COPING WITH THE PEDIATRIC COPING LITERATURE: INNOVATIVE APPROACHES TO MOVE THE FIELD FORWARD

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COPING WITH THE PEDIATRIC COPING LITERATURE: INNOVATIVE APPROACHES TO MOVE THE FIELD FORWARD

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Table of Contents

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EDITORIAL
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Editorial: Coping With the Pediatric Coping Literature: Innovative Approaches to Move the Field Forward

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Keywords: coping, pediatric, chronic conditions, chronic illness, childhood and adolescence

Editorial on the Research Topic

Coping With the Pediatric Coping Literature: Innovative Approaches to Move the Field Forward

INTRODUCTION TO COPING

Chronic illnesses, injuries, and other health conditions (herein "conditions") such as sickle cell disease, chronic pain, and burns are life-disrupting challenges for children and their families. Coping strategies can be defined as "cognitive and behavioral efforts to manage specific external or internal demands that are appraised as taxing or exceeding the resources of a person" (Lazarus, 1991, p. 112). In the context of chronic pediatric health concerns, children and their caregivers/parents must cope with the management of the condition itself, its indirect impact and associated treatment on their daily life (e.g., effect on school engagement), in addition to unrelated "everyday" stressors (e.g., parenting, peer conflict) (Turner-Cobb, 2013). Despite a substantive body of literature exploring coping strategies and quality of life in children living with a chronic condition, several theoretical and empirical gaps remain, including a large number and variable application of coping frameworks or models together with vague and inconsistent operationalization of coping strategies. For instance, Rudolph et al. (1995) proposed a conceptualization of coping that distinguishes between coping responses, goals, and outcomes. Coping responses are actions initiated in relation to a perceived stressor, while the goals are the reasons behind the engagement in a coping response, and the outcomes are the consequences of the coping response. Yet, these different components of coping have been used interchangeably in the context of pediatric chronic health conditions, with assessment or conceptualization of each aspect of coping varying substantially within and across health concerns. Consequently, the goal of this Research Topic *"Coping with the Pediatric Coping Literature: Innovative Approaches to Move the Field Forward"* was to bundle innovative and cutting-edge research that increases our understanding of coping strategies and their underlying mechanisms within pediatric chronic health conditions.

COPING WITH THE PEDIATRIC COPING LITERATURE

The Research Topic contains nine original articles illustrating the variety of innovative work being conducted around pediatric coping. This Research Topic of articles covers a wide range of populations [i.e., pediatric burn survivors (Snider et al.), chronic pain (Balter et al.; Caes et al.; Nabbijohn et al.; Wakefield et al.), spina bifida (Ohanian et al.), sickle cell disease (Hood et al.), inflammatory bowel disease (Reed et al.), and Median Arcuate Ligament Syndrome

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(Stiles-Shields et al.)], across various methodologies [i.e., cross-sectional questionnaire studies (Snider et al.; Hood et al.; Reed et al.), longitudinal evaluations (Ohanian et al.), diary assessments (Caes et al.), scoping reviews (Nabbijohn et al.), and qualitative explorations (Wakefield et al.)]. Including a focus on children and their parents, these articles highlight the important role many coping responses [e.g., concealment (Wakefield et al.), primary control coping (Ohanian et al.), distraction (Snider et al.), catastrophic thinking (Caes et al.; Reed et al.)] play in understanding how a pediatric chronic condition can impact a child's functioning and wellbeing, as well as the need to adopt an integrative biopsychosocial approach to the assessment, formulation, and management of pain coping that goes beyond focusing on pain intensity (Hood et al.). Beyond expanding the theoretical understanding of pediatric coping mechanisms, two articles within the Research Topic also explored interventions to support the uptake of adaptive coping skills (Balter et al.; Ohanian et al.). In particular, these studies highlight how interventions aimed at coping responses are feasible, even in a population with high prevalence of comorbid psychiatric disorders (Stiles-Shields et al.), and can lead to improvements in pain experience, socioemotional functioning, and psychosocial inflexibility (Balter et al.). Nabbijohn et al. present a comprehensive scoping review that gives an overview of the state of the art of the coping literature in the context of pediatric chronic pain. This review highlights four current challenges the pediatric chronic pain coping literature faces, and provides clear suggestions on how to overcome current gaps in the field: (a) lack of theory, conceptual clarity, and conceptual consistency across research; (b) need for diverse methodologies, along with better consistency or standardization in measurement across research; (c) inclusion of diverse populations to understand variations in coping approaches; and (d) improved conceptualization and measurement of proactive coping through theory testing/validation and revision. Arguably, these gaps are reflected in the pediatric health coping literature more broadly.

The Research Topic highlights key questions that remain for future research, aligning with trends seen across the broader pediatric coping literature. Firstly, the research focuses on coping *responses* and their *outcomes*, with little known about coping *goals* or the reasons why children and/or parents engage in these coping responses. A better understanding of the reasons or motivations to engage in a particular coping response is crucial for a comprehensive understanding of the benefits of a coping response and how these benefits could be situationally specific (Carver and Scheier, 2001). Secondly, due to the predominant focus on coping by the child, there is a clear need for family-based assessment of coping processes accounting for the dynamic and interrelated coping of family members. Thirdly, all articles focus on coping with condition-related issues and their indirect impact on daily functioning, but none examine coping with everyday stressors unrelated to the health issue. As human beings with multiple roles, responsibilities, and facets to our identity, we need to adopt a correspondingly integrative view of coping. Lastly, most articles explore potentially harmful coping responses (e.g., concealment, catastrophic thinking). This over-focus on risk factors parallels most research also targeting specific conditions, such as chronic pain. The combined examination of risk and resilience factors is an important avenue for future research, and various models, such as the Resilience-Risk Model for Pediatric Chronic Pain (Cousins et al., 2015), have been put forward to stimulate such research and inform treatment approaches.

AUTHOR CONTRIBUTIONS

LC drafted the first version of the editorial, with CMM and CLD providing feedback and additions on structure and content. All authors approved the final version.

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ORIGINAL RESEARCH
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Neuropsychiatric Symptoms in Pediatric Chronic Pain and Outcome of Acceptance and Commitment Therapy

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Considerable heterogeneity among pediatric chronic pain patients may at least partially explain the variability seen in the response to behavioral therapies. The current study tested whether autistic traits and attention-deficit/hyperactivity disorder (ADHD) symptoms in a clinical sample of children and adolescents with chronic pain are associated with socioemotional and functional impairments and response to acceptance and commitment therapy (ACT) treatment, which has increased psychological flexibility as its core target for coping with pain and pain-related distress. Children and adolescents aged 8–18 years ($N = 47$) were recruited. Patients and their parents completed questionnaires pre- and post-ACT of 17 sessions. Correlational analyses and mixed-effects models were used to assess the role of autistic traits and ADHD symptoms in pretreatment functioning and ACT-treatment response. Outcome variables were degree to which pain interfered with daily activities (i.e., pain interference, sleep, and physical and school functioning), socioemotional functioning (i.e., depressive symptoms, emotional, and social functioning), psychological inflexibility, and pain intensity. Autistic traits and ADHD symptoms, pain frequency, and pain duration were measured at pretreatment only. Higher autistic traits were associated with greater pain interference, higher depression, and greater psychological inflexibility. Higher ADHD symptomatology was associated with greater pretreatment pain interference, lower emotional functioning, greater depression, and longer duration of pain. Across patients, all outcome variables, except for sleep disturbances and school functioning, significantly improved from pre- to post-ACT. Higher autistic traits were associated with greater pre- to post-ACT improvements in emotional functioning and sleep disturbance and non-significant improvements in pain interference. ADHD symptomatology was not associated with treatment outcome. The current results showed that neuropsychiatric symptoms in pediatric chronic pain patients are associated with lower functioning, particularly pain interfering with daily life and lower socioemotional functioning. The results suggest that not only pediatric chronic pain patients low in neuropsychiatric symptoms may benefit from ACT, but also those high in autism traits and ADHD symptoms. With the present results in mind, pediatric chronic pain patients higher in autistic traits may actually derive extra benefit from ACT. Future research could assess whether increased psychological

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flexibility, the core focus of ACT, enabled those higher in autism traits to cope relatively better with pain-related distress and thus to gain more from the treatment, as compared to those lower in autism traits. Moreover, to address specific effects of ACT, inclusion of an appropriate control group is key.

Keywords: pediatric chronic pain, autism spectrum disorder, attention-deficit hyperactivity disorder, acceptance and commitment therapy, socioemotional functioning

INTRODUCTION

Pediatric chronic pain is often accompanied by mental health comorbidities such as depressive and anxiety symptoms, which contribute to lower quality of life and worse daily functioning (i.e., school absenteeism, social withdrawal). Such comorbid problems posit additional physical and emotional burdens for patients and their families (Gold et al., 2009; Miller and Cano, 2009; Hoftun et al., 2011). Chronic pain in childhood and adolescence further incurs a high risk for development of widespread pain, psychological comorbidities, and lower functional status later in life (Hassett et al., 2013). Hence, there is great need for interventions that effectively improve functioning in pediatric chronic pain patients.

While nearly half of the pediatric chronic pain patients suffer from comorbid mental health disorders including mood and anxiety disorders (Vinall et al., 2016; Fisher et al., 2018), neuropsychiatric disorders such as autism and attention-deficit/hyperactivity disorder (ADHD) seem to go unrecognized (Lipsker et al., 2018; Low Kapalu et al., 2018). Lipsker et al. (2018) recently showed that clinically significant levels of autism traits and/or ADHD symptoms were indicated in more than a quarter of children and adolescents with chronic pain. Similarly, chronic pain occurs in a large proportion of individuals diagnosed with clinical autism and/or ADHD (Asztély et al., 2019; Whitney and Shapiro, 2019).

Meta-analyses have shown that psychological therapies, particularly those based on cognitive behavior therapy (CBT), can improve functioning in chronic pain in children, adolescents, and adults (Eccleston et al., 2002; Palermo et al., 2010; Veehof et al., 2011, 2016; Fisher et al., 2014; Hann and McCracken, 2014; Öst, 2014; Hughes et al., 2017). However, little is known about how comorbid neuropsychiatric problems may be associated with treatment outcome (Coffelt et al., 2013; Harrison et al., 2014; Liossi and Howard, 2016; Vinall et al., 2016). Studies suggest that comorbid emotional distress predicts acceptance and commitment therapy (ACT) response, but the results are inconsistent, and the direction of the association has varied between studies (Gilpin et al., 2017). Higher pretreatment psychological distress in different types of chronic pain has been associated with both greater and poorer improvements in functioning and pain interference after psychological treatment (Zautra et al., 2008; Trompetter et al., 2016; Tseli et al., 2019). Also no relationship between distress and psychological treatment outcome has been reported (Broderick et al., 2016; Wetherell et al., 2016). Because distress is a broad umbrella term for adversities, more specific characterization of comorbid problems is warranted to understand for whom treatment is likely to work and to better tailor treatments to individual needs. Moreover, most of these studies that addressed the role of emotional distress in ACT response have been conducted with adults, leaving largely unexplored the relationship between emotional distress and ACT outcome in pediatric chronic pain.

Poor socioemotional functioning is a characteristic of many of the psychiatric disorders comorbid to chronic pain (i.e., autism, ADHD, depression, and anxiety) (Liossi and Howard, 2016; Smith and White, 2020). Psychological interventions targeting socioemotional functioning may thus be specifically beneficial for patients with greater levels of autism traits and ADHD symptoms. ACT, a development within CBT, is such a psychological treatment for chronic pain with potential to improve emotional distress and affective symptoms (i.e., reduce anxiety or depressive symptoms) and to reduce pain interfering with daily life (Veehof et al., 2011, 2016; Hann and McCracken, 2014; Öst, 2014; Pahnke et al., 2014). ACT aims to improve daily functioning by strengthening active and accommodative coping strategies through increasing psychological flexibility. Patients are encouraged to engage in behaviors that are in line with personal and meaningful values. In this process, acceptance of what cannot be changed (e.g., pain) and recognition of the things that can be changed, such as behaviors that serve valued ends, are emphasized. By helping the patient to recognize and acknowledge negative thoughts (e.g., "going to school will make my pain worse"), the therapist helps the patient to distance oneself from the thoughts rather than by discussing whether or not the thoughts are correct (Hayes and Wilson, 1994). Increased psychological flexibility can thus serve as a coping mechanism to improve dealing with pain and pain-related distress (Vowles et al., 2014b).

The empirical support for ACT in adults with unspecific chronic pain is considered as strong (Vowles et al., 2014a). A growing body of evidence suggests that ACT may have a similarly positive impact on children and adolescents with chronic pain (e.g., Kanstrup et al., 2016), although this notion is preliminary due to a limited number of studies performed in pediatric chronic pain patients. For example, in two clinical pilot studies, ACT improved adolescent functioning (Kanstrup et al., 2016; Martin et al., 2016). A case example of an adolescent with chronic pain, as well as a few studies including children or adolescents, likewise documented improvements in daily life functioning (e.g., school attendance) following ACT (Wicksell et al., 2005; Gauntlett-Gilbert et al., 2013; Ghomian and Shairi, 2014).

However, studies addressing the effect of ACT on socioemotional outcomes, such as depressive symptoms, emotional and social functioning, and sleep, are limited in pediatric chronic pain patients (reviewed in Fisher et al., 2014). This is of particular interest in the context of neurodevelopmental disorders such as autism disorder and ADHD, which appear to be comorbid to pediatric chronic pain to a larger extent than in healthy populations and have socioemotional dysfunction as a core feature (Lipsker et al., 2018). In a study of Pahnke et al. (2014), involving a sample of 13–21-year-old high-functioning students with autism, increased prosocial behavior and reduced stress, hyperactivity, and emotional distress were observed after ACT treatment. Individuals with autism often exhibit rigidity and a need for rule-governed behaviors (Leekam et al., 2011), which has been suggested to contribute to chronic pain (Beeckman et al., 2019). Therefore, ACT's focus on strengthening coping strategies may further enhance its effects in pediatric chronic pain patients high in traits and symptoms of neurodevelopmental disorders (Simons et al., 2008; Wicksell et al., 2011). Furthermore, executive dysfunctions, such as cognitive inflexibility and attentional deficits, are prevalent in autism and ADHD, although with a different focus between the disorders, and have likewise been linked to how patients cope with pain (Corbett et al., 2009; Berryman et al., 2014; Craig et al., 2016). The biopsychosocial model of Compas and Boyer (2001) highlights the importance of attentional processes in coping with pain. Shifting focus away from pain and sustaining attention to favorable coping strategies are important neurocognitive processes for effective coping. Executive dysfunction, or comorbid problems associated with executive dysfunction, may thus augment pain processing and impede the utilization of coping strategies. Together, this suggests that ACT may be particularly beneficial for chronic pain patients high in autism traits and/or ADHD symptomatology, partially due to its ability to improve psychological flexibility, which is regarded a component of executive functioning. However, the impact of such comorbid neurodevelopmental disorders in pediatric chronic pain on treatment outcome is yet unknown. Despite that acceptance-based therapies demonstrate promising results for improving socioemotional functioning and pain interference in chronic pain, considerable interpatient variability in the response to ACT exists (e.g., Hann and McCracken, 2014). Insight into what works for whom is crucial for patient–treatment matching to maximize treatment outcome and thereby limit the impact of chronic pain and comorbid problems on the patient's emotional, cognitive, and physical development (Vlaeyen and Morley, 2005; Edwards et al., 2016).

To this end, the primary aim of the current single-arm trial was to assess the association between comorbid neuropsychiatric symptoms (i.e., autism traits and ADHD symptoms) in pediatric chronic pain patients and (1) pre-ACT functioning and (2) change from pre- to post-ACT. Outcomes were defined as the degree to which pain interfered with daily activities (i.e., pain interference, sleep, school and physical functioning), socioemotional functioning (i.e., depressive symptoms, emotional and social functioning), psychological inflexibility, and pain intensity.

METHODS

Treatment

The intervention consisted of a standard face-to-face ACT-based treatment. The treatment consisted of 17 sessions involving four phases with different but interrelated treatment objectives: (1) preparing for behavioural change (sessions 1–3), (2) shifting perspective (sessions 4–6), (3) acceptance and cognitive defusion (sessions 7 and 8), and (4) values-oriented behavioral activation (sessions 9–17). In every session, participants were given individualized home assignments related to the treatment content and to their own specific challenges, and these outcomes were discussed at the beginning of the following session. Five ACT-trained psychologists delivered the treatment. All psychologists were continuously supervised by a senior researcher with extensive experience using ACT for pediatric chronic pain. To promote a uniform approach among the treatment providers, patients were discussed with all therapists during clinical supervision meetings. No other therapist monitoring measures were utilized. For a detailed description of the ACT protocol, see Kanstrup et al. (2016). A parent support program was embedded in the treatment and comprised four parent sessions and one joint session including the parents and the patient. The objective of the parent program was to enhance the parents' ability to support their child to improve functioning, by means of pain education, contingency management including clarification of own values, and the use of acceptance skills to manage their own distress due to their child's pain. For the purpose of the present study, a single group of patients in which all received active treatment was studied.

Participants

Participants in this convenience sample were 47 children and adolescents (8–18 years old, 33 girls) with chronic pain, recruited via a tertiary pain clinic. Patients referred to the clinic because of chronic debilitating pain were considered eligible for the study if they (1) suffered from pain for more than 6 months, (2) reported insufficient effects of previous pain treatments, and (3) reported substantial pain-related disability. Patients were not considered as eligible if they had psychiatric comorbidity that required immediate intervention; substantial risk for suicide or substantial cognitive dysfunction; insufficient proficiency in Swedish; other ongoing or planned treatments (i.e., within the next 6 months); and pain that was fully explained by a pathophysiological process, e.g., cancer. The study was approved by the ethical review board in Stockholm, Sweden. All participants (parents and children) provided written informed consent to participate in the study.

Assessment

Data collection took place pre- and post-ACT. Eligibility was assessed in a semistructured clinical interview during the first visit. Patient's functioning was assessed through patient- and parent-reported questionnaires (see below).

Measures

Questionnaires were completed by the children, except for the Social Responsiveness Scale (SRS) and the Conners Third Edition (Conners-3). The following questionnaires were completed at

pretreatment and posttreatment, as part of a clinical routine using paper and pencil.

Pain Interference

The six-item Pain Interference Index (PII) was used to assess the influence of pain on behavior or to what extent pain impacts everyday functioning (e.g., schoolwork, leisure activities, sleep). Items are rated on a scale from 0 (not at all) to 6 (completely), and the maximum score is 36. A higher score indicates more pain interference. The Swedish version of PII has shown sensitivity to change (Wicksell et al., 2010). The Cronbach α was 0.83.

Insomnia Severity

The Insomnia Severity Index (ISI) is a seven-item measure that assesses the individual's subjective perception of sleep complaints, rated on a five-point Likert scale and a maximum score of 28. A higher score suggests more severe insomnia (Bastien et al., 2001). Internal consistency was high in the current sample (Cronbach $\alpha = 0.87$).

Quality of Life

The Pediatric Quality of Life Inventory (PedsQL) is a 23-item measure that evaluates the child's quality of life in four areas of functioning: physical functioning (eight items), emotional functioning (five items), social functioning (five items), and school functioning (five items). Two age versions were used; one for children 8–12 years old and one for adolescents 13–18 years old. Cronbach α's for the respective subscales ranged from 0.66 to 0.86. The instrument uses a five-point Likert scale from 0 (never a problem) to 4 (almost always a problem) to indicate severity. A higher score indicates better functioning (maximum score is 100) (Bastiaansen et al., 2004).

Depression

The Center for Epidemiological Studies–Depression Scale Children (CES-DC) (Faulstich et al., 1986) consists of 20 items concerning feelings and actions relevant for depressive disorder, rated on a scale from 0 (not at all) to 4 (a lot). The maximum score is 60. Higher scores indicate higher levels of depression. CES-DC has shown adequate psychometric properties and the ability to discriminate depressive disorder in a Swedish population of adolescents (Olsson, 1997). The overall Cronbach α was 0.80.

Psychological Inflexibility

The Psychological Inflexibility in Pain Scale (PIPS) is a measure of psychological inflexibility, measuring avoidance of pain and fusion with pain thoughts (Wicksell et al., 2010). The PIPS consists of 12 items rated on a scale from 1 (never true) to 7 (always true), with scores ranging from 12 to a maximum of 84. Higher scores indicate greater levels of psychological inflexibility. Internal consistency (Cronbach α) of the total scale was 0.89 in the current sample.

Pain Intensity

The Lubeck Pain-Screening Questionnaire (LPQ) evaluates the prevalence and consequences of pain during the previous 3 months (Roth-Isigkeit et al., 2004). As single items of the LPQ have been shown to be valid and reliable measures of pain, the current study used "How strong is your main pain usually?" to construct a pain intensity variable. The item was rated on a visual analog scale (VAS) from 0 ("hardly noticeable pain") to 100 ("strongest imaginable pain").

The following questionnaires were completed at pretreatment only, as part of a clinical routine using paper and pencil.

Autism Traits

The SRS-Parent report is a 65-item parent report for children and adolescents (4–18 years of age) that measures the severity of autism spectrum symptoms. Scoring is on a four-point Likert scale. Higher scores indicate a higher degree of social impairment. SRS scores of 60 T or higher indicate a level of autistic social impairment that is clinically significant (Constantino and Gruber, 2014). *T* scores are gender-corrected scores (gender of the child). Standardized *T* scores with mean = 50 (SD = 10) were used in the current study. Internal consistency (Cronbach α) of the total scale is 0.94 (parent rated) (Constantino and Gruber, 2005).

Attention-Deficit/Hyperactivity Disorder

The Conners-3 ADHD Index 10-item subscale (i.e., Conners Hyperactivity Index) of the Conners 3rd Edition–Parent (Conners-3-P, 110 items) was used to obtain the parent's observation about the most prominent symptoms of ADHD over the last month in their child (Conners, 2008). This 10-item scale is recommended as a quick and valid research and clinical screening tool in large samples. It has been shown to accurately differentiate children with ADHD from those without the clinical diagnosis (Chang et al., 2016). Items are based on *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition* criteria and scored on a four-point Likert scale. Standardized *T* scores with mean = 50 (SD = 10) were used in the current study. *T* scores are gender- and age-corrected scores. A higher score indicates more symptoms. ADHD scores of 65 T or greater indicate a level of ADHD that is clinically significant. Internal consistency (Cronbach α) of the Conners-3 Hyperactivity Index is 0.94 (parent rated) (Kumar and Steer, 2003).

Duration and Frequency of Pain

Two items of the LPQ were used to assess frequency of pain ("How often do you have this pain?") and duration of pain ("How long have you had pain?") (Roth-Isigkeit et al., 2004). Each item was rated on a five-point scale. As indicated earlier, pain intensity was assessed pre- and post-ACT on a VAS.

Sample Size

Based on previous studies of ACT in chronic pain patient groups, medium ($d = 0.6$) to large ($d = 0.8$) within-group effect sizes were expected for pain interference and pain intensity. A small to medium effect size was expected for depression (Wicksell et al., 2007, 2009; Gauntlett-Gilbert et al., 2013). Power analysis using a power of 80% and an α of 0.05 suggests that a total sample between 15 and 34 is adequate to detect medium to large effect sizes. As attrition was expected and to take into account the possibility of small to medium effect sizes, we aimed to recruit a minimum of 45 patients.

TABLE 1 | Descriptive characteristics of the study population.

Characteristic	
N	47
Age (years) ($n = 47$)	
Mean (SD)	14.8 (2.2)
Range	9.5–17.9
Gender (% girls)	70
Pain characteristics	
Pain duration	
Every now and then (%)	0
1–3 months (%)	2.1
<1 month (%)	0
>3 months (%)	6.4
>6 months (%)	17.0
>12 months (%)	53.2
Pain frequency	
<1 × per month (%)	0
1 × per month (%)	0
2–3 × per month (%)	4.3
1 × per week (%)	4.3
Multiple times per week (%)	14.9
Every day	55.3
Pain intensity (0–100) (SD)	57.2 (13.1)
Autistic traits (SRS) ($n = 38$)	
Mean (SD)	47.3 (8.8)
Range	34–71
Clinically significant level (%)	13.2
ADHD symptoms (Conners-3) ($n = 40$)	
Mean (SD)	55.3 (12.6)
Range	46–89
Clinically significant level (%)	22.5

Statistical Analysis

Analyses were conducted in a series of steps in SPSS version 24 (SPSS Inc., Chicago, IL, USA) and JASP (version 0.13, JASP Team, 2020). Means and standard deviations were calculated for descriptive purposes. Correlational analyses were used to assess the relationship of pretreatment functioning with level of autistic traits and with ADHD symptoms. Mixed models were used to estimate the change from pre- to post-ACT for pain interfering with daily activities (insomnia; ISI), school and physical functioning (subscales of the PedsQL), pain interference (PII), socioemotional functioning (depressive symptoms; CES-D), emotional and social functioning (subscales of the PedsQL), psychological inflexibility (PIPS), and pain intensity (item of LPQ). Then, mixed-model regressions were conducted for autistic traits (SRS) and ADHD symptoms (Conners-3-P) separately to assess associations between autism/ADHD and ACT outcomes. Model simplicity and likelihood ratio tests were used to select appropriate covariance structures. Data for the main measures were analyzed using timepoint (pretreatment and posttreatment) as a repeated and fixed factor and subject as a random factor. Continuous autism/ADHD *T* scores were added as a fixed factor. The effects of interest were main effects of time and autism/ADHD and interaction effects of time × autism/ADHD. Subsequently, each model with a significant interaction effect controlled for gender and age (fixed factors) (results are shown in the **Supplementary Materials**). Variables were *Z*-transformed before analysis yielding standardized regression coefficients. The pre- to post-ACT changes analyses were repeated with a between-subjects factor, dividing patients into above or below clinically significant levels of autism traits and/or ADHD symptoms based on the criteria for clinically significant *T* scores (i.e., 60 T or higher for autism traits, 65 T or higher for ADHD) (results are shown in **Supplementary Materials**). Alpha Values were set at 0.05 throughout. In addition to traditional null hypothesis significance testing, Bayes factors (BF_{10}) were calculated using Bayesian correlation analyses with default prior probabilities. Bayes factors provide relative evidence of both the null (H0) and alternative (HA) hypothesis, compared to the conclusions about the null hypothesis proffered by traditional null hypothesis significance testing.

RESULTS

Sample Characteristics

Sample characteristics are presented in **Table 1**. Data on autistic traits and ADHD symptoms were available for 38 and 40 patients, respectively. See also **Supplementary Table 1** for an overview of the missing variables separately for pre- and post-ACT. No information regarding reasons for attrition was obtained. Clinically significant levels of autism traits and ADHD symptoms occurred in 13.2% ($n = 5$) and 22.5% ($n = 9$) of the patients, respectively. Ten percent ($n = 4$) of the patients scored greater than clinically significant levels for both autism traits and ADHD symptoms. The current data are based on partially the same sample as was used in Lipsker et al. (2018).

Relationships Between Autism/ADHD and Pretreatment Functioning

Autism

As shown in **Table 2**, at pretreatment, patients with higher levels of autism reported greater pain interference, higher depression, greater psychological inflexibility, and lower physical and social functioning, although the latter two were non-significant. Bayesian statistics revealed anecdotal (Bayes factor 1–3), moderate (Bayes factor 3–10), and strong (Bayes factor 10–30) evidence in favor of the alternative relative to the null hypothesis (Wagenmakers et al., 2017).

ADHD

At pretreatment, patients with higher levels of ADHD symptoms reported greater pain interference, higher depression, lower emotional functioning, and longer duration of pain. The significant relationships between autism/ADHD and pretreatment functioning are displayed in **Figure 1**. An overview of all correlation analyses results (both statistically significant and non-significant) and the corresponding Bayes factors are

TABLE 2 | Pearson correlation coefficients, 95% confidence intervals (CIs), and Bayes factors (BF_{10}) of each pretreatment outcome with autism traits and with ADHD symptoms.

	Autism traits			ADHD symptoms		
	r	BF_{10}	95% CI	*r*	BF_{10}	95% CI
Pain interference	0.430**	6.67	0.10–0.64	0.377*	2.80	0.07–0.62
Physical functioning	−0.298#	0.99	−0.56–0.02	−0.211	0.44	−0.50–0.12
School functioning	−0.180	0.36	−0.47–0.15	−0.271	0.74	−0.54–0.05
Insomnia	0.120	0.26	−0.22–0.44	0.097	0.24	−0.24–0.42
Depression	0.406*	4.43	0.10–0.64	0.342*	1.43	0.03–0.60
Emotional functioning	−0.249	0.60	−0.53–0.08	−0.413*	4.95	−0.65–−0.11
Social functioning	−0.313#	1.18	−0.58–0.01	−0.256	0.64	−0.53–0.07
Psychological inflexibility	0.520***	38.48	0.24–0.72	0.291	0.92	0.01–0.60
Pain intensity	0.109	0.25	−0.23–0.42	−0.022	0.21	−0.35–0.31
Pain frequency	−0.001	0.21	−0.33–0.32	−0.038	0.21	−0.36–0.29
Pain duration	−0.130	0.27	−0.44–0.29	−0.624***	723.41	−0.79–−0.38

$^{***}p < 0.001$, $^{**}p < 0.01$, $^{*}p < 0.05$, $^{\#}p < 0.07$.

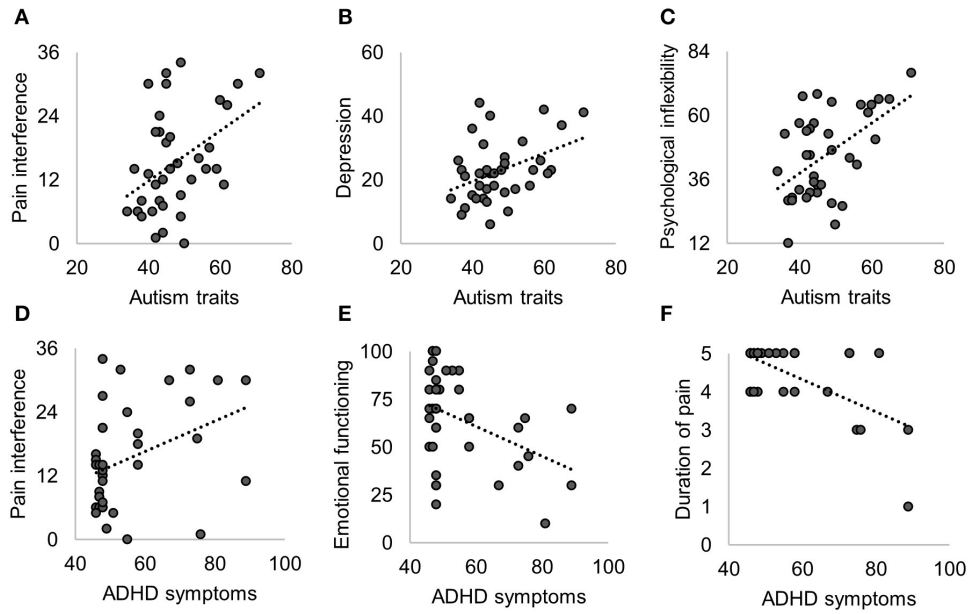

FIGURE 1 | Significant Pearson correlations of pretreatment functioning with autism traits **(A–C)** and with ADHD symptoms **(D–F)**. Non-significant correlations are shown in **Table 2** and **Supplementary Figure 1**.

shown in **Table 2**. All non-significant correlation plots are shown in **Supplementary Figure 1**.

Pre- and Post-act Comparisons

As can been seen in **Figure 2**, statistically significant improvements from pre- to post-ACT were found for pain interference [$t_{(1,41)} = -4.13$, $\beta = -0.69$, 95% confidence interval (CI) [−1.03, −0.35], $p < 0.001$], physical functioning [$t_{(1,41)} = 3.42$, $\beta = 0.57$, 95% CI [0.23, 0.91], $p = 0.001$], depression [$t_{(1,44)} = -4.46$, $\beta = -0.65$, 95% CI [−0.95, −0.36], $p < 0.001$], emotional functioning [$t_{(1,44)} = 2.70$, $\beta = 0.35$, 95% CI [0.09, 0.61], $p = 0.010$], social functioning [$t_{(1,38)} = 2.45$, $\beta = 0.42$, 95% CI [0.07, 0.76], $p = 0.019$], psychological inflexibility [$t_{(1,42)} = -5.13$, $\beta = -0.83$, 95% CI [−1.16, −0.50], $p < 0.001$], and pain intensity [$t_{(1,43)} = -2.50$, $\beta = -0.45$, 95% CI [−0.82, −0.09], $p = 0.016$]. Insomnia and school functioning did not significantly change from pre- to post-ACT.

Associations Between Autism Traits/ADHD Symptoms and Treatment Outcome

Autism

As shown in **Figure 3** and **Table 3**, significant time × autism interactions were evident for insomnia severity and emotional functioning, indicating that patients higher in

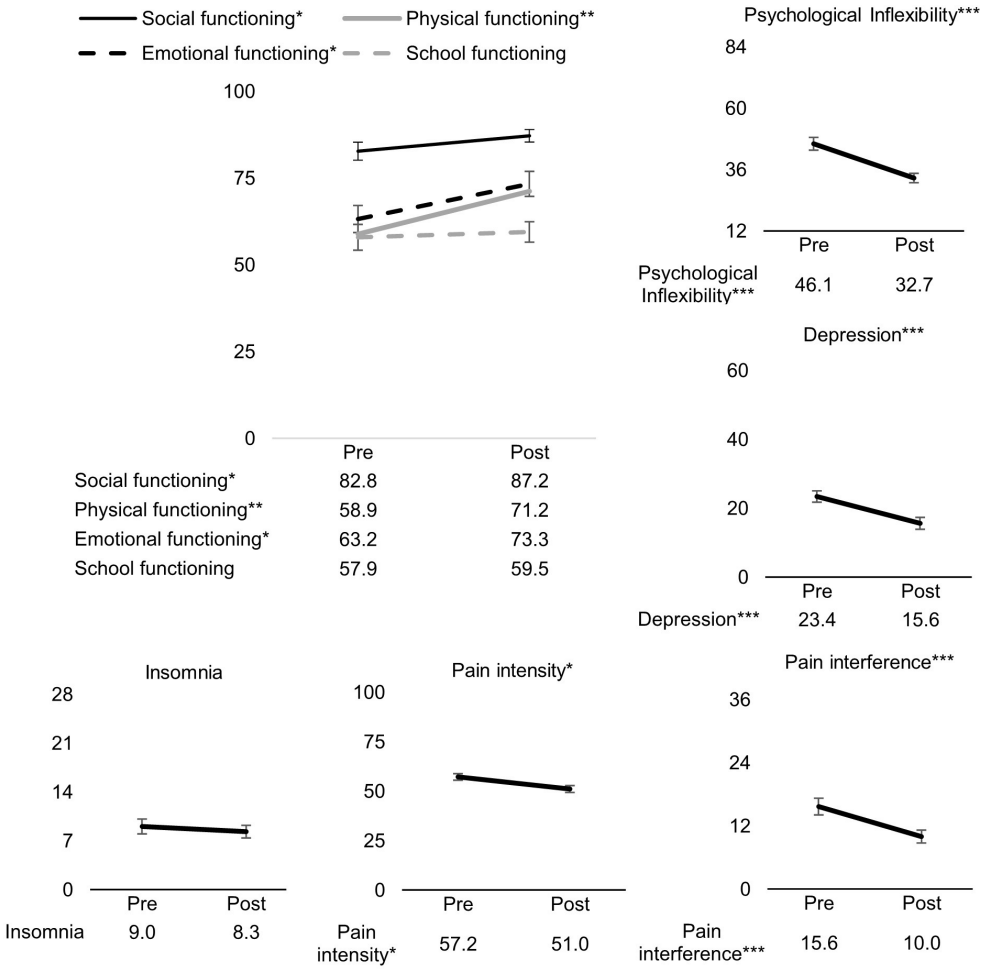

FIGURE 2 | Pre- to post-ACT changes for all outcome measures. Values on the *y*-axes are mean scores pre- and post-ACT. Errors bars represent standard error of the mean. ***$p < 0.001$, **$p < 0.01$, *$p < 0.001$.

autism trait showed greater improvements in insomnia and emotional functioning. Similarly, those higher in autism trait showed greater improvements in pain interference, although non-significant. Controlling for gender and age did not alter the direction of the uncorrected results (see **Supplementary Materials** for age- and gender-adjusted results). Similar results were found when patients were divided into below and above clinically significant levels of autism or ADHD (see **Supplementary Figure 2**).

ADHD

No significant time × ADHD interactions were evident (see **Table 3** for an overview of all results).

DISCUSSION

In the current study, pediatric chronic pain patients showed improvements from pre- to post-ACT in interference of pain with daily activities, socioemotional functioning, psychological inflexibility, and pain intensity, contributing to the growing body of evidence showing that acceptance-based treatments improve functioning in pediatric chronic pain. Those with greater neuropsychiatric traits and symptoms (autism and ADHD) benefited at least to the same degree from ACT as compared to those lower in neuropsychiatric traits and symptoms. In fact, those higher in autism traits showed greater improvements in insomnia and emotional functioning. However, because of the lack of a control treatment, it cannot be concluded that improvements resulted from ACT, as they might have arisen from non-specific effects of ACT, natural improvements placebo effects, or because of changes in patient characteristics. Nevertheless, clinical improvements of such a large extent are unlikely to be spontaneous. Moreover, improvements were consistent with the ACT model and consonant with previous evidence (e.g., Hann and McCracken, 2014; Veehof et al., 2016; Hughes et al., 2017), in that improvements in psychological inflexibility, pain interference, and functioning were achieved.

Greater improvements in insomnia and emotional functioning were seen in those with greater levels of autism. It

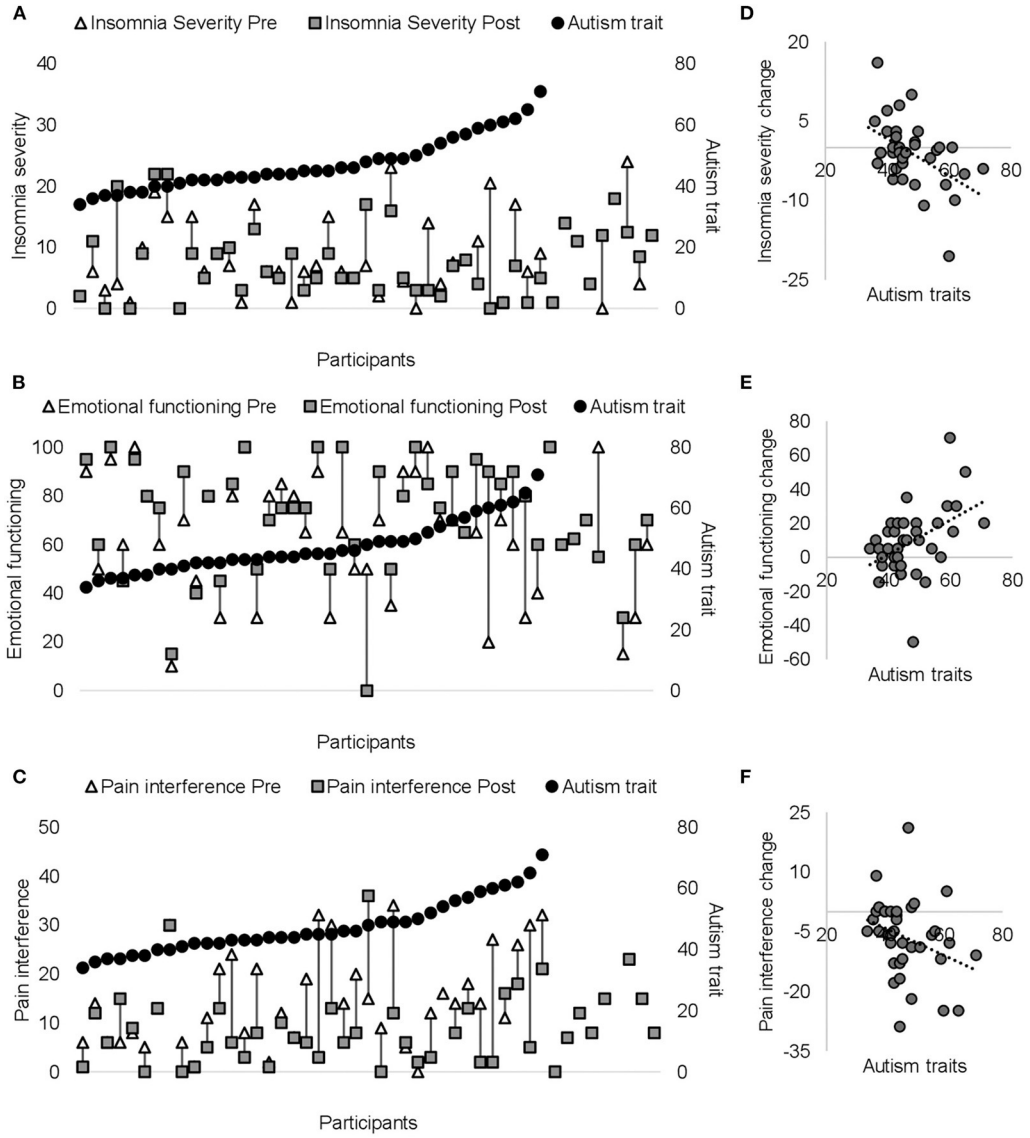

FIGURE 3 | Pre- (white triangles) and post-ACT scores (dark gray squares) are plotted on the primary (left) *y*-axis for each patient for insomnia **(A)**, emotional functioning **(B)**, and pain interference **(C)**. Black circles on secondary *y*-axis represent individual autism scores (presented from the lowest to the highest score). The right panel **(D–F)** shows significant correlations between autism traits and change score (from pre- to post-ACT) for insomnia **(D)** emotional functioning **(E)**, and pain interference **(F)**. Negative insomnia severity/pain interference change scores indicate greater improvements in insomnia/pain interference. Positive emotional functioning change scores indicate greater improvements in emotional functioning.

has been proposed that pediatric chronic pain can disrupt sleep, which can, independently of pain, promote physiological and psychological changes that exacerbate pain, suggesting that pain and sleep operate in a bidirectional manner (Lewin and Dahl, 1999; Valrie et al., 2013). Besides that sleep disturbances are commonly reported by pediatric chronic pain patients (Valrie et al., 2013), such disturbances are also a prevailing problem in autism and ADHD (Cortese et al., 2009; Elrod and Hood, 2015). Considering that poor sleep compromises the emotional, cognitive, and behavioral development of adolescents with chronic pain (Palermo and Kiska, 2005), improvements of sleep in this young chronic pain population with greater autism traits are highly encouraging. Greater autism traits were further associated with lower pre-ACT functioning, highlighting the possibility that there was a greater room for improvement in those with greater autism traits. However, this unlikely explains the added benefit with higher autism traits as insomnia and emotional functioning were not associated with autism traits before ACT. Keeping the behavioral problems associated with autism in mind, such as deficits in executive functions and associated social challenges (Weiss et al., 2018), a perhaps more reasonable explanation for the positive association between

TABLE 3 | Results of the mixed regression analyses of autism traits/ADHD symptoms and change from pre- to post-ACT for all outcomes (autism/ADHD*time); 95% CI = 95% confidence interval; $^{***}p < 0.001$, $^{**}p < 0.01$, $^{*}p < 0.05$, $^{\#}p < 0.07$.

