implications for Clinical Practice**

This study provides several implications for clinical practice. Most interventions deliver cognitive behavioral therapy only to children and adolescents with pain, (e.g.,(e.g.,Eccleston, Yorke, Morley, Williams, & Mastroyannopoulou, 2003). When parents are incorporated into treatment, the focus of their involvement has centered on modifying parent behaviors (i.e., solicitous/protective behaviors) in an attempt to increase functioning in the child (Levy et al., 2010). A recent review found that psychological interventions delivered to parents of children with chronic pain improved parent behaviors and mental health (Law, Fisher, Eccleston, & Palermo, 2019). Our findings support theoretical models suggesting that both parent cognitive/affective and behavioral variables impact child functioning (Simons et al., 2015). In fact, even though there were significant associations between parent variables and child functioning, only pain catastrophizing and miscarried helping behavior were additionally associated with pain intensity in this review. Treatment providers should therefore be mindful to take parent cognitive/affective variables into account, as accurate assessment of these variables may have important implications for treatment of parents and for functional outcomes in children.

### **Recommendations for Future Research**

Effort should be made by researchers to fully report demographic data for the parents. Many studies included in this review did not report on age or sex of the parent. Also, when utilizing

a measure with multiple factor structures and/or short-form versions, researchers should report which factor structure or version they are utilizing, including which items were combined to form each factor, and how they calculated reported scores for the measure (e.g., for the ARCS). This will aid in ease of interpretation across studies moving forward. Additionally, it is important to be aware of which parent version of a measure is used (e.g., parent proxy for their child versus a parent's own report) and to clearly report that. In terms of psychometrics, future research is needed on temporal stability (test- re-test reliability) as well as sensitivity to change of the measures. Related, for clinical practice it would be very relevant to calculate clinical indices, such as the reliable change index (Jacobson & Truax, 1992). Attempts to identify differences between pain conditions in this population may also be important to investigate and could be used to provide focus for interventions. Finally, while we did not specifically aim to exclude measures of the broader family system, no pain-specific measures of family functioning were found. This is an important area for future development.

In summary, our findings demonstrate that there is an abundance of measures available, with adequate reliability overall. However, we also identified skewness in use, inconsistent use of measure subscales and scoring, and a lack of psychometrics on temporal stability. Synthesizing data across studies revealed small effects between parent responses and child functioning, and even smaller and/or absent effects on child pain intensity. This further supports the value of parent-targeted treatment within pediatric chronic pain populations, but also highlights gaps in the current literature and the need for more consistent and thorough use of the measures.



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**Figure legends**

**Figure 1.** PRISMA flow chart of the systematic search.

**Figure 2:** Visual overview of the results from the meta-analysis, showing which correlations between parent measures and child outcomes were observed to be significant. Thicker lines represent larger effects. Note that only analyses that could be conducted (i.e., number of studies  $k \geq 3$ ) are depicted, and only child pain intensity and child functional disability are included (i.e., child school functioning and activity limitations are not depicted). ES = effect size.