	Autism traits			ADHD symptoms		
	β	*t*	95% CI	*β*	*t*	95% CI
Pain interference	−0.32#	−1.96	−0.65–0.01	−0.15	−0.89	−0.49–0.19
Physical functioning	0.10	0.69	−0.19–0.38	0.05	−0.32	−0.24–0.33
School functioning	−0.08	0.68	−0.16–0.33	0.07	0.59	−0.17–0.32
Insomnia	−0.46**	−3.01	−0.76–−0.15	−0.14	−0.84	−0.48–0.20
Depression	−0.24	−1.70	−0.53–0.05	0.06	0.41	−0.24–0.35
Emotional functioning	0.36**	3.09	0.12–0.60	0.22	1.76	−0.03–0.48
Social functioning	−0.09	0.54	−0.25–0.44	−0.01	−0.06	−0.36–0.33
Psychological inflexibility	−0.21	−1.30	−0.54–0.12	0.01	0.06	−0.33–0.36
Pain intensity	−0.05	−0.34	−0.37–0.27	0.03	0.19	−0.29–0.35

autism traits and treatment outcome is that these individuals in particular may benefit from the structure that a behavioral-based treatment provides as well as ACT's methodology to increase awareness and understanding of one's own thoughts and feelings. In speculation, such treatment may provide help in directing attentional resources to adaptive behaviors.

Greater improvement in selective symptoms was not seen in patients higher in ADHD symptoms. In part similar to autism, the level of ADHD was associated with lower pre-ACT functioning in some domains (i.e., pain interference, depression, emotional functioning, and duration of pain), but this did not lead to greater improvements after treatment. Despite that autism and ADHD depend in part on a shared neural dysfunction and often co-occur (Ghirardi et al., 2018; Kernbach et al., 2018), autism and ADHD show considerable differences in symptom profiles, and autistic traits are not common in ADHD (Mayes et al., 2012). For example, children with autism show more prominent deficits in cognitive flexibility as compared to children with ADHD (Corbett et al., 2009). A similar pattern was observed in the current study, in which psychological flexibility (which overlaps with cognitive flexibility; Whiting et al., 2017) was associated with autism traits only. Although speculative at this point, improvements in psychological flexibility, the core target of ACT, enabled those higher in autism traits to cope better with negative private experiences such as pain and distress and gain more from the treatment (Compas et al., 2017). ACT may have potentially also improved specific autism-related traits. Indeed, ACT improved prosocial behavior and emotional distress in a clinical sample of children with autism spectrum disorder (Pahnke et al., 2014), suggesting that ACT may positively affect behaviors connected to autistic traits. Considering that functioning improved from pretreatment to posttreatment in not only those low but also those high in autism traits and ADHD symptoms, ACT could potentially be a suitable treatment for improving functioning in individuals with such neuropsychiatric traits.

The current results need to be seen in the light of several limitations. First, lack of a control group receiving a comparator (e.g., physical therapy) or no (e.g., waiting list) treatment can be considered as a major limitation, as pre- to post-ACT changes may be due to specific treatment components but could also reflect natural improvement of functioning, expectation (because the patients and their parents could not be blinded), or developmental changes in the patient due to becoming older. Second, this study was conducted with a limited sample size, and null findings should thus be interpreted with some caution. In particular, the number of patients above cutoff for clinically significant autism ($n = 5$) or ADHD ($n = 9$) is low. Thus, also positive findings should be interpreted with caution, and the generalizability of the findings to clinical samples of autism and ADHD remains unknown until larger controlled studies using diagnostic testing have been performed. Future controlled clinical trials may also monitor treatment fidelity. Third, findings from this study, and related studies (Lipsker et al., 2018; Low Kapalu et al., 2018), suggest that the combination of pediatric chronic pain and neurodevelopmental issues is common and presents significant challenges to the child and his/her family. Future research could evaluate the role of neurodevelopmental symptoms in aspects such as dropout and treatment adherence. Fourth, ACT was explicitly aimed at creating sustained changes, and even though improvements were observed directly post-ACT, whether these persist long-term, in addition to the specificity of ACT, requires further examination. Future studies may address these limitations by conducting larger controlled studies including chronic pain patients as well as patients diagnosed with autism and/or ADHD. Addressing whether severity of autism traits and ADHD symptoms can be reduced in response to ACT as well as which factors enabled chronic pain patients, and specifically those higher in autism trait, to gain from ACT can provide mechanistic links connecting improved functioning with ACT. We suggest here two potential factors that deserve further scrutiny: psychological flexibility and attention to interoceptive cues. First, as discussed previously, improvements in psychological flexibility, the core target of ACT, may enable greater gains from the ACT sessions. Intermediate assessment of psychological flexibility may shed light onto the time course of improvement and the possibility to gain more from each ACT session. Second, it has been suggested that autism patients disproportionally allocate attention recourses to internal rather than to external cues (Schauder et al., 2015). Whether a shift in attention away from interoceptive cues and sustaining attention toward

helpful coping strategies can boost patients' ability to gain more from the treatment is a question that future research may consider.

Taken together, even though the results should be viewed in light of its limitations, the results of the current study suggest that pediatric chronic pain patients higher in autistic traits could have an extra benefit from ACT and those higher in ADHD symptomology benefit to the same degree as those low in ADHD symptomology.

DATA AVAILABILITY STATEMENT

The dataset of this article is accessible on request from the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Regional ethical review board in Stockholm: 2013/231-31-4. Written informed consent to participate in this study was provided by both the participant and the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

CW, RW, and ML conceived the experiment. CW data collection. LB statistical analysis. LB, CW, RW, and ML writing. All authors reviewed the manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2021.576943/full#supplementary-material

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"If It Ever Really Hurts, I Try Not to Let Them Know:" The Use of Concealment as a Coping Strategy Among Adolescents With Chronic Pain

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Objective: Despite considerable evidence of chronic pain in adolescents, and its adverse consequences for their health and well-being, less is known about pain-related stigma that these youth face, such as pain disbelief by others. Adolescents with chronic pain may conceal their symptoms as a coping strategy to avoid pain-related stigma, contributing to further social isolation and disruptions in medical treatment. In the current study, we used focus group methodology to examine adolescent motivations for using concealment and the possible benefits and harmful consequences of this form of coping.

Materials and Methods: Five focus groups of 3–5 adolescents (ages 12–17) with chronic pain conditions (N = 18) were conducted as a part of a larger study to evaluate the impact of, and reaction to, pain-related stigma. Patients were recruited from an outpatient pediatric pain management clinic. Transcripts of focus group sessions were analyzed using directed content analysis for the main study, yielding anticipatory stigma and concealment categories. These categories were then explored using inductive content analysis for the current study.

Results: Adolescents described engaging in concealment of their pain symptoms. Our analysis revealed three social motivations for concealment: (1) avoidance of judgment; (2) avoidance of being a social burden; and (3) desire to be treated normally, and two harmful consequences of concealment: (1) social isolation and (2) cognitive burden.

Conclusion: Disbelief of pain symptoms may exacerbate the social isolation and disease-related burden in this population. Clinical implications of concealing pain symptoms are discussed, and points of intervention are proposed.

Keywords: adolescents, chronic pain, concealment, coping, stigma

INTRODUCTION

Chronic pain in youth has been on the rise over the past 25 years, with a prevalence of approximately one in every 4–5 adolescents (King et al., 2011). Despite the prevalence of pediatric chronic pain, diagnostic ambiguity commonly exists for youth with chronic pain complaints (Betsch et al., 2017; Neville et al., 2019; Tanna et al., 2020). Youth with chronic pain, and their families, experience this diagnostic uncertainty in the context of repeated negative medical findings (Neville et al., 2019), alongside providers who may not feel confident in their management of pediatric chronic pain (Neville et al., 2020). Many parents have also indicated feeling that their child's pain symptoms are often dismissed by pediatric providers (Igler et al., 2020). The invisibility of chronic pain symptoms has led to adolescents with chronic pain experiencing pain-related stigma, specifically in the form of others indicating that they do not believe their pain is "real," and/or believing that the person is making up their symptoms (Wakefield et al., 2018).

Stigma occurs when an individual has a specific attribute that is socially undesirable (Goffman, 1963) that can lead to discrimination by others. There are different types of stigma including felt, anticipatory, and internalized (Major et al., 2018). Felt stigma occurs when the stigmatized individual perceives that they are being treated unfairly based on their stigmatizing attribute. Within the context of pediatric chronic pain, felt stigma is experienced when youth with chronic pain perceive that others are negatively judging them or not believing their pain symptoms. Anticipatory stigma refers to negative judgment or treatment that individuals with stigmatized identities expect to experience if others become aware of their stigmatized condition.

Internalized stigma (also called self-stigma) occurs when the stigmatized individual engages in self-blame and applies negative stereotypes to themselves. The anticipation of pain-related stigma may motivate adolescents with chronic pain to conceal their condition from others. While the literature is limited, all of these forms of stigma have been reported by youth with chronic pain conditions (Wakefield et al., 2018; Laird et al., 2020).

When faced with possible negative judgment or treatment by others, individuals who experience stigma may attempt to hide their stigmatizing identity, especially if this identity can be concealed (Quinn and Earnshaw, 2013), such as chronic pain. This coping strategy is known as "concealment," and research on its impact on health outcomes has been mixed (Camacho et al., 2020). Whereas the lack of disclosure of illness may allow individuals with concealable stigma identities to avoid potential negative treatment by others (Quinn and Chaudoir, 2009; Barned et al., 2016; Benson et al., 2017; Galano et al., 2017), some evidence suggests that concealment leads to poorer physical and psychological well-being (Frable et al., 1998; Quinn et al., 2017; Laird et al., 2020), possibly due to the consequences of internalizing distress. Concealment also prevents access to social support, which is a protective factor in adapting to chronic pain (Ross et al., 2018).

For youth with chronic pain conditions, the empirical evidence on disclosure or concealment of pain symptoms is limited. Some research has linked concealment of chronic pain to negative physical health outcomes (Laird et al., 2020), worse psychological well-being, and lower pain tolerance (Uysal and Lu, 2011; Laird et al., 2020). For example, Uysal and Lu (2011) observed that healthy young adults who demonstrated greater levels of concealment exhibited lower pain tolerance in response to a cold pressor task, indicating that concealment may lead to heightened reactivity to pain stimuli. The inhibition or internalization of emotions that occurs when one engages in concealment can also have a physiological cost, as demonstrated in individuals with trauma (Pennebaker and Beall, 1986). Collectively, these initial findings demonstrate the potential harm that may result from concealing chronic pain conditions to others, but less is known about the experience of pain-related stigma and motivations among adolescents with chronic pain to use concealment as a coping strategy.

To begin to address these gaps in this emerging field of study, the present study aimed to examine the nature of concealment as a coping strategy used by adolescents with primary chronic pain, using focus group methodology. Focus group methodology was used in this case to develop themes that described the experiences and coping behavior of the adolescents with chronic pain without the constraints on responding imposed by established questionnaires or interviews. Thus, focus groups allow the emergence of responses that might not become clear using other means of data collection (Morgan and Krueger, 1993). Due to the benefits of peer connections in the context of social difficulties, and the ability to understand similarities and differences in experiences, focus group methodology was justified. The use of focus groups also provides in-depth descriptions of motivations for concealment as well as observed benefits and harmful consequences. Due to the qualitative nature of the study, there were no specific hypotheses developed *a priori*; however, it was expected that the adolescents in this study would discuss the use of concealment given the previous literature on concealment in the context of health-related stigma (Quinn, 2018).

MATERIALS AND METHODS

Participants

The current study was a secondary analysis of data collected in a larger study evaluating pain-related stigma among adolescents with chronic pain, which was approved by the Institutional Review Board at Connecticut Children's. The recruitment and procedures of the current study and larger study are the same. Patients with chronic pain receiving treatment in a tertiary multidisciplinary pain management clinic at a midsize children's hospital were screened for eligibility and approached by research personnel either in person during their outpatient clinic visit or via phone shortly after (within 2 weeks) their appointment. Inclusion criteria for the study was adolescent patients, ages 12–17 years, with a documented diagnosis of chronic pain. Exclusion criteria included the presence of a comorbid chronic medical condition, such as juvenile rheumatoid arthritis or diabetes, in order to reduce the potential stigma bias attributed to non-chronic or identifiable pain conditions. Additionally, adolescents who were not fluent in English were excluded because the focus

groups were conducted in English. Data collection occurred from December 2017 to October 2019.

Of the 25 adolescents approached for the study, 18 agreed to enroll in the study. The seven patients who did not enroll were interested, but had scheduling conflicts with the focus group dates. No patients were excluded based on study eligibility criteria. Of those who participated, 88.89% were female (5.56% male, 5.56% gender fluid) and the mean age was 15.33 years (SD = 1.28, range = 12–17). Regarding pain conditions, 13 of the 18 had pain in more than one location (72.22%). Additional demographic information is presented in **Table 1**.

Procedures

Parental/legal guardian written informed consent and adolescent assent were obtained prior to the meeting of the focus group. The groups were conducted in person by a child clinical psychologist trained in qualitative interviewing and experience working with adolescents with chronic pain. The focus groups were conducted using a semi-structured format. Group rules were discussed prior to the start of the focus group. Rules included show respect for different experiences, maintain confidentiality of content discussed following the group, and participants need only answer questions to the extent to which they are comfortable.

TABLE 1 | Demographic information for adolescent participants (*N* = 18).

Demographic characteristics	Number of participants	Percentage of sample (%)
Gender		
Female	16	88.89
Male	1	5.56
Gender fluid	1	5.56
Race/ethnicity		
White, Non-Hispanic	11	66.11
White, Hispanic	3	16.67
Multi-race, Non-Hispanic	2	11.11
Black, Hispanic	1	5.56
Other, Hispanic	1	5.56
Pain diagnoses		
Amplified musculoskeletal Pain syndrome	11	66.11
Abdominal pain	6	33.33
Pain amplification syndrome	5	27.78
Back pain	4	22.22
Chest pain	2	11.11
Irritable bowel syndrome	2	11.11
Complex regional pain syndrome	1	5.56
Fibromyalgia	1	5.56
Neck pain	1	5.56
Migraine	1	5.56

A question route was developed prior to the focus groups (see **Table 2**) as suggested by Krueger and Casey (2015) to provide a sequential question structure that introduced the concept of pain-related stigma gradually. The key questions targeted five primary areas: (1) participants' experiences with initial diagnosis and treatment by medical providers; (2) participants' perceived reactions and level of support from teachers/school staff, family members, and peers; (3) participants' perceptions of exclusion or being treated unfairly; (4) participants' feelings of being ashamed because of pain; and (5) participants' perceptions of being teased or negatively judged because of pain. After participants responded to each question, the interviewer probed for additional information to gain more insight into their experiences.

To encourage feedback from all participants, the focus group facilitator invited any adolescent who had not shared on a topic area, if they felt comfortable, to include their experiences before moving on to another question. All participants shared their experiences in each topic area. A total of five focus groups were conducted and each focus group consisted of 3–4 participants. The duration of focus groups ranged from approximately 36–106 min.

Focus groups were conducted until no additional stigma information was presented by participants and identified during coding for the larger study. The focus groups were recorded. The recordings were transcribed and the transcriptions were reviewed for accuracy. The transcripts were then reviewed by two independent coders using directed content analysis (Hsieh and Shannon, 2005) who obtained 90.34% agreement (552 code agreements out of 611 total codes). The 59 discordant codes were discussed between the coders to determine consensus.

Stigma theory was used *a priori* to develop a stigma-based codebook, which included anticipatory stigma (defined as the perception of negative future reactions to their chronic pain by others) and concealment, defined as intentionally hiding or not disclosing chronic pain symptoms to others. Responses that included these two categories were extracted from the data for a more focused analysis for the current study using inductive content analysis by one coder. This coder engaged in repeated reading and rereading of the transcripts to develop codes that were organized into categories that reflected emerging themes for the current study. The transcripts were revisited on a regular basis to confirm that the codes and themes were reflected in the data (Hsieh and Shannon, 2005). The research literature on concealment in youth with chronic health conditions was also reviewed to provide additional validation of these categories. The coders for the larger and current study were trained and received supervision in qualitative research analyses.

RESULTS

The primary study analyses indicated that pain-related stigma experiences were reported by all participants and experienced across diverse social situations (i.e., medical providers, school

personnel, family members, and peers). Sources of pain-related stigma included disease invisibility and diagnostic uncertainty. In response to these pain-related experiences, anticipatory stigma and concealment of pain symptoms emerged as consequences of perceived pain-related stigma from others. Anticipatory stigma was a reason for participants to conceal their symptoms. In total, 46 responses were included in the analysis of the current study. All participants except one described the use of concealment. The participant who did not use concealment was the only male in our study and he shared an indifference about whether others knew of his chronic pain status.

Inductive content analyses yielded two main themes of responses related to concealment: (1) social motivations to conceal pain, and (2) harmful effects of concealment. Within social motivations to conceal pain, three categories emerged: (1) avoidance of judgment; (2) avoidance of being a social burden; and (3) desire to be treated normally. Harmful consequences of concealment had two categories: (1) social isolation and (2) cognitive burden. These categories are further described below.

Social Motivations to Conceal Pain

Avoidance of Judgment

Participants discussed several ways that concealment of their pain symptoms had social benefits. The most commonly described social motivation for concealment of pain symptoms was avoidance of judgment from others. In reaction to previous experiences of negative interactions from others, most participants in all the focus groups shared that they hid their pain to avoid negative judgment. The majority expressed using concealment as a coping strategy to avoid judgments with school staff. For example, one 15-year-old female shared, "I wouldn't tell [my teachers] I'm having a whole bunch of pain. . .I would get on the bad side of some teachers." Many participants reported that they had experienced negative interactions with teachers and other school personnel when attempting to engage in school accommodations or services for their pain symptoms. They described teachers or school nurses being "frustrated" with them or questioning the need for the accommodation and, as a result, the adolescent engaged in concealment to avoid these interactions. For example, the same 15-year-old stated, "I told this one teacher at the beginning. . .he like got really pissed at me, really fast, so I just told him and he again, he didn't really care or understand. So I just pretty much gave up with that."

A few participants in all of the focus groups described concealment as a way to avoid judgments from family members. One female participant (age 15 years) shared

> "It was more like I was judged by pretty much everyone, even my family and my siblings. My grandmother thought I was just faking it, and pretty much everyone thought that I'm doing it for attention, that it's not real. It's all in my head. More like, I was more like, I felt like I learned to stop telling everyone, more like I kept it inside."

A few participants also described the experience of others believing that they were fabricating their pain symptoms within their own families. Concealment of pain symptoms was described as a way to avoid this judgment.

Specifically regarding peers and friendships, a few participants described using concealment to avoid judgment from peers, but more participants described concealment in the context of avoiding discomfort experienced in reaction to the lack of understanding or empathy. An example of this experience was described by a 15-year-old female participant:

> "I've tried not to tell most of my friends, especially since they don't understand what it is. This year, actually I moved, I have a 504 [school accommodations plan] where I can like sit, like so I can just get out of the classroom, just in case I have to take a walk or ask for a drink of water, and I'll be out for probably like half an hour, 20 min, and this kid didn't understand so I told him to look it up. He didn't understand, so pretty much, like I don't tell anyone, only one person probably knows about it."

A few participants in each of the focus groups discussed concealment in the context of medical provider interactions. For

TABLE 2 | Focus group question sequence.

Question type	Focus group question
Introductory question	Let us go around the room and have everyone introduce themselves by saying what school you go to, grade you are in, and, if you feel comfortable, where you experience pain and how long you have had pain?
Transition question	Has there been challenges you have experienced related to your pain? Tell me more about them?
Key question	Tell me about how your chronic pain was diagnosed. What was your experience of how doctors initially reacted to your pain?
Key question	How have teachers and school nurses reacted to your pain? In what ways have they been supportive? In what ways have they not been supportive?
Key question	Tell me about how other students and friends reacted to your pain? In what ways have they been supportive? In what ways have they not been supportive?
Key question	How do your parents and/or other members of your family support or do not support your pain condition?
Key question	Tell me about any time you have felt excluded or treated unfairly by others because of pain?
Key question	Tell me about any time you have been made to feel ashamed of your pain?
Key question	Tell me about any time you have been teased or judged negatively because of your pain?
Ending question	Of all the topics we have discussed today, which of them would you say are most important to you?
Ending question	If you were to ask other teens about these experiences, what type of questions would you ask? How would you ask them?

example, due to the perception that medical providers would dismiss her pain symptoms, a 15-year-old female participant stated, "They kind of look at you. And you look fine. You look like you're just sitting there, but sometimes you just don't really want to show them how you feel I guess. And they rely on what they see; not really how you are feeling I guess."

Avoidance of Social Burden

Another social motivation to conceal their symptoms was the perception that disclosing their pain symptoms to others would be too much of a burden on others. Social burden was described as both feeling like their pain condition would be difficult to hear and that, over time, others would stop supporting them. All participants of one focus group and a few in two other focus groups described this social burden in the context of family members and friendships. As a result, some participants concealed their pain symptoms to avoid feeling like a hardship on others. For example, one 16-year-old female participant stated:

> "I'm probably ashamed to tell anyone if I have like a new pain because I feel like I've already had so much pain that it's my problem and no one else should have to deal with it. . .it seems like every time I tell someone, it just adds something else they have to deal with, with everything else they have."

In addition to feeling like talking about their pain symptoms would be too much for others to manage initially, participants also described the perception that others might stop supporting them over time. The anticipation of reduced support from others motivated participants to conceal their pain. For instance, as a different 16-year-old female participant expressed, "you have these things going on and you can't complain about every second of every day because otherwise everyone gets pissed off at you."

Desire to Be Treated Normally

The third social motivation to conceal pain involved hiding pain symptoms in an attempt to be socially accepted and seen as "normal." This social motivation was described in by some participants in most of the focus groups with head nodding by all participants when it was described. For example, a 15-year-old female participant also stated, "you don't want to be treated like, kind of like, not like you're a patient, but you want to be treated as if you're like just regular." Regarding services in school, a different 15-year-old female participant stated, "when you have a 504 plan [school accommodations], sometimes you kind of forget about what you have, or you try to ignore it, because you want to be as normal as possible and fit in with everybody, so you don't want to bring it up." Participants described the desire to feel "normal" mostly in the school setting due to the context of their peers.

Harmful Consequences of Concealment

Participants also discussed ways in which concealment was harmful to them in the context of their pain symptoms. Two subcategories emerged regarding negative aspects of concealment. First, participants shared that concealment interfered with their ability to either develop friendships or receive support from others. Second, participants reported that it was cognitively exhausting to constantly keep their symptoms to themselves.

Social Isolation

Participants described ways in which concealment led to further social isolation from others. Social isolation was described by all participants generally, but a few participants in most of the focus groups described it in the context of their use of concealment. The lack of understanding of the stress of their symptoms contributed to loneliness and social isolation from others. A gender fluid 14-year-old adolescent (she/her preferred pronoun) shared,

> "In my school I don't have like any friends and like if I did, they probably wouldn't like if they probably want do a lot of stuff but like I can't like I can't go to the movies all the time because I can't walk around. . .like then I would feel upset because like every day I come home and cry my eyes out because I have no friends."

This participant experienced significant social isolation that was complicated by the perception that peers would not want to be this persons' friend due to her pain symptoms, which was very distressing to her. This experience was shared following another female participant (age 12 years) who discussed a "disconnection" due to her inability to describe her pain to her friends. The majority of participants also shared the challenges with social isolation in either not having friendship or experiencing distance from current friendships due to the lack of understanding regarding their chronic pain.

Cognitive Burden

In order to successfully conceal pain symptoms, adolescents with primary chronic pain need to be vigilant to cues that may reveal their pain symptoms and attempt to hide those cues. This process creates extra cognitive effort for individuals who attempt to conceal their pain, which has been noted in the coping literature (e.g., Wegner and Lane, 1995). Cognitive burden was described in some of the participants in all of the focus groups. As an example, one 15-year-old female participant shared, "Like you get good at covering for pain or just like not showing it . . .I think that's why I'm tired all the time because I'm pushing myself to like cover it up I guess." Another female participant (age 16 years) similarly shared, "But you can't hide like it all the time because sometimes it gets to be too much, so it's sort of regulating that bit." These examples demonstrate that concealment can contribute to a cognitive burden for adolescents with primary chronic pain due to the cognitive effort that this concealment requires.

DISCUSSION

Our findings in these focus groups suggest that concealment is used as a coping strategy in the context of pain-related stigma in our sample of adolescents with chronic pain. Both perceived social benefits and negative consequences of concealment were described. Specifically, social motivations to conceal chronic pain symptoms included (1) avoidance of judgment, (2) avoidance of feeling like a burden, and (3) a desire to feel normal. These

findings parallel previous evidence of social motivations to conceal disease status observed in other pediatric populations (Sunil George and Lambert, 2015; Galano et al., 2017).

Our sample of adolescents with chronic pain discussed ways that the concealment of their pain symptoms from others was socially beneficial to them. The most endorsed reason was the avoidance of judgment by others, which included medical providers, school personnel, peers and family members. Thus, a primary motivation to hide chronic pain symptoms may be avoiding felt stigma experiences. In particular, the use of concealment as a coping strategy was perceived to be helpful in avoiding the experience of social rejection or exclusion by others, which is consistent with previous research on social relationships and youth with chronic pain (Forgeron et al., 2013). Concealment was also used to avoid a lack of understanding from peers, which has been noted to be a social difficulty for youth with chronic pain (Stinson et al., 2014). Prior research has also documented the fear of social rejection in the context of other stigmatized chronic health populations (Kaushansky et al., 2017), and peer victimization has also been documented in adolescents with chronic pain (Forgeron et al., 2010; Fales et al., 2016). Thus, our findings indicate the need for additional research to better understand pain-related stigma, concealment, and peer relationships among adolescents with chronic pain.

There was also a sense that adolescents with chronic pain felt that their physical and emotional needs were considered a burden to others. The experience of feeling like a burden has been identified as one aspect of internalized stigma (Vervoort et al., 2014). In our sample, adolescents described their perceptions that others within their social environment would not have the capacity to handle their needs or lose interest in their friendship over time due to their chronic pain. This notion was a motivating factor in their concealment of chronic pain symptoms, and suggests the importance of future work to better understand the ways in which adolescents with chronic pain may internalize stigma. Moreover, adolescents with chronic pain were motivated to conceal their pain symptoms out of a desire to be treated normally, which has emerged as a theme in other qualitative research in youth with chronic pain (Meldrum et al., 2009). If peers were not aware of the pain that the adolescent was experiencing, the adolescent could experience feeling "normal." Future psychological interventions should target communication and advocacy regarding the chronic pain needs to increase support from others.

Whereas adolescents in our sample identified several beneficial aspects of concealment as a coping strategy, there were several descriptions that indicated that concealment also may have harmful consequences; specifically, social isolation and increased cognitive burden. Social relationships for adolescents with chronic pain can be challenging, and it appears that concealment of pain symptoms may allow for some feelings of normalcy and social support. Social support is a protective factor in the management of chronic health conditions in young populations. However, adolescents with chronic pain who conceal their conditions may inadvertently create further social isolation for themselves by silently suffering in their pain alone. This social isolation has negative health implications for youth with chronic pain (Steele et al., 2002; Forgeron et al., 2010). Clinical interventions should focus on improving social connectedness between adolescents with chronic pain and their primary support groups, specifically friends and family members, in order to reduce social isolation and consequential negative health outcomes.

Another negative consequence of concealment described by our sample was an increase in cognitive burden needed to manage concealment. Cognitive burden in the context of stigma concealment has also been mentioned among individuals of other stigma identities (Quinn, 2018). Adolescents with chronic pain need to stay vigilant to social cues that they may be devalued due to their pain by others, as seen in other stigmatized conditions (Chaudoir and Quinn, 2010). This vigilance can contribute to cognitive and emotional fatigue as the adolescent strains to internalize their pain experience and attempt to behave as if not in pain. The findings of this study revealed the possibility of this process, but more research is needed to determine the presence and frequency of this cognitive vigilance, and its impact on daily and school functioning in adolescents with chronic pain, particularly their psychological wellbeing, concentration and school performance. Additional research is also needed to assess the cognitive burden of concealment in the context of intersecting pain-related stigma and other stigmatized identities (Benson et al., 2017), such as youth who identify as a sexual or gender minority.

Finally, a potential negative implication of concealment that was not discussed by participants in our study, is the potential for medical neglect. If adolescents are concealing their pain episodes, they may fail to seek or receive appropriate or adequate medical care when needed. It may be that social consequences (e.g., peer rejection) of disclosing their pain symptoms are more salient to adolescents than potential medical consequences, especially given their developmental stage and the importance of social relationships and peer acceptance during this time period. Nevertheless, it seems warranted for future research to examine links between concealment of chronic pain and medical consequences/quality of care in adolescents.

Several limitations of this study should be noted. Our sample consisted of mostly female adolescents and only one male and one gender fluid participants, which limits our ability to describe stigma concealment perspectives across gender identifications. Future research should prioritize samples with increased gender and racial/ethnic diversity to determine how experiences of pain-related stigma differ for adolescents across diverse backgrounds. We also did not capture more specific information about the frequency and intensity of their pain condition, which may have provided insight into how concealment may be experienced differently based on disease characteristics. The analysis of the current study is also limited by the exploration of codes that were extracted from the larger study. It is possible that more directed questions about concealment may increase the depth of knowledge on this coping strategy. A limitation for inductive content analysis is that the analysis can be subject to coder bias, which was managed by continual reference

to the data. Due to the cross-sectional nature of the study, we could describe the experience of adolescents with chronic pain in the context of pain-related stigma and concealment, but could not directly link this coping strategy to health outcomes. We also could not include the experience of adolescents who suffer with chronic pain who have not received a formal diagnosis from a pain clinic, for which concealment from pain-related stigma may be intensified.

While our qualitative study suggests the presence of, and motivations for, concealment among adolescents with chronic pain in the context of pain-related stigma, future research should examine in what context or relationships adolescents with chronic pain may feel comfortable disclosing their health status or symptoms. The decision to seek support from others has been studied in other populations with concealable stigmas (Barned et al., 2016; Roberts et al., 2020) and pediatric chronic conditions (Barned et al., 2016). The decision to disclose health status can vary based on situational contexts, which creates challenges in the measurement of stigma concealment due to its existence along a continuum (Pihlaskari et al., 2020). Further, it is important to note that communicating about disclosure of chronic medical conditions can be challenging even with less stigmatizing conditions (Roberts et al., 2020), and the role of communication difficulties related to one's health status may be a contributing factor in lack of disclosure (Remedios and Snyder, 2018; Puhl et al., 2019). A greater understanding of these communication barriers in adolescents with chronic pain may lead to appropriate interventions to improve communications and outcomes.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available because the transcripts are not available publicly due the need to keep the participants anonymous and protect health information. Requests to access the datasets should be directed to EW, ewakefield@connecticutchildrens.org.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Connecticut Children's IRB. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

EW, ML, RP, and WZ developed the purpose and design of the study. EW contributed to the implementation of the study, conducted the analyses and wrote the manuscript with input from all authors. All authors contributed to the article and approved the submitted version.

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Passive Coping Associations With Self-Esteem and Health-Related Quality of Life in Youth With Inflammatory Bowel Disease

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Pediatric patients with inflammatory bowel disease (IBD) may experience chronic stress related to disease symptoms and treatment, with negative consequences to their health-related quality of life (HRQOL). Lower HRQOL among pediatric patients with IBD has been associated with worse disease-related symptoms and psychological functioning, while higher HRQOL has been associated with more adaptive coping with disease symptoms and treatment. In addition, patients' self-esteem may impact the selection and use of coping strategies through global cognitions about their abilities and perceived competence. The current study seeks to extend existing research on HRQOL in youth with IBD by examining cross-sectional associations among self-esteem and passive coping strategies. Youth ages 9–18 with IBD (n = 147) rated their HRQOL using a disease-specific measure, typical strategies used to cope with pain or GI symptoms, and their general self-esteem. Mediation analyses were performed using regression-based techniques and bootstrapping. Results indicated that greater self-esteem was positively associated with HRQOL but negatively associated with passive coping. Controlling for disease activity, age, and gender, significant indirect effects were found in the relation between self-esteem and HRQOL through passive coping. Multiple mediation analyses using the three passive coping subscales found that self-esteem was indirectly associated with HRQOL through its effects specifically on catastrophizing as a passive coping strategy. Results suggest that pediatric patients' general self-esteem can impact their HRQOL through passive coping and specifically, maladaptive cognitions (e.g., catastrophizing). Interventions aimed at addressing both self-esteem and catastrophizing as a passive coping strategy may offer promise for improving HRQOL in youth with IBD.

Keywords: coping, quality of life, inflammatory bowel disease, self-esteem, pediatrics

INTRODUCTION

Youth diagnosed with inflammatory bowel disease (IBD) may experience significant and chronic stress related to the unpredictable, painful, and potentially embarrassing symptoms associated with an IBD diagnosis. As such, psychological and behavioral functioning among youth with IBD may be impacted in a variety of ways compared to healthy peers, including reduced health-related quality of life (HRQOL; Greenley et al., 2010). HRQOL encompasses an individual's subjective perception of their physical, mental, and social functioning in relation to their health (Center for Disease Control and Prevention, 2018). Within the pediatric IBD population, lower HRQOL has been associated with increased disease activity, increased pain perception, and greater depressive symptoms (Herzer et al., 2011; Claar et al., 2017). Given the array of challenges faced by youth with IBD and associated relations to poorer HRQOL, there is a great need to understand and promote effective coping with the disease and health-related distress within this population.

Coping refers to efforts to manage stress in the face of adverse experiences, including pain or disease-related distress. The disability-stress-coping model (Wallander and Varni, 1998) theorizes that a youth's adjustment or distress related to the disease is impacted by both risk and resilience factors. Risk factors include disease activity or symptoms, functional disability related to disease, and psychosocial stressors, while resilience factors include available family or social supports and the use of effective coping strategies. Within this model, coping is theorized to mediate the relations between psychosocial stress and adjustment to medical conditions. Notably, not all cognitive or behavioral coping strategies are adaptive. Adaptive coping strategies, such as active problem solving or social support seeking, are distinct from maladaptive coping, including pain catastrophizing, avoidance, or behavioral disengagement. Adaptive coping strategies are thought to be more organized, flexible, and problem-focused and thus have a positive impact on patient long-term functioning while maladaptive strategies are more disorganized and rigid and are frequently characterized by greater perceived helplessness, thus having a more negative impact on long-term functioning (Skinner et al., 2003). Importantly, maladaptive coping strategies may impede the development of other more adaptive coping resources through negative appraisal of self or situation (Skinner et al., 2003). As such, it is critical to understand and assess coping strategies, particularly among vulnerable populations, including children and adolescents with IBD.

The extent to which patients with IBD engage in maladaptive coping strategies may have important implications for their quality of life, and there is some evidence that youth with IBD may use maladaptive coping strategies more frequently than their healthy peers. For example, compared to healthy peers, youth with IBD have reported greater use of avoidance as a maladaptive behavioral coping strategy (Van der Zaag-Loonen et al., 2004). Notably, pain catastrophizing, a maladaptive cognitive coping strategy, demonstrated the strongest impact on HRQOL among youth with IBD, even when compared to assessments of disease severity and psychological distress (De Carlo et al., 2019). In contrast, among youth using adaptive cognitive coping strategies, including those with greater optimism and perceived control, greater social functioning was reported, while those reporting less perceived control reported poorer social and emotional functioning (Van der Zaag-Loonen et al., 2004). These findings underscore the impact of coping strategies on HRQOL outcomes within the pediatric IBD population. However, other aspects of youth functioning may influence coping; thus, it is not yet clear how to best promote the use of adaptive cognitive and behavioral coping strategies to improve HRQOL among these youth.

Self-esteem is an important perception of the self, varying between individuals, which may relate to the extent to which youth engage in adaptive coping. Self-esteem includes one's perceived competence and worth regarding their abilities, skills, and qualities, further motivating one's cognitions and behaviors (Mruk, 2013). Self-esteem is particularly relevant to the coping literature, as the selection and use of either adaptive or maladaptive coping strategies may vary based on cognitions or behaviors influenced by self-esteem. Self-esteem may be further impacted by self-perceptions of appearance, social acceptance, and perceived competence in managing stress, elements which may be uniquely impacted by IBD symptoms and disease course. Interestingly, a recent meta-analysis founds no significant difference in self-esteem between healthy youth and youth with IBD, or youth with IBD compared to youth with other chronic illnesses (Greenley et al., 2010). However, greater disease severity was significantly related to lower self-esteem among adolescents with IBD (Lindfred et al., 2008), suggesting self-esteem may have disease-specific relevance for youth with IBD. Self-esteem has been shown to relate to multiple important outcomes among youth with IBD, including emotional functioning, psychological adjustment, and HRQOL (De Boer et al., 2005). However, the mechanism through which self-esteem relates to these outcomes among youth with IBD is not yet known, nor is it clear how to effectively intervene and improve HRQOL in these youth.

One such possibility is that improved self-esteem may relate to more effective and active cognitive and behavioral coping mechanisms and thus promote better HRQOL, while poorer self-esteem is associated with more maladaptive coping mechanisms and thus poorer HRQOL. Among adults with IBD, greater self-esteem has been associated with greater perceived self-efficacy in coping with life demands (Opheim et al., 2020). As such, self-esteem may impact the differential use of adaptive or maladaptive coping mechanisms in response to disease symptoms and treatment. However, no such studies have examined relations between self-esteem and coping among youth with IBD. Further, an examination of the relations among self-esteem, coping strategies, and resulting impacts on HRQOL appears particularly warranted to further inform interventions aimed at improving HRQOL among youth with IBD. The current study sought to extend existing research on HRQOL in youth with IBD by examining associations between self-esteem and specific maladaptive, passive coping mechanisms as they relate to perceived HRQOL. We hypothesized that the

relation between self-esteem and HRQOL among youth with IBD would be mediated by youth's reported use of passive coping strategies, such that poorer self-esteem would relate to poorer HRQOL through greater use of maladaptive, passive coping mechanisms.

MATERIALS AND METHODS

Participants

The sample consisted of 147 children and adolescents diagnosed with IBD and their parents. Participants were enrolled in a randomized controlled trial (RCT) of a cognitive-behavioral intervention for pediatric IBD patients (Levy et al., 2016). The present paper examines a distinct set of research questions using cross-sectional data collected at baseline prior to randomization or intervention. Interested readers can refer to our previously published papers examining disease and psychosocial characteristics in these participants at baseline (Langer et al., 2014; Van Tilburg et al., 2015; Reed-Knight et al., 2018). Of the 210 dyads enrolled in the RCT, 63 were excluded from the current analysis subsample for the following reasons: baseline assessment not complete (20 dyads), child data missing for assessments included in the current investigation (38 children), and non-independence among siblings as the family had two children with IBD enrolled in the RCT (5 dyads).

Participants were recruited from the gastroenterology departments of Seattle Children's Hospital in Seattle, WA, and Mary Bridge Children's Hospital in Tacoma, WA. All procedures were approved by the Institutional Review Boards of both institutions. Inclusion criteria consisted of (1) child age of 8–18 years, (2) child diagnosed with IBD for at least 3 months, (3) child medically approved to engage in normal daily activities at the time of recruitment, (4) child and parent participant had cohabitated for at least the past 3 months, and (5) child and parent English fluency. Exclusion criteria included (1) child diagnosed with a chronic disease other than IBD and (2) developmental disability requiring full-time special education or impairing the ability to participate in study procedures.

Measures

Child-Reported Measures

Children and adolescents completed a battery of questionnaires *via* telephone for the baseline assessment, administered by a nurse researcher. To facilitate comprehension, answer choices were mailed to participants in advance of the telephone session.

Self-Esteem

Children's self-esteem was assessed using the Global Self-Worth subscale from The Self-Perception Profile for Children (Harter, 2012). The Global Self-Worth subscale, conceptualized as analogous to overall self-esteem by Harter, assesses a respondent's general sense of worth as a person. The subscale evaluates how much one likes oneself as a person, is happy with the way one is leading one's life, and is generally happy with the way one is, as a human being. Each of the six items on the Global Self-Worth subscale asks respondents to pick which of two descriptions of a person he or she is most like and rate whether the chosen description is "Really True for Me" or "Sort of True for Me." Each item is then scored on a four-point scale from 1 to 4, where a score of 1 indicates the lowest perceived competence and a score of 4 reflects the highest level of competence. The mean rating is then calculated, with higher scores in the range of 1–4 representing higher perceived self-esteem. The internal consistency for the Global Self-Worth subscale in the current sample was good (α = 0.82).

Passive Coping

Children's coping with abdominal pain or other GI symptoms was measured with the Pain Response Inventory (PRI; Walker et al., 1997). The PRI consists of three higher-order scales (active, passive, and accommodative coping). As passive coping has been associated with poorer outcomes in patients with abdominal pain in past research (Walker et al., 2005; Van Tilburg et al., 2015), the current study focused on this domain. The Passive Coping scale consists of Catastrophizing (five items, such as "Think to himself or herself that it's never going to stop"), Self-Isolation, (five items, such as "Try to be alone."), and Behavioral Disengagement (five items, such as "Give up trying to feel better"). Parents rated how often their child employed each of these coping strategies on a 0–4 (never to always) scale when experiencing abdominal pain or other GI symptoms. Items on the Passive Coping scale are summed and averaged to obtain a mean subscale score, which can range from 0 to 4.00. The internal consistency for the PRI within the current sample was good (α = 0.90).

Health-Related Quality of Life

Health-related quality of life was assessed using the 35-item child-report IMPACT-III (Otley et al., 2002). The IMPACT-III is an IBD-specific measure of HRQOL validated in children and adolescents ages 8–17. It is comprised of six domains: bowel symptoms, systemic symptoms, emotional functioning, social functioning, body image, and treatment/interventions. Each item [e.g., *How often did you have to miss out on certain things (hobbies, play, parties) because of your inflammatory bowel disease in the past 2 weeks?*] has five response options, with response labels that differ from item-to-item. To gain a comprehensive representation of children's HRQOL, the present study utilized the total score, which was calculated by summing all 35 items, with higher scores representing better HRQOL. The possible range for the total HRQOL score is 35–175. In the present study, the internal consistency for the IMPACT-III total score was excellent (α = 0.93).

Pediatric Gastroenterologist Completed Measures

Each patient's treating gastroenterologist completed an index of disease activity for study purposes during the patient recruitment visit.

Disease Activity

Disease activity was assessed *via* the Pediatric Crohn's Disease Activity Index (PCDAI; Hyams et al., 1991) for children with Crohn's disease and *via* the Pediatric Ulcerative Colitis Activity Index (PUCAI; Turner et al., 2007) for children with ulcerative colitis. The PCDAI is a rating of clinical disease activity which incorporates patient report, laboratory values, physician examination results, and growth parameters. PCDAI scores range from 0 to 100. Scores <10 reflect disease remission; scores of 11–30 reflect mild disease activity; and scores >30 reflect moderate to severe disease activity. The PUCAI is a 6-item validated measure of disease activity based on patient reports of symptoms. The PUCAI is completed by pediatric gastroenterologists and scores range from 0 to 85. Scores <10 reflect remission; scores of 10–34 reflect mild disease activity; scores of 35–64 reflect moderate disease activity; and scores of 65 or higher reflect severe disease activity. For the purposes of this study, participants were categorized as experiencing remission/no disease activity or mild/moderate/severe disease activity in **Table 1**. As has been done in past research, a continuous variable representing disease activity using scores from the PCDAI and the PUCAI was also created in order to control for disease activity in mediation analyses (Reed-Knight et al., 2016). Since the PCDAI and the PUCAI are rated on different scales, PUCAI scores were converted to the PCDAI's 0 to 100 total scaling by multiplying each PUCAI score by (100/85), resulting in a combined, continuous measure of disease activity for analyses.

Analyses

Descriptive statistics and correlation analyses were used to describe the sample in terms of demographics and study variables. Mediation analysis was performed using Hayes' PROCESS macro, a regression-based path analytic technique (Hayes, 2017). Using an ordinary least squares framework, PROCESS estimates direct and indirect effects in multiple mediator models. To test mediation hypotheses, PROCESS uses bootstrapping to construct confidence intervals for indirect effects through the repeated sampling of the dataset. Findings are based on 5,000 bias-corrected bootstrapped samples. In the event that zero does not lie within the 95% confidence interval for the bootstrapped results for indirect effects, we can conclude that the indirect effect is significantly different from zero and that mediation is demonstrated (Preacher and Hayes, 2004). In the mediation model, disease activity was included as a continuous covariate after computing a combined disease activity score using PCDAI and PUCAI values as described above. Analyses were conducted using IBM Statistical Package for the Social Sciences 26.0.

TABLE 1 | Sample characteristics (n = 147 pediatric patients with IBD).

Characteristic	Child
Age, *M* (SD)	13.88 (2.53)
Age, range	9–18
Gender, *n* (%) female	71 (48.3)
Ethnicity, *n* (%) hispanic	6 (4.1)
Race, *n* (%) caucasian	127 (86.4)
Disease, *n* (%)	
Crohn's disease	101 (68.7)
Ulcerative colitis	46 (31.3)
Disease activity, *n* (%)	
Quiescent	99 (67)
Mild/moderate/severe	48 (33)

RESULTS

Descriptive Results

Descriptive data for the sample are provided in **Table 1**. The majority of the sample was classified as having inactive disease based on clinical disease activity indices (67%), and the remaining was classified as experiencing active disease at the time of data collection (33%). Patients classified as experiencing active disease reported significantly lower disease-specific HRQOL (M = 122.24, SD = 20.51) compared to those classified as experiencing inactive disease (M = 135.45, SD = 17.22), $t(145) = 4.24$, $p < 0.001$. No differences in self-esteem or passive coping were observed based on disease activity. No differences in self-esteem based on gender were observed, but males reported significantly higher disease-specific HRQOL (M = 134.54, SD = 18.35) compared to females (M = 124.70, SD = 20.06), $t(145) = 3.10$, $p = 0.002$. On the Passive Coping subscales, females reported higher levels of self-isolation (M = 1.11, SD = 1.05 vs. M = 0.80, SD = 0.87) compared to males, $t = -1.95$, $p = 0.05$. In addition, females reported higher levels of behavioral disengagement (M = 0.75, SD = 0.86 vs. M = 0.48, SD = 0.60), $t = -2.23$, $p = 0.028$, and catastrophizing (M = 1.12, SD = 0.86 vs. M = 0.79, SD = 0.72), $t = -2.54$, $p = 0.01$, compared to males. Correlations among study variables are provided in **Table 2**.

Simple Mediation Model

We first examined associations between child-reported self-esteem, disease-specific HRQOL, and passive coping strategies using the Passive Coping scale from the PRI. Disease activity, child age, and child gender were included as covariates given their significant relations with disease-specific HRQOL and passive coping strategies (**Table 2**). In a simple mediation analysis, self-esteem was indirectly associated with HRQOL through its effect on passive coping. Self-esteem was negatively associated with the use of passive coping strategies ($a = -0.40$), and youth who reported greater use of passive coping strategies reported lower HRQOL ($c = -11.18$). A bias-corrected bootstrap confidence interval for the indirect effect controlling for disease activity, child age, and child gender (ab = 4.43) based on 5,000 bootstrap samples was entirely above zero (2.09–7.08).

Multiple Mediator Model

As simple mediation analysis supported a relationship between self-esteem and disease-specific HRQOL through the Passive Coping scale, we evaluated a multiple mediator models using the three Passive Coping subscales: Catastrophizing, Behavioral

TABLE 2 | Correlations among study variables and descriptive statistics (n = 147).

S.No		1	2	3	4	5	6	7	8	*M* (SD)	Observed range
1	Self-esteem	1.00	−0.42**	−0.35**	−0.41**	−0.30**	0.48**	−0.15	−0.17*	3.29 (0.65)	1.17–4.00
	PRI: Passive										
2	Coping total		1.00	0.82**	0.84**	0.84**	−0.60**	0.07	0.28**	0.84 (0.70)	0–3.47
3	PRI: Self-Isolation			1.00	0.50**	0.47**	−0.44**	−0.04	0.37**	0.95 (0.97)	0–4.00
4	PRI: Behavioral Disengagement				1.00	0.67**	−0.52**	0.13	0.18*	0.61 (0.75)	0–3.80
5	PRI: Catastrophizing					1.00	−0.56**	0.11	0.12	0.95 (0.80)	0–3.20
6	HRQOL						1.00	−0.25**	−0.38**	129.79 (19.76)	68.00–161.00
7	Disease activity							1.00	0.01	10.29 (13.82)	0–76.47
8	Child age								1.00	13.88 (2.53)	9–18

*$p < 0.05$ and **$p < 0.01$. *PRI, pain response inventory.*

Disengagement, and Self-Isolation. **Figure 1** displays the full model with unstandardized B weights for the path coefficients. Multiple mediation analyses conducted using ordinary least squares path analysis found that self-esteem was indirectly associated with HRQOL through its effects specifically on catastrophizing as a passive coping strategy controlling for disease activity, child age, and child gender.

In the full model with the three Passive Coping subscales, self-esteem was negatively associated with self-isolation ($a_1 = -0.44$, $p < 0.01$), behavioral disengagement ($a_2 = -0.42$, $p < 0.01$), and catastrophizing ($a_3 = -0.32$, $p < 0.01$). Only catastrophizing was negatively associated with disease-specific HRQOL ($b_3 = -8.56$, $p < 0.01$). Mediated effects were demonstrated by significant indirect effects in the relation between self-esteem and disease-specific HRQOL through catastrophizing [path a_3b_3 point estimate = 2.76, standard error (SE) = 1.21, and 95% CI = 0.75 to 5.43]. The full model including mediators and controlling for disease activity, child age, and child gender accounted for 54% of the variance in disease-specific HRQOL; self-esteem alone controlling for disease activity, child age, and child gender accounted for 40% of the variance in disease-specific HRQOL.

DISCUSSION

Maladaptive coping strategies, including catastrophizing and avoidance, are known to be associated with poorer HRQOL in patients with IBD (De Carlo et al., 2019), though the higher-order factors driving the use of particular coping strategies are less understood. In this study, we sought to investigate relations between global self-esteem, a higher-order construct representing one's perceived competence and abilities, and disease-specific HRQOL. As expected, higher global self-esteem was associated with higher HRQOL. We hypothesized that global levels of self-esteem may impact the differential use of coping strategies in response to coping with IBD. To evaluate this, we examined passive coping, a maladaptive coping style characterized by catastrophizing, behavioral disengagement, and self-isolation. After finding supports for a model in which global self-esteem is associated with HRQOL through passive coping, we sought to further specify this relationship by examining the three passive coping subscales and interestingly found that catastrophizing alone accounted for the indirect effect in the relation between self-esteem and disease-specific HRQOL.

Results build upon work in adults with IBD, where greater self-esteem has been associated with greater perceived self-efficacy in coping with life demands (Opheim et al., 2020). In the present study, youth with greater global self-esteem were less likely to endorse passive coping through catastrophizing, in which they have negative expectations for their ability to manage pain or symptoms. Results suggest that as youth report higher global perceptions of their competence and abilities they experience lower use of passive coping strategies overall and fewer catastrophic thoughts about their abilities to manage pain or symptoms, both of which were associated with poorer HRQOL.

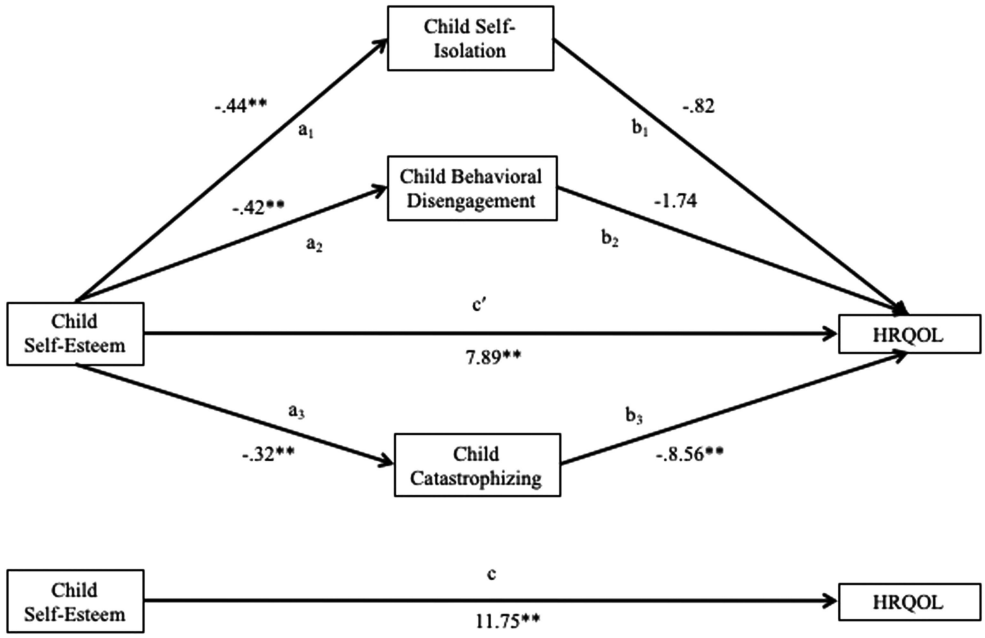

FIGURE 1 | Multiple meditation model of HRQOL. a_3b_3, significant indirect effect. Disease activity, child age, and child gender included as covariates. $^{*}p < 0.05$, $^{**}p < 0.01$.

Though not specific to their disease or coping, global self-esteem seems to represent an overarching, higher-order factor impacting the way in which youth think about and cope with the distress of IBD. We controlled for disease activity, child age, and child gender given the relatively large age range of our sample and observed differences in disease-specific HRQOL based on disease activity and child gender, as well as higher levels of passive coping observed in females relative to males. Past research conducted in the Netherlands also founds that male youth with IBD reported higher levels of disease-specific HRQOL compared to females (Van der Zaag-Loonen et al., 2004), and higher levels of passive coping have been observed in females compared to males in non-IBD samples (Hampel and Petermann, 2005). It is well known that girls are more prone to internalizing symptoms from adolescence onwards, and results suggest that passive coping may be at least one risk factor placing girls at increased risk for negative psychological symptoms.

Cognitive-behavioral interventions targeting passive coping and catastrophizing typically address these maladaptive thoughts and behaviors directly, teaching youth to engage in more adaptive ways of thinking about and responding to their pain or symptoms. Such interventions, including work conducted by authors of the current study, have shown strong results in increasing functioning and quality of life for patients with IBD (Levy et al., 2016). However, the present study suggests that addressing global beliefs about overall competence, abilities, and self-worth may offer an additional avenue for improving HRQOL. Such an approach would be more consistent with classic applications of cognitive therapy for depression, in which core beliefs are the ultimate treatment target to change patient's underlying, maladaptive beliefs about their self-worth (Compton et al., 2004). For a patient with poor global self-esteem, identifying and addressing behavioral deficits in functioning leading to negative self-evaluations, as well as maladaptive thought patterns, may offer an avenue for impacting how he or she approaches to pain and symptom-related thoughts and ultimately improving HRQOL. In addition, research is needed to test whether improvements in functioning and adaptive coping for chronic pain patients treated with exposure-based therapies extend to patients with IBD (Vlaeyen and Crombez, 2019; Lalouni et al., 2020).

Limitations of this study should be considered when interpreting results, which inform future research directions. First, the use of cross-sectional and observational data precludes us from making conclusions regarding causation. Future research should utilize a longitudinal study design to determine how self-esteem and passive coping are temporally related to disease-specific HRQOL and how these factors impact one another over time. Further, empirical exploration of whether interventions aimed at improving self-esteem subsequently decrease the use of passive coping strategies and increase HRQOL is warranted. In the present study, the majority of children was experiencing inactive disease, and results may not generalize to children with active disease. Future research should examine associations between self-esteem, passive coping, and disease-specific HRQOL among children experiencing active disease to determine how disease activity may impact observed relations. Telephone administration of child study questionnaires as opposed to self-administration may have impacted responses if children underestimated symptoms due to social desirability or embarrassment. However, past research has demonstrated comparability between phone interviews and

in-person interviews for the assessment of emotional symptoms (Rohde et al., 1997). Additionally, we are unfortunately unable to report data on recruitment rates or potential differences between participants and those who were approached for participation but declined, as these data points were not collected. Finally, the present sample was primarily Caucasian, which may limit generalizability to youth with IBD from different ethnicities. As such, future research is needed to better understand how self-esteem and the use of passive coping strategies are related to HRQOL among youth of different races and ethnicities. While the present model accounted for over half of the variance in disease-specific HRQOL, future research should explore other factors influencing HRQOL and use of specific coping strategies, such as catastrophizing among youth with IBD, including the potential role of family and caregivers' own coping and the impact of environmental and social contexts on children's coping with IBD. Such work would be a logical extension of the disability-stress-coping model (Wallander and Varni, 1998) informed by the current study.

This study provides initial evidence using cross-sectional data that global self-esteem, a higher-order construct representing one's perceived competence and abilities, is associated with the extent to which youth utilize passive coping strategies and engage in catastrophizing, as well as disease-specific HRQOL. Results suggest that a child's global self-esteem may impact the degree to which adaptive or maladaptive coping strategies are utilized to manage the stressors associated with IBD. As interventions are developed to promote adaptive coping for patients with IBD, it will be important to measure the extent to which beliefs about one's abilities and competence are associated with uptake of adaptive or maladaptive coping strategies and whether targeting low self-esteem increases utilization of adaptive coping strategies.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available because the data continue to be utilized for manuscripts and presentations. Requests to access the datasets should be directed to rlevy@uw.edu.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Seattle Children's Hospital and Mary Bridge Children's Hospital. Written informed consent to participate in this study was provided by the participants' legal guardian/ next of kin.

AUTHOR CONTRIBUTIONS

BR, MT, and RL contributed to the conception and design of the study. BR performed the statistical analysis. BR and KR wrote the first draft of the manuscript. All authors contributed to manuscript revision, read, and approved the submitted version.

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Parental Catastrophizing and Goal Pursuit in the Context of Child Chronic Pain: A Daily Diary Study

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Background: Despite daily variability in children's chronic pain experiences, little is known about how parents' emotions and goals toward their child's pain are influenced by these daily changes. This diary study examined how daily child pain intensity (as perceived by parents) moderates the associations between parental catastrophic thoughts about child pain on the one hand, and daily parental distress and parents' goals with regard to their child's pain (pain control vs. activity engagement) on the other hand.

Method: Participants were 25 parents of 20 different children ($N = 18$; 90% girls). Children, aged 8–14 years ($M = 9.5$, $SD = 2.09$), experienced either chronic headache or functional abdominal pain with an average pain duration of 22.5 months ($SD = 24.5$ months). Daily parental responses (i.e., perceived child pain intensity, distress and goal endorsement) were collected through a 3-week daily diary (resulting in 413 valid diary reports). Parents completed the Pain Catastrophizing Scale for Parents prior to starting the diary (PCS-P general) and a daily measure (PCS-P daily) included in the diary. To account for the interdependence of the data, the data were analyzed using multilevel modeling.

Results: Perceived daily child pain intensity moderated the impact of parental general and daily catastrophic thoughts on parents' daily distress. Only for parents experiencing low general catastrophic thoughts an increase in distress was observed on days when they perceived their child's pain intensity as high. For all parents, high levels of perceived child pain intensity were related to more distress on days where parents reported high levels of catastrophic thinking (i.e., PCS-P daily). Perceived daily child pain intensity also moderated the impact of parental general catastrophic thinking on parents' daily endorsement of goals. Parents with high levels of general catastrophic thinking reported a lower focus on child pain control on days when child pain intensity was perceived to be low. Parents with low general catastrophic thinking reported lower endorsement of the activity engagement goal on days where the child's pain intensity was perceived to be low.

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Conclusion: These findings highlight the complexity of daily fluctuations in parental distress and goals regarding their child's pain. Clinical implications and future directions are critically assessed.

Keywords: chronic pain, parents, diary, catastrophizing, goals, distress

INTRODUCTION

Chronic pain in children is a common and serious health- and developmental problem that has a major impact on the child's daily living (Palermo, 2000; Oddson et al., 2006). Prevalence of chronic pain in children ranges between 11 and 38%, increases with age, and is higher in girls as opposed to boys (Perquin et al., 2000; King et al., 2011). The most frequently examined types of chronic pain in children are abdominal pain, headache, back pain, and musculoskeletal pain (King et al., 2011). Children who report a high frequency of pain and high current pain generally report a lower quality of life (Oddson et al., 2006). Furthermore, children experiencing chronic pain have a higher risk for developing psychiatric disorders, especially anxiety disorders or depression, compared to children without chronic pain complaints (Palermo, 2000; Machnes-Maayan et al., 2014). However, many children function well despite the presence of chronic pain. Whether and how much pain-related disability a child with chronic pain experiences, depends, among other things, on several child characteristics. For example, children with higher levels of anxiety sensitivity and fear of pain report more pain-related disability (Martin et al., 2007). However, to fully understand children's pain-related disability it is crucial to also take into account the role of how parents respond to and cope with their child's pain experiences (Goubert and Simons, 2013).

Childhood chronic pain can be considered a substantial stressor for parents for which they need to find appropriate coping approaches to avoid or minimize pain-related disability. According to the Interpersonal Fear Avoidance model (IFAM; Goubert and Simons, 2013), a pain-specific cognitive process, important to understand how parents cope with and respond to their child's pain, is catastrophic thinking (Goubert et al., 2006). Catastrophic thinking is the tendency to focus upon the threat value of the pain stimuli, to exaggerate the threat value and to negatively evaluate one's own (or one's child's) abilities to handle the pain (Sullivan et al., 2001). Parental catastrophic thinking is thus an exaggerated negative mental set or response that emerges during painful experiences of one's child and might have negative consequences. Catastrophic thinking can be assessed as either a trait or state characteristic, both of which will be examined within this study. We will use the term "general" to refer to trait levels of catastrophizing and "daily" for state levels of catastrophizing. There is, indeed, an abundance of evidence supporting the assumptions of the IFAM by showing how parental catastrophic thinking is related to heightened feelings of distress and maladaptive coping responses, such as pain-attending/protective behaviors, which in turn are related to more pain and disability in children (Goubert and Simons, 2013).

To enhance understanding of parental coping approaches and their impact on child functioning, comprehension of parents' underlying goals is needed. Based on the tenets of motivational theories on goal-directed behavior (Riediger and Freund, 2004), it is likely to assume that parents have multiple goals for their children, such as pain relief, attending school, socializing with friends, and engaging in leisure activities (e.g., sports, arts; Rasmussen et al., 2006). Not all goals might be compatible with each other, in which case parents need to prioritize some goals over other (Riediger and Freund, 2004). In line with the above-described evidence, parents with high levels of catastrophic thinking might be more protective toward their child in pain because they prioritize the goal to relieve their child's pain over the other goals they have for their child (Caes et al., 2012). However, little is known as to why this is the case. To gain a better understanding of parental coping mechanisms to deal with their child's pain, more research is needed to explore parental goals when faced with child (chronic) pain, and how the endorsement of different goals varies depending on daily child pain characteristics. On a daily basis, parents may want to pursue the goal to control or relieve their child's pain as well as the goal to encourage their child toward engagement in daily activities (such as attendance at school or leisure activities) in the presence of pain. While for some occasions, both goals could be achieved through engagement in the same coping approach (e.g., promoting distraction), in other circumstances this might not be possible leading parents to perceive these two goals as potentially incompatible. Hence, a careful balance in pursuing these two goals may have important clinical consequences in terms of adequate child functioning and development.

As any goal, the goal of controlling pain can be attained by different parental responses, such as comforting or distracting their child (Carver and Scheier, 2001; Riediger and Freund, 2004; Rasmussen et al., 2006). The adaptive or maladaptive impact of different parental behaviors upon child functioning might depend on the extent to which behavior is primarily and inflexibly driven by the parental goal for pain control at the expense of other important aspects/goals in their child's life. Specifically, although the use of coping strategies, such as distraction, could be motivated by the goal of controlling child pain, engaging in distraction may also reflect parental attention for other aspects of child functioning despite the pain. This could explain the positive influence of this coping strategy on child functioning (Gonzalez et al., 1993; Sweet and McGrath, 1998; Blount et al., 2008; MacLaren Chorney et al., 2009). In contrast, parental protective responses, such as allowing the child to stay home from school, may reflect a strong priority of parents to reduce pain even if this negatively impacts their child's daily functioning substantially. Further research is needed to investigate how parents flexibly

attune between child pain needs (i.e., pain control) and non-pain needs and how this translates into behavior.

Given the fluctuating nature on a daily basis of chronic pain and the mutual influence between child pain characteristics and parental responses proposed in the IFAM (Goubert and Simons, 2013), it is plausible to assume that the way parents attune to their child's needs is likely to fluctuate and depend on daily pain characteristics of the child. Indeed, preliminary evidence, using a vignette methodology (Caes et al., 2012), revealed that higher levels of child pain intensity are related to parents attaching more priority to pain control goals. Nevertheless, research on the role of catastrophic thinking in understanding how parents cope with their child's pain is limited on focusing mostly on general tendencies, while little is known about the daily fluctuations in parental catastrophic thinking and its subsequent impact on parental distress and behavioral tendencies/underlying goals. The usage of diary methodologies is lacking in this context but could offer a unique insight into and capture these daily fluctuations in childhood pain experiences and how this impacts parental coping mechanisms on a daily basis.

Consequently, the primary aim of the current diary study, in children (8–14 years) with chronic headache or chronic functional abdominal pain, was to examine the associations between parental catastrophizing about the pain of their child, both in general and on a daily level, on the one hand, and daily parental distress and daily parental goals of child pain control vs. child activity engagement on the other hand. We expected that parents with high levels of catastrophic thoughts about their child's pain, compared to parents low on catastrophic thinking, would (1) experience more daily distress, (2) endorse the goal to relieve the pain of their child to a higher extent on a daily basis, but the goal to engage their child in activity engagement (in the presence of pain) less. While no difference in the direction of the associations was expected for general vs. daily parental catastrophic thinking, we expected that the hypothesized associations would be stronger for daily levels of catastrophizing compared to general levels (Durand et al., 2017; see **Figure 1**). As a second aim, the role of daily child pain intensity, as reported by the parent, was examined (further referred to as "perceived daily child pain intensity"). We expected perceived daily child pain intensity, to be (1) positively associated with daily parental distress and the daily parental goal to control their child's pain, and (2) negatively associated with the daily parental goal to engage their child in activity engagement. Furthermore, we hypothesized that higher perceived child pain intensity would strengthen the associations between (general and daily) parental catastrophizing on the one hand, and parental distress and the extent to which parents endorse child pain control or activity engagement goals on the other hand (see **Figure 1**).

MATERIALS AND METHODS

The study is part of the G-GiCPP project (Ghent—Goals in Chronic Pediatric Pain—Project) that consists of two parts; a cross-sectional questionnaire part, followed by an optional 3-week diary part, completed by both children with chronic pain and their parents. The current manuscript focuses on reporting the findings from the diary as completed by the parents and on parental catastrophizing as measured during the questionnaire study prior to starting the diary. The study was approved by the Ethics Committees of the Faculty of Psychological and Educational Sciences of Ghent University, University Hospital Ghent, AZ Maria Middelares Ghent, and General Hospital Nikolaas, Belgium.

Participants

Families with children with chronic headaches or chronic functional abdominal pain were recruited through four Flemish hospitals (University hospital Ghent, Maria Middelares Ghent, Jan Palfijn Gent, general hospital Nikolaas). To be eligible for study participation, children had to be aged between 4 and 16 years, children had to be diagnosed by their physician with chronic headache or chronic functional abdominal pain, and both parent and child had to be Dutch-speaking. Children with a developmental disorder, mental delay or migraine were excluded. Families who met the inclusion criteria were informed about the study by their physician. Eighty-two families agreed to be contacted for study participation, of which 48 families (58%; with data from 62 parents) participated in the first part of the study (i.e., the questionnaire part). The main reason for non-participation was parent-reported lack of time. Of the 48 families (covering 62 parents) that completed the questionnaires, 24 families (50%; with data from 32 parents, 52%), also completed the diary part. Due to methodological reasons (i.e., < completed <50% or 11 days of the diary entries) 7 parents were excluded from the analyses. The final sample consisted of 25 parents (5 mother-father dyads, 14 mothers only, 1 father only) of 20 different children. Demographics of the final sample of 25 parents, across 20 children, are presented in **Table 1**.

Data Collection Procedure

All families who were interested in participation and gave written consent to transfer their contact details to the researchers received a phone call with further detailed information about the study. If after receiving all necessary information parents were interested to participate, a home visit was scheduled. During the home visit, the study information was repeated, and parents provided informed consent for themselves and for the child. Children older than 12 years provided, in addition, written informed consent for themselves. In a first phase all parents and children (≥9 years) completed questionnaires under supervision of a research assistant and the procedure of the diary study was explained. Following the home-visit, parents were sent an email containing a secured weblink to the online diary (LimeSurvey software). Parents were asked to complete the diary every evening for 21 consecutive days. Diaries were completed during school weeks and started shortly after the home visit. All families received 30 EUR as a compensation for participation-related costs.

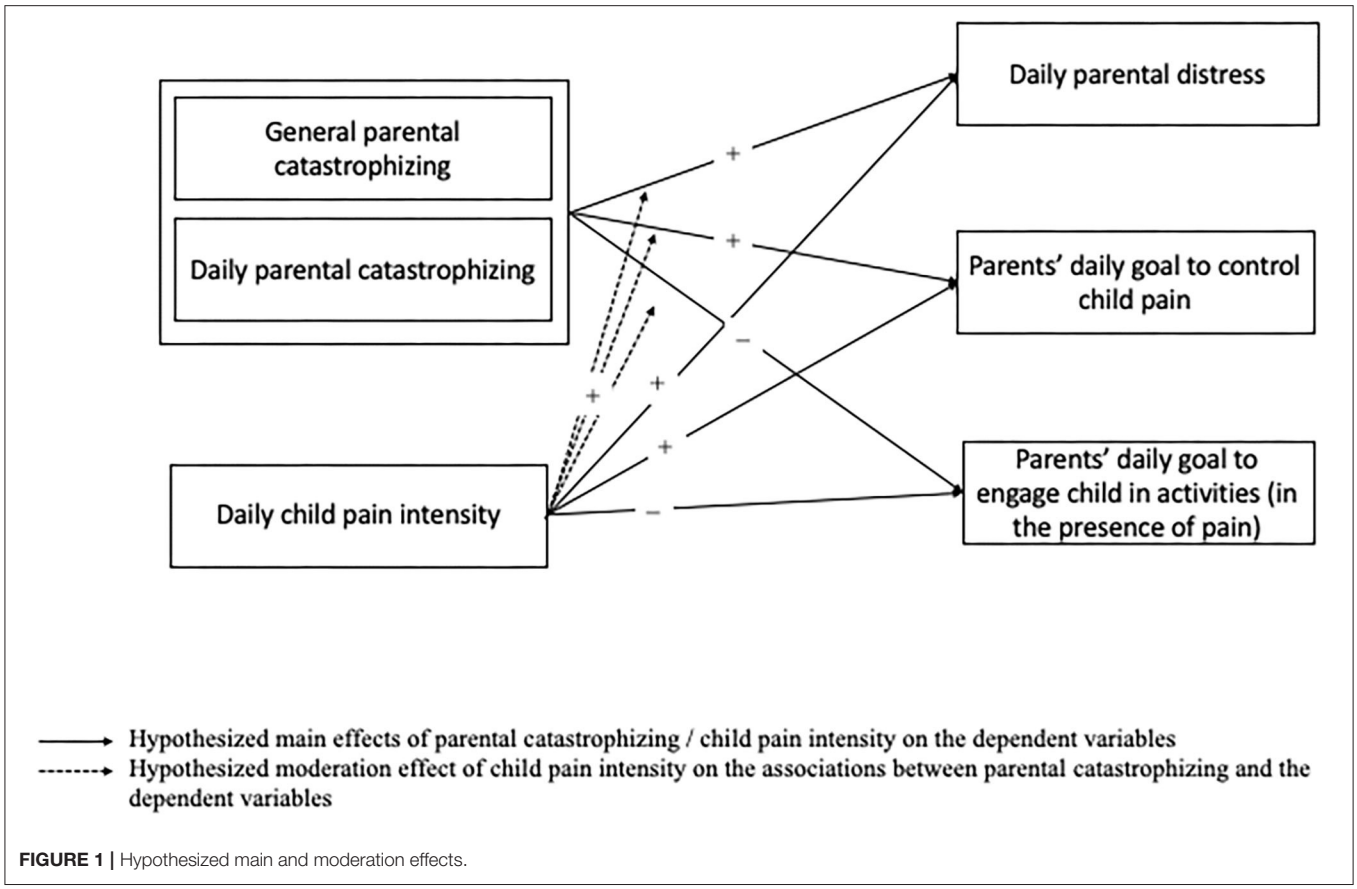

FIGURE 1 | Hypothesized main and moderation effects.

Questionnaire Measures

General parental catastrophizing about their child's pain was assessed by the Dutch Pain Catastrophizing Scale for Parents (general PCS-P; Goubert et al., 2006), an adaptation of the adult Pain Catastrophizing Scale (PCS; Sullivan et al., 1995). The PCS-P encompasses 13 items, rated on a 5-point Likert Scale (0 = not at all, 4 = extremely). Parents were asked to rate how frequently they experience different thoughts and feelings when their child is in pain (e.g., "When my child is in pain, I become afraid that the pain will get worse"). The PCS-P contains three subscales, assessing *rumination* (ruminate about the pain), *magnification* (the thought that something serious will happen due to the pain), and *helplessness* (the feeling that you cannot stand it anymore because of the pain), and yields a total score that ranges between 0 and 52, with higher scores indicating more catastrophic thoughts. The PCS-P demonstrated adequate internal consistency in the current study ($\alpha = 0.90$).

Diary Measures

Below is an overview of the constructs assessed by the diary. All diary items were rated on a 7-point Likert-scale (for example, 0 = not at all, 6 = a lot). For constructs assessed by multiple items, the mean of the respective items was calculated, resulting in a total score ranging from 0 to 6. Level-specific reliabilities were estimated based upon a multilevel confirmatory factor analysis framework. Within-parent, between-parent, and between-couple alphas are reported in **Table 2** (Geldhof et al., 2014).

Perceived daily child pain intensity was reported by the parents and measured by one item: "How much pain did your child experience today, on average, according to you?"

Daily parental distress when confronted with their child's pain was assessed by means of seven items, based upon the theory of Batson et al. (1987). Parents were asked to report to what extent they experienced different feelings that day, related to the pain of their child (i.e., "Indicate to what extent you experienced the emotions listed below today in response to your child's headache or abdominal pain: worried, anxious, upset, sad").

Daily parental catastrophic thoughts about their child's pain was measured by means of the PCS-P daily (Durand et al., 2017), which includes a selection of 3 items from the PCS-P (Goubert et al., 2006) adapted for use in a daily context. For each subscale of the PCS-P, the PCS-P daily contains one item (i.e., Rumination: "Today, to what extent did you kept thinking about how much pain your child experienced?"; Magnification: "Today, to what extent did you think that, because of the pain, something serious might happen to your child?"; Helplessness: "Today, to what extent did you think, because of the pain of your child, you would not be able to stand it anymore?").

Daily parental goals of child pain control and child activity engagement were assessed by asking parents to report on the degree to which they focused on either relieving their child's

TABLE 1 | Demographic characteristics of the final sample of 25 parents, across 20 children.

		N (%)	Mean	Range	*SD*
Child (*N* = 20)					
Age (years)			9.7	8–14	1.49
Pain duration (months)			22.53	2–96	24.36
Type of pain	Chronic headache	5 (25.0%)			
	Chronic functional abdominal pain	14 (70.0%)			
	Unknown	1 (5.0%)			
Medication	Takes medication	11 (55.0%)			
	Pain medication	5 (25.0%)			
	No medication	9 (45.0%)			
Nationality	Belgian	20 (100%)			
Gender	Girls	18 (90.0%)			
	Boys	2 (10.0%)			
Marital status of parents	Married/cohabiting	18 (90.0%)			
	Divorced	1 (5.0%)			
	Blended family	1 (5.0%)			
Parent (*N* = 25)					
Age			41.03	35–48	3.56
Gender	Mother	20 (76.9%)			
	Father	6 (23.1%)			
Education	Highly educated (>18 years)	20 (76.9%)			
	High school	4 (15.4%)			
	Middle school	2 (7.7%)			

TABLE 2 | Level-specific reliabilities of the diary variables.

Construct	Within parent α	Between parent α	Between couple α
Parental distress when confronted with the child's pain	0.77	0.94	0.85
Parental catastrophic thinking about their child's pain	0.82	0.83	0.88

Reliabilities were estimated by a multilevel confirmatory factor analysis framework (Geldhof et al., 2014).

pain "To what extent were you focused on relieving your child from his/her pain today?") or encouraging their child to engage in activities (i.e., "To what extent did you encourage your child to engage in his/her daily activities today, even if he/she could experience pain by doing so?"). The items were formulated based on the items used in the vignette study by Caes et al. (2012), which used adjusted items of the Chronic Pain Acceptance Questionnaire (CPAQ-8; Fish et al., 2010). We know of no other existing questionnaire measuring parental goal engagement in the context of pain.

Parents completed in total 485 end-of-day diary observations. In case of multiple records on the same day, the first completions were deleted ($N = 4$). Diaries completed after 10 AM the next day or before 4 PM the same day were deleted ($N = 16$) (Nezlek, 2012). The records of 7 participants (for a total of 65 records) were excluded from the analyses due to completing less than half of the requested 21 entries (<11 valid records). Finally, 413 records were included in the analyses, representing 85% of the available records and including reports across 25 parents. On average, parents completed 16 diaries (range 12–21).

Analyses

The data of the present study are hierarchically nested within 3 levels of assessment: perceived daily pain intensity, parental daily catastrophic thoughts, daily distress, and daily pain control and activity engagement goal (Level 1) are nested within the participating parents (Level 2), which are nested within a particular family (Level 3). To account for this interdependence and hierarchical nesting of the data, the data were analyzed by means of multilevel modeling (HLM version 6.01, Raudenbush et al., 2004). Multilevel modeling is the preferred method for this data structure as it allows more precise parameter estimates compared with more traditional statistical methods (e.g., repeated-measures analyses of variance; Kenny et al., 2006, Nezlek, 2001), and can deal appropriately with missing data (Hox, 2010). Importantly, multilevel modeling allows for a mother and father of the same child to be identified as different participants while accounting for the dependency in their data due to being parents of the same child (Kenny et al., 2006).

For each of the three dependent variables (i.e., parental distress, pain control goal pursuit and activity engagement goal pursuit) the same set of analyses was performed to test

the hypotheses. In a first step, a baseline model, without any predictors was run to calculate the level of variance in the dependent variables accounted for by the variables between parents (Level 2) and within parents (Level 1). In the second step, we added the Level 3 control variables child age, child sex and pain duration. These demographic variables were included in the models as evidence in the literature highlights that pain experiences, and parental responses vary depending on the child's age, sex and pain duration (Cohen et al., 2010; Palermo et al., 2014; Boerner et al., 2015). The categorical variable child sex was dummy coded (0 = male; 1 = female) and added to the model uncentered. The continuous variables child age and pain duration were standardized before adding to the model. In the third step, the impact of Level 1 variables was assessed by adding the standardized, group-mean centered scores for PCS-P daily and perceived daily child pain intensity. In the fourth step, the influence of parental general catastrophic thinking was evaluated by adding the standardized, grand-mean centered Level 2 variable PCS-P general. In the last and fifth step, the slopes for Level 1 variables were set to random on Level 2 and if the random error term was significant, two interaction terms were added: the cross-level interaction between perceived child pain intensity and PCS-P general and the Level 1 interaction between perceived child pain intensity and PCS-P daily. If the random error term was not significant ($p < 0.05$; Nezlek, 2001), the slopes were fixed. The slopes for all Level 1 and 2 variables were fixed on the third level because dyads do not have enough lower-level units to allow for the slopes to vary (Kenny et al., 2006). Full maximum likelihood estimation was applied for each step in this build-up strategy. The effect size *r* was calculated for all significant effect, with an *r* value of 0.10 reflecting a small effect, an *r* value of 0.30 reflecting a medium effect and an *r* value of 0.50 reflecting a large effect (Kenny et al., 2006).

RESULTS

The Impact on Daily Parental Distress

The intercept model indicated that 11.22% of the variance in parental distress was accounted for by variables on the third level (between families or child characteristics), 62.78% by variables on the second level (parent characteristics), and 38.32% by variables on the first level (within parents or daily characteristics). The model exploring the main effects of the variables across all levels (steps 2–4) revealed a significant positive effect of daily parental catastrophic thinking (PCS-P daily) [$\gamma_{200} = 0.40$; $t_{(352)} = 11.36$; $p < 0.001$, $r = 0.41$] and perceived daily child pain intensity [$\gamma_{100} = 0.22$; $t_{(352)} = 5.76$; $p < 0.001$, $r = 0.23$]. No main effect of general parental catastrophic thinking (PCS-P general) was found. The random error terms for perceived daily child pain intensity and PCS-P daily were significant, so the interaction terms (perceived daily pain intensity*PCS-P daily and perceived daily pain intensity*PCS-P general) were added in step 5. Both interaction terms were significant.

A significant positive interaction was found between daily levels of parental catastrophizing and perceived daily child pain intensity [PCS-P daily; $\gamma_{300} = 0.11$; $t_{(350)} = 6.47$; $p < 0.001$, $r = 0.26$]. This interaction reveals that on days that parents catastrophize a lot about their child's pain, their distress is strongly influenced by the daily level of perceived child pain intensity with higher levels of perceived child pain intensity related to more parental distress. On days that parents experience low levels of catastrophic thoughts; parental distress remains lower compared to days on which parents catastrophize a lot and the perceived level of the child's pain intensity does not add much to explaining this distress level (See **Figure 2**).

In contrast, a significant negative interaction between general levels of parental catastrophic thoughts and perceived daily child pain intensity [PCS-P general; $\gamma_{110} = -0.12$; $t_{(350)} = -4.26$; $p < 0.001$, $r = 0.18$] was found. This finding indicates that for parents with low general levels of catastrophic thoughts about their child's pain, increasing levels of perceived daily child pain intensity were related with increased levels of daily parental distress. However, for parents with high general levels of catastrophic thinking, the perceived level of daily child pain intensity did not add much explanation to the experienced level of daily parental distress (see **Figure 3**). Results for the final hierarchical linear model assessing the impact of parental catastrophic thinking and perceived child pain intensity on parental distress are presented in **Table 3**.

The Impact on Parents' Daily Endorsement of the Pain Control Goal

The intercept model indicated that 25.57% of the variance in the pain control goal was accounted for by variables on the third level (between families or child characteristics), 73.05% by variables on the second level (parent characteristics), and 1.38% by variables on the first level (within parents or daily characteristics). The model exploring the main effects of the variables across all levels (steps 2–4) revealed a significant positive effect of daily parental catastrophic thinking (PCS-P daily) [$\gamma_{200} = 0.43$; $t_{(352)} = 5.87$; $p < 0.001$, $r = 0.30$] and perceived daily child pain intensity [$\gamma_{100} = 0.96$; $t_{(352)} = 12.07$; $p < 0.001$, $r = 0.54$]. No main effect of general parental catastrophic thinking (PCS-P general) was found. The random error terms for perceived daily child pain intensity and PCS-P daily were significant, so the interaction terms (perceived daily pain intensity*PCS-P daily and perceived daily pain intensity*PCS-P general) were added in step 5.

Only the interaction between perceived daily child pain intensity and general levels of parental catastrophic thoughts (PCS-P general) was found to be significant [$\gamma_{110} = 0.15$; $t_{(350)} = 2.39$; $p < 0.05$, $r = 0.18$]. This significant positive interaction reveals no differences between parents with low and high levels of general catastrophizing on days that parents perceive high levels of pain intensity in their child, with all parents showing higher endorsement of the pain control goal on such days. However, on days where perceived child experiences of pain intensity is low, parents with high levels of general catastrophizing show a much more reduced focus on pain control compared to parents with low levels of general catastrophic thinking (see **Figure 4**). Results for the final hierarchical linear model assessing the impact of parental catastrophic thinking and perceived child pain intensity on parental endorsement of the pain control goal are presented in **Table 3**.

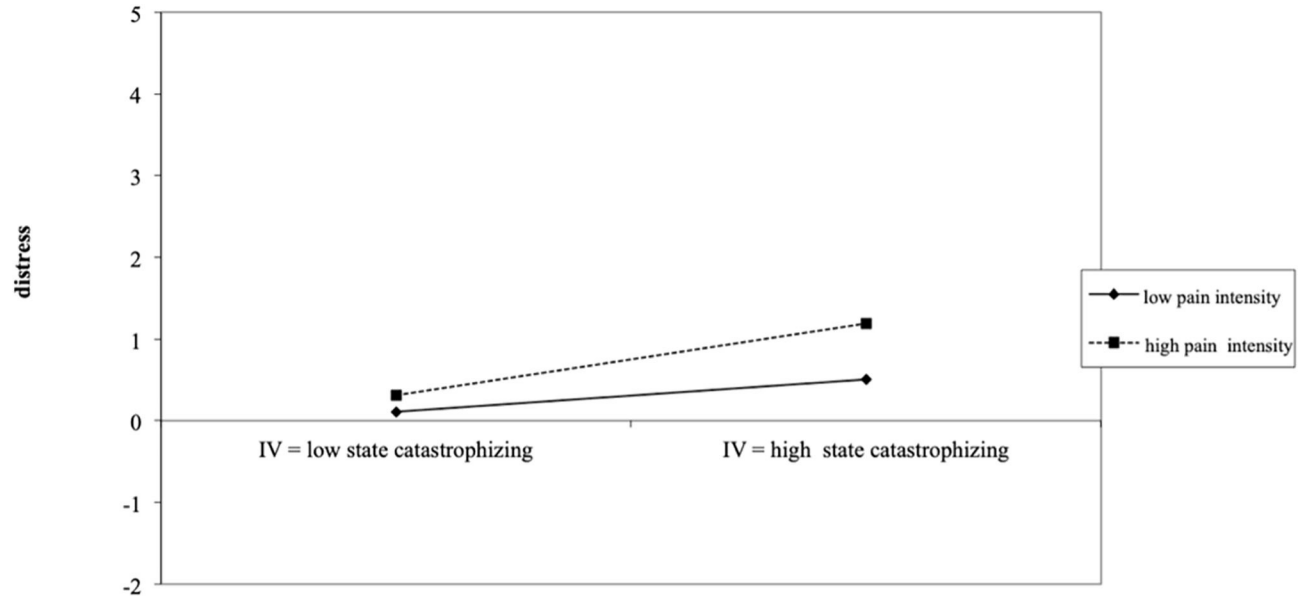

FIGURE 2 | Depiction of how the significant interaction between daily parental catastrophizing and perceived daily child pain intensity influences parental daily distress experiences.

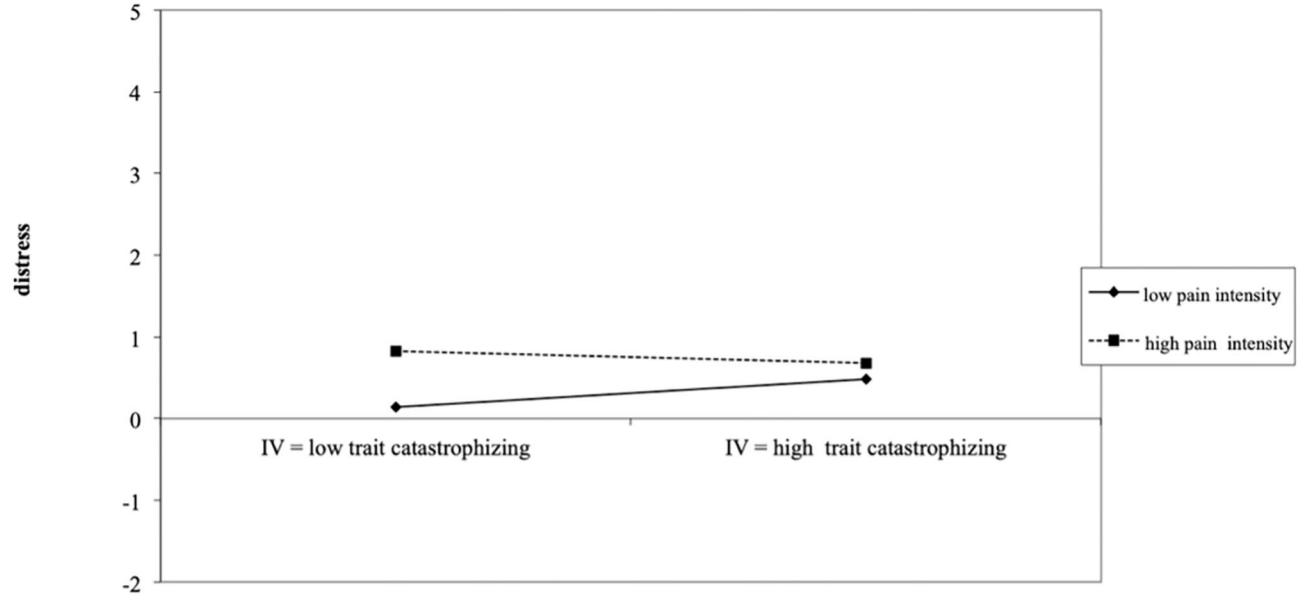

FIGURE 3 | Depiction of how the significant interaction between daily parental catastrophizing and perceived daily child pain intensity influences parental daily distress experiences.

Impact on Parents' Daily Endorsement of the Activity Engagement Goal

The intercept model indicated that 49.52% of the variance in parental priority for the activity engagement goal was accounted for by variables on the third level (between families or child characteristics), 46.44% by variables on the second level (parent characteristics), and 4.03% by variables on the first level (within parents or daily characteristics). The model exploring the main effects of the variables across all levels (steps 2–4) revealed a significant negative effect of child sex [$\gamma_{001} = -2.56$; $t_{(15)} = -2.46$; $p < 0.05$, $r = 0.44$], which indicates that parents endorse the activity engagement goal less for girls compared to boys. Furthermore, significant positive effect of daily parental catastrophic thinking (PCS-P daily) [$\gamma_{200} = 0.29$; $t_{(352)} = 2.89$;

TABLE 3 | Final hierarchical linear models of daily parental distress, endorsement of pain control goal and endorsement of activity engagement goal.

	Parental daily distress			Parental endorsement of pain control goal			Parental endorsement of activity engagement goal		
Variable	**β coeff.**	**SE**	**T**	**β coeff.**	**SE**	**T**	**β coeff.**	**SE**	**T**
Intercept (γ000)	0.16	0.25	0.64	2.46	0.36	6.85***	4.48	0.94	4.75***
Child sex (γ001)	0.40	0.27	1.45	−1.09	0.39	−2.78*	−2.17	1.01	−2.14*
Child age (γ002)	0.21	0.11	1.90#	0.11	0.16	0.66	0.12	0.42	0.30
Pain duration (γ003)	0.02	0.07	0.33	0.11	0.11	1.07	0.11	0.28	0.39
PCS-P general (γ010)	0.05	0.09	0.56	−0.08	0.12	−0.68	0.13	0.30	0.42
PCS-P general* Perceived pain intensity (γ110)	−0.12	0.03	−4.26***	0.15	0.06	2.39*	−0.30	0.08	−3.59***
Perceived pain intensity (γ100)	0.22	0.04	6.08***	0.94	0.08	11.75***	−0.42	0.11	3.87***
PCS-P daily (γ200)	0.32	0.04	8.18***	0.41	0.09	4.72***	0.32	0.12	2.74**
Perceived pain intensity*PCS-P daily (γ300)	0.12	0.02	6.47***	−0.02	0.04	−0.44	0.09	0.05	1.61

*#p = 0.08, *p < 0.05, **p < 0.01, ***p < 0.001.*

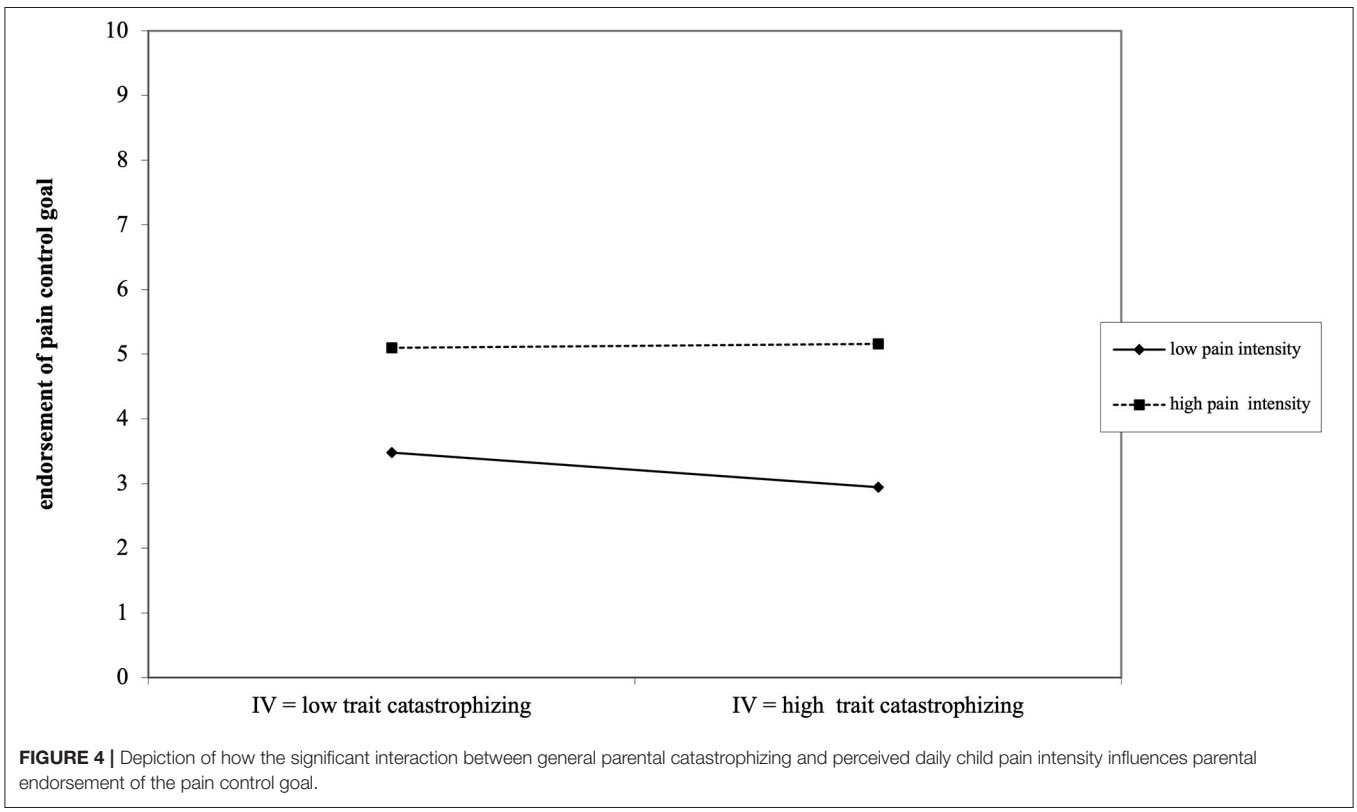

FIGURE 4 | Depiction of how the significant interaction between general parental catastrophizing and perceived daily child pain intensity influences parental endorsement of the pain control goal.

$p < 0.01$, $r = 0.15$] and perceived daily child pain intensity [$\gamma_{100} = 0.41$; $t_{(352)} = 3.70$; $p < 0.001$, $r = 0.19$] was found. No main effect of general parental catastrophic thinking (PCS-P general) was found. The random error terms for perceived daily child pain intensity and PCS-P daily were significant, so the interaction terms (perceived daily pain intensity*PCS-P daily and perceived daily pain intensity*PCS-P general) were added in step 5.

Only the interaction between perceived daily child pain intensity and general levels of parental catastrophic thoughts (PCS-P general) was found to be significant [$\gamma_{110} = -0.30$; $t_{(350)} = -3.59$; $p < 0.001$, $r = 0.18$]. This significant negative interaction reveals that for parents with high general levels of catastrophic thoughts about their child's pain, the pursuit of the activity engagement goal is always moderately high and not influenced by perceived daily levels of child pain intensity. However, for parents with low general levels of catastrophic thinking, the focus on activity engagement shows more flexibility depending on the level of perceived child pain intensity: the lower the perceived child pain intensity, the lower the motivation to encourage activity engagement in their child (see **Figure 5**). Results for the final hierarchical

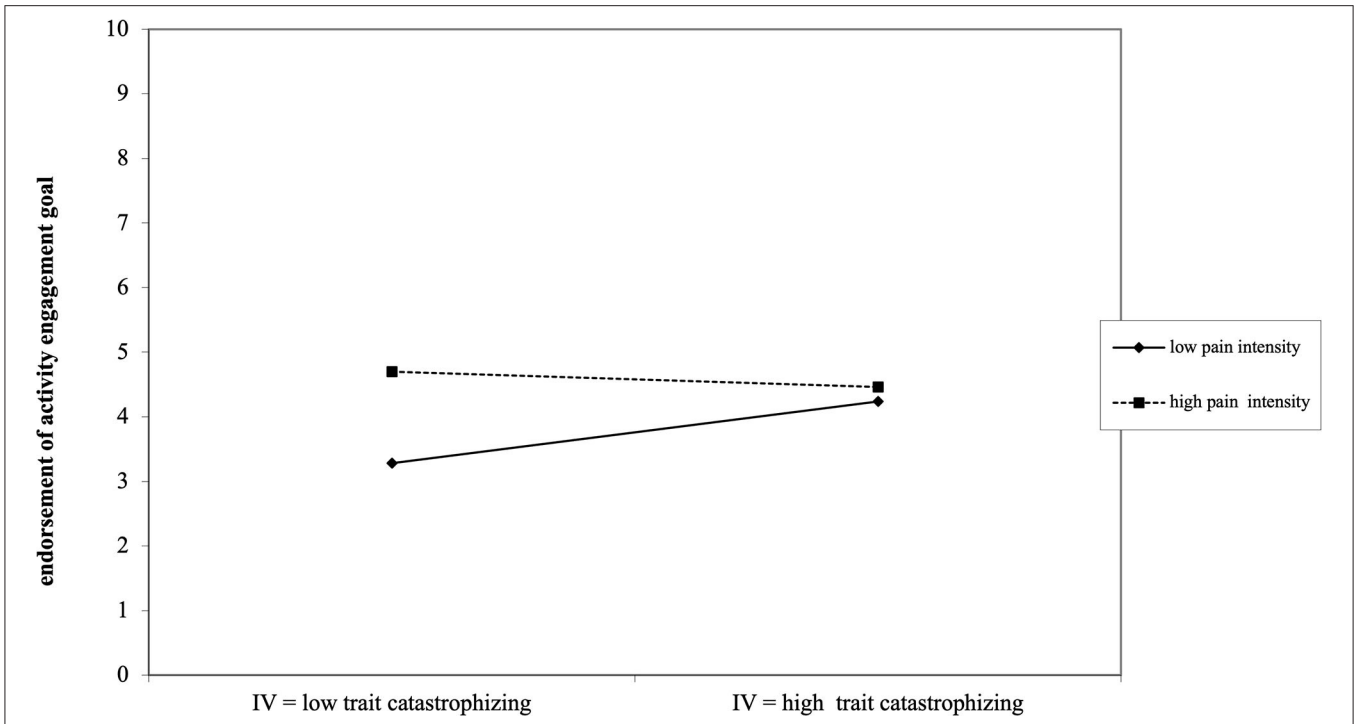

FIGURE 5 | Depiction of how the significant interaction between general parental catastrophizing and perceived daily child pain intensity influences parental endorsement of the activity engagement goal.

linear model assessing the impact of parental catastrophic thinking and perceived child pain intensity on parental endorsement of the activity engagement goal are presented in **Table 3**.

DISCUSSION

Given the critical role parents play in understanding childhood chronic pain experiences, the current diary study explored how parental catastrophic thinking about their child's pain influences parents' daily experiences of distress and endorsement of pain control and/or activity engagement goals for their child, and how these associations were influenced by daily perceptions in fluctuations of child pain intensity. For parental levels of distress, our findings revealed that both the impact of general and daily parental catastrophic thoughts was moderated by perceived child's pain intensity levels. Daily fluctuations in perceived child pain intensity had the strongest impact for parents with low levels of general catastrophic thoughts about their child's pain (as measured by the PCS-P general), as increasing levels of perceived daily child pain intensity were related with increased levels of daily parental distress. However, for daily levels of parental catastrophic thoughts, the level of perceived child pain intensity mostly modulated parental distress on days where parents catastrophized a lot about their child's pain: on days where parents endorsed high levels of catastrophic thoughts, high levels of perceived child pain intensity were associated with higher levels of parental distress.

With respect to parents' daily goal pursuit, the findings illustrate how parents focus on both pain control and activity engagement on days when they have higher levels of catastrophic thoughts about their child pain. This was unrelated to the perceived levels of child pain intensity on that day. On the other hand, the impact of general catastrophic thoughts about their child's pain (i.e., PCS-P general) was influenced by the level of perceived pain intensity: on days where low levels of child pain intensity were perceived, a reduced focus on pain control was reported by parents with high levels of catastrophic thoughts (with no substantial change reported for the focus on activity engagement goals), while parents with low catastrophic thoughts rather reported a reduced focus on child activity engagement (with no substantial change reported for the pain control goal).

In sum, these results highlight how parental coping with their child chronic pain fluctuates considerably from day to day, with the impact of parental general tendencies (e.g., general levels of catastrophic thinking) moderated by daily perceived fluctuations in the child's pain experiences. Such daily differences underscore the need for continuous and situation specific assessment for a comprehensive understanding of how parents cope with their child's chronic pain. It is also interesting to note though that the variability in parents' daily coping responses was situated on different levels depending on the type of coping responses (i.e., emotional distress or goal endorsement). Indeed, for parental distress most variability was observed between days within the same parent, while for the endorsement of the pain control and activity engagement goal the variability was mostly observed between parents/families. This indicates

that parental distress experiences are likely mostly influenced by situational characteristics of a particular day, while goal endorsement is mostly impacted by more stable characteristics of the parent and/or couple. While these preliminary observations need further confirmation, these findings could be of clinical relevance as it allows providing parents an insight on how their emotional responses are variable and changeable. Insight in such variability provides opportunities to teach parents appropriate emotion regulation techniques which would allow them to manage their daily changing levels of distress and engage in more optimal responses.

Parental Daily Distress Experiences

For distress, the results are largely confirming accumulating evidence revealing how parents with high levels of catastrophic thoughts about their child experience heightened feelings of distress (e.g., Caes et al., 2012), as well as how daily levels of catastrophizing contribute stronger to parental emotional distress compared to general levels (Durand et al., 2017). However, our findings further untangle the complexity of these associations by demonstrating the strong moderating impact of daily fluctuations in parental perceptions of their child's pain experience, in particular pain intensity. Contrary to our hypotheses, this moderating impact was different for general vs. daily parental catastrophic thinking. As such our findings call into question the association between parental levels of general and daily catastrophic thinking, and possibly suggest that general and daily levels of catastrophic thinking influence parental responses independently.

The findings for daily levels of parental catastrophic thoughts are in line with our hypotheses, and previous literature, revealing how parental perceptions of high child pain intensity were associated with increased parental distress experiences on days where parents report high levels of catastrophic thinking. This finding further supports the assumptions of the model of empathy in the context of pain (Goubert et al., 2005) as well as the Interpersonal Fear Avoidance Model (Goubert and Simons, 2013) underscoring the interplay of child and parental characteristics in determining how parents emotionally cope with their child's chronic pain experiences. To our knowledge, however, this is the first study exploring these interpersonal interactions on a daily basis, thereby highlighting how the interplay of interpersonal characteristics may not necessarily be stable but fluctuate from day to day. It is likely to assume that these daily fluctuations may present a barrier and explain observed difficulties in skill generalization from the treatment sessions to the home context (Jensen et al., 2003; Guite et al., 2014). Clinically this can have important implications, as it not only emphasizes the need for situation-specific assessment to gather a comprehensive picture of parental coping responses but also the need to ensure any skills parents are taught to reduce their distress and catastrophic thoughts take into account such substantial situational influences in their responses. For instance, our findings highlight the need to encourage parents to practice emotion regulation and cognitive exercises aimed at challenging their catastrophic thoughts in various different situations in their home environment.

However, contrary to our expectations, parental perception of high child pain intensity contributed to more distress only in parents reporting low levels of general catastrophic thinking. While on face value this is unexpected, **Figure 3** demonstrates how parents high on general thinking report high levels of distress independent of their perception of child pain intensity. Consequently, there is more variance and situational differences in the distress experiences of parents low in general catastrophic thinking. These results further corroborate the findings from a vignettes study in parents (Caes et al., 2012), which revealed that parents with high levels of general catastrophic thinking show less flexibility, compared to parents low in catastrophizing, in how they cope with their child's pain experience depending on the particular pain characteristics, e.g., short term vs. long-term pain or mild vs. intense pain. Therefore, it appears that parents with low levels of catastrophic thinking attune their feelings of distress appropriately to the particular pain experience of their child, while parents with high levels of general catastrophic thoughts rather show a sustained mild to high distress response, even in low threatening situations (i.e., low child pain intensity). Such sustained experiences of distress entail the risk of engaging in maladaptive responses toward their child pain (e.g., overprotectiveness, Caes et al., 2012) and developing clinical depression (e.g., decreased response to rewards and anhedonia) through elevated activity of the hypothalamus- pituitary-adrenal (HPA) axis (Willner, 2017). To avoid this chronic experience of distress, it is vital for parents with a high general tendencies to catastrophize about their child's pain to increase their distress tolerance by learning how to appropriately regulate their emotional coping responses in accordance with their child's specific pain characteristics (Guite et al., 2018). For instance, possible adaptive regulation strategy to mitigate the impact of parental distress and catastrophic thinking is mindfulness. Indeed, there is growing evidence on how mindfulness, or being focused on the present moment, in parents of a child with a chronic illness (e.g., diabetes) is an asset which allows parents to adjust their level of distress and protectiveness toward their child to the child's current needs (Van Gampelaere et al., 2019).

Parental Daily Goal Endorsement

The findings with respect to parental goal endorsement are more complex and not completely in line with our hypotheses. For instance, the role of daily parental catastrophic thinking on their goal endorsements was not associated with the perceived level of the child's pain intensity on that particular day and revealed a counterintuitive simultaneous endorsement of both pain control and activity engagement goals on days parents report high levels of catastrophic thinking (as measures by the PCS-P daily). Based on previous evidence (Caes et al., 2012) and motivational theories on goal incompatibility, which highlight the need to prioritize one goal over the other (Riediger and Freund, 2004), it would be expected that the goals of pain control and activity engagement are deemed incompatible by parents and hence decisions on which goal will be prioritized would need to be made. Following such preliminary evidence (Caes et al., 2012) we expected that on days when parents reported high levels of catastrophic thinking

(i.e., high levels of PCS-P daily), the endorsement of pain control goals would be stronger compared to the endorsement of activity engagement goals. It is possible, that this unexpected equal goal endorsement of potentially incompatible goals is related to the formulation of the items assessing parental goal endorsement. In particular, the item assessing activity engagement includes the subphrase *"even if he/she could experience pain by doing so"* which could have influenced parents' interpretation of the item in an unintended way. Indeed, this subphrase could have led parents to interpret this item as follows: "*I had to encourage my child to engage in his/her normal activities because he/she was in pain and hence did not want to engage in those activities by themselves*". Given this potentially unintended interpretation, the positive association found between parents' activity engagement goal endorsement and daily parental catastrophic thinking could be a reflection of how parental catastrophic thoughts (in this case the daily levels) impact how much parents attune their coping responses to the child's pain characteristics. Following this reasoning, it is likely to assume that on days where parents report lower levels of catastrophic thoughts, they feel less threatened by the child's pain, and hence can accordingly attune their responses as reflected in a lesser need to encourage activity engagement. Alternatively, social desirability could also play a role in explaining this unexpected association. Future research is needed to entangle this complexity.

As for the impact of general or general levels of parental catastrophic thinking, a moderation by the fluctuations in perceived child pain intensity levels was observed with parents who report low levels of catastrophic thinking showing adjustments in the endorsement of the activity engagement goal. As explained above, the potentially unintended interpretation of the activity engagement goal endorsement item could explain the lower endorsement of the activity engagement goal on days the child's pain intensity was perceived as low, especially by parents with low general levels of catastrophic thinking. Indeed, the need to stimulate such engagement in daily activities could be reduced when the child is perceived to experience less pain as the child engages in those activities by themselves without the parents needed to encourage them. Particularly for parents with low levels of catastrophic thinking, who might be better at attuning their coping responses more to the child's pain characteristics, could a reduced perceived level of child pain intensity be related to lowering their endorsement of activity engagement that is relative to the child's pain experiences.

Although the above-described findings align with the proposition that parents with high levels of catastrophic thinking are less flexible in adjusting their goals (Caes et al., 2012), the reduced endorsement of the pain control goal by parents with high general of catastrophic thinking—compared to parents with low general levels—on days when the perceived child pain intensity is low challenges this proposition. It is not entirely clear how to interpret this finding and further confirmation of these findings in larger, more heterogenous samples, is needed to gain a comprehensive understanding of the factors that influence parental goal endorsements. The current study is the first to assess these associations on a daily basis in parents caring for a child with chronic pain, compared with the existing evidence stemming from a vignette methodology in parents of pain-free children, which could contribute to the differences. For instance, it is possible that the flexibility of parental coping responses might be dependent on the type of coping responses (i.e., emotional vs. goals and/or behavioral responses) and whether we focus on general vs. situation-specific or daily changing) parental characteristics. While speculative, and requiring testing, it is possible lower endorsement of the pain control goal in parents with high general catastrophic thinking on days with low perceived pain intensity is an attempt to regulate their continued heightened feelings of distress and compensate for their heightened focus on pain control goals on high intensity days. Such explanation would further strengthen the need for providing parents with high levels of catastrophic thinking the tools to appropriately regulate their feelings of distress.

A research avenue that can potentially shed light on the complexity of how perceived child pain intensity and parental catastrophic thinking influence parental goal endorsement, is to explore how these different and potentially incompatible parental goals translate into different parental behaviors. It is reasonable to assume that parental behavior toward their child pain is driven by multiple goals and, vice versa, a particular goal can be achieved through various different behaviors (Rasmussen et al., 2006). For instance, in order to reach their goal of reducing their child's pain parents could comfort their child or stimulate their child to distract themselves by engaging in different activities (e.g., watching a movie). Consequently, depending on the specific behavioral coping strategy the parent engages in, the goal of pain control and activity engagement are not necessarily incompatible on a behavioral level and can be achieved by the same behavioral response. There is growing evidence on how parents who catastrophize about their child's pain tend to engage in protective behavioral responses (e.g., reassuring, comforting and paying attention to their child's pain) at the expense of engaging the child in their daily activities (e.g., attending and engaging with school; Caes et al., 2011; Sieberg et al., 2011). Therefore, the goal of controlling their child's pain and engaging their child in activities might be incompatible for parents experiencing high levels of catastrophic thinking given that they do not naturally perceive coping-promoting responses, such as distraction, as a possible way to reduce their child's pain, leading. On the other hand, for parents low in catastrophic thinking achieving their pain control and activity engagement goal could be more compatible, given their tendency to engage more frequently in, and therefore perceive, coping-promoting behaviors as an approach to reduce their child's pain. However, as highlighted by the current study's findings, this incompatibility might also be influenced by daily fluctuations in parental catastrophic thoughts and perceptions of their child's pain intensity.

Limitations

It is important to consider our findings in light of several limitations. The study was conducted in a relatively small and homogenous sample, limiting the generalizability of the findings in various ways. The relatively small sample prevented us from conducting intricate prospective analyses and rather limited our analytic approach to cross-sectional analyses. Furthermore, only

for a handful of families we were able to collect data from both parents, making comparisons and explorations of potential differences between mothers and fathers impossible. Reflective of the typical population distribution of patients attending a pediatric chronic pain clinic, our sample comprised mostly of girls. Hence, recruiting through pediatric chronic pain clinics gives an over-representation of girls and to fully understand boy's experiences alternative recruitment routes might need to be explored. Additionally, the average age of the children in our sample was on the lower end (i.e., 9.7 years of age) of our target range (i.e., 8–14). This has important implications for our findings as younger children are more dependent on their parents, which could substantially influence parental goals as well as coping responses. Replication within an older sample of adolescents with chronic pain is advised. Lastly, given our focus on children with chronic headache or functional abdominal pain, the results may not be applicable to other pain conditions or different pain locations (e.g., musculoskeletal pain, sickle cell disease).

Beyond the limitations related to generalization of the findings, it is important to recognize that the level of daily child pain intensity was reported by the parent, rather than the child, thereby potentially introducing a reporting bias and preventing us from inferring causal effects. Previous evidence indeed highlights discrepancies between parent and child reports on the child's functioning, particularly when parents endorse high levels of catastrophic thinking (Birnie et al., 2020). Specifically, heightened levels of parental catastrophic thoughts have been found to be related to increased perception of child's pain intensity and disability (Birnie et al., 2016, 2020). While our findings align with a study utilizing child report (e.g., Neville et al., 2020), further research is needed to disentangle this complex interrelation between parental perceived pain intensity and catastrophic thinking.

CONCLUSION

Despite these limitations, the findings underscore the importance of including parents into clinical pain management programs for pediatric chronic pain, with a focus on understanding the underlying mechanisms of and situational influences on parental engagement in maladaptive coping strategies. Accumulating evidence indeed shows promise of actively including parents within interdisciplinary pediatric pain management programs, by for instance addressing parental problem-solving skills (Palermo et al., 2016), parental distress tolerance and resilience (Russell et al., 2020), and parental psychological flexibility (Kemani et al., 2018). However, the observed daily fluctuations in parental emotional response and goal endorsements—and how these are associated with parental general and daily catastrophic thinking as well as child's daily pain intensity—reveals a strong need to ensure the acquisition of pain management skills within these dedicated pain management programs generalizes from the therapy to the home environment.

DATA AVAILABILITY STATEMENT

The datasets for this study are available upon requested by contacting the first and corresponding author (Line Caes).

ETHICS STATEMENT

The study was approved by the Ethics Committees of the Faculty of Psychological and Educational Sciences of Ghent University, University Hospital Ghent, AZ Maria Middelares Ghent, and General Hospital Nikolaas, Belgium. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

LC designed the study procedures, assisted with data collection, took the lead on data analyses, and wrote up the results and discussion section. CG assisted with data collection and contributed substantially to the write up of the manuscript, taking the lead on writing up the methods section, and providing feedback on all other sections. EV, MV, and KK assisted with recruitment of the patients in their respective clinics and provided feedback on all sections of the manuscript. LG assisted with the design of the study procedures and contributed substantially to the write up of the manuscript, taking the lead on writing up the introduction section, and providing feedback on all other sections. All authors contributed to the article and approved the submitted version.

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Stress and Coping in Youth With Spina Bifida: A Brief Longitudinal Study in a Summer Camp Setting

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Introduction: It is well established that youth with chronic conditions experience elevated levels of stress; the manner in which they respond to or cope with this stress is likely to impact both health and psychosocial outcomes. The current study examined stress and coping in youth and young adults with spina bifida (SB) using the response to stress questionnaire-SB version (RSQ-SB; Connor-Smith et al., 2000).

Methods: Data were collected as part of a camp-based psychosocial intervention for children (ages 7–13), adolescents (ages 14–19), and young adults (ages 20–38) with SB. Participants completed the RSQ-SB as well as questionnaires assessing demographics and condition severity. Data were collected prior to camp (T1) and 1 month (T2) after camp ended. Self-report data were collected from adolescents and young adults; parents of children and adolescents reported on their child's stress and coping. Ratios of primary control coping, secondary control coping, disengagement coping, involuntary engagement, and involuntary disengagement coping were calculated. Descriptive statistics and *t*-tests were utilized to describe coping and stress responses and to determine potential change over time. *T*-tests were also used to compare youth and parent reported coping styles with those of youth with type 1 diabetes (T1D) and sickle cell disease (SCD). Associations between demographic/disease factors and coping styles were also examined.

Results: Parent and youth report indicated that youth with SB tend to use primary control coping. Youth with SB use more primary control coping and less disengagement coping compared to youth with SCD and youth with T1D. Few significant changes in coping were found between T1 and T2. IQ and socioeconomic status were significantly associated with coping styles.

Conclusion: Youth with SB use more primary control coping compared to other coping methods and as compared to other pediatric populations. Future studies should examine mechanisms by which primary control coping is advantageous for youth with SB. Future interventions should be more focused on promoting adaptive coping behaviors and be tailored to developmental age and access to resources.

Keywords: coping, stress, spina bifida, chronic condition, camp

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INTRODUCTION

Spina bifida (SB) is a relatively common congenital birth defect, affecting approximately 3 in every 10,000 births in the United States (Centers for Disease Control and Prevention, 2011). Due to the failed closure of the neural tube in early pregnancy, most individuals with SB must contend with multiple medical complications, including neurogenic bowel and bladder, ambulation challenges, and social and cognitive deficits. Medical complications often require daily medical care tasks (e.g., catheterization) in order to avoid secondary conditions as well as ongoing management by a number of medical providers. With regard to psychosocial challenges, youth with SB report reduced quality of life, lower self-image, internalizing symptoms, and reduced social contact (Kazak and Clark, 1986; Holmbeck et al., 2003; Copp et al., 2015). While there is a paucity of literature examining the impact of stress in SB, youth with SB often face many disease-related stressors. Stressors experienced by youth with SB are varied and may encompass medical, psychosocial, and family factors.

More generally, the larger pediatric literature has consistently demonstrated that youth with chronic conditions experience significantly elevated levels of stress (Compas et al., 2012). Chronic conditions can require a demanding medical regimen, have taxing symptomatology, and can be accompanied by painful or time-consuming medical procedures. In addition, chronic illnesses can result in reduced school attendance and difficulties with social functioning (Alderfer and Kazak, 2006; Boles, 2017). Furthermore, each condition is characterized by different condition-related stressors (Compas et al., 2012); for example, a child undergoing cancer treatment may struggle with stress related to how treatment has altered their appearance (Zebrack and Isaacson, 2012), whereas a child with diabetes may struggle with stress related to their daily self-management tasks (Rechenberg et al., 2017). A child with SB, on the other hand, may struggle with stress related to ambulation and shunt malfunction.

In response to stress, one may experience automatic stress responses, such as emotional and physiological arousal and stress-conditioned behaviors, or one may respond with a coping behavior. Research considers coping to be a controlled and volitional response; coping is purposeful and directed toward addressing the stressor (Compas et al., 2012). Adaptive coping has been shown to result in better psychosocial functioning, stronger quality of life, and better health outcomes in youth with chronic conditions (Grey et al., 2000; Szigethy et al., 2007). The current literature has produced multiple coping frameworks, which are characterized by various subtypes of coping (e.g., problem-focused coping vs. emotion-focused coping, approach vs. avoidance, etc.; Compas et al., 2012). Coping frameworks that are commonly used with pediatric populations often include active or primary control coping, accommodative or secondary control coping, and avoidant or disengagement coping (Walker et al., 1997; Connor-Smith et al., 2000; Compas et al., 2012).

Stress and coping in pediatric chronic illness can often be challenging to assess, especially taking into account different types of stressors that vary in frequency across chronic illness populations. The response to stress questionnaire (RSQ) was developed by Connor-Smith et al. (2000), and is one of the most frequently used measures of coping responses in pediatric chronic illness populations (Compas et al., 2012). The RSQ assesses specific sources of stress and the use of coping strategies, as well as involuntary stress responses pertaining to a specific stressor (i.e., pediatric cancer, SB), regardless of the success of those coping strategies. The RSQ is valid and reliable, has been supported by several confirmatory factor analyses (Compas et al., 2012), and can be easily completed by youth and/or adults who are involved in the stressful situation (Connor-Smith et al., 2000). Responses to the RSQ comprise five factors: primary control coping, secondary control coping, disengagement coping, involuntary engagement, and involuntary disengagement. Researchers can then compute proportion scores for individuals, indicating coping responses most frequently utilized in relation to the stressor in question.

The first and primary dimension of this measure delineates between involuntary and voluntary responses, with involuntary stress responses considered to occur outside of conscious awareness and reflect individual temperament or conditioned responses to stressors (Connor-Smith et al., 2000). Involuntary and voluntary behaviors are then further subdivided into engagement and disengagement responses wherein engagement responses approach or address the stressor and disengagement responses avoid the stressor. Within voluntary engagement responses, there are two coping types: primary control coping and secondary control coping. Primary control coping strategies attempt to alter the stressor itself or one's emotional response to that stressor, such as emotional regulation, emotional expression, and problem solving. Secondary control coping strategies are strategies that support adaptation to the stressor, such as acceptance, distraction, cognitive restructuring, and positive thinking (Connor-Smith et al., 2000). Disengagement coping strategies, avoid the stressor, and resultant emotional reactions; examples include, denial, avoidance, and wishful thinking. Finally, there are two involuntary domains: involuntary engagement and involuntary disengagement. An individual experiencing involuntary engagement might have intrusive thoughts or ruminate on the stressor. Further, involuntary disengagement is characterized by emotional numbing, inaction, escape, and cognitive interference (Connor-Smith et al., 2000).

The RSQ coping styles have been associated with multiple outcomes in both typically developing (TD) youth and youth with chronic conditions. Indeed, with regard to psychopathology, a 2017 meta-analysis examining coping in childhood and adolescence in response to various stressors [e.g., interpersonal stress, cancer, type 1 diabetes (T1D), etc.] demonstrated that disengagement coping was associated with increased internalizing and externalizing symptoms, whereas both primary and secondary control coping were associated with fewer internalizing and externalizing symptoms (Compas et al., 2017). These coping styles have also been associated with several physical and psychological outcomes in youth with chronic conditions. Indeed, for youth with T1D, use of primary control coping was positively associated with social competence, quality of life, and low HbA1c. Additionally, this study found that secondary control coping was associated with better social

competence and quality of life. In contrast, disengagement was associated with lower social competence and higher HbA1c (Jaser and White, 2011). For youth with chronic abdominal pain, use of secondary control coping has been associated with lower levels of somatic complaints and symptoms of depression and anxiety; the inverse relationship was found with disengagement coping in this population (i.e., more disengagement coping was related to worse somatic complaints and increased symptoms of depression and anxiety; Compas et al., 2012). The positive associations between both primary and secondary control coping and healthy adjustment in chronic illness populations are likely a reflection of the fact that most chronic illnesses include both controllable and uncontrollable stressors, with some having fewer controllable stressors vs. others (Compas et al., 2012). Controllable stressors may be best addressed with primary control coping, whereas secondary control coping would be the most effective strategy to address uncontrollable stressors. With respect to SB, use of secondary control coping strategies, such as acceptance, might be more beneficial when faced with the stress of "not being able to do what others can do," whereas use of primary control coping, such as problem solving, might be more helpful for "parents bugging me about taking care of myself." On the other hand, disengagement or avoidance of the aforementioned stressors might result in emotional distress, self-isolation, poor peer relations, reduced independence, and improper self-management and medical adherence.

There is currently very limited literature on how youth with SB cope with stress. Of the few existing studies, research has suggested that coping behaviors develop similarly (i.e., coping socialization) in youth with SB and TD youth (McKernon et al., 2001), and that youth with SB and TD youth employ problem-focused coping, an adaptive coping behavior, with similar frequency when asked to think about a stressful peer encounter (Monsen, 1992). Finally, another study found that adults with SB who received an executive functioning (EF) intervention demonstrated significant gains in adaptive coping strategies (decreased avoidant-focused and increased task-focused coping; Stubberud et al., 2014). To our knowledge, no study has examined (1) how youth with SB respond to illness-related stress, (2) if there are differences between how youth with SB and youth with other pediatric conditions respond to illness-related stress, and (3) the effectiveness of a psychosocial intervention that teaches adaptive coping strategies to youth with SB.

Therefore, the first aim of the current study was to examine coping methods and stress responses in youth with SB using the response to stress questionnaire-SB (RSQ-SB), and to compare our study sample's responses to that of other pediatric populations that contend with similar illness-related stressors (e.g., samples with self-management concerns, complex medical regimens, or neurocognitive difficulties). For this study, we chose to examine differences between youth with SB and youth with T1D and sickle cell disease (SCD). Similar to SB, both of these populations contend with complex medical regimens. Specifically, the treatment regimen for T1D typically requires checks for blood glucose levels, dietary restrictions, insulin injections, and other related medical tasks (American Diabetes Association, 2016). Likewise, youth with SCD often manage medication regimens or adhere to complex therapies (e.g., blood transfusions for anemia) as well as engage in preventative behaviors to reduce the likelihood of having a pain crisis (i.e., staying hydrated, avoiding sudden temperature changes; Center for Disease Control and Prevention, 2020). Further, both youth with SB and SCD struggle with neurocognitive deficits. Youth with SCD, regardless of cerebral infarct history, typically have lower IQs compared to their TD peers (Schatz et al., 2002; Prussien et al., 2018). Likewise, while intellectual functioning in youth with SB tends to fall in the low average to average range, there is significant variability within the condition; indeed, 20–25% of individuals with SB-myelomeningocele and hydrocephalus are estimated to have an intellectual disability (Copp et al., 2015). Regarding cognitive functioning in T1D, although there is some evidence for cognitive differences between youth with T1D and TD youth (Kirchhoff et al., 2017), particularly in cases of poorly controlled insulin, these are not often considered to be clinically significant. Further, cognitive differences may not be as prominent in youth with SCD vs. SB. Therefore, the unique cognitive profile in SB should be considered when examining comparisons with T1D and SCD youth. Nevertheless, given similarities in the condition-related stressors amongst these populations, comparisons between SB, and T1D and SCD will provide valuable insights into the nature of coping in SB.

The second aim of this study was to examine relations between stress and coping responses and condition and demographic factors in this population. While there are mixed findings with regard to demographic factors and coping, some studies have found that younger children employ more primary control vs. secondary control coping strategies, a difference which may be related to certain cognitive skills needed to engage in secondary control coping (Weisz et al., 1994; Thomsen et al., 2002). Interestingly, other studies have not found any age-related differences (Compas et al., 2012). As previously noted, intellectual functioning is highly variable among youth with SB. Thus, many children with SB have different developmental ages vs. their chronological age as a result of this condition (Copp et al., 2015). Furthermore, it is also important to consider the role of EF in relation to coping, as EF is challenging for youth with SB (Brown et al., 2008) and some studies have found associations between EF skills and greater use of both primary and secondary control coping (Compas et al., 2012). Therefore, it was hypothesized that younger participants will use more primary control strategies compared to older participants. Further, youth with lower levels of intellectual functioning were expected to use both primary and secondary control coping less compared to youth with higher intellectual abilities.

It is also important to examine youth coping responses longitudinally and determine what types of interventions promote positive coping in this population. Therefore, the third aim of the current study was to examine potential differences in coping behaviors after attending a camp-based psychosocial intervention. Camp Independence is a summer camp for children, adolescents, and young adults with SB. It is an accessible camp that promotes recreation as well as social and independence skill building. At this camp, youth enjoy typical summer camp

experiences (e.g., swimming, archery) in a safe environment with same age peers with SB. In addition to engaging in recreation activities, youth also participate in a psychosocial intervention for 1 h per day, which addresses independence, social skills, emotional wellness, and self-care. This intervention is tailored to three different age groups (i.e., 7–13; 14–19; and 20+). The main themes of these 4 days include (1) taking care of your relationships, (2) self-care, (3) living with SB, and (4) taking care of SB. Previous studies have found Camp Independence to result in improved management of SB responsibilities and increased independence in SB task management 1 month post-intervention (O'Mahar et al., 2010; Holbein et al., 2013). More generally, medical camps have been found to lead to improvements in psychosocial functioning, including improvements in self-esteem, emotional functioning, and coping (Hunter et al., 2006; White et al., 2016). Given the aforementioned findings regarding the success of Camp Independence and other medical camp interventions in improving youth psychosocial functioning, as well as Camp Independence's focus on increasing multiple forms of adaptive coping (i.e., both primary and secondary), it was hypothesized that youth would report an increase in primary and secondary control coping strategies post-intervention.

MATERIALS AND METHODS

Participants

Participants consisted of campers aged 7–38 years, who attended a weeklong overnight summer camp in 2019. Camp Independence exclusively serves individuals with SB, is located in northern Illinois, and is funded in part by the Illinois Spina Bifida Association (ISBA). Camp Independence is also associated with the YMCA. Camp/study inclusion and exclusion criteria were as follows: individuals needed to be at least 7 years old to apply and those with severe allergies or unpredictable health conditions (e.g., uncontrollable seizures) were ineligible. Camp sessions were separated into three age groups: Group A (children, age 7–13; M = 10.94, SD = 1.73), Group B (adolescents, ages 14–17; M = 15.35, SD = 1.35), and Group C (young adults, ages 18–38; M = 26.96, SD = 5.60). The current study used data from two time points: Time 1 (T1; pre-intervention) and Time 2 (T2; 1-month follow up post-intervention). For this study, data were collected from parents of campers in groups A and B and from campers in groups B and C at both time points. All campers were approached to participate in the study; however, it was not required that everyone at camp participate in this study. Further, regardless of whether or not the camper decided to participate in the study and fill out questionnaires, all campers attended the daily-intervention workshop, as it was embedded into the camp programming.

Procedure

This study was approved by the Institutional Review Board at Loyola University Chicago. Participants were recruited *via* flyers given at regularly scheduled doctor's visits as well as print and online information disseminated by the ISBA. Questionnaires, and assent and consent forms were mailed to enrolled campers' homes prior to the start of camp. Parent consent and camper consent/assent were completed either before the beginning of camp *via* mailed forms or were completed on the first day of camp. With regard to the timing of the questionnaires, questionnaires at T1 were completed prior or on the first day of camp, and T2 questionnaires were completed 1 month after the end of camp.

At T1, parents and campers completed questionnaires assessing demographic and medical information as well as various aspects of camper functioning. Throughout camp, trained research assistants administered brief cognitive assessments to obtain an estimate of intellectual functioning. At T2, parents and campers again completed questionnaires that examined the same domains of functioning as were assessed at T1 and requested feedback regarding the intervention and camp activities in general.

Intervention

A description of the 2010 version of the intervention can be found in Holbein et al. (2013). Since that time, the programming has undergone minor changes and improvements. The intervention includes 1-h daily workshops, which occurred during 4 days of the weeklong camp program. Each day of the intervention had its own theme related to promoting better overall psychosocial functioning. The intervention was composed of the following themes (1) taking care of your relationships, (2) taking care of yourself, (3) living with SB, and (4) taking care of SB. Further, this workshop was tailored to be developmentally appropriate with regard to the three different age groups. For example, the intervention involved more games and examples related to siblings for the younger age groups, whereas, with the older group, the intervention was more didactic in nature and included programming related to, for example, romantic relationships. Day 1 focused on social skill building, Day 2 focused on self-care, relaxation, and coping, Day 3 focused on discussing SB with others, and Day 4 focused on developing SB knowledge, and taking responsibility for SB tasks. Day 2, the focus of this study, involved affect recognition, developing positive coping skills and avoiding negative stress responses, as well as psychoeducation regarding recognizing depression and anxiety and how to ask for help.

Although this invention was not created specifically to promote coping skills, it taught both primary control coping and secondary control coping. Such coping skills were predominately derived from cognitive behavioral, dialectical behavioral, and mindfulness related orientations. Primary control coping skills included problem solving and emotion regulation. Problem solving was typically oriented around self-management (e.g., catheterization) and communication skills (talking with friends). Emotion regulation skills included teaching distress tolerance skills (Dialectical behavioral therapy), specifically the Soothe with Six Senses lesson. Secondary Control coping skills included distraction, cognitive restructuring, and positive thinking. Youth were taught to identify pleasurable activities (e.g., spending time with friends, exercising, relaxation) and engage in those activities when feeling sad or anxious (i.e., distraction). Relaxation strategies included deep breathing,

guided visualization, and progressive muscle relaxation. With regard to cognitive restructuring, youth were taught how to make coping statements/cards (i.e., "This is hard but I have gotten through it before!") With regard to positive thinking, youth were also taught that when they felt down to "look for the positives," such as remembering three positive things that happened that day prior to experiencing the stressor. Finally, we also provided psychoeducation on the negative impact of disengagement coping and provided the aforementioned alternatives to encourage the use of adaptive coping strategies.

Measures

Medical and Demographic Variables

Parents and young adults completed questionnaires assessing demographic information and medical history. Demographic information included child/adolescent/young adult age, gender, race, ethnicity, and family household income. Medical information included lesion level, type of SB, shunt status, and ambulation method. Participants indicated their family household income using a 21-point scale; the scale ranged from under $10,000 per year to over $200,000 per year, using increments of $10,000.

Response to Stress Questionnaire

Parents, adolescents, and young adults completed the RSQ-SB version at T1 and T2 (Connor-Smith et al., 2000). This questionnaire measures coping and involuntary stress responses. It has been adapted to capture the experience of coping and stress for a variety of populations (e.g., pediatric cancer, autism, school stress). It has been translated into four languages, including Spanish; the Spanish and English versions of this questionnaire were used for this study. Similar to other forms of the RSQ, the RSQ-SB starts with a checklist of stressors related to a specific domain of stress which was, in this case, SB. With those stressors in mind, the participant is then asked to indicate how often they use certain coping methods or how often they experienced an involuntary stress response, using a Likert scale (range 1–4). Parents reported on their *child's* use of coping methods or how often their *child* experienced an involuntary stress response. In addition to the Likert ratings, participants were also often asked how they used a given strategy (e.g., after rating how often they "let someone or something know how I feel" they are asked to "check all that you talk to," which includes parent, teacher, friend, God, pet, etc.). The coping and stress responses fall into five factors: primary control coping, secondary control coping, disengagement, involuntary engagement, and involuntary disengagement. Factor ratios, or how much a person engages in behaviors that fall into a specific factor, were calculated by dividing the score of each factor by the total score for the RSQ. This methodology is intended to control for response bias and individual differences in the rate of item endorsement (Vitaliano et al., 1987).

Cognitive Ability

To estimate cognitive ability, the Vocabulary and Matrix Reasoning subtests from the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999) were administered to campers. These two subtests yield an estimated Full-Scale IQ (FSIQ). For campers who had completed this testing within the past 2 years while participating in the camp intervention, their prior WASI score was extracted for analyses.

Data Analyses

Descriptive statistics, including mean and standard deviation (SD), were used to address Aim 1 of this study. These analyses included the means and SDs of parent and youth report of the ratios of coping styles. Further, to address Aim 1 we also chose two study samples (T1D and SCD) against which to compare use of primary control coping, secondary control coping, and disengagement coping. Means, SDs and sample size were extracted from Jaser et al. (2017) and Prussien et al. (2018) to conduct unpaired samples *t*-tests. For comparisons, we used the current study's T1 data. In Jaser et al. (2017), 117 youth (ages 10–16) with T1D completed the self-report RSQ, and in Prussien et al. (2018), 44 parents of youth with SCD (ages 6–16) completed the RSQ-parent report about their child. As previously noted, this choice was guided by an examination of current literature on coping in pediatric populations to assess which populations struggle with similar concerns to that of youth with SB. We also examined the stressors listed in the RSQ for these populations and determined that there were many similarities in item content [e.g., "dealing with diabetes care," "having to go to the hospital or clinic so often (for Sickle Cell Disease care)"].

However, it is also important to note several differences in the demographics across these samples. Specifically, there is a larger age range in the current study's sample vs. both the T1D sample and the SCD sample. Sample race and ethnicity could not be directly compared given limited information in both of the comparison samples. However, race was predominately White, non-Hispanic, in Jaser et al. (2017; T1D) and Black in Prussien et al. (2018; SCD; participants in the study identified as African American); race was predominately White, non-Hispanic, in the current study sample. With regard to family income, in Jaser et al. (2017; T1D) family income was controlled for in analyses but was not explicitly reported. Instead they reported that their study sample had a "fairly high socioeconomic status and income." On the other hand, the majority of the SCD sample (67.4%) in Prussien et al. (2018) reported family income to be less than $50,000 per year. Therefore, as our sample's mean income was around $95,100 per year (SD = $49,100), comparisons should take into account differences in socioeconomic status. Further, regarding intellectual functioning, there were no relevant data on the T1D sample. However, as previously noted, cognitive functioning is not typically reduced in T1D; therefore, we would expect intellectual functioning in that sample to be higher. Further, while it was not possible to directly compare intellectual functioning between the SB sample and the SCD sample, no statistical differences were detected [$t(114) = 1.96$, $p > 0.05$] between our sample's FSIQ score ($M = 87.26$, SD = 18.42, range: 55–136) and the verbal comprehension standard score ($M = 93.39$, SD = 12.08, range = 62–123) reported in Prussien et al. (2018). Finally, it should also be noted that youth with SB may struggle with difficulties seen in those with intellectual and physical disabilities, as well as chronic

health conditions. However, SB is often considered a "snowflake" condition (i.e., symptomatology in SB is quite variable; Stiles-Shields et al., 2019), therefore, these comparison samples were determined to be *appropriate* comparison groups rather than perfect comparison groups.

To address Aim 2, bivariate Pearson correlation coefficients were computed to examine associations between the demographic/disease factors and coping. Due to differences in sample size, independent-samples Kruskal–Wallis tests were also conducted to examine associations between coping, and SB type, shunt status, lesion level, and, race and ethnicity. To address Aim 3 paired sample *t*-tests were performed to compare parent report of coping and stress response style at T1 vs. T2 and youth report of coping and stress response style at T1 vs. T2 (T2 = post-camp intervention).

RESULTS

Preliminary Analyses

A total of 77 families provided RSQ data at T1, including 46 parents and 45 youth. There were 15 families for whom we had both parent and youth RSQ data, and therefore we used a composite score based on both parent and youth report. At T2, a total of 48 families provided RSQ data, including 29 parents and 25 youth. There were six families for whom we had both parent and youth RSQ data; therefore, we again used a composite score based on both parent and youth report. Other studies using the RSQ have similarly adopted this technique of collapsing across reporters to reduce the number of analyses (see Vreeland et al., 2019). With regard to attrition, there were no statistically significant differences between those families who participated at T2 ($n = 48$) and those who did not participate at T2 ($n = 29$) with regard to child age, child sex, child IQ, child lesion level, family income, or primary or secondary control coping ratios.

Among participating families at T1, most youth were female ($n = 44$, 57.9%) and White ($n = 57$, 75.0%). The majority of parents were female ($n = 38$, 82.6%). Youth ranged in age from 7 to 38 years old, with a mean child age of 18.43 (SD = 8.6) and had an average IQ of 87.26 (SD = 18.42, range: 55–136). Yearly family income averaged approximately $95,100 per year (SD = $49,100). In terms of medical characteristics, youth most often had myelomeningocele SB ($n = 63$, 82.9%), lumbar lesion levels ($n = 41$, 53.9%), and a shunt present ($n = 58$, 76.3%). See **Table 1** for additional descriptive information regarding demographic and medical characteristics.

TABLE 1 | Descriptive demographic and medical information for sample, $n = 76$.

Variable	*n*	%
Child sex		
Female	44	57.9
Male	30	39.5
Missing	2	2.6
Parent sex[a]		
Female	38	82.6
Male	4	8.7
Missing	4	8.7
Child race/ethnicity		
White	57	75.0
African American	5	6.6
Hispanic/Latino	9	11.8
Asian American	1	1.3
Biracial (African American and White)	1	1.3
Missing	3	3.9
Child type of SB		
Myelomeningocele	63	82.9
Occulta	2	2.6
Lypomeningocele	1	1.3
Meningocele	2	2.6
Not sure	8	10.6
Child lesion level		
Sacral	13	17.1
Lumbar	41	53.9
Thoracic	3	3.9
Not sure	18	23.7
Missing	1	1.3
Shunt status		
Present	58	76.3
Not present	16	21.1
Missing	2	2.6
	***M* (SD)**	***n***
Child age (range: 7–38 years)	18.43 (8.6)	75[b]
Child IQ (range: 55–136)	87.26 (18.42)	72[b]
Yearly family income (range: 2–21)	10.51 (5.91)	41[a]

Yearly family income was reported on a 21-point scale, from <$10,000 per year to >$200,000 per year, with each point on the scale representing increments of $10,000. For this sample, family income ranged from $10,000 to >$200,000+ per year with a mean of ∼$95,100 and a standard deviation of ∼$49,100.

[a]*Sample size is reduced for these characteristics because we only obtained parent report for these characteristics, and approximately half of the sample was over the <18 without a parent-report version.*

[b]*Sample size is reduced for these characteristics because of missing data/assessments.*

Aim 1: Descriptive Information Regarding Coping Measure and Comparison to Other Pediatric Samples

Descriptive information (including means and SDs) for each subscale of the RSQ at T1 and T2 by reporter (i.e., parent and youth) are provided in **Table 2**. At T1, the ratio of primary control coping was greatest and significantly higher than the ratio of secondary control coping for both parents [$t(45) = 2.68$, $p = 0.010$] and youth [$t(44) = 2.05$, $p = 0.047$]. While the tendency to engage in primary control coping vs. secondary control coping was maintained for youth at T2 [$t(24) = 2.19$, $p = 0.04$], there was no statistically significant difference in parent endorsement of primary vs. secondary control coping at T2 [$t(28) = 1.07$, $p = 0.29$]. The ratio of primary control coping was also higher than the ratio of disengagement coping for both parents [$t(45) = 5.975$, $p < 0.001$], and youth [$t(44) = 5.262$, $p < 0.001$]. This tendency to engage in primary control coping vs. disengagement coping was maintained at T2, as reported by

both youth [$t(24) = 4.058$, $p < 0.001$] and parents [$t(28) = 3.065$, $p = 0.005$].

Descriptive information and results from unpaired samples *t*-tests comparing youth with SB to youth with SCD and T1D can be found in **Table 3** (Note: all SB data were from T1). With regard to comparisons with SCD, parents of youth with SB reported a significantly higher ratio of primary control coping [$t(88) = 4.15$, $p = 0.00$] and significantly lower ratio disengagement coping than parents of youth with SCD [$t(88) = 6.39$, $p = 0.00$; Prussien et al., 2018]. There was no statistically significant difference when comparing the ratio of secondary control coping in parent report of youth with SB vs. parent report of youth with SCD [$t(88) = 1.67$, $p = 0.10$]. With regard to comparisons with T1D, youth with SB reported a significantly higher ratio of primary control coping vs. youth with T1D [$t(160) = 6.49$, $p = 0.00$], and significantly lower ratios of secondary control coping [$t(160) = 2.63$, $p = 0.01$] and disengagement coping [$t(160) = 3.91$, $p = 0.00$] vs. youth with T1D (Jaser et al., 2017).

TABLE 2 | Parent- and youth-reported RSQ scores at T1 and T2.

Variable	Time 1		Time 2			
	M	SD	*M*	SD	*t* (df)	*p*-Value
Parent report						
Ratio primary control coping	0.41	0.35	0.38	0.31	0.62 (28)	0.54
Ratio secondary control coping	0.21	0.23	0.26	0.20	−1.65 (28)	0.11
Ratio disengagement coping	0.07	0.07	0.13	0.17	−2.14 (28)	0.04
Ratio involuntary engagement coping	0.23	0.24	0.14	0.10	1.63 (28)	0.11
Ratio involuntary disengagement coping	0.09	0.09	0.09	0.07	−0.09 (28)	0.93
Youth report						
Ratio primary control coping	0.35	0.26	0.41	0.34	−1.21 (24)	0.24
Ratio secondary control coping	0.22	0.19	0.20	0.14	1.44 (24)	0.16
Ratio disengagement coping	0.10	0.10	0.09	0.07	−0.37 (24)	0.71
Ratio involuntary engagement coping	0.20	0.17	0.16	0.12	1.17 (24)	0.26
Ratio involuntary disengagement coping	0.14	0.14	0.14	0.12	−0.24 (24)	0.82

M, mean; SD, standard deviation.

TABLE 3 | Parent- and youth-reported RSQ scores for youth with SB, SCD, and TD1.

Variable	SB		SCD			
	M	SD	*M*	SD	*t* (df)	*p*-Value
Parent report						
Ratio primary control coping	0.41	0.35	0.19	0.03	4.15 (88)	0.00
Ratio secondary control coping	0.21	0.23	0.27	0.06	1.67 (88)	0.10
Ratio disengagement coping	0.07	0.07	0.14	0.02	6.39 (88)	0.00
	SB		**TD1**			
	M	SD	*M*	SD	*t* (df)	*p*-Value
Youth report						
Ratio primary control coping	0.35	0.26	0.19	0.04	6.49 (160)	0.00
Ratio secondary control coping	0.22	0.19	0.27	0.05	2.63 (160)	0.01
Ratio disengagement coping	0.10	0.10	0.14	0.03	3.91 (160)	0.00

SCD, sickle cell disease; TD1, type 1 diabetes; M, mean; SD, standard deviation.

Aim 2: Associations Between Demographic and Medical Characteristics and Coping

Table 4 displays correlation results between child demographic and medical characteristics and composite coping scores at T1 and T2. Child age was significantly correlated with involuntary disengagement in T2, such that older children were more likely to engage in involuntary disengagement than younger children ($r = 0.32$, $p = 0.02$). Child IQ was significantly associated with the ratio of primary control coping at T2, such that lower IQ was associated with more primary control coping ($r = -0.31$, $p = 0.03$). IQ was also associated with the ratio of secondary control coping at T2, such that higher IQ was associated with more secondary control coping ($r = 0.40$, $p = 0.005$). Finally, family income was significantly associated with the ratio of secondary control coping at T1, such that higher family income correlated with more secondary control coping ($r = 0.48$, $p = 0.001$). There were no significant associations between child gender, SB type, shunt status, or lesion level and any of the coping subscales.

Aim 3: Examine the Utility of a Camp Based Psychosocial Intervention With Regard to the Promotion of Healthy Coping Behaviors

According to parent-report, results showed no significant differences in the ratio of primary control coping, secondary control coping, involuntary engagement coping, and involuntary disengagement coping from T1 to T2. However, there was a significant increase in the ratio of disengagement coping from T1 to T2 [$t(28) = -2.14$, $p = 0.04$]. According to youth report, results showed no significant differences across all five coping and stress response styles between T1 and T2 (**Table 2**).

DISCUSSION

Research has repeatedly documented the advantages of adaptive coping in pediatric populations. To our knowledge, no study to date has examined how youth with SB cope with the stressors that accompany this condition. Therefore, the purpose of this study was to gain foundational knowledge regarding how youth with SB cope with disease-related stressors. We also aimed to determine whether youth with SB cope in a manner similar to youth with other chronic conditions (i.e., T1D and SCD). Further, given the complexity of this condition we sought to understand relations between coping behaviors and condition-related factors (e.g., lesion level, shunt status, etc.). We also

TABLE 4 | Correlations between composite RSQ scores and demographic and medical variables.

Variables	1	2	3	4	5	6	7	8	9	10	11	12	13	14
1. Child age	1													
2. Child gender	−0.09	1												
3. Child IQ	−0.25*	−0.06	1											
4. Family income	0.22	0.14	0.04	1										
5. T1 primary control	−0.08	0.21	0.03	−0.26	1									
6. T1 secondary control	0.15	−0.11	−0.09	0.48**	−0.57**	1								
7. T1 disengagement	0.22	−0.09	−0.06	0.12	−0.43**	0.15	1							
8. T1 involuntary engagement	−0.21	−0.11	0.11	−0.13	−0.51**	−0.17	−0.08	1						
9. T1 involuntary disengagement	0.16	−0.10	−0.06	0.13	−0.45**	0.00	0.27*	−0.03	1					
10. T2 primary control	−0.04	−0.09	−0.31*	−0.01	−0.11	−0.16	−0.16	0.31*	0.05	1				
11. T2 secondary control	0.02	0.04	0.40**	−0.05	0.03	0.23	0.09	−0.24	−0.04	−0.72**	1			
12. T2 disengagement	−0.15	0.07	0.08	−0.22	0.16	−0.05	0.12	−0.19	−0.09	−0.49**	−0.00	1		
13. T2 involuntary engagement	0.01	0.05	0.26	0.30	0.08	0.12	−0.01	−0.23	0.01	−0.79**	0.56**	0.08	1	
14. T2 involuntary disengagement	0.32*	0.09	0.00	0.21	−0.03	0.09	0.24	−0.13	0.03	−0.61**	0.23	0.06	0.55**	1

Correlation is significant at the 0.05 level.
***Correlation is significant at the 0.01 level.*

examined relations between demographic factors (e.g., age, gender, SES, etc.) and coping in this population. We hypothesized that younger children would engage in more primary control coping compared to older children. Further, we hypothesized that those with lower intellectual functioning would report using less primary and secondary control coping. Finally, we examined the utility of a brief psychosocial intervention within a camp environment for promoting positive coping behaviors 1-month post-intervention. It was hypothesized that use of both primary and secondary control coping would increase post-intervention. We addressed these aims using the recently created SB version of the RSQ, a well-validated and commonly used measure of coping, particularly for pediatric populations (Compas et al., 2012).

Regarding our first objective, examining how youth with SB respond to illness-related stress, results revealed that youth with SB tend to utilize primary control coping strategies when addressing SB-related stress. Indeed, both parent and youth report indicated that youth with SB used primary control coping strategies at T1 significantly more than secondary control coping and disengagement coping. These results suggest that when confronted with a stressor youth with SB often try to alter the stressor itself or their emotional response to that stressor (i.e., emotional regulation, emotion expression, problem solving). This is an interesting finding given the intractable nature of many stressful aspects of SB (e.g., self-management, having to go to clinic often). However, while problem solving may not be always *as* useful for youth with chronic conditions, emotion regulation or expression might be a useful strategy for youth with SB. Further, these findings overall demonstrate that youth with SB tend to use an *adaptive* coping strategy when faced with illness-related stress, which represents an area of strength. Nevertheless, we must also consider the fact that, with regard to parent report, primary control coping may be more easily recognized in their children vs. secondary control coping. Indeed, secondary control coping involves more internal processes, which may not be readily seen by parents observing their child's response to stress.

Results also suggested that youth with SB may cope differently with illness-related stressors compared to youth with other chronic conditions. Indeed, according to parent report, youth with SB utilize more primary control coping and less disengagement coping than youth with SCD (Prussien et al., 2018). Further youth self-report indicated that youth with SB may utilize more primary control coping, and less secondary control coping and disengagement coping than youth with T1D (Jaser et al., 2017). Overall, these results further demonstrate that youth with SB have a strong tendency to use primary control coping more than other methods, even in comparison to other pediatric populations that contend with similar illness-related stressors.

Our second objective was to examine associations between coping and a variety of medical and demographic characteristics. Interestingly, this study did not find any significant associations between coping and disease factors. This finding may be due, in part, to the homogeneity of our sample with regard to certain disease characteristics (e.g., most of the sample had myelomeningocele SB and a shunt). With regard to demographic factors, contrary to hypotheses, we found no differences between younger and older youth with regard to the use of primary and secondary control coping. However, results did indicate that older participants were more likely to engage in involuntary disengagement (e.g., emotional numbing, escape) than younger participants. This may suggest that as youth become older, become more aware of disease stressors, and become more responsible for their care, they become emotionally overwhelmed, and unintentionally engage in maladaptive stress responses. It will be important for future studies to explore this finding further, as disengagement coping has been found to be associated with increased internalizing symptoms (Compas et al., 2017). Therefore, this finding may highlight an important area for intervention, wherein the teaching of positive coping skills could mitigate risks for developing depressive and anxious symptomatology, which are associated both with this developmental period (Hyde et al., 2008) and having SB (Holmbeck and Devine, 2010).

Our hypothesis, that those with lower intellectual functioning would use less primary and secondary control coping, was partially supported. Specifically, lower IQ was significantly associated with more use of primary control coping strategies (in contrast to our hypothesis), whereas higher IQ was associated with using more secondary control coping strategies (in line with our hypothesis). Compas et al. (2012) suggested that older age may be related to more use of secondary control coping due to the cognitive demands of reappraisal, acceptance, etc. Therefore, this finding may reflect coping differences in relation to developmental age. It may also be related to the EF deficits commonly found in SB (Brown et al., 2008). Strong EF has been found to be associated with more use of secondary control coping in multiple pediatric populations (Campbell et al., 2009; Desjardins et al., 2018). Further, another study found that stronger verbal comprehension was related to increased use of secondary control coping in youth with SCD (Prussien et al., 2018). They hypothesized that cognitive reappraisal relies on internal self-speech, which is more easily accessible to those with stronger verbal abilities. While youth with SB have relative strengths in verbal abilities compared to non-verbal abilities (Dennis and Barnes, 2010), they tend to struggle with more complex verbal skills that involve integrating information and applying previously acquired knowledge (Dennis et al., 2006), skills that likely underlie the ability to engage in secondary control coping.

Moreover, higher family income was positively associated with secondary control coping. Research on associations between SES and different coping styles have produced mixed results. One study found that cognitive coping strategies, a core component of secondary control coping, were associated with more symptoms of anxiety and depression in a lower SES group, potentially indicating that these strategies may not be as effective in high stress environments (Perzow et al., 2021). However, other studies have found no differences in coping strategies in relation to SES (Gage-Bouchard et al., 2013). Still, others have argued that secondary control coping strategies may be particularly helpful for low SES adolescents, due to the unchangeable nature of the stressors they face (DeCarlo Santiago and Wadsworth, 2008). Overall, future research is necessary to understand the relationship between SES and coping in this population in order to inform the development of effective interventions.

The final aim of the study was to determine the utility of a weeklong camp-based intervention in promoting adaptive coping. In contrast to our hypothesis, no differences were found in terms of primary or secondary control coping for both parent and youth report from T1 to T2. According to parent report, the ratio of disengagement coping appeared to increase significantly from T1 to T2. Still, this was a relatively small change and indicates that coping was generally stable over time. Therefore, this intervention, which has demonstrated positive effects on independence skills, and individual self-care and social goals (Holbein et al., 2013), did not significantly improve coping style. This finding likely reflects the fact that the intervention at Camp Independence is not a "coping intervention"; indeed, adaptive coping is only addressed on 1 day of this weeklong intervention. Further, this finding may again point to the difficulties in using parent report for assessing coping in a child; it may be quite difficult for a parent to recognize change in their child's chosen coping strategies. These findings also support past research that has found coping styles to be generally stable over time (Kirchner et al., 2010; Shirkey et al., 2011).

Limitations

This study had several strengths including the use of a well-validated measure of coping that had yet to be used in this population, a large age range, multiple reporters, and a longitudinal design. Nevertheless, there were several limitations that should be noted. First, the study was overall descriptive in nature. Second, the study's sample size was small and predominately White, which limited our ability to test the generalizability of our findings. Further, our ability to accurately assess potential differences in coping styles with regard to certain demographic variables (i.e., age, IQ, income) was also hindered by a further reduced sample size due to missing data. Attrition from T1 to T2 is another important limitation to note. Our reduced sample size likely impacted on our longitudinal analyses, and left this study vulnerable to Type II error. Further, the drop in the sample size due to attrition was primarily amongst the youth self-report vs. parent report. Indeed, as previously noted, there are some concerns with parents ability to report on their child's coping; therefore, the loss of self-reported coping from T1 to T2 likely impacted our ability to detect statistically significant differences in coping over time. Further, our data were collected 1 month apart, which may not have been enough time to detect meaningful changes in coping. Future studies should repeat these analyses using a longer-term longitudinal design. Finally, the intervention discussed in this study only had 1 day dedicated to teaching about adaptive coping strategies; therefore, the non-significant findings with regard to change over time in coping style should be interpreted with caution. Such a lack of change in coping likely reflects the limitations associated with this intervention in terms of improving coping skills, rather than a reduced potential for skill improvement with a more focused intervention in this population.

CONCLUSION

The results of this study have several important clinical implications for promoting the psychosocial wellbeing of youth and young adults with SB. This was the first study to systematically examine coping styles in youth with SB. Research has consistently demonstrated that the use of adaptive coping strategies leads to improvement in social, emotion, and medical outcomes in pediatric chronic illness populations (Compas et al., 2012). This study was able to highlight areas of risk and resilience that can be built upon to create effective and targeted interventions. Results from this study indicated that youth with SB predominately use primary control coping strategies. Further, our findings suggest that youth with SB respond to illness-related stressors in a different manner compared to other pediatric populations (SCD and T1D). Specifically, youth with SB appear to use more primary control coping and less disengagement coping compared to SCD and T1D, and less secondary control coping compared to youth with T1D. Therefore, this study identified

that youth with SB have a tendency to utilize an adaptive coping strategy, highlighting a significant strength that can be built upon in the context of clinical intervention (e.g., focusing on primary control coping in therapy). Future studies should examine the effectiveness of clinical interventions that promote the use of primary control coping (e.g., modules of CBT such as problem solving) for reducing illness-related stress in youth with SB. Supporting the use of primary control coping may contribute to improving psychosocial and medical outcomes in this population.

Nevertheless, given that there are many intractable stressors in this condition, future studies should also examine the utility of using primary control coping in this population (i.e., some SB-related stressors may be less responsive to primary control coping) in order to develop interventions that utilize this strength (i.e., tendency toward primary control coping) when appropriate and teach secondary control coping strategies for stressors that are less likely to respond to primary control coping. Further, coping strategies were associated with intellectual functioning, such that lower IQ was associated with more frequent use of primary control coping and higher IQ was associated with more frequent use of secondary control coping. Therefore, it will be important to consider developmental age in addition to the nature of the stressor itself when creating clinical interventions. For example, for youth with lower IQ, it may be more effective to focus on promoting primary control coping strategies, such as emotion regulation, when faced with unchangeable stressors vs. acceptance, which is a more complex construct/strategy. Further, when one teaches secondary control coping to youth with lower cognitive functioning, it may be beneficial to focus on strategies that are less complex (distraction vs. cognitive restructuring). Relatedly, as EF deficits are common in this population, increased structure when teaching adaptive coping is essential. Helpful strategies may include: reminders on the patient's phone, visual cue cards to prompt the patient to engage in coping strategies, having a simple menu of coping skills ready and accessible to use, involvement of parents in therapy to facilitate the practicing of coping skills in between sessions. This study also found that higher SES was associated with more secondary control coping. Future studies should continue to examine the relationship between SES and coping styles in this population in order to develop actionable interventions for youth from diverse socioeconomic backgrounds. Finally, this study found that coping styles in youth with SB did not change significantly over time, further demonstrating the consistency of coping styles in pediatric populations.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Loyola University Chicago Institutional Review Board. Written informed consent to participate in this study was provided by the participant or the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

DO and TK contributed to conception and design of the study and performed the statistical analysis. OC and KS performed integral literature reviews. DO, OC, TK, KS, and GH wrote sections of the manuscript. GH, OC, and KS edited the manuscript. MS organized the database. All authors contributed to manuscript revision, read, and approved the submitted version.

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Biopsychosocial Predictors of Quality of Life in Paediatric Patients With Sickle Cell Disease

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Sickle cell disease (SCD) refers to a group of inherited blood disorders with considerable morbidity that causes severe pain, reduces life expectancy, and requires significant self-management. Acute painful episodes are the hallmark of SCD, but persistent daily pain is also highly prevalent in this population. Characterising the impact and experience of SCD-related morbidity (i.e., sleep disruption, frequent emergency department visits, cognitive dysfunction) on health-related quality of life (HRQOL) requires multiple assessment methods to best capture the underlying mechanisms. To gain a greater understanding of the effect of common symptom categories on HRQOL and to determine potential pain coping targets, the present study investigated whether demographic, socioeconomic, sleepiness, pain burden, frequency of emergency department (ED) visits, and cognition predicted HRQOL in a paediatric sample of patients with SCD. Our study was a secondary analysis of baseline assessment data of children with SCD aged 8–15 years ($n = 30$) in the Prevention of Morbidity in Sickle Cell Anaemia Phase 2b (POMSb2) randomised controlled clinical trial of auto-adjusting continuous positive airways pressure. Patients completed cognitive testing (IQ, Processing Speed Index, Delis-Kaplan Executive Function Scale (DKEFS) Tower, Conner's Continuous Performance Test), sleepiness (Epworth Sleepiness Scale), and HRQOL (PedsQL Sickle Cell Module) at baseline. Patients reported pain burden (Sickle Cell Pain Burden Inventory-Youth) each month over 8 visits. Caregivers provided demographic information and reported their child's executive function (Behavioural Rating Inventory of Executive Function) at baseline. Data from our analysis demonstrated that demographic factors (i.e., age, gender, level of neighbourhood deprivation) and treatment variables (i.e., hydroxyurea use) did not independently predict HRQOL, and laboratory values (i.e., haemoglobin, haematocrit, mean oxygen saturation) were not significantly correlated

with HRQOL (*ps* > 0.05). However, sleepiness, pain burden, ED visits, and executive dysfunction independently predicted HRQOL ($R^2 = 0.66$) with large effects ($\eta^2 = 0.16$ to 0.32). These findings identify specific, measurable symptom categories that may serve as targets to improve HRQOL that are responsive to change. This knowledge will be useful for multimodal interventions for paediatric patients with SCD that include sleep management, pain coping strategies, and executive function training.

Keywords: executive function, pain burden, sleep, emergency department visit, coping

INTRODUCTION

Sickle cell disease (SCD) is a group of genetic disorders that affect the structure and oxygen-carrying capacity of haemoglobin in red blood cells (Rees et al., 2010). SCD can impact multiple systems, with symptoms including an increased risk of infection, cognitive complications, and damage to the organs and bones (Redding-Lallinger and Knoll, 2006; Booth et al., 2010; DeBaun and Kirkham, 2016). Globally, it is estimated that 300,000 infants with SCD are born each year, with the majority born in Sub-Saharan Africa (Piel et al., 2013; Kato et al., 2018). Patients with SCD experience recurrent acute painful episodes with persistent pain between episodes (Dampier et al., 2002; Smith et al., 2008). The occurrence, location, severity, and duration of pain episodes and persistent pain can vary substantially in an individual patient tending to worsen with age (Ballas, 2020). These types of pain can sometimes have precipitating factors or objective signs specific to the individual patient (e.g., infection, fever, obstructive sleep apnoea, dehydration) and changes in the environment associated with increased admissions for pain in the population (e.g., increased wind speed, rainfall) (Piel et al., 2017). Importantly, unpredictable fluctuations can also lead to uncertainty (Gil et al., 2003). As such, patients with SCD have a unique pain profile (Yawn et al., 2014) that can negatively impact quality of life (Sil et al., 2016). Patients with SCD generally experience poorer quality of life than national norms and other patients with chronic conditions (i.e., cystic fibrosis, asthma, haemodialysis patients) with worsening quality of life as pain increases (McClish et al., 2005).

For patients with SCD, coping with pain and the associated complex treatment regimen (e.g., frequent hospital contacts) is often associated with comorbid psychological symptoms (e.g., depression, anxiety) (Benton et al., 2007; Graves et al., 2016), impaired family functioning (Oliver-Carpenter et al., 2011), chronic fatigue (Ameringer et al., 2014), and sleep disturbances (Kaleyias et al., 2008), all of which may be exacerbated by potentially challenging socioeconomic and environmental factors. Given the aetiology of pain in paediatric patients with SCD, assessing morbidity requires an integrative biopsychosocial approach to evaluating, formulating, and managing pain. To move beyond just measuring pain intensity, information on a wide range of relevant dimensions (i.e., biological, psychological, and sociocultural factors) using multiple assessment methods is needed to capture the patients' specific experiences and understand the causes, contributors, and effects of pain (Liossi and Howard, 2016).

Evaluation and treatment utilising the biopsychosocial approach enables patients to actively manage their condition and improve coping resources. Coping refers to the dynamic processes (behavioural and psychological) that people employ to manage or reduce stress when faced with adverse experiences, including pain or disease-related distress (Skinner and Zimmer-Gembeck, 2007). Pain coping is often categorised into strategies defined as adaptive (e.g., active activities like exercise, meditation, listening to music) and maladaptive (e.g., catastrophising). A similar dichotomy has been posited between "approach vs. avoidant" coping strategies, which describe engaging with or avoiding pain, respectively (Van Damme et al., 2008) and "problem-focused vs. emotion-focused" strategies, which describe efforts to control and emotionally manage pain, respectively (Lazarus and Folkman, 1984).

Consideration of potential cultural factors may account for some variation in pain coping strategies used by patients with SCD. For example, Oliver-Carpenter et al. (2011) found that children and adolescents with SCD used passive (e.g., resting, drinking fluids) rather than positive coping attempts (e.g., active coping strategies) most often. These mostly avoidant or passive pain coping strategies are related to more emergency department visits, higher pain intensity, and lower activity levels (Gil et al., 1991; Anie et al., 2002). Additionally, children with SCD have been shown to experience pain that affects sleep patterns and how they cope with pain (e.g., behavioural distraction or catastrophising) (Graves and Jacob, 2014). However, it has also been shown that patients with SCD with and without abnormal sensory patterns (e.g., enhanced pain sensitivity) frequently use positive approaches (e.g., seeking social support) for coping with pain (Hyacinth et al., 2020).

Distinctions between different types of coping strategies are not as clear cut as they may seem; however, as adaptiveness can vary with context, culture, and for the individual. Further, both types of strategies can be helpful for some patients in some settings. For example, recent studies have demonstrated that spiritual coping strategies influence health outcomes depending on whether they are more positive (e.g., seeking comfort and strength) or more negative (e.g., spiritual doubts) (Reynolds et al., 2013). Because Black individuals engage in more spiritual coping strategies (e.g., prayer) than White individuals and because most commonly used measures of coping conceptualise prayer as a passive/avoidant strategy (Meints et al., 2016) the potential adaptiveness of spirituality can be obscured.

Psychosocial interventions that empower patients with chronic conditions to learn coping skills can improve their ability

to manage and reduce persistent pain (Somers et al., 2018). Addressing the biopsychosocial factors related to persistent pain is particularly important for paediatric patients with SCD as they often lack the necessary skills and confidence to effectively manage their disease (Abel et al., 2015; Stollon et al., 2015). Study endpoints for psychosocial interventions for paediatric patients with SCD often aim to improve health-related quality of life (HRQOL) (Anderson et al., 2018; Adegbolagun et al., 2020). HRQOL is a multidimensional construct that includes health risks, conditions, functional status, physical and mental health, and social support (Ferrans et al., 2005). Specific to patients with SCD, better HRQOL is related to improved treatment, psychosocial and psychological outcomes and fewer hospital contacts (Thornburg et al., 2011; Beverung et al., 2015a; Sil et al., 2016; Hood et al., 2020b). In relation to pain coping, mediation analyses have shown that the relationship between pain and physical HRQOL was mediated by emotion-focused avoidance pain coping. Specifically, emotion-focused avoidance coping was related to worse pain and, in turn, decreased physical HRQOL (Lim et al., 2019). Relatedly, negative thinking mediated the role between pain and internalising psychological symptoms (Barakat et al., 2008). Finally, in a study of adults with SCD, affective coping strategies (e.g., anger and fear self-statement, isolation) significantly predicted poor HRQOL (Anie et al., 2002).

In addition to overall poorer HRQOL (Panepinto et al., 2013), cognitive challenges, particularly in the domains of processing speed (Stotesbury et al., 2018), attention (Daly et al., 2012), and executive function (Hood et al., 2019) are frequently observed in patients with SCD and have also been suggested as efficacious endpoints for clinical trials in this population (Farrell et al., 2019). The aetiology of cognitive difficulties in patients with SCD are multi-faceted (Prussien et al., 2020). There is some evidence for relationships between cognitive dysfunction and pain (Connolly et al., 2019), sleep (Marshall et al., 2009), pain coping (Ludwig et al., 2018), and HRQOL (Allen et al., 2017; Hardy et al., 2018). Two randomised control trials (RCT) have begun to utilise smartphone technology to improve pain outcomes (e.g., reduce emergency department visits) by targeting sleep hygiene techniques, pain tracking, and self-management (Schatz et al., 2015; Palermo et al., 2018). Nevertheless, more work is needed to better understand the relative impact of these symptom categories on HRQOL, along with the potential impact of cognitive difficulties. Moreover, studies have often considered one or two symptoms or aspects of SCD-related morbidity rather than considering multiple potential biopsychosocial factors in one paediatric cohort.

Given that utilisation of pain coping strategies improve the management of these symptom categories and complications (i.e., sleep, cognition, ED visits) and that pain coping interventions often aim to improve HRQOL, the purpose of our secondary analysis is to assess whether biopsychosocial factors that are frequent pain coping targets predict HRQOL. Gaining a deeper understanding of these factors will allow for targeted interventions for paediatric patients with SCD. Through the inclusion of multiple assessment tools that are often used when examining the effects of pain coping, we collected baseline data in the paediatric arm of the Prevention of Morbidity in Sickle Cell Disease Phase 2b (POMS2b) trial (Howard et al., 2018), we aimed to determine whether demographic, socioeconomic, sleep, pain burden, emergency department visits, and cognition predicted HRQOL in patients with SCD. We focused on cognitive data in domains where patients with SCD experience the most profound challenges (i.e., executive function, attention, and processing speed). The present study aims to understand what symptom categories and complications have the most significant effect on HRQOL for paediatric patients with SCD and provide knowledge for future multimodal interventions.

MATERIALS AND METHODS

Procedures

This study reports data from a *post hoc* secondary analysis of paediatric data ($n = 30$) collected before randomisation for the single-blind, randomised, controlled phase II trial (POMS2b). Baseline included all visits (i.e., cognitive testing and completion of caregiver and self-report) up to and including the day of randomisation (see **Figure 1** for a study timeline). The POMS2b trial compared 6-months of overnight auto-adjusting continuous positive airways pressure (APAP) with standard care ($n = 30$) to standard care alone ($n = 30$) in paediatric patients (children $\leq$16 years of age) and adults with sickle cell anaemia (HbSS genotype). The trial aimed to determine whether the intervention was safe and whether there were posttreatment improvements in cognition and brain structure. Descriptions of the study design and findings have been published elsewhere (Howard et al., 2018, 2019; Slee et al., 2019).

Participants were recruited through sickle cell clinics at the Guy's and St Thomas' Hospital and King's College Hospital in the United Kingdom (UK), serving ∼460 and 480 patients with SCD, respectively. Trained PhD and MSc neuropsychology students administered cognitive testing in a quiet room at University College London. Inclusion criteria for the paediatric arm of the POMS2b trial were being 8 to 15.99 years of age, having the ability to speak and understand English, a diagnosis of sickle cell anaemia (HbSS genotype). Exclusion criteria included mean overnight saturation of <90% for >30% of the night (i.e., meeting current criteria for overnight oxygen therapy), current or prior experience with overnight respiratory support, respiratory or decompensated cardiac failure, hospital admission for acute sickle complications within the past 1 month and more than 6 hospital admissions in the last 12 months, receiving chronic transfusions, or contraindications to APAP therapy or MRI. The study received ethical approval from the NRES Committee East of England—Cambridge South (14/EE/0163).

Measures

Demographics and Clinical Characteristics

Caregivers reported on child demographics. We obtained medical information through electronic medical chart review, including identifying if patients received common disease-modifying SCD treatments (i.e., hydroxyurea). Active medical problems documented in electronic medical records were reviewed to obtain neurological complications such as stroke history, abnormal magnetic resonance imaging (MRI; indicative

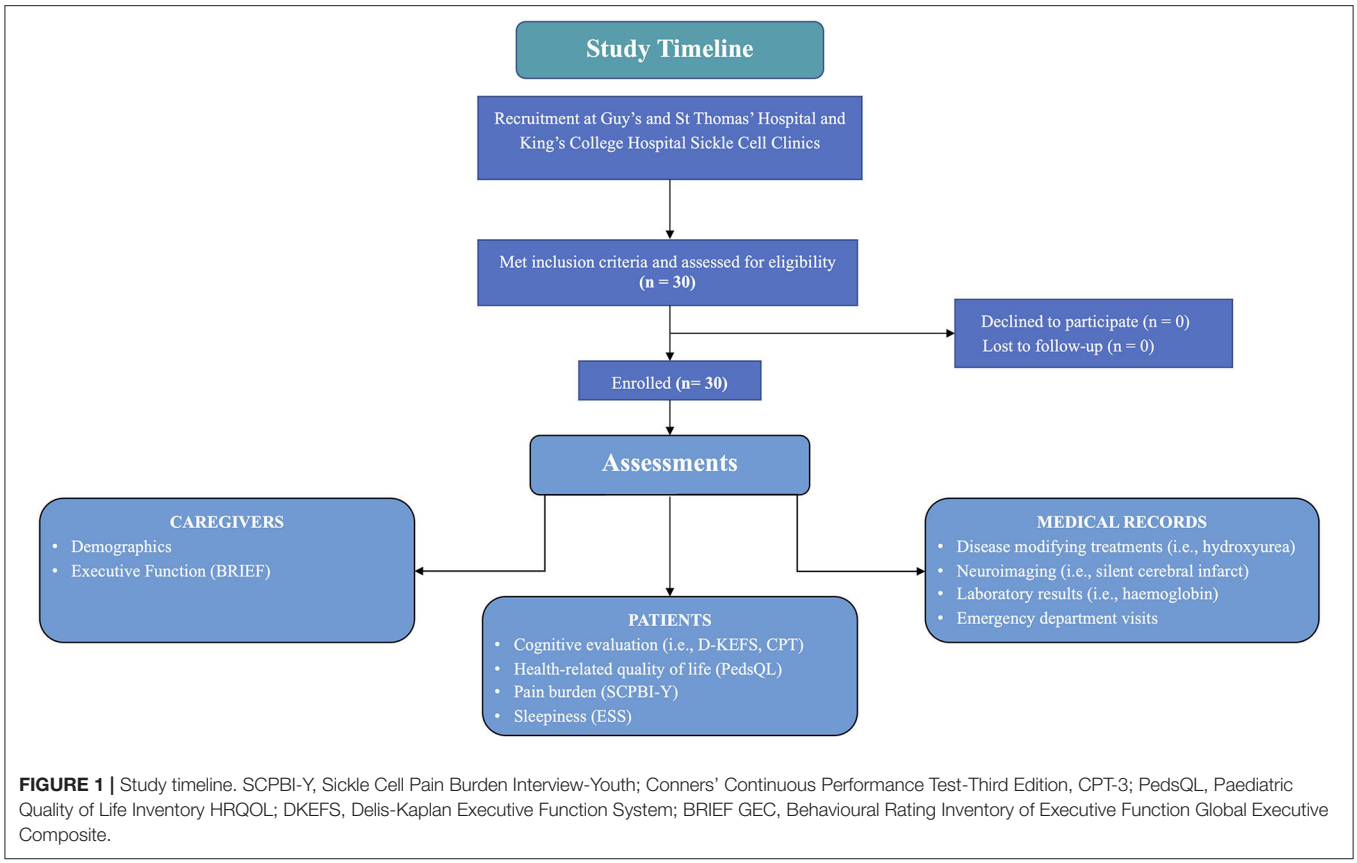

FIGURE 1 | Study timeline. SCPBI-Y, Sickle Cell Pain Burden Interview-Youth; Conners' Continuous Performance Test-Third Edition, CPT-3; PedsQL, Paediatric Quality of Life Inventory HRQOL; DKEFS, Delis-Kaplan Executive Function System; BRIEF GEC, Behavioural Rating Inventory of Executive Function Global Executive Composite.

of structural brain abnormalities), medical laboratory results (i.e., mean overnight oxygen saturation, haemoglobin, haematocrit), and the number of emergency department (ED) visits within the past year.

The English Indices of Deprivation are widely used open-source data that provide official government measures of relative deprivation in England (Ministry of Housing, Communities & Local Government, 2019). The Index of Multiple Deprivation (IMD) defines deprivation as encompassing seven different domains which are weighted as follows (i.e., income [22.5%], employment [22.5%], education [13.5%], health [13.5%], crime [9.3%], housing/services barriers [9.3%], and living environment [9.3%]), and is obtained through the patient's home postcode. These domains each have multiple components. For example, the housing and services barriers domain includes household overcrowding, homelessness etc. Neighbourhoods are ranked on a relative rather than absolute scale according to their level of deprivation relative to that of other areas. Zones are grouped into 5 bands (quintiles), each containing 20% of the data zone, with "1 = most deprived, 5 = least deprived."

Paediatric Quality of Life Inventory Sickle Cell Disease Module (PedsQL)

Participants completed the PedsQL (Panepinto et al., 2013) at baseline, which is a 43-item multidimensional rating scale that assesses HRQOL in children with SCD. Caregivers rated how much of a problem an issue had been for their child on a 5-point scale of "Never" to "Almost Always." Responses were reverse-scored and linearly transformed to a 0–100 scale (0 = 100, 1 = 75, 2 = 50, 3 = 25, 4 = 0). Total scores were then computed as the sum of the items divided by the number of items answered. Total scores and clinical classifications (81–100 = high levels of HRQOL, 61–80 = intermediate levels HRQOL, and 0–60 = poor HRQOL related to pain) were used in analyses (Beverung et al., 2015b).

Sickle Cell Pain Burden Interview-Youth (SCPBI-Y)

We assessed pain burden through the SCPBI-Y. Participants completed the SCPBI-Y at baseline, which is a 7-item multidimensional interview assessing the impact of pain on functional ability, sleep, school, and mood over the past month. Each item is rated on a 5-point Likert scale from "none = 0" to "every = 4." Scores range from 0 = no pain burden to 28 = severe pain burden. Scores from 8 months of visits were averaged and used in the present study to improve the reliability of the estimate and to best capture the experience of chronic or persistent pain (i.e., 12 weeks or more).

Epworth Sleepiness Scale (ESS)

Patients completed the Epworth Sleepiness Scale (ESS) (Johns, 1991) at baseline, which is an 8-item questionnaire that assesses the chance of dozing off or falling asleep while engaged in eight different activities. Responses are rated on a 4-point scale of 0 = "would never doze" to 3 = "high chance of dozing." The ESS

total score is the sum of all items and can range from 0 to 24. The higher the ESS score, the higher the child's daytime sleepiness.

Delis-Kaplan Executive Function System (D-KEFS) Tower Test

Participants completed the D-KEFS (Delis et al., 2001) at baseline. The D-KEFS comprises 9 tests with normed scores for children aged 8 to 18 years that represent domains of executive function. The Tower subtest was used in the present study and primarily assesses planning and problem-solving, as well as learning rules, inhibiting impulsivity, and maintaining instructional sets. Participants have to build a series of progressively more difficult towers using disks of various sizes in as few moves as possible across three pegs, following pre-specified rules and within a time limit. The Tower test is discontinued after three consecutive failures. Tower raw scores were converted to standardised scores (M= 10, SD = 3) and examined using the summary score (i.e., Total Achievement Score), which is the sum of achievement points earned for all completed items. A higher total achievement score represents better overall executive function.

The Conners' Continuous Performance Test-Third Edition

Participants completed the Conners' Continuous Performance Test-Third Edition (CPT-3) (Conners, 2000) at baseline, which is a 14 min computerised test of sustained attention and impulsivity where letters are displayed on a computer monitor one at a time. Participants observe the stimuli and press the space bar as quickly as possible after each letter, except for the letter X. Outcome measures used in the present study are omission errors which represent the number of times the target was presented, but the participant did not respond (i.e., distractibility) and commission errors which represent the number of times the participant responded but no target was present (i.e., impulsivity). Higher T-scores (> 65) indicate clinical concern (i.e., poorer sustained attention).

Behavioural Rating Inventory of Executive Function

Caregivers completed the 86-item Behavioural Rating Inventory of Executive Function (BRIEF) at baseline. The BRIEF assesses EF behaviour in the school and home environments (Gioia et al., 2000). The BRIEF scales include: Inhibit, Shift, Emotional Control, Self-Monitor, Initiate, Working Memory, Planning/Organisation, Task Monitoring, and Organisation of Materials. The Global Executive Composite (GEC), which incorporates scores from all scales, was used in our analyses. Higher T-scores (>65) indicate clinical concern (i.e., greater executive dysfunction).

Wechsler Tests

Participants completed tests at baseline. The Wechsler Abbreviated Scale of Intelligence (WASI) (Wechsler, 1999) which was used to estimate the intelligence quotient (IQ; Mean = 100, SD = 15) and the Wechsler Intelligence Scale for Children Fourth-Edition (WISC-IV) (Wechsler, 2003) was used to measure the Processing Speed Index (PSI) using the scores from the Symbol Search, Coding or Cancellation (substituted when required) subtests.

Statistical Analyses

The R statistical package was used to conduct analyses (Team, 2016). Descriptive statistics and summary scores with percentages described the overall sample. Visual inspection of our data using histograms along with skew and kurtosis analyses indicated that the number of ED visits within the past year and pain burden were both positively skewed (i.e., mostly lower values). Therefore, these two variables were log-transformed. One-sample *t*-tests assessed differences in sample cognitive scores and the normative population mean. Bivariate Pearson correlations assessed linear relationships between continuous predictors of interest. Two hierarchical linear regression analyses were conducted assessing predictors of HRQOL. To test the most parsimonious model, we only included potential predictors in models if correlations with HRQOL were above $r = 0.2$. To increase the power of our models, demographic [i.e., age, gender, deprivation (IMD)] and medical variables (i.e., receiving hydroxyurea) were only included in linear regression models if they were significant predictors of HRQOL ($ps < 0.05$).

To reduce multicollinearity, we conducted two separate regression models assessing whether psychosocial measures and tests of executive function (i.e., BRIEF GEC) and attention (i.e., CPT commissions errors) predicted HRQOL over and above the influence of symptom categories (i.e., sleepiness, pain burden, ED visits). We considered $p < 0.05$ two-tailed as statistically significant. Partial eta squared (η_p^2) was the measure of effect size used for the linear regression analyses. $\eta^2 = 0.01$, 0.06, and 0.14 represented small, medium, and large effect sizes, respectively (Cohen, 1988). Bias and corrected 95% confidence intervals were used as interval estimates to adjust for possible bias and skewness.

RESULTS

Participant Characteristics

Baseline demographic characteristics are displayed in **Table 1**. The mean age of participants was 12.3 years (40% female). Most participants lived in the most deprived regions in the UK. Nearly half had silent cerebral infarction on MRI. Less than a quarter had a pre-existing diagnosis of asthma. The number of ED visits within the past year was low, with 86% of participants having zero visits. Just over half of the sample were receiving hydroxyurea. Cronbach's alpha's for patient-reported outcome measures ranged from 0.66 to 0.98, demonstrating adequate to excellent internal consistency (see **Table 2**).

Descriptive data from patient-reported outcomes indicated some variability in overall pain burden across study visits, but scores were generally low in the sample (see **Figure 2**). Sleepiness scores were generally low. Data further revealed that 37% of the sample had poor-to-intermediate HRQOL, and nearly 14% of the sample had clinically elevated BRIEF scores (indicative of executive dysfunction) (see **Table 3**). The cognitive scores of the overall sample indicate that IQ and CPT-3 omission errors were firmly in the average range and were not significantly lower than the normative population mean. In contrast, the PSI and DKEFS

TABLE 1 | Characteristics of POMS2b paediatric patients at baseline before randomisation.

Characteristics	*N* = 30
Age	
Mean (SD)	12.3 (2.1)
Range	8–15
	***N* (%)**
Sex	
Female	12 (40%)
Male	18 (60%)
IMD quintile (1 = most deprived)	
1	13 (43.3%)
2	15 (50%)
3	2 (6.7%)
4	0 (0%)
5	0 (0%)
Genotype	
HbSS	30 (100%)
Silent cerebral infarct on MRI	
No	16 (53.3%)
Yes	14 (46.7%)
Prescribed hydroxyurea	
No	14 (46.7%)
Yes	16 (53.3%)
Diagnosis of asthma	
No	23 (76.7%)
Yes	7 (23.3%)
	Mean (SD)
Mean oxygen saturation (%)	95.8 (2.8)
Haemoglobin level (g/l)	89.3 (11.9)
Haematocrit level (volume %)	27.2 (3.9)
	Median (range)
Number of ED visits in the past year	0 (0–2)

SD, Standard deviation; IMD, Index of Multiple Deprivation; MRI, Magnetic Resonance Imaging; ED, Emergency department.

TABLE 2 | Patient-reported outcomes for POMS2b paediatric participants at baseline before randomisation.

Measures	α	*N* = 30
		Mean (SD)
Sickle Cell Pain Burden Interview-Youth	0.90	1.0 (1.7)
Range		0–28
Epworth Sleepiness Scale	0.66	4.6 (3.4)
Range		0–24
PedsQL Sickle Cell Disease Module	0.98	79.3 (21.3)
Range		0–100
PedsQL clinical classifications [*N* (%)]		
Poor		8 (26.7%)
Intermediate		3 (10%)
High		19 (63.3%)
BRIEF GEC T scores	0.93	51.5 (9.5)
Range		36–68
BRIEF classifications		
Within normal		25 (86.2%)
Clinically elevated		4 (13.79%)

α, Cronbach's alpha; PedsQL, Paediatric Quality of Life Inventory; BRIEF GEC, Behavioural Rating Inventory of Executive Function Global Executive Composite. Higher T-scores are of greater clinical concern.

Tower total achievement scores were poorer than the population mean. CPT-3 commission errors were also significantly lower; however, only higher scores are indicative of impulsivity (see **Table 3**).

Correlation Analyses

Initial bivariate correlations demonstrated that HRQOL was not correlated with any baseline laboratory values (i.e., mean oxygen saturation, haemoglobin, or haematocrit) ($ps > 0.05$). Regarding patient-reported outcomes, HRQOL was significantly negatively correlated with the SCPBI-Y ($r = -0.35$) and the ESS ($r = -0.57$), indicating that those with poorer HQROL had higher pain burden and more sleepiness. In terms of cognition, HRQOL was significantly negatively correlated with CPT-3 commission errors ($r = -0.33$) and the BRIEF GEC ($r = -0.48$), indicating that those with poorer HQROL had more impulsivity and greater executive dysfunction (see **Figure 3** for a correlation matrix of all potential predictors).

Hierarchical Regression Analyses

Preliminary analyses indicated that no demographic [i.e., age, gender, deprivation (IMD)] or medical variables (i.e., hydroxyurea) were significant predictors of HRQOL ($ps > 0.05$), so they were not considered in subsequent linear regression models. As correlation analyses determined that the CPT-3 commission errors (impulsivity) and BRIEF GEC (executive dysfunction) were correlated with HRQOL, we assessed only these measures of cognition in regression analyses (a priori cut-off of $r > 0.2$).

Each regression model determined if attention and executive function predicted HRQOL within models that included ESS (sleepiness), SCPBI-Y (pain burden), and ED visits within the past year. The variance-inflation factors (VIF) in regression analyses were within the acceptable range (1.14 to 1.62), indicating that correlations between independent predictors are not inflating the regression coefficient due to multicollinearity. Our regression analyses indicated that ESS (sleepiness) and ED visits within the past year were significant predictors of HRQOL in both models, including CPT-3 commission errors and the BRIEF. Scores on the SCPBI-Y (pain burden) approached significance ($p = 0.06$).

We found that CPT-3 commission errors did not significantly predict HRQOL ($p = 0.84$). However, the regression analysis including the BRIEF GEC was the most explanatory (overall model fit $R^2 = 0.66$) and demonstrated that the BRIEF GEC significantly predicted HRQOL ($p = 0.04$). Effect sizes for all predictors were large ($\eta^2 = 0.16$–0.32) (see **Table 4**). Based on our data, with an estimated mean T score for the BRIEF GEC of 51.50 in our sample, for every 0.74 *increase* in the BRIEF T-score, there was a corresponding

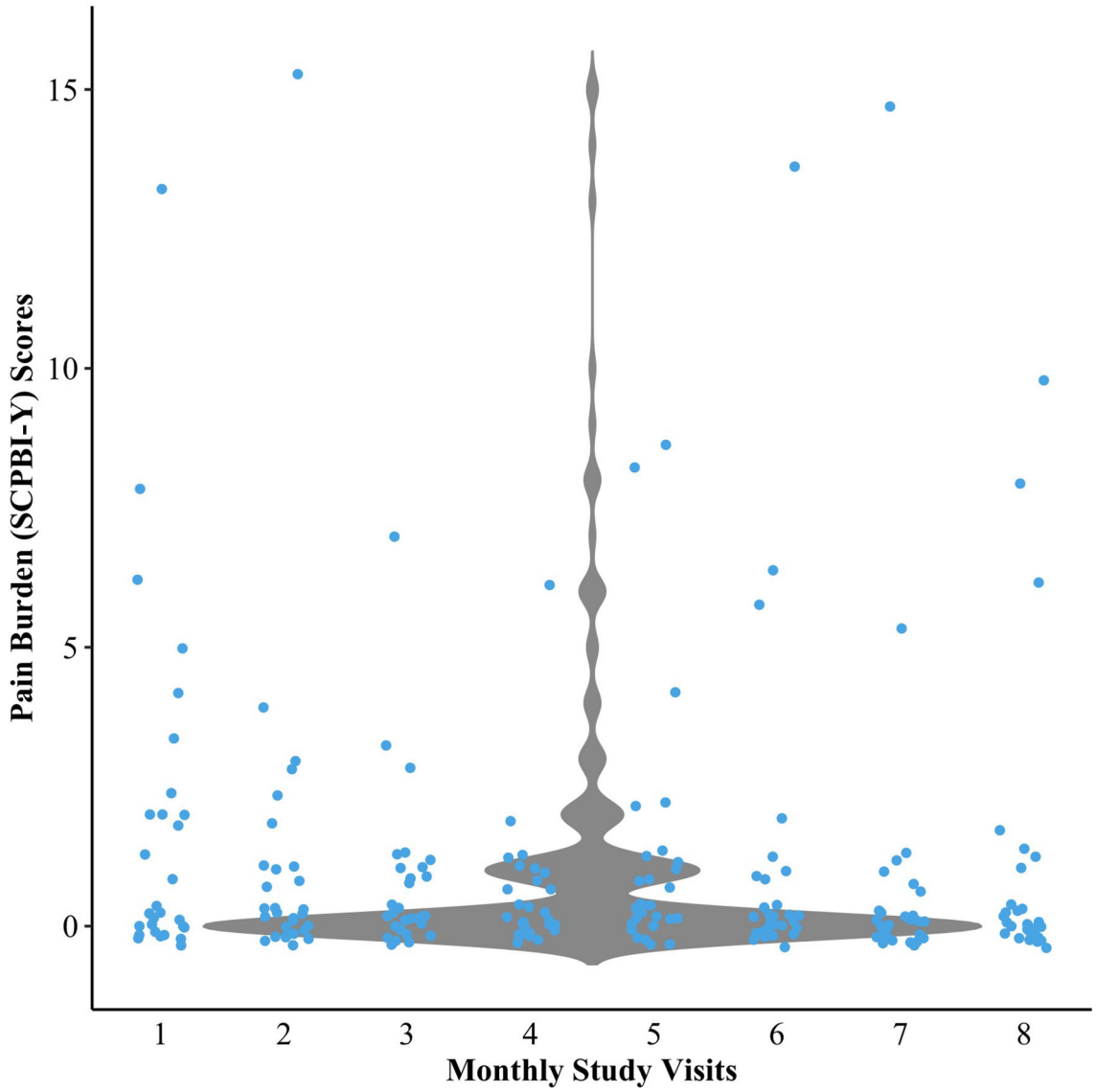

FIGURE 2 | Violin plot demonstrating the variability in pain burden scores over 8 study visits. SCPBI-Y, Sickle Cell Pain Burden Interview-Youth.

1-point *decrease* in HRQOL indicating that as executive dysfunction increased, HRQOL decreased for paediatric patients with SCD.

DISCUSSION

Our *post hoc* analysis of data from the POMS2b RCT demonstrated sleepiness, pain burden, ED visits, and executive dysfunction predicted HRQOL. All effect sizes were large and clinically significant. Although impulsivity was negatively correlated with HRQOL ($r = -0.33$), it was not a predictor within regression models. Demographic factors (i.e., age, gender, deprivation) and treatment variables (i.e., hydroxyurea use) did not independently predict HRQOL and laboratory values (i.e., haemoglobin, haematocrit, mean oxygen saturation) were not significantly correlated with HRQOL.

Patients with SCD often experience marked inequities influenced by geographic region, population density, socioeconomic status, care and pain outcomes, and disease management, along with reduced access and use of comprehensive specialist clinics (Mensah et al., 2019). Our findings did not show an association between these factors and HRQOL, but our patients were from a single geographical region and attended a comprehensive SCD clinic, which may explain why these factors had less of an influence on HRQOL. These sociocultural factors *should* be incorporated into the design, development, and implementation of skills-based programmes to ensure equitability and access. Within the context of pain coping

TABLE 3 | Cognitive scores of POMS2b paediatric participants at baseline before randomisation.

Cognitive scores	Sample $N = 30$	Normative population mean	Mean difference	Difference between the sample and the normative population mean		
				t	*p*	Cohen's *d*
	Mean (SD)					
Intelligence Quotient (IQ)	99.7 (13.2)	100 (15)	0.30	−0.14	0.89	0.03
Processing Speed Index (PSI)	93.9 (13.8)	100 (15)	6.1	2.38	**0.03**	0.44
DKEFS Tower achievement score	8.9 (2.0)	10 (3)	1.1	−3.00	**0.01**	0.55
CPT-3 omission errors	54.0 (14.7)	50 (10)	4	1.48	0.15	0.27
CPT-3 commission errors	46.4 (8.5)	50 (10)	3.6	2.24	**0.03**	0.42

SD, Standard deviation; DKEFS, Delis-Kaplan Executive Function System; Conners' Continuous Performance Test-Third Edition, CPT-3; IQ and PSI, Standard Score; DKEFS, Scaled Score; CPT-3, T-score; Higher T-scores are of greater clinical concern. Significant differences are highlighted in bold.

interventions, however, the symptom targets that predicted HRQOL in the present study are defined, measurable, and most importantly, amenable to change.

Pain is often referred to as the "hallmark of SCD," given that painful episodes and persistent pain frequently occur throughout the lifespan and are often the first presentation of the disease (Dampier et al., 2017). Although this focus is warranted, other pain coping targets (i.e., sleep) also play a role in HRQOL. Our data revealed the strong relationship between sleepiness and HRQOL and suggest incorporating measures to explore coping in relation to sleep difficulties (e.g., sleep-disordered breathing) and sleep hygiene behaviours (e.g., bedtime routine) may be a valuable addition to coping interventions for children with SCD. Our findings add to a growing body of literature that has demonstrated that more sleep disturbances (i.e., parasomnias and movement at night) were related to greater daytime sleepiness (Kölbel et al., 2020) and that there is a bi-directional relationship between poor night-time sleep quality and higher daytime pain in youth with SCD (Valrie et al., 2020). Expanding upon the influence of sleep, fatigue (a feeling of tiredness or exhaustion) has also been shown to be more strongly related to cognitive deficits in children with SCD (Rogers et al., 2017). In a non-SCD adult sample of patients with fibromyalgia, more sleep disturbances were associated with a reduced use of pain coping strategies (Theadom et al., 2007). Although further research is necessary with a paediatric SCD sample, sleepiness, cognitive function, and HRQOL are probably overlapping aspects of daytime function that may reduce the ability to cope with demands throughout the day, limiting the ability to use pain coping resources.

Although the number of ED visits within the past year was low in our sample, we did show that attending the ED had a large negative influence on HRQOL. This finding aligns with previous research in adults with SCD, where high ED utilisers experienced more distress and poorer HRQOL (Aisiku et al., 2009). Increased ED visits likely reflect increased pain, but increased ED visits also have an independent effect on HRQOL. Coping with the challenges of frequent hospital contacts and ED visits can be burdensome for patients with SCD. Being a disease that impacts a predominately Black population, SCD places patients at increased risk of racialised disparities. Patients with SCD report that race affects the quality of care and interpersonal relationships with hospital staff (Nelson and Hackman, 2013). Despite these significant barriers, a coping skills training programme has been shown to reduce ED visits in paediatric patients with SCD (Broome et al., 2001).

In non-SCD paediatric pain populations, multimodal interventions are effective for chronic pain management. They have demonstrated improvements in pain intensity, functional disability, anxiety, depression, catastrophising, school attendance, school functioning, and pain acceptance, with similar effectiveness seen whether the interventions were delivered in inpatient or outpatient settings. Successful non-pharmacological components of these interventions include group-based cognitive behavioural therapy (CBT), acceptance and commitment therapy (ACT), biofeedback, and acupuncture (Liossi et al., 2019). A wide diversity of non-pharmacological interventions have been assessed in adult and paediatric populations with SCD, including many of the successful components described. Other interventions that have also been tested include complementary therapies (e.g., prayer, guided imagery, music therapy) and physically-based therapies (e.g., massage therapy, yoga, aquatic rehabilitation) (Williams and Tanabe, 2016), as well adjuvant technology (e.g., virtual reality, smartphone app), which have been shown to improve self-efficacy and self-management about successfully managing SCD-related complications (Agrawal et al., 2019; Crosby et al., 2020, 2021; Hood et al., 2020a). However, most of these interventions are not multimodal, have primarily assessed acceptability and feasibility, and have been conducted with small samples at a single institution (Williams et al., 2018). Future interventions will need to be on a larger scale, include randomisation, and incorporate a biopsychosocial approach with significant patient involvement to increase their engagement in improving pain-coping resources and HRQOL.

In light of the fact that executive function difficulties have frequently been demonstrated for patients with SCD (Hood et al., 2019, 2020b; Prussien et al., 2019) and that executive function was predictive of poorer HRQOL in this study, we would suggest the utilisation of multimodal interventions that include components

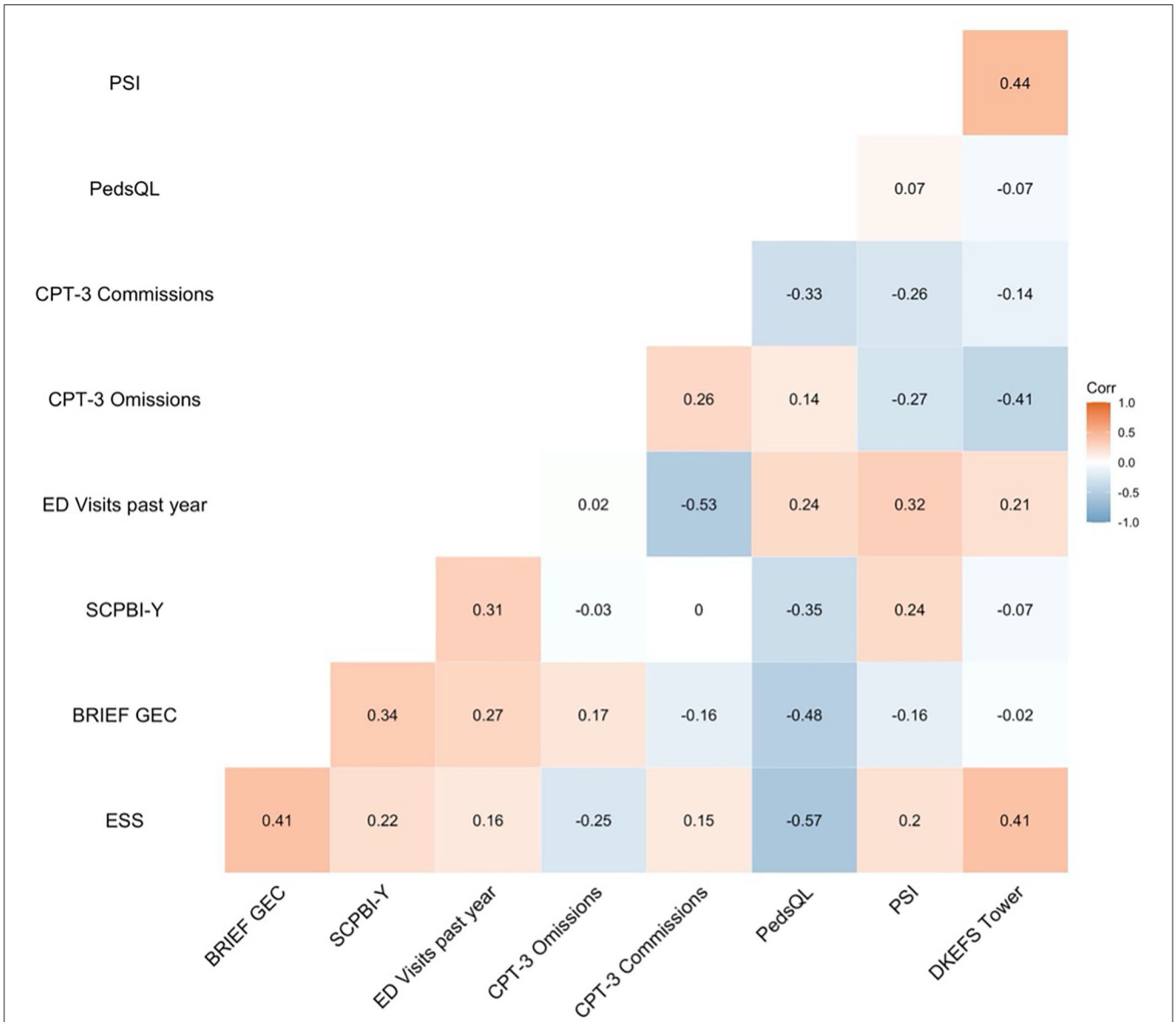

FIGURE 3 | Bivariate correlations of patient-reported outcomes, emergency department visits, and cognition. SCPBI-Y, Sickle Cell Pain Burden Interview-Youth; Conners' Continuous Performance Test-Third Edition, CPT-3; ED, emergency department; PedsQL, Paediatric Quality of Life Inventory HRQOL; DKEFS, Delis-Kaplan Executive Function System; BRIEF GEC, Behavioural Rating Inventory of Executive Function Global Executive Composite.

of executive function training or monitoring. Generally, studies assessing how to improve executive function in patients with SCD have not incorporated pain assessment, coping, or management (Marshall et al., 2009; Hood et al., 2020b). However, one small pilot study did demonstrate that participants who completed more of the executive function training programme had lower pain impact scores on the PedsQL, providing a promising avenue for future research (Hardy et al., 2016).

Across the symptom categories identified here, adequate cognitive function is required to process and work through any potential hurdles. Patients with SCD experience specific challenges in the cognitive domains that, if improved, would make managing pain and their treatment regimen easier. Cognitive deficits in processing speed, attention, and executive function make it more difficult to quickly process information from medical providers and maintain sustained attention over long periods. Further, the integration of data from multiple sources along with balancing needs and priorities also requires the ability to process higher-order information. Our study found that executive dysfunction measured through caregiver-report was related to HRQOL, but executive, attention, and processing speed test scores were not associated. Difficulties in completing everyday behaviours in the school and home environment (e.g., time management, organisation) may have

TABLE 4 | Two regression model with health-related quality of life (PedsQL) as the dependent variable and sleepiness (ESS), pain burden (SCPBI-Y), ED visits within the past year, with either executive function (BRIEF GEC) or CPT commission errors as predictors.

Effect	*B*	*B* 95% CI [LL, UL]	*β*	*SE*	*r*	*Partial* η^2	*p*	Fit
(Intercept)	134.46	[100.05, 168.87]						
Epworth Sleepiness Scale	−2.93	[−4.86, −0.99]	−0.46	0.90	−0.57	0.32	**0.005**	
Sickle Cell Pain Burden	−11.74	[−24.00, 0.53]	−0.29	1.61	−0.35	0.16	0.06	
Number of ED visits in the past year	34.41	[13.54, 55.29]	0.49	5.72	0.23	0.36	**0.003**	
BRIEF Global Executive Composite	−0.74	[−1.46, −0.02]	−0.33	0.33	−0.48	0.18	**0.04**	
								R^2 = **0.633**
								95% CI [0.25, 0.73]
(Intercept)	102.71	[60.01, 145.40]						
Epworth Sleepiness Scale	−3.44	[−5.48, −1.40]	−0.55	0.98	−0.58	0.36	**0.002**	
Sickle Cell Pain Burden	−14.04	[−27.27, −0.81]	−0.35	6.36	−0.37	0.18	**0.03**	
Number of ED visits in the past year	30.04	[2.06, 58.02]	0.42	13.45	0.23	0.19	**0.03**	
CPT-3 Commission Errors	−0.07	[−0.98, 0.85]	−0.03	0.44	−0.33	0.01	0.88	
								R^2 = **0.552**
								95% CI [0.14, 0.67]

B represents unstandardised regression weights. LL and UL indicate the lower and upper limits of a confidence interval, respectively. β, standardised regression weights; SE, Standard Error; r, the zero-order correlation; Partial η^2*, partial eta squared. Significant differences are highlighted in bold (p < 0.05). ED, Emergency department; BRIEF GEC, Behavioural Rating Inventory of Executive Function Global Executive Composite; CPT-3, Conners' Continuous Performance Test-Third Edition.*

a greater effect on HRQOL, emphasising the need for future studies to incorporate ecologically valid measures (Berg et al., 2012). Adaptations to pain coping interventions to account for and better accommodate these cognitive challenges could include clear communication that can be understood the first time it is read or heard. Using plain or jargon-free language is not "dumbing things down"; instead, it ensures information is accessible for the intended audience. Other modifications could include integrating frequent and immediate feedback during components of pain coping skills training, shorter, more frequent sessions, and breaking down tasks and materials (e.g., handouts) into small, manageable steps.

Some limitations influence the present study's findings that should be addressed. Although a strength of research was that our sample was drawn from an RCT, and pain burden was measured each month over 8 months, our data primarily focuses on baseline assessments. More robust and longitudinal research designs are needed to thoroughly appreciate the consequences of sleepiness, pain burden, ED utilisation, and executive dysfunction on paediatric patients with SCD. It is also possible that including pain burden scores collected after assessing HRQOL could have influenced results. Additionally, our sample size was small, so we did not have the power to detect smaller effects, and possible interactions between variables (e.g., pain burden and sleep) and analyses of specific PedsQL subscales could not be assessed. Replication in a larger sample would provide greater clarity. This study was conducted at large, urban institutions in the UK, and the cohort may not be representative of the paediatric patients internationally. Although we collected data to assess hydroxyurea use, adherence to this treatment was not included in our study protocol. Our study did not include a direct measure of pain coping, which would have allowed us to test the relationship with our variables of interest. However, not having a specific pain coping measure allowed us to consider whether more generic coping scales that assess a range of outcomes might be more useful, given the complexity of the challenges faced by patients with SCD.

Our study aimed to elucidate the symptom categories influencing HRQOL to determine targets for future multimodal pain coping and HRQOL interventions. We found that demographic and medical variables were weakly related to HRQOL; whereas, sleepiness, pain burden, ED visits, impulsivity, and executive dysfunction were more strongly related to HRQOL. Notably, almost all of the strongly correlated variables (except impulsivity) independently predicted HRQOL. Although thoughtfulness about sociocultural factors is critically needed to develop programmes and retain patients with SCD in research and clinical practise, we have shown that specific symptom-based targets for pain coping are measurable and may be responsive to change through intervention. As we consider helping to improve the lives of this paediatric population, it is necessary to expand beyond the measurement of pain intensity and instead integrate the assessment of measurable symptom categories that may serve as targets to improve HRQOL that are responsive to change.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available because the datasets generated and analysed during the current study are available from the corresponding author on reasonable request and in accordance with ethical restrictions imposed by the Ethics Committees that approved this study. Requests to access the datasets should be directed to fenella.kirkham@ucl.ac.uk.

ETHICS STATEMENT

The study received ethical approval from the NRES Committee East of England – Cambridge South (14/EE/0163). Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

FK is the chief investigator, she conceived the study, grant application, and protocol development. CL made significant contributions to the design and implementation of the study. AH drafted this work and managed all revisions. AH, HS, MK, JK, and AS contributed to the study design and development of the proposal. BI, MP, JH, SC, SH, and MA made contributions to the conception of the study, the design of the work, and data acquisition. All authors read and approved the final manuscript.

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The Measurement and Conceptualization of Coping Responses in Pediatric Chronic Pain Populations: A Scoping Review

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Background: Pediatric chronic pain is a prevalent condition that requires significant coping to encourage optimal functioning; however, relevant research is vast, heterogeneous, and difficult to interpret. To date, no attempt has been made to map and summarize the measurement and conceptualization of coping responses in the context of pediatric chronic pain.

Objectives: A scoping review was conducted to map and summarize the participant characteristics, methodologies, theoretical frameworks, and measures used to assess coping responses in youth with chronic pain. The extent to which authors used definitions and examples of coping responses (conceptual clarity) as well as consistently used measures (measurement consistency) and their corresponding conceptualizations (conceptual consistency) relative to how they were intended to be used were assessed.

Methods: Searches were conducted through MEDLINE (PubMed) and PsycINFO. Following title/abstract screening, full-text extractions were performed on 125 English-language publications on coping in youth with chronic pain.

Results: Of the 125 studies, only 12.8% used a theoretical framework to explain the coping responses assessed, and even fewer (7.2%) used theory to guide measure selection. Conceptual clarity was rated "low/very low" (i.e., no definitions and/or examples) for 47.2% of studies. The majority of studies were conducted in the United States (67%) and a preponderance of White and female participants was sampled. The research primarily used quantitative methods (85%) and cross-sectional designs (67%). Parent- or self-report questionnaires were the most common methods for assessing coping (86%). Of the 95 studies that utilized one of the 14 questionnaires with known psychometric properties, 33.7 and 55.8% had one or more discrepancies for conceptual and measurement consistency, respectively.

Conclusions: This review highlights the lack of clear descriptions and theoretical frameworks of coping responses for pediatric chronic pain. Inconsistencies in the measurement and conceptualization of coping responses limit research and clinical

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advancements. As a field, we need to strive toward using well-developed theory to create fewer, more well-established standardized measures with clearly defined coping responses. Opportunities for qualitative and observational research in more diverse patient populations should be considered for theory construction and measure validation.

Clinical Trial Registration: https://osf.io/xvn2a/?view_only=eff04e0c0b9649be89d403b10e9ff082.

Keywords: coping, coping responses, pediatric chronic pain, scoping review, conceptualization and measurement

INTRODUCTION

Pediatric chronic pain refers to persistent or recurrent pain in infants, children, and adolescents (herein "youth") that lasts for more than 3 months and lacks an adaptive purpose (Treede et al., 2015). Given the long-lasting and unpredictable nature of chronic pain, youth rely on coping to manage their pain and its impact (Peres and Lucchetti, 2010). Coping is defined as the use of intentional and effortful thoughts or behaviors to manage the internal and external demands of stressful situations or experiences (Compas et al., 2014). Rudolph et al. (1995) proposed a model that conceptualizes coping to occur in a sequence of events (i.e., a "coping episode") consisting of coping responses, goals, and outcomes. Within this model, coping responses are defined as mental or physical actions initiated in relation to a perceived stressor (Rudolph et al., 1995). Coping goals are the reasons for engaging in a particular coping response, and coping outcomes are the specific consequences of a coping response. Coping responses may serve different goals and be related to different outcomes across time and situations (Skinner et al., 2003). Therefore, our ability to understand the efficacy and implementation of a coping episode depends on successfully identifying and measuring coping responses. Although there are other models of coping (Skinner et al., 2003; Stanisławski, 2019), this review uses the abovementioned terminology to summarize the literature.

The coping literature is vast, heterogenous, and difficult to interpret. In an effort to better understand this literature, reviews have summarized and critically evaluated the measurement and conceptualization of coping in adult chronic pain (Peres and Lucchetti, 2010), childhood chronic illness (Rudolph et al., 1995; Compas et al., 2012), and general stress (Jensen et al., 1991; Skinner et al., 2003; Garcia, 2010; Stanisławski, 2019); however, there is no review of coping in pediatric chronic pain. This is a critical gap considering that coping responses may vary by health condition as well as age (Garcia, 2010; Compas et al., 2012). For example, chronic pain is often perceived as less predictable and controllable than other chronic illnesses (Compas et al., 2012). Consequently, youth with chronic pain may rely more on coping responses aimed at *adapting* to their pain such as "distraction" or "acceptance" rather than efforts to *change* their pain or emotional response to the pain (e.g., "problem-solving", "emotional regulation"). Moreover, developmental changes in perceived control and competence, in combination with sociocultural factors (e.g., culture and gender), influence the way people choose and implement various coping responses (Compas et al., 1991; Compas, 1998). For instance, children and young adolescents may be less capable of managing their emotional reactions to their pain and depend more directly on their caregivers to cope than older adolescents and adults (Skinner and Zimmer-Gembeck, 2016). Therefore, a comprehensive review of the existing literature on coping in the context of pediatric chronic pain is needed to identify and evaluate measures and conceptualizations of coping responses specific to this population.

In the broader coping literature, several important gaps limit the ability to communicate effectively about coping and consolidate research on effectiveness. With regard to conceptual gaps, there is a lack of consensus about how best to classify and define coping responses (Skinner et al., 2003; Stanisławski, 2019). As such, coping has been conceptualized using over 400 coping responses organized within more than 100 different typologies (Skinner et al., 2003). Typologies of coping are generally hierarchical and multidimensional organization systems where specific coping responses are unidimensional lower-order categories nested within more complex and abstract higher-order categories of coping. Correspondingly, the most widely used conceptualizations of lower- and higher-order coping responses in the pediatric coping literature include coping *strategies* (i.e., specific and discrete cognitive, emotional, and/or behavioral responses, such as "planning" or "distraction") and coping *styles* (i.e., a set of coping strategies that fulfill a specific function showing relative stability over time and situations, such as "problem-focused" or "emotion-focused" coping), respectively (Stanisławski, 2019). The extent to which other terminologies have been used remains unclear. Furthermore, the organization of lower-order into higher-order categories is typically accomplished according to their intended function (i.e., the coping goal) and topological distinction (i.e., descriptive categories that are concerned with *how* children cope) (Skinner et al., 2003). However, given that some lower-order coping responses include multiple functions and behaviors, the use of a classification system may contribute to inconsistencies across coping measures. For example, within "problem-focused" coping (i.e., responses aimed at modifying or eliminating the stressor) and "emotion-focused" coping (i.e., responses aimed at managing the emotions aroused by the stressor), the response of "planning" (identified as a lower-order category on scales assessing coping) can be functional for both "problem-focused" and "emotion-focused" coping by guiding problem-solving and calming negative emotions, respectively. Although theories and definitions can guide more consistent

conceptualizations of coping responses, they are seldom used (Garcia, 2010). Addressing conceptual inconsistencies (i.e., varied terminologies/categorizations) and ambiguities is critical as they limit our ability to draw conclusions from research and make real-world applications.

Existing reviews also highlight the limitations of having too many measures of coping. Within the general pediatric coping literature, at least 52 measures of a child coping have been identified (i.e., 38 self-report measures, eight observational measures, and six caregiver-report measures) (Blount et al., 2008). The existence of a large number of measures that vary in content and structure contributes to an excessive number of coping responses and makes it difficult to compare and consolidate research findings across studies. In addition, the lack of clear theories and descriptions of coping responses makes it difficult to understand the similarities and differences across measures.

The *timing* in which coping is assessed in relation to the identified stressor is also not well-understood. Within pediatric chronic pain, proactive coping would include efforts or goals undertaken in advance of a painful episode to prevent it or reduce its severity (e.g., practicing daily mindfulness), and reactive coping would include responses to experiencing pain (e.g., listening to music to distract from pain). Distinguishing between proactive and reactive coping is particularly important for intervention planning. For instance, assessments of proactive coping may be more relevant to interventions aimed at facilitating lifestyle changes. In contrast, interventions aimed at helping youth learn how to cope in response to pain may require assessments of what coping responses are used during painful situations (Ho, 2019). The extent to which studies have appropriately considered the timing of coping in response to pain in the selection and interpretation of coping measures has not yet been examined.

In sum, our ability to perform research and effectively communicate about how youth cope with pain is limited by: (i) the vastness of the literature; (ii) the use of unclear and inconsistent terminologies and categorizations of coping responses; and (iii) inconsistencies in how coping responses are measured across studies. **Figure 1** illustrates content, organizational, and definitional challenges by comparing the subscales of the pain response inventory (PRI) (Walker et al., 1997) and the pain coping inventory (PCQ) (Reid et al., 1998), the two most frequently cited (Web of Science citation count: PRI = 197 articles and PCQ = 164) and well-established measures of pain-related coping in youth (Blount et al., 2008). The extent to which these challenges apply in the field of pediatric chronic pain remains unclear. Thus, a scoping review was conducted to systematically map the measurement and conceptualization of coping in populations with pediatric chronic pain. Specific research questions were as follows:

1) *In whom, what, why, and how are coping responses measured*? Specific characteristics of interest were: (i) The "who"—the sample characteristics (i.e., gender, age, ethnicity, and pain conditions) and study characteristics (i.e., country, year); (ii) the "what"—research methodologies employed (i.e., study design, type of data); (iii) the "why"—applications of theoretical frameworks by study authors; and (iv) the "how"—types of coping measures used and their characteristics (i.e., the name of the measure, purpose, response options, internal reliability of subscales, assessment of proactive vs. reactive coping, parent or youth report, the types of coping responses captured by the subscales, and coping structure used).
2) *How have coping responses been conceptualized in the literature*? This was examined based on: (i) the operationalized definitions and/or descriptions of coping responses used; (ii) the clarity with which authors defined or described the coping responses measured; (iii) the terminology used to classify coping responses within higher- and lower-order categories (e.g., coping strategies vs. coping styles); and (iv) the extent to which the selection of coping measures was grounded in a clear theoretical or empirical rationale (herein "concept guided").
3) *Are coping responses measured and conceptualized consistently relative to their intended purpose*? This review evaluated the extent to which: (i) the appropriate validation studies were cited; (ii) measures were used consistently (i.e., measurement consistency); and (iii) concepts were described consistently with what the original scale purports to measure (i.e., conceptual consistency).

METHODS

This scoping review was developed using the methodological framework put forward by Arksey and O'Malley (2005) and further refined by the Joanna Briggs Institute (Peters et al., 2019) and was written in accordance with the PRISMA-ScR reporting guidelines (Tricco et al., 2018). A scoping review approach allows for a descriptive account of the available information in a particular field and is ideal for broadly defined research questions and heterogeneous sources of evidence, which can then guide future narrower systematic reviews (Sucharew, 2019). The literature reviewed was derived from a larger review of positive psychological factors in the context of pediatric chronic/recurrent pain (see **Supplementary Materials**); only peer-reviewed articles pertaining to youth pain-related coping were included in this study. The study protocol and materials are available *via* Open Science Framework (https://osf.io/xvn2a/?view_only=eff04e0c0b9649be89d403b10e9ff082).

Eligibility Criteria

Population

Studies that examined a pediatric population (youth 0–18 years of age). Studies with participants above the age of 18 were retained only if they included a pediatric sample that extended into young adulthood with a maximum age of 21 (Hardin and Hackell, 2017). Studies that examined adult, both pediatric and adult (above 21 years of age), and/or animal populations were excluded.

Concept

The main concept of interest is youth pain-related coping, which includes the use of coping responses by youth to manage pain

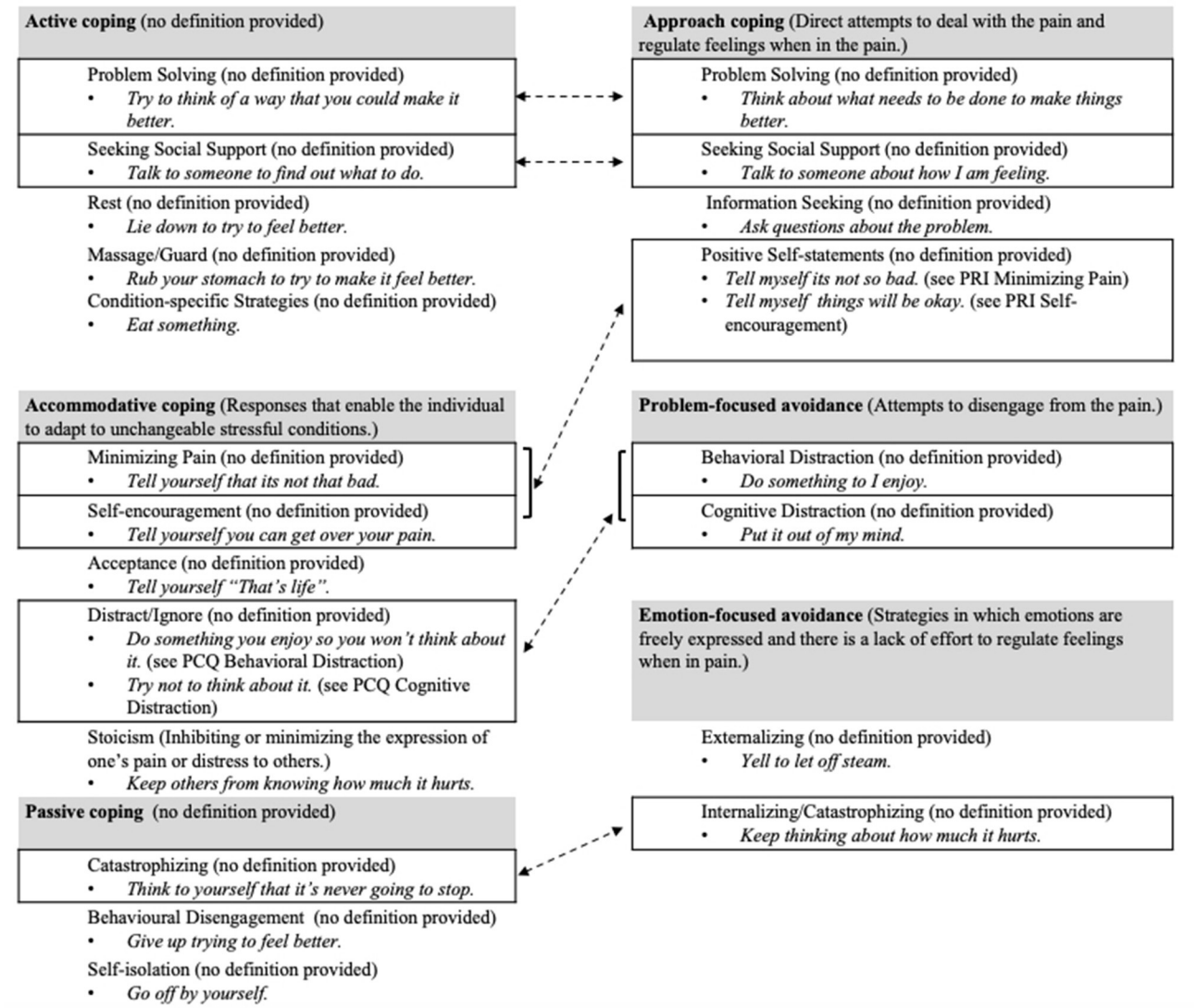

FIGURE 1 | Using the PRI (Walker et al., 1997) and the PCQ (Reid et al., 1998), this figure illustrates the inconsistencies in content (e.g., items, subscales) and organization (categorizations of coping responses) as well as the use of definitions across measures. The gray boxes indicate the higher-order factors, and the lined boxes indicate subscales that appear conceptually similar across measures. Example items are provided in italics. Dotted lines connect subscales that appear to be similar on each measure and demonstrate differences in their categorization within higher-order categories. Of note, the PRI and PCQ assess three different higher-order factors. Some lower-order coping responses either sound the same (e.g., "problem-solving") or consist of similar items (e.g., "positive self-statements", "minimizing pain", and "self-encouragement"), but the extent to which these seemingly similar subscales should be conceptualized the same way is unclear due to the lack of clear definitions and inconsistent categorizations within higher-order factors [i.e., "problem solving" as "active coping" (PRI) vs. "approach coping" (PCQ); "seeking social support" as "active coping" (PRI) vs. "approach coping" (PCQ); "catastrophizing" as "passive coping" (PRI) vs. "emotion-focused avoidance" (PCQ); "positive self-statements" (conceptually similar to "self-encouragement" and "minimizing pain" combined) as "accommodative coping" (PRI) vs. "approach coping" (PCQ); and "distract/ignore" (conceptually similar to "cognitive distraction" and "behavioral distraction" combined) as "accommodative coping" (PRI) vs. "problem-focused avoidance" (PCQ)].

or pain-related stressors. Pain catastrophizing in the context of coping (i.e., the tendency to magnify the threat associated with pain and to forecast negative outcomes) (Quartana et al., 2009) was excluded because it has been well-documented as a non-adaptive cognitive–affective response to chronic pain that has been reviewed on its own (Vervoort et al., 2006; Leung, 2012). The exclusion of catastrophizing is also consistent with previous reviews, which have conceptualized catastrophizing as a unique psychological factor rather than a type of coping response (Sharma et al., 2020).

Context

Studies including youth with chronic pain conditions (i.e., persistent and/or recurrent pain) were included. Examples

of included pediatric chronic pain conditions are sickle cell disease, juvenile arthritis, complex regional pain syndrome, and abdominal pain, among others. In line with other pediatric chronic pain reviews (Lewandowski et al., 2010; King et al., 2011; Eccleston et al., 2014), studies that exclusively examined a diverse pediatric chronic illness group (e.g., non-chronic pain disorders such as diabetes and asthma) or cancer-related pain were excluded. Finally, studies that examined "recurrent pain" in healthy samples drawn from community or school samples of children were excluded.

Types of Evidence Sources

English language articles containing original data published in scientific journals were included. All non-peer-reviewed publications (e.g., dissertations), reviews, commentaries, editorials, and chapters were excluded. Non-English language studies were excluded due to feasibility (i.e., the lack of translators on the research team).

Stage One: Database Searches and Screenings

Electronic literature searches were conducted by a lead researcher (RMT or ANN) in MEDLINE (PubMed) and PsycINFO in three phases between March 26, 2015 and August 20, 2020. Databases were selected in accordance with Cochrane Review recommendations for reviews in the fields of medicine and psychology (Higgins and Green, 2008). All citations were uploaded into EndNote, and the titles and abstracts were screened for inclusion prior to conducting full-text screenings and extractions. Upon completion of a training period (i.e., 100 articles), title/abstract screenings were completed by two reviewers within EndNote, including undergraduate research assistants and/or a lead researcher (RMT or ANN), and inter-rater reliability was calculated based on percentage agreements.

The first two phases (March 26, 2015; September 4, 2018) employed double coding for 20% of all articles identified and had inter-rater reliability of 89% at the abstract level. In the third phase (August 20, 2020), all title/abstract screenings were double-coded and had an inter-rater agreement of 98.3%. Articles were then screened at the full-text level by two reviewers. All included articles were checked by the lead researcher (ANN) prior to inclusion, and any discrepancies were resolved through discussion.

Stage Two: Data Extraction

Information was extracted on study details (e.g., the year of study, country), participant characteristics (e.g., gender, age), and methodological characteristics (e.g., study designs, types of data collected). In addition, characteristics of the measures used (e.g., number of items, the content/structure, timing of coping assessed) and the terminologies and conceptualizations of the coping responses assessed (e.g., theoretical frameworks, definitions/descriptions, categorization of coping types) were extracted from the included studies. The appropriateness of the measure selected in each included study of the review was evaluated based on whether the concepts assessed were in alignment with the stated research objectives/hypotheses and/or theoretical framework of the authors (i.e., a concept-guided approach), which is a recommended approach for developing and validating knowledge (Coster, 2013; Boateng et al., 2018). In order to map the current published research literature, study authors were not contacted for missing information.

A standardized data extraction form was piloted on a random sample of 10 included articles and modified as required based on feedback from other reviewers. A final revision and the pilot of the extraction spreadsheet were completed on July 10, 2020. All extractions were completed by the primary researcher (ANN) and were reviewed and verified by a second reviewer (an undergraduate research assistant). The number of disagreements between reviewers per article ranged from 0 to 4 out of 45 decisions made per article ($Mdn = 1.00$, $M = 0.99$, $SD = 1.08$) and was resolved through discussion between the two reviewers or adjudication by a third researcher (CMM) as needed. The following subsections will outline key areas of consideration and procedures used in the data extraction process.

Stage Three: Comparisons With Original Scale Development Studies

Scale development/validation studies were identified from the database search ($n = 4$) as well as *via* snowball searching (i.e., using the included studies as a starting point and pursuing references for assessment tools cited by the study authors; $n = 20$). Appropriateness of the measure selected, as well as the accuracy and clarity of the use of the coping measures and concepts, was determined by examining information from the scale development studies. An "overview document" (see **Supplementary Materials** for a sample) of the scale development studies was developed for each measure that included information on the participant characteristics, theoretical background, measure characteristics, and definitions of coping responses. Each overview document was checked by a lead researcher (ANN) prior to being used.

For studies that used a questionnaire with published information on its psychometric properties, comparisons between each included study and the respective scale development study/studies were completed to determine measurement consistency, conceptual consistency, and conceptual clarity (see **Table 1** for more information). For conceptual clarity and consistency, the use of both definitions (i.e., a statement that describes the meaning of a concept) and examples (i.e., describing a specific behavior or naming subtypes of coping responses) were evaluated because they are both important elements of a well-established concept (Gerring, 1999) and have been used to explain coping in previous research (Garcia, 2010). The rating scales employed were adapted from recommendations by the Cochrane Review Groups to maximize the simplicity and clarity of the coding schemes (Lundh and Gøtzsche, 2008). These rating scales are not an assessment of methodological quality but rather highlight when our ability to directly interpret, consolidate, and/or compare research findings may be limited. All three rating scales were evaluated by two reviewers (an undergraduate research assistant and ANN), and agreement was assessed using percent agreement.

TABLE 1 | Operationalization of measurement consistency, conceptual consistency, and conceptual clarity (adapted from the Cochrane Review Groups recommendations) (Lundh and Gøtzsche, 2008).

Domain	Description	[a]Ratings
Measurement consistency	The extent to which the author appropriately used the measure selected.	**High:** The use of the coping measure employed was fully consistent with the scale development study. **Low:** There is at least one discrepancy in how the coping measure was used relative to the scale development study. **Unclear:** Unable to evaluate measurement consistency due to insufficient information (e.g., missing more than two relevant characteristics)
Conceptual consistency	The extent to which the [b]descriptors of coping used in a particular study are consistent with the descriptors that were proposed by the scale development study.	**High:** The descriptors of all coping constructs were fully consistent with the scale development study. **Low:** There is at least one discrepancy in how coping was described between the study and scale development study. **Unclear:** Unable to evaluate conceptual consistency due to insufficient information (e.g., there are no descriptors of the coping constructs in the study or the corresponding scale development study).
Conceptual clarity	The extent to which coping constructs were defined or described using examples.	**High:** All relevant coping terms were clearly defined AND potential applications of the coping construct were provided. **Moderate:** All relevant coping terms were either clearly defined OR potential applications of the coping construct were provided. **Low:** Some relevant coping terms were defined and/or potential applications were provided. **Very Low:** No relevant coping terms were defined, and no potential applications were provided.

[a] *Ratings are used to indicate the presence of discrepancies that may impact our ability to directly interpret, consolidate, and/or compare research findings. These ratings do not reflect an assessment of the quality of the research.*
[b] *Descriptors = refer to the use of definitions and/or examples.*

High agreement was found between reviewers for ratings of conceptual clarity (91.2%), measurement consistency (88.4%), and conceptual consistency (89.5%).

Data Synthesis

The results were summarized using a combination of descriptive numerical and narrative summaries in accordance with the research questions. All numerical descriptive statistics (means and proportions) were conducted using SPSS version 26. Weighted means were used to summarize participant characteristics (i.e., age and gender) to account for sample size differences.

RESULTS

As shown in **Figure 2**, database searching identified 37,172 potential articles encompassing a wider range of positive psychological factors associated with adjustment in youth with chronic pain. After the removal of duplicates and articles that did not meet the inclusion criteria for this review at the title/abstract level of screening, 1,159 articles remained. Of these articles, 129 articles met the eligibility criteria for full-text review. Scale development/validation studies identified from the database search ($n = 4$) were reviewed separately and used as a reference to evaluate the measurement and conceptual consistency of research studies in the field. Thus, 125 peer-reviewed articles related to coping and pediatric chronic pain were included in this review (see **Supplementary Table 1** for a list of included studies) (Moher et al., 2009).

Aim 1: The Who, What, Why, and How of Measuring Pain-Related Coping in Youth

Table 2 provides an overview of the included studies; see **Supplementary Table 2** for the participant, study, and methodological characteristics of each individual study. Studies were published between 1991 and 2020. The vast majority of studies (88%) were conducted in the USA ($n = 84$; 67.2%) and countries in Western Europe ($n = 26$; 20.8%).

Who: Sample Characteristics

The articles included 13,474 youth who were predominately female (*weighted mean* = 65%, *weighted SD* =0.12%). The participants ranged from 3 to 20 years old (*weighted mean* = 12.8 years, *weighted SD* = 2.25).

The participants presented with a wide range of pain conditions (**Table 2**), with the most frequent being sickle cell disease (SCD; 33.6%) and abdominal pain (16.0%). A heterogenous chronic pain sample was used in 21.6% of studies. Only 78 studies (62.4%) reported on the ethnic and/or racial composition of the participants. When reported, the participants were predominately White ($n = 41$; 54.5%). The only exception was studies on sickle cell disease, wherein the majority of the participants were African American or Black ($n = 32$; 41.0% of studies that reported on ethnicity/race).

What: Study Design and Data Collection Method

The majority of included studies examined youth pain-related coping using a cross-sectional ($n = 85$; 67.2%) approach. Of the studies that measured coping as an outcome of a cognitive, behavioral, or physical intervention for youth with chronic pain ($n = 34$), 51.5% ($n = 17$) had a control or comparison group. Additional 16 studies used a control or comparison group without examining the effects of an intervention.

Why: Theoretical Frameworks

Just over a third of included studies identified a theoretical conceptualization of coping ($n = 45$; 36.%). Of these 45 studies, the majority ($n = 29$) referred to theoretical frameworks of

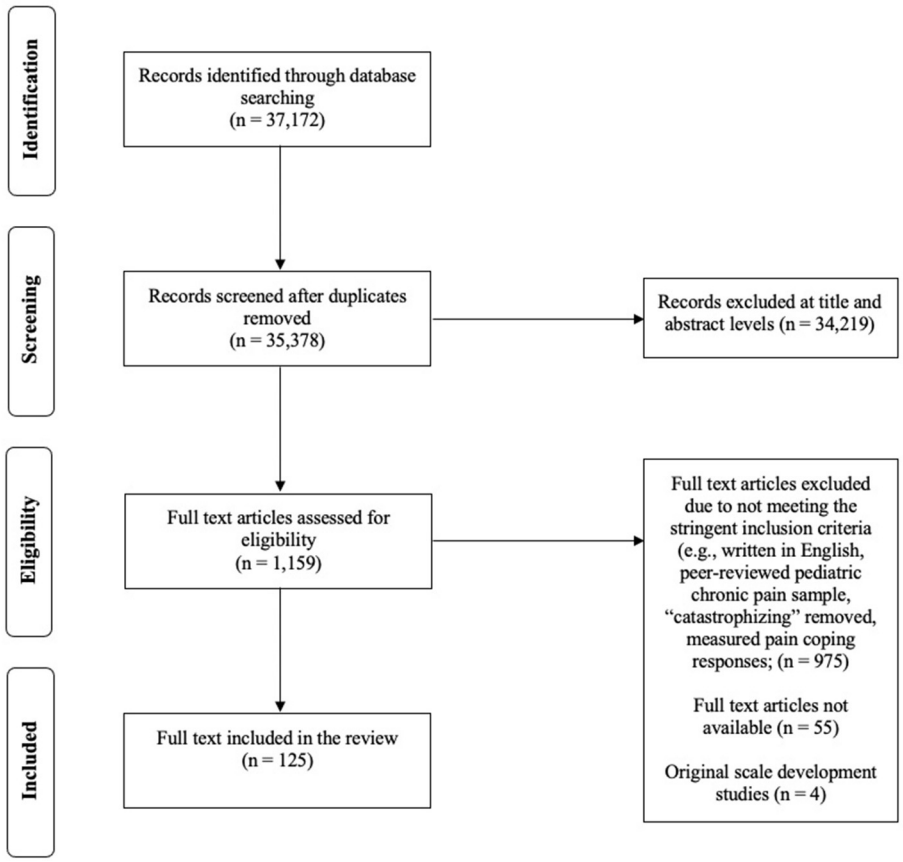

FIGURE 2 | Included a study flow chart following PRISMA guidelines.

the coping process (e.g., the biopsychosocial model, risk-resistance model, transactional stress, and coping model) as opposed to conceptualizations of specific coping responses. Five theoretical models conceptualizing specific coping responses were identified within 16 included articles. The most common theory cited was the cognitive-appraisal theory of Lazarus and Folkman ($n = 8$), which suggests that coping responses can be categorized as being "problem-focused" (i.e., coping that is aimed at managing or altering the problem causing the distress) or "emotion-focused" (i.e., coping that is directed at regulating emotional responses to the problem) (Lazarus and Folkman, 1984). Following a similar dichotomous structure, the approach- vs. avoidance framework purposed by Roth and Cohen was used in one study, conceptualizing coping responses as cognitive and emotional strategies that are oriented either toward or away from the stressor, respectively (Roth and Cohen, 1986). Alternatively, five studies referred to the control-based model as a more complex, hierarchical classification system that distinguishes between voluntary vs. involuntary (i.e., unconscious vs. intentional/volitional responses) and engagement vs. disengagement processes (i.e., oriented toward or away from the stressor) (Compas et al., 1991). Within the control-based model, coping is characterized as voluntary responses that can be distinguished by engagement vs. disengagement responses. Engagement responses are further distinguished by their goals: "primary-control-engagement" coping involves attempts to alter emotions (e.g., emotion regulation) or the stressor itself (e.g., problem-solving), whereas "secondary-control-engagement" coping includes efforts to become accustomed to the stressor by modifying cognitions or regulating attention (e.g., acceptance). In contrast, "disengagement" coping involves both removing oneself from the stressor and removing oneself from his or her emotions related to the stressor.

As an alternative to the abovementioned hierarchical classifications of coping responses, one study referred to the typology of Walker and colleagues to explain meaningful patterns of coping responses that are used by youth with chronic abdominal pain and their association with different levels of emotional and physical distress (i.e., coping profiles) (Walker et al., 2008). For example, using cluster analytic techniques with the PRI scales, patients identified as "engaged copers" were characterized by using high levels of "distraction" and "social support-seeking" and associations with lower levels of depressive symptoms and disability, representing an overall adaptive pattern of coping responses. As such, coping profiles

TABLE 2 | Overview of characteristics of the included studies ($N = 125$).

Characteristics		No. of studies (%)	Characteristics (cont.)		No. of studies (%)
Geographic region	USA	84 (67.2)	*Predominant ethnicity/race (>50% of sample)*	Caucasian	41 (32.8)
	[a]Europe	26 (20.8)		[d]African American	32 (27.2)
	Canada	7 (5.6)		[e]Other	3 (2.4)
	Australia	3 (2.4)		Not reported	47 (37.6)
	[b]Rest of the world	5 (4.0)			
			Methods	Quantitative	108 (84.6)
Year	1991–1995	12 (9.6)		Qualitative	13 (10.4)
	1996–2000	14 (11.2)		Mixed	4 (3.2)
	2001–2005	19 (15.2)			
	2006–2010	28 (22.4)	*Informant*	Child	93 (75.2)
	2011–2015	24 (19.2)		Parent	5 (4.0)
	2016–2020	28 (22.4)		Multi-informant	21 (16.8)
				Behavioral	1 (0.8)
Sample size	<50	52 (41.6)		Not specified	5 (4.0)
	50–100	33 (26.4)			
	101–500	37 (29.6)	*Design*	Case study/series	2 (1.6)
	501–1,000	3 (2.4)		Cross-sectional	84 (67.2)
				Longitudinal	24 (19.2)
Age-range	Children	10 (8.0)		RCT	12 (9.6)
	(<12 years)			[f]Other	3 (2.4)
	Adolescent	28 (22.4)	*Control group*		
	(>12 years)			Chronic pain	23 (18.4)
	Both	85 (68.0)		Non-chronic pain	11 (8.8)
	Not specified	2 (1.6)		None	91 (72.8)
Pain condition	SCD	42 (33.6)	*Intervention used*	Yes	34 (27.2)
	Abdominal	20 (16.0)		No	91 (72.8)
	Arthritis	15 (12.0)			
	Headache	14 (11.2)	*Timing of coping*	Reactive	45 (36.0)
	Fibromyalgia	4 (3.2)		Proactive	6 (4.8)
	Multiple	27 (21.6)		Both	5 (4.0)
	[c]Other	3 (2.4)		Not reported	69 (55.2)

SCD, sickle cell disease; RCT, randomized control trail.

[a]*Countries in Europe included The Netherlands (n = 7), Germany (n = 6), UK (n = 5), Sweden (n = 2), Spain (n = 2), Denmark (n = 2), Italy (n = 1), Hungary (n = 1), and Norway (n = 1).*

[b]*Other countries included Brazil (n = 2), India (n = 1), Jamaica (n = 1), and Lebanon (n = 1).*

[c]*Other pain conditions include complex regional pain syndrome (n = 1;0.8), non-cardiac chest pain (n = 1;0.8), and systemic lupus erythematosus (n = 1;0.8).*

[d]*All 32 studies that focused predominantly on an African American sample were concerned with a diagnosis of sickle cell disease.*

[e]*Other predominant ethnic/racial groups include Hispanic (n = 1), East Indian (n = 1), and Lebanese/Palestinian (n = 1).*

[f]*Other designs include non-RCT pre-post design (n = 1), retrospective chart review (n = 1), and ethnography (n = 1).*

were conceptualized to retain information about the specific coping responses while capturing the relationship between coping responses and outcomes.

How: Types of Measures

The vast majority of studies employed only quantitative measures ($n = 108$; 86.4%; see **Table 2**), primarily questionnaires ($n = 105$; see **Table 3**). Other questionnaires that did not have a peer-reviewed and/or English development/validation study include: the child and adolescent coping inventory ($n = 1$) (Harris et al., 1991); sick-role adoption index ($n = 1$) (Barbarin et al., 1999); Utrecht coping list ($n = 1$) (Westendorp et al., 2017); stress and coping questionnaire for children and adolescents ($n = 1$) and other coping scales derived by the authors of the included study ($n = 3$).

Other quantitative measures included structured daily diaries ($n = 6$), structured interviews using closed-end questions ($n = 2$), observations of child behavior ($n = 1$), or an unspecified measure ($n = 1$). Of the quantitative studies, five studies (4.6%) reported using multiple quantitative measures (e.g., a questionnaire and the daily pain and activity diary). Qualitative measures for assessing coping responses included semi-structured interviews with open-ended questions ($n = 9$), unstructured interviews ($n = 2$), drawings ($n = 1$), written narrative tasks ($n = 1$), and clinician judgements based on a retrospective chart review of clinical notes and patient interactions ($n = 1$).

TABLE 3 | A list of the questionnaires identified with clear development studies available, reporting psychometrics, ordered by most to least frequently used measure.

Measure/No. of items	Population (P)/Informant (I)	Timing	Subscales/Factors (Cronbach's alpha, if provided)	Response options/Scoring	[a]No. of studies	[b]Citation accuracy (%)
Pain Coping Questionnaire (PCQ); 39 items (Reid et al., 1998) Translations: Danish; 36 items (Thastum et al., 1999) Dutch; 39 items (Bandell-Hoekstra et al., 2002) Catalan; 39 items (Huguet et al., 2009) Finnish; 39 items (Marttinen et al., 2018)	P: Healthy youth and youth with recurrent (headache, arthritis) pain, ages 7–17 I: Youth and parent form	Reactive	Approach (0.89) Information seeking (0.79) Problem solving (0.86) Seeking social support (0.86) Positive self-statements (0.82) Problem-focused avoidance (0.86) Behavioral distraction (0.78) Cognitive distraction (0.85) Emotion-focused avoidance (0.85) Externalizing (0.81) Internalizing/Catastrophizing (0.82)	Five-point scale (1 = Never; 5 = Very often)/Factor and subscale scores are derived by computing the means across items or subscales, respectively.	28	96.4
Coping Strategies Questionnaire for SCD (CSQ-SCD); 80 items (Gil et al., 1991) Alternate versions: Original CSQ; 50 items (Rosenstiel and Keefe, 1983)	P: Youth with SCD, ages 7–17 I: Youth and parent form	Reactive	Coping attempts Diverting attention (0.72) Reinterpret pain (0.67) Ignoring pain sensations (0.70) Calming self-statements (0.72) Increased behavioral activity (0.55) Negative thinking Catastrophizing (0.76) Fear self-statements (0.70) Anger self-statements (0.67) Isolation (0.69) Passive adherence Resting (0.72) Taking fluids (0.89) Praying and hoping (0.67) Heat, cold, massage (0.66)	Seven-point scale (Never to Always)/Factor and subscale scores are derived by computing the means across items or subscales, respectively.	21	66.7
Pain Response Inventory (PRI); 60 items (Walker et al., 1997)	P: School-aged sample and youth with recurrent abdominal pain; ages ranged from 8 to 23 I: Youth form	Reactive	Active coping Problem solving Seeking social support Rest Massage/Guard Condition-specific strategies Passive coping Behavior disengagement Self-isolation Catastrophizing Accommodative coping Acceptance Minimizing pain Distract/Ignoring pain Stoicism	Five-point scale (Never to Always)/Factor and subscale scores are derived by computing the means across items or subscales, respectively.	19	100
Pediatric Pain Coping Inventory (PPCI) 41 items (Varni et al., 1996) Alternate version: PPCI–Revised (German) (Hechler et al., 2008)	P: Children and adolescents with musculoskeletal pain, ages 5–16 I: Child, adolescent and parent forms	Reactive	Cognitive self-instruction (0.77) Seek social support (0.74) Strive to rest and be alone (0.73) Cognitive refocusing (0.68) Problem-solving self-efficacy (0.67)	Three-point scale (0 = Not at all, 3 = Often)/Subscale scores are derived by computing the means across items.	7	100
KidCope Adolescent version; 10 items (Spirito et al., 1988) Child version; 15 items (Spirito et al., 1991)	Adolescent: P: High school sample and chronic pain patients, ages 12–18 Child: P: Children, ages 9–13 I: Youth	Reactive	Problem-solving Distraction Social support Social withdrawal Cognitive restructuring Self-criticism Blaming others Emotional regulation Wishful thinking Resignation	Adolescent: Four-point scale (Not at all to Almost all the time) Child: Yes or no Scoring was not specified	6	16.7

(Continued)

TABLE 3 | Continued

Measure/No. of items	Population (P)/Informant (I)	Timing	Subscales/Factors (Cronbach's alpha, if provided)	Response options/Scoring	[a]No. of studies	[b]Citation accuracy (%)
Response to Stress Questionnaire (RSQ); 57 items (Connor-Smith et al., 2000)	P: College and high school students and adolescents with recurrent abdominal pain; ages across samples ranged from 12 to 19 I: Youth and parent forms	Reactive	Primary control engagement Problem solving Emotional regulation Emotional expression Secondary control engagement Positive thinking Cognitive restructuring Acceptance Distraction Disengagement coping Denial Avoidance Wishful thinking	Four-point scale (1 = Not at all, 4 = A lot)/Scoring was not specified	5	80
Coping Strategies Inventory (CSI); 72 items (Tobin et al., 1984, 1989)	P: College students I: Self-report	Reactive	Engagement (0.90) Problem-focused engagement (0.87) Problem-solving (0.82) Cognitive restructuring (0.83) Emotion-focused engagement (0.92) Social support (0.89) Express emotions (0.89) Disengagement (0.89) Problem-focused disengagement (0.81) Problem-avoidance (0.72) Wishful thinking (0.78) Emotion-focused disengagement (0.90) Self-criticism (0.81) Social withdrawal (0.94)	Five-point Likert scale (Not at all to Very much)/Factor and subscale scores are derived by computing the sum across items or subscales, respectively.	4	75
The Children's Coping Strategies Checklist (CCSC);45 items (Ayers et al., 1996)	P: School-aged children, ages 8-13 I: Youth	Reactive	Active coping Cognitive decision making Direct problem solving Seeking understanding Positive cognitive restructuring Avoidance coping Cognitive avoidance Avoidant action Distraction Distracting action Physical release of emotion Support Emotion-focused support Problem-focused support	Four-point scale (Never to Most of the time)/Factor and subscale scores are derived by computing the means across items or subscales, respectively.	2	100
Adolescent Coping Style and Behavior (A-COPE); 54 items (Patterson and McCubbin, 1987)	P: Community samples of youth (non-chronic pain), ages 11–18 I: Youth	Reactive	Venting Feelings (0.75) Seeking Diversions (0.75) Developing self-reliance and Optimism (0.69) Developing social support (0.75) Solving family problems (0.75) Avoiding problems (0.71) Seeking spiritual support (0.72) Investing in close friends (0.76) Seeking professional support (0.50) Engaging in demanding activity (0.67) Being humorous (0.72) Relaxing (0.60)	Five-point scale (Never to Most of the time)/Factor and subscale scores are derived by computing the sum across items or subscales, respectively.	1	100
Children's Headache Assessment Scale (CHAS) Versions: Original; 30 items (Budd and Kedesdy, 1989) Revised; 44 items (Budd et al., 1994)	Original: P: Youth with headaches, ages 7-16 Revised: P: Youth with headaches, ages 6-16 I: Parent form	Reactive	Original: Coping response (0.64) Revised: Physical Antecedents and Quiet Coping	Six-point scale (0 = Never, 6 = Always)/Subscale scores are derived by computing the means across items.	1	100

(Continued)

TABLE 3 | Continued

Measure/No. of items	Population (P)/Informant (I)	Timing	Subscales/Factors (Cronbach's alpha, if provided)	Response options/Scoring	[a]No. of studies	[b]Citation accuracy (%)
How I Coped Under Pressure Scale (HICUPS); 45 items (Ayers et al., 1996)	P: School sample, ages 9-13 I: Youth	Reactive	Active coping Cognitive decision-making (0.71) Direct problem-solving (0.71) Seeking understanding (0.74) Positive cognitive restructuring (0.62) Avoidance coping Cognitive avoidance (0.61) Avoidant action (0.64) Distraction Distracting action (0.65) Physical release of emotion (0.65) Support Emotion-focused support (0.60) Problem-focused support (0.57)	Four-point scale (Not at all to A lot)/Factor and subscale scores are derived by computing the sum across items or subscales, respectively.	1	100
The Schoolagers Coping Strategies Inventory (SCSI); 30 items (Ryan-Wenger, 1990)	P: Community sample (10% with a chronic health condition, e.g., asthma, allergies), ages 8-12 I: Youth	Reactive	Social support Avoidant Emotional Distracting Cognitive Aggressive motor Physical exercise Isolating Aggressive verbal Relaxation Habitual Spiritual Other	Three-point scale (frequency)/Scoring was not specified	1	100
Ways of Coping Checklist; 64 items (Folkman and Lazarus, 1980)	P: Adults, ages 45-64 I: Self-report	Unclear	Problem-focused coping (0.80) Emotion focused coping (0.81)	Yes or No/Scoring was not specified	1	100
Religious Coping (R-COPE); 105 items (Pargament et al., 2000)	P: College sample who encountered a negative life event, ages 18-38; hospitalized adults with a moderately severe medical illness, ages 55-97 I: Self-report	Not reported	Benevolent religious reappraisal (0.91) Punishing God reappraisal (0.92) Demonic reappraisal (0.90) Reappraisal of God's power (0.78) Collaborative religious coping (0.89) Active religious surrender (0.92) Passive religious deferral (0.83) Pleading for direct intercession (0.84) Religious focus (0.84) Purification/forgiveness (0.93) Spiritual connection (0.81) Spiritual discontent (0.88) Marking religious boundaries (0.61) Seeking support from clergy (0.90) Religious helping (0.90) Interpersonal religious discontent (0.82) Religious direction/conversion (0.94)	Four-point scale (Not at all to A great deal)/Subscale scores are derived by computing the means across items.	1	100

The scale characteristics and reliability estimates provided are based on the original scale development study.

[a]*A list of each individual study and the study characteristics can be found in* ***Supplementary Table 2****.*

[b]*Citation accuracy refers to the extent to which authors of the included studies cited the correct scale development study.*

Overall, 75.2% of included studies indicated using child self-report ($n = 93$), and the remaining studies (16.8%) used multiple informants ($n = 20$ child and parent; $n = 1$ child, parent, siblings, and clinicians), parent-reported responses ($n = 5$; 4%), behavioral data ($n = 1$;0.8%), or did not specify ($n = 5$; 4%). Of the 56 studies that explicitly specified the timing of the coping responses assessed in relation to pain, 80.3% indicated measuring reactive coping responses ($n = 45$) and with the remainder reporting on proactive coping ($n = 6$) or both ($n = 5$). Current measures/approaches used to assess proactive coping consisted open-ended interviews ($n = 5$) and daily diary tools ($n = 6$; see **Supplementary Table 3**).

Aim 2: Conceptualizations of Coping Responses

A total of 168 coping responses were identified across all included studies (see **Supplementary Table 4** for a full list). The following sections summarize: (i) the terminology used for classifying coping responses; (ii) the extent to which coping responses were clearly defined and/or described (conceptual clarity); and (iii) the extent to which authors used a concept-guided approach to measurement selection.

Terminology for Classifying Coping Responses

Across the 125 included studies, 21.6% ($n = 27$) reported on only "coping strategies". Of these studies, the term "coping strategies" was occasionally interchanged with "coping behaviors" or "coping skills". Furthermore, 38.4% ($n = 48/125$) of the included studies reported on higher-order categories of coping only, of which 75% ($n = 36/48$) did not provide a label for the higher-order categorization. Only seven of these 48 studies (14.5%) explicitly referred to the higher-order categories as "coping styles". In addition, the term "coping style" was used interchangeably with the following terms: "coping patterns", "coping approaches", "dimensions of coping", "coping subtypes" "coping subthemes", "domains of coping", "coping potential", or "coping response". In addition, 6.4% ($n = 8/125$) of included studies used a term other than coping strategy or style to categorize coping responses (e.g., coping mechanism, coping behavior, coping skill) and 17.6% ($n = 22/125$) reported on multiple levels of coping responses (e.g., coping strategies and coping styles). Sixteen percent of the included studies ($n = 20/125$) did not provide a specific term to classify the coping responses assessed.

Conceptual Clarity

The majority of the 125 studies were rated as "very low" ($n = 40$; 32.0%) or "low" ($n = 19$; 15.2%) for conceptual clarity. Forty-seven studies (37.6%) were rated "moderate" because they provided either a definition or specific examples of coping behaviors for each coping response, but not both. As such, it was less common for studies to demonstrate "high" conceptual clarity by including both examples and a specific statement defining the coping responses used ($n = 19$; 15.2%).

Measurement Selection

A concept-guided approach to measurement selection was used in only 33.3% of included studies ($n = 43$) as demonstrated by providing a clear and consistent rationale for the chosen measurement tool. Of these 43 studies, 20.9% ($n = 9$) clearly mapped the measurement tool selected to a theoretical framework of coping responses (as described in Aim 2 above). The remaining 79.1% of studies ($n = 34$) were rated as concept guided on the basis that the measure employed clearly mapped onto the study questions, hypotheses, and/or objectives.

Aim 3: Evaluations of Measurement and Conceptual Consistency (Questionnaires Only)

The majority of studies (76.0%; $n = 95$) assessed coping responses using questionnaires with known psychometric properties (**Table 3**). Measurement and conceptual consistency were assessed for studies that used questionnaires with known psychometric properties because of their widespread use and the availability of previous scale development or validation studies for comparison. As such, **Table 4** presents the list of coping responses, corresponding descriptors, and empirically derived classifications assessed by questionnaires with known psychometric properties.

Primary, Secondary, and Tertiary Factor Levels

There were inconsistencies in the terminology used to classify coping responses. For example, higher-order coping responses were referred to by various terms, such as "coping styles", "coping patterns", or "coping approaches" (see Section Terminology for Classifying Coping Responses for other examples). Thus, exploratory and/or confirmatory factor analysis results from the original scale development studies used represent the hierarchical classifications of coping responses. The terms "primary", "secondary", and "tertiary" correspond to first-, second-, and third-order-factor levels (**Table 4**). Primary coping responses are made up of related items on a questionnaire. Within multidimensional scales, primary coping responses load onto secondary coping responses; and secondary coping responses load onto tertiary coping responses. For example, the PCQ (Reid et al., 1998) consists of eight primary coping responses (problem-solving, information-seeking, seeking social support, positive self-statements, behavioral distraction, cognitive distraction, externalizing, and internalizing) that each loads onto one of the three secondary factors (approach, problem-focused avoidance, and emotion-focused avoidance); **Table 3** presents the factor structure of each questionnaire. In summary, there were 86 primary coping responses (e.g., acceptance, cognitive refocusing), 17 secondary coping responses (e.g., active coping, passive coping), and two tertiary coping responses (i.e., engagement, disengagement). Two coping responses were categorized into multiple levels, depending on the measure used (see **Table 4**): "avoidance" and "disengagement".

Overall Measurement Consistency

In sum, 55.8% ($n = 53$) of studies were rated "low", and 23.2% ($n = 22$) were rated "high" for measurement consistency. Measurement consistency was rated as "unclear" for 21.1% of studies ($n = 20$) due to a lack of information about the scale characteristics (i.e., studies that were missing more than two relevant scale characteristics). The most common characteristics missing from articles included the number of items, response options, scoring procedure, and/or the timing of coping strategy use in relation to the pain onset (e.g., proactive vs. reactive).

Overall Conceptual Consistency

In terms of conceptual consistency, 35.8% ($n = 34$) of studies were rated "high" (i.e., no discrepancies), and 33.7% ($n = 32$) of studies were rated "low" (i.e., one or more discrepancies). The remaining 30.5% ($n = 29$) of studies were rated as "unclear" because the authors did not provide any descriptors to allow for comparison.

TABLE 4 | A list of coping responses (in alphabetical order) and their corresponding descriptors (if available) conceptualized by questionnaires with a clear scale development study identified (see **Table 3** for the list of questionnaires).

Coping concept	Measure	Descriptor(s)	[a]Factor level
Acceptance	PRI	No definition was provided. Conceptualized as a coping strategy under "accommodative coping".	Primary
	RSQ	No definition was provided. Conceptualized as a coping strategy under "secondary control coping".	
Accommodative coping	PRI	"Strategies, such as positive reappraisal and acceptance, that enable the individual to adapt to unchangeable stressful conditions" (Walker et al., 1997, p. 392). Includes acceptance, self-encouragement, minimizing pain, distract/ignoring pain, and stoicism.	Secondary
Active coping	CCSC/ HICUPS	"Strategies in which the child is focused on the stressful event, either to change the situation or to think about it more positively" (Ayers et al., 1996, p. 929). Includes cognitive decision making, direct problem-solving, seeking understanding, and positive cognitive restructuring.	Secondary
	PRI	No definition was provided. Consists of problem-solving, social support, rest, massage/guard, and condition-specific strategies.	
Active religious surrender	RCOPE	"An active giving up of control to God in coping" (Pargament et al., 2000, p. 522)	Primary
Aggressive motor	SCSI	No definition was provided.	Primary
Aggressive verbal relaxation	SCSI	No definition was provided.	Primary
Anger self-statements	CSQ-SCD	No definition was provided. Conceptualized as a pattern of "negative thinking".	Primary
Approach coping	PCQ	"Direct attempts to deal with the pain and the use of active methods to regulate feelings when in the pain" (Reid et al., 1998, p. 84). Includes information seeking, problem solving, seeking social support, and positive self-statements.	Secondary
Avoidance	CCSC/ HICUPS	"Strategies that attempt to manage emotion by trying to avoid or stop thinking about the problem entirely" (Ayers et al., 1996, p. 930). Includes avoidant actions and cognitive avoidance.	Primary (RSQ)/ Secondary (CCSC/ HICUPS)
	RSQ	No definition was provided. Conceptualized as a coping strategy under "disengagement" coping.	
Avoidant/Avoidant actions/Avoiding problems	A-COPE	"Coping behaviors that involve the use of substances (e.g., drinking beer, smoking) as a way to escape or avoiding persons or issues which cause problems (e.g., staying away from home, telling self the problem is not important)" (Patterson and McCubbin, 1987, p. 174).	Primary
	CCSC/ HICUPS	"This includes behavioral efforts to avoid the stressful situation by staying away from it or leaving it" (Ayers et al., 1996, p. 930). Conceptualized as a coping strategy under "avoidance" coping.	
	SCSI	No definition was provided.	
Behavior disengagement	PRI	No definition was provided. Conceptualized as a coping strategy under "passive coping".	Primary
Behavioral distraction	PCQ	No definition was provided. Conceptualized as a coping strategy under "problem-focused avoidance" coping	Primary
Being humorous	A-COPE	"Coping behaviors focused on not taking the situation too seriously by joking or making "light" of it" (Patterson and McCubbin, 1987, p. 174).	Primary
Benevolent religious reappraisal	RCOPE	"Redefining the stressor through religion as benevolent and potentially beneficial" (Pargament et al., 2000, p. 522).	Primary
Blaming others	KIDCOPE	No definition was provided.	Primary
Calming self-statements	CSQ-SCD	No definition was provided. Conceptualized as a coping strategy under "coping attempts".	Primary
Cognitive	SCSI	No definition was provided.	Primary
Cognitive avoidance	CCSC/ HICUPS	"This includes efforts to avoid thinking about the problem. It includes the use of fantasy or wishful thinking or imagining that the situation was better. It refers to cognitive activity and not behaviors one does to avoid thinking about it" (Ayers et al., 1996, p. 930). Conceptualized as a coping strategy under "avoidance" coping.	Primary
Cognitive decision making	CCSC/ HICUPS	"This refers to planning or thinking about ways to solve the problem. It includes thinking about choices, thinking about future consequences, and thinking of ways to solve the problem. It is not simply thinking about the problem but thinking about how to solve it. It involves the planning and not the execution of actions to solve the problem" (Ayers et al., 1996, p. 930). Conceptualized as a coping strategy under "active" coping.	Primary
Cognitive distraction	PCQ	No definition was provided. Conceptualized as a coping strategy under "problem-focused avoidance" coping	Primary
Cognitive refocusing	PPCI	"An active cognitive process to focus one's attention away from pain perception, rather than simply distraction which may imply a more reactive cognitive response to external stimuli" (Varni et al., 1996, p. 148).	Primary

(Continued)

TABLE 4 | Continued

Coping concept	Measure	Descriptor(s)	[a]Factor level
Cognitive restructuring	CSI	"Includes cognitive strategies that alter the meaning of the stressful transaction as it is less threatening, is examined for its positive aspects, is viewed from a new perspective, etc". (Tobin et al., 1984, p. 2). Conceptualized as a coping strategy under "problem-focused engagement".	Primary
	KIDCOPE	No definition was provided.	
	RSQ	No definition was provided. Conceptualized as a coping strategy under "secondary control" coping.	
Cognitive self-instruction	PPCI	"Internal self-statements that deal with the child's pain at a cognitive level" (Varni et al., 1996, p. 143).	Primary
Collaborative religious coping	RCOPE	"Seeking control through partnership with God in problem-solving" (Pargament et al., 2000, p. 522).	Primary
Condition-specific strategies	PRI	"Coping strategies specific to abdominal pain, such as going to the bathroom" (Walker et al., 1997, p. 393). Conceptualized as a coping strategy under "active coping".	Primary
Coping attempts	CSQ-SCD	"Children high on this factor appeared to cope with pain in an active fashion using a variety of cognitive and behavioral coping strategies" (Gil et al., 1991, p. 658). Consists of diverting attention, reinterpret pain, ignoring pain sensations, calming self-statements, and increased behavior activity.	Secondary
Coping response	CHAS	"Thoughts or actions by the child during headaches to help manage them" (Budd and Kedesdy, 1989, p. 3).	Primary
Demonic reappraisal	RCOPE	"The stressor is defined as the work of the devil" (Pargament et al., 2000, p. 522).	Primary
Denial	RSQ	No definition was provided. Conceptualized as a coping strategy under "disengagement" coping.	Primary
Developing self-reliance and optimism	A-COPE	"Coping behaviors focused upon the direct efforts by the adolescent to be more organized and in charge of the situation, as well as to thinking positively about what is happening to him or her (e.g., organizing your life, making your own decisions)" (Patterson and McCubbin, 1987, p. 174).	Primary
Disengagement	CSI	"Strategies that are likely to result in disengaging the individual from the person/environment transaction. Feelings are not shared with others, thoughts about situations are avoided, and behaviors that might change the situation are not initiated" (Tobin et al., 1984, p. 4). Consists of problem avoidance, wishful thinking, social withdrawal, and self-criticism.	Tertiary (CSI/ Secondary (RSQ)
	RSQ	"Responses oriented away from a stressor or one's reactions" (Connor-Smith et al., 2000, p. 977). Includes avoidance, denial, and wishful thinking.	
Distraction/Distract and ignoring pain/Distracting/ Diverting attention	CCSC/ HICUPS	"These strategies are represented by the categories of physical release of emotions and distracting actions. The underlying similarity between these two dimensions of distraction strategies is that the child or adolescent uses some other activity or stimulus to distract themselves from dealing with or thinking about the problem situation" (Ayers et al., 1996, p. 952).	Primary
	CSQ-SCD	No definition was provided. Conceptualized as a coping strategy under "coping attempts".	
	KIDCOPE	No definition was provided.	
	PRI	No definition was provided. Conceptualized as a coping strategy under "accommodative coping".	
	RSQ	No definition was provided. Conceptualized as a coping strategy under "secondary control" coping.	
	SCSI	No definition was provided.	
Distracting actions	CCSC/ HICUPS	"This includes efforts to avoid thinking about the problem situation by using distracting stimuli, entertainment, or some distracting activity. If the distracting activity involves more than moderate physical exertion it should not be included here" (Ayers et al., 1996, p. 930). Conceptualized as a coping strategy under "distraction" coping.	Primary
Emotion-focused avoidance	PCQ	"Strategies in which emotions are freely expressed and strategies that reflect a lack of effort to regulate feelings when in pain" (Reid et al., 1998, p. 84). Includes internalizing/catastrophizing and externalizing.	Secondary
Emotion-focused coping	Ways of coping checklist	"Cognitive and behavioral efforts directed at reducing or managing emotional distress" (Folkman and Lazarus, 1980, p. 225).	Primary
Emotion-focused disengagement	CSI	"Shutting oneself and one's feelings off from others and criticizing or blaming oneself for what happened" (Tobin et al., 1984, p. 4). Includes social withdrawal and self-criticism.	Secondary
Emotion-focused engagement	CSI	"Items reflect open communication of feelings to others and increased social involvement, especially with family and friends. These coping efforts are focused on the individual's emotional reaction to the stressful situation" (Tobin et al., 1984, p. 3). Includes express emotions and social support.	Secondary
Emotion-focused support	CCSC/ HICUPS	"This involves other people in listening to feelings or providing understanding to help the person be less upset" (Ayers et al., 1996, p. 930). Conceptualized as a form of "support seeking".	Primary
Emotional	SCSI	No definition was provided.	Primary
Emotional regulation	KIDCOPE	No definition was provided.	Primary
	RSQ	No definition was provided. Conceptualized as a coping strategy under "primary control" coping.	

(Continued)

TABLE 4 | Continued

Coping concept	Measure	Descriptor(s)	[a]Factor level
Engagement	CSI	"Attempts by the individual to engage the individual in efforts to manage the stressful person/environment transaction. Through these coping strategies individuals engage in an active and ongoing negotiation with the stressful environment" (Tobin et al., 1984, p. 4). Consists of problem-solving, cognitive restructuring, social support, and express emotions.	Tertiary
Engaging in demanding activity	A-COPE	"Coping behaviors in which poses a challenge from the adolescent to excel at something or achieve a goal such as strenuous physical activity, improving oneself, or working hard on schoolwork" (Patterson and McCubbin, 1987, p. 174).	Primary
Expressing emotions/ Express emotions/ Emotional expression	CSCC/ HICUPS	"This involves the overt expression of feelings either by an action to express feelings, a verbal expression of feelings, or simply an overt release of emotion. It is a solitary activity and does not include discussing feelings with another person. It also does not include inappropriately acting out feelings by threatening or hurting another person" (Ayers et al., 1996, p. 929).	Primary
	CSI	"Releasing and expressing emotions" (Tobin et al., 1984, p. 2). Conceptualized as a coping strategy under "emotion-focused engagement".	
	RSQ	No definition was provided. Conceptualized as a coping strategy under "primary control" coping.	
Externalizing	PCQ	No definition was provided. Conceptualized as a coping strategy under "emotion-focused avoidance" coping	Primary
Fear self-statements	CSQ-SCD	No definition was provided. Conceptualized as a pattern of "negative thinking".	Primary
Habitual	SCSI	No definition was provided.	Primary
Heat, cold, massage	CSQ-SCD	No definition was provided. Conceptualized as a coping strategy under "passive adherence".	Primary
Ignoring pain sensations	CSQ-SCD	No definition was provided. Conceptualized as a coping strategy under "coping attempts".	Primary
Increased behavioral activity	CSQ-SCD	No definition was provided. Conceptualized as a coping strategy under "coping attempts".	Primary
Information seeking	PCQ	No definition was provided. Conceptualized as a coping strategy under "approach" coping	Primary
Internalizing/ Catastrophizing	CSQ-SCD	No definition was provided. Conceptualized as a pattern of "negative thinking".	Primary
	PCQ	No definition was provided. Conceptualized as a coping strategy under "emotion-focused avoidance" coping.	
	PRI	No definition was provided. Conceptualized as a coping strategy under "passive coping".	
Interpersonal religious discontent	RCOPE	"Expressing confusion and dissatisfaction with the relationship of clergy or members to the individual in the stressful situation" (Pargament et al., 2000, p. 524).	Primary
Investing in close friends	A-COPE	"Coping behaviors that involve seeking closeness and understanding from a peer (e.g., be with a boyfriend)" (Patterson and McCubbin, 1987, p. 174).	Primary
Isolating/Isolation	SCSI	No definition was provided.	Primary
	CSQ-SCD	No definition was provided. Conceptualized as a pattern of "negative thinking".	
Marking religious boundaries	RCOPE	"Clearly demarcating acceptable from unacceptable religious behavior and remaining within religious boundaries" (Pargament et al., 2000, p. 523).	Primary
Massage/Guard	PRI	No definition was provided. Conceptualized as a coping strategy under "active coping".	Primary
Minimizing pain	PRI	No definition was provided. Conceptualized as a coping strategy under "accommodative coping".	Primary
Negative thinking	CSQ-SCD	"Children high on this factor appeared to engage in negative thinking patterns including catastrophizing and self-statements of fear and anger, as well as isolation in response to pain" (Gil et al., 1991, p. 658). Consists of catastrophizing, fear self-statements, anger self-statements, and isolation.	Secondary
Passive coping	PRI	No definition was provided. Includes behavior disengagement, self-isolation, and catastrophizing.	Secondary
Passive adherence	CSQ-SCD	"Children high on this factor seemed to rely on concrete coping strategies typically recommended by health care professionals for SCD pain management (e.g., increasing fluid intake, resting), and it appears, they perceive that using these strategies is sufficient for controlling and decreasing their pain" (Gil et al., 1991, p. 658). Consists of resting, taking fluids, praying/hoping, and heat/cold/massage.	Secondary
Passive religious deferral	RCOPE	"Passive waiting for God to control the situation" (Pargament et al., 2000, p. 522).	Primary
Physical exercise	SCSI	No definition was provided.	Primary
Physical release of emotion	CCSC/ HICUPS	"This includes efforts to physically work off feelings with physical exercise, play, or efforts to physically relax. There needs to be at least a moderate amount of physical exertion involved, so that very light physical activity for a child (e.g., walking) would not be included here" (Ayers et al., 1996, p. 930). Conceptualized as a coping strategy under "distraction" coping.	Primary
Pleading for direct intercession	RCOPE	"Seeking control indirectly by pleading to God for a miracle or dive intercession" (Pargament et al., 2000, p. 522).	Primary

(Continued)

TABLE 4 | Continued

Coping concept	Measure	Descriptor(s)	[a]Factor level
Positive cognitive restructuring	CCSC/ HICUPS	"This refers to thinking about the situation in a more positive way. It includes thoughts that minimize the problem or the consequences of the problem. Acceptance that one can live with the situation the way it is optimistic thinking and an example of positive cognitive restructuring" (Ayers et al., 1996, p. 929). Conceptualized as a coping strategy under "active" coping.	Primary
Positive self-statements	PCQ	No definition was provided. Conceptualized as a coping strategy under "approach" coping.	Primary
Positive thinking	RSQ	No definition was provided. Conceptualized as a coping strategy under "secondary control" coping.	Primary
Praying and hoping	CSQ-SCD	No definition was provided. Conceptualized as a coping strategy under "passive adherence".	Primary
Primary control engagement	RSQ	"Altering objective conditions such as stressor or one's emotional response to stressor" (Connor-Smith et al., 2000, p. 977). Includes problem solving, emotional regulation, and emotional expression.	Secondary
Problem avoidance	CSI	"The denial of problems and the avoidance of thoughts or action about the stressful event" (Tobin et al., 1984, p. 2). Conceptualized as a coping strategy under "problem-focused disengagement".	Primary
Problem-focused avoidance	PCQ	"Attempts to disengage from the pain" (Reid et al., 1998, p. 84). Includes cognitive and behavioral distraction.	Secondary
Problem-focused coping	Ways of coping checklist	"Cognitive problem-solving efforts and behavioral strategies for altering or managing the source of the problem" (Folkman and Lazarus, 1980, p. 225).	Primary
Problem-focused disengagement	CSI	"Items reflect denial, avoidance, and an inability or reluctance to look at the situation differently. They reflect cognitive and behavioral strategies to avoid the situation" (Tobin et al., 1984, p. 4). Includes problem avoidance and wishful thinking.	Secondary
Problem-focused engagement	CSI	"Items involve cognitive and behavioral strategies to change the situation or to change the meaning of the situation for the individual. These coping efforts are focused on the stressful situation itself" (Tobin et al., 1984, p. 4). Includes problem-solving and cognitive restructuring.	Secondary
Problem-focused support	CCSC/ HICUPS	"Use of other people as resources to assist in seeking solutions, seeking advice/information or direct task assistance" (Ayers et al., 1996, p. 930). Conceptualized as a form of "support seeking".	Primary
Problem-solving/Direct problem solving	CCSC/ HICUPS	"This refers to efforts to change the problem situation by changing the self or by changing the environment. It involves what one does, not what one thinks"(Ayers et al., 1996, p. 929). Conceptualized as a coping strategy under "active" coping	Primary
	CSI	"Behavioral and cognitive strategies designed to eliminate the source of stress by changing the stressful situation" (Tobin et al., 1984, p. 2). Conceptualized as a coping strategy under "problem-focused engagement".	
	KIDCOPE	No definition was provided.	
	PCQ	No definition was provided. Conceptualized as a coping strategy under "approach" coping	
	PRI	No definition was provided. Conceptualized as a coping strategy under "active coping".	
	RSQ	No definition was provided. Conceptualized as a coping strategy under "primary control" coping.	
Problem-solving self-efficacy	PPCI	"An adaptive coping strategy consisting of both concrete problem-solving (e.g., put ice or heat on the sore spot) and cognitive statements regarding one's ability to resolve the pain problem (e.g., know that I can do something to main the pain or hurt feel better)" (Varni et al., 1996, p. 149).	Primary
Punishing God reappraisal	RCOPE	"Redefining the stressor as a punishment from God for the individual's sins" (Pargament et al., 2000, p. 522).	Primary
Purification/forgive-ness	RCOPE	"Searching for spiritual cleansing through religious actions" (Pargament et al., 2000, p. 523).	Primary
Reappraisal of God's power	RCOPE	"Redefining God's power to influence the stressful situation" (Pargament et al., 2000, p. 523).	Primary
Reinterpret pain	CSQ-SCD	No definition was provided. Conceptualized as a coping strategy under "coping attempts".	Primary
Relaxing	A-COPE	"Coping behaviors which focus on ways to reduce tension such as daydreaming, listening to music, or riding around in a car" (Patterson and McCubbin, 1987, p. 174–175).	Primary
Religious direction/ conversion	RCOPE	"Looking to religion for assistance in finding a new direction for living when the old one may no longer be viable/looking to religion for a radical change in life" (Pargament et al., 2000, p. 524).	Primary
Religious focus	RCOPE	"Engaging in religious activities to shift focus from the stressor" (Pargament et al., 2000, p. 523).	Primary
Religious helping	RCOPE	"Attempting to provide spiritual support and comfort to others" (Pargament et al., 2000, p. 524).	Primary
Resignation	KIDCOPE	No definition was provided.	Primary
Rest/Resting	CSQ-SCD	No definition was provided. Conceptualized as a coping strategy under "passive adherence".	Primary
	PRI	No definition was provided. Conceptualized as a coping strategy under "active coping".	
Secondary control engagement	RSQ	"Focused on adaptation to the problem" (Connor-Smith et al., 2000, p. 977). Includes positive thinking, cognitive restructuring, acceptance and distraction.	Secondary
Seeking diversions	A-COPE	"Coping behaviors focused upon the adolescent's efforts to keep busy and engage in relatively sedate activities that are a way to escape from or forget about the sources of tension and stress such as sleeping, watching TV or reading" (Patterson and McCubbin, 1987, p. 174).	Primary

(Continued)

TABLE 4 | Continued

Coping concept	Measure	Descriptor(s)	[a]Factor level
Seeking professional support	A-COPE	"Coping behaviors directed at getting help and advice from a professional counselor or teacher about difficult problems" (Patterson and McCubbin, 1987, p. 174).	Primary
Seeking social support/ Developing social support/Social support	A-COPE	"Coping behaviors directed at efforts to stay emotionally connected with other people through reciprocal problem solving and expression of affect (e.g., helping others solve their problems, taking to a friend about one's feelings, apologizing to others)" (Patterson and McCubbin, 1987, p. 174).	Primary
	CSI	"Includes items that refer to seeking emotional support from people, one's family, and one's friends" (Tobin et al., 1984, p. 2).	
	KIDCOPE	No definition was provided.	
	PCQ	No definition was provided. Conceptualized as a coping strategy under "approach" coping	
	PPCI	"The child seeks aid, comfort, or understanding from parents, peers, and others" (Varni et al., 1996, p. 143).	
	PRI	No definition was provided. Conceptualized as a coping strategy under "active coping"	
	SCSI	No definition was provided.	
Seeking spiritual support/ Seeking support from clergy	A-COPE	"Coping behaviors ffocused on religious behaviors (e.g., praying, going to church) or talking to clergy" (Patterson and McCubbin, 1987, p. 174).	Primary
	RCOPE	"Searching for comfort and reassurance through God's love and care" (Pargament et al., 2000, p. 524).	
Seeking understanding	CCSC/ HICUPS	"This includes cognitive efforts to find meaning in a stressful situation or to understand it better. It involves seeking understanding of the situation and not seeking to put a positive interpretation on the situation" (Ayers et al., 1996, p. 929). Conceptualized as a coping strategy under "active" coping.	Primary
Self-criticism	CSI	"Blaming oneself for the situation and criticizing oneself" (Tobin et al., 1984, p. 3). Conceptualized as a coping strategy under "emotion-focused disengagement".	Primary
	KIDCOPE	No definition was provided.	
Self-isolation	PRI	There was no definition provided. Conceptualized as a coping strategy under "passive coping".	Primary
Social withdrawal	CSI	No definition was provided. Conceptualized as a coping strategy under "emotion-focused disengagement".	Primary
	KIDCOPE	No definition was provided.	
Solving family problems	A-COPE	"Coping behaviors focused on working out difficult issues with family members (e.g., talk to parent about what bothers you) and doing things with the family" (Patterson and McCubbin, 1987, p. 174).	Primary
Spiritual/Spiritual connection	SCSI	No definition was provided.	Primary
	RCOPE	"Searching for comfort and reassurance through God's love and care" (Pargament et al., 2000, p. 523).	
Spiritual discontent	RCOPE	"Expressing confusion and dissatisfaction with God's relationship to the individual in the stressful situation" (Pargament et al., 2000, p. 523).	Primary
Stoicism	PRI	"Inhibiting or minimizing the expression of one's pain or distress to others" (Walker et al., 1997, p. 393). Conceptualized as a coping strategy under "accommodative coping".	Primary
Strive to rest and be alone	PPCI	"Attempts to rest or socially withdrawal" (Varni et al., 1996, p. 149).	Primary
Support seeking	CCSC/ HICUPS	No definition was provided. Comprised of emotion-focused- and problem-focused support.	Secondary
Taking fluids	CSQ-SCD	No definition was provided. Conceptualized as a coping strategy under "passive adherence".	Primary
Ventilating feelings	A-COPE	"Coping behaviors focused upon the adolescent's expression of frustrations and tensions such as yelling, blaming others, saying mean things, and complaining to friends or family" (Patterson and McCubbin, 1987, p. 174).	Primary
Wishful thinking	CSI	"Cognitive strategies that reflect an inability or reluctance to reframe or symbolically alter the situation. The items involve hoping and wishing that things could be better" (Tobin et al., 1984, p. 3). Conceptualized as a coping strategy under "problem-focused disengagement".	Primary
	KIDCOPE	No definition was provided.	
	RSQ	No definition was provided. Conceptualized as a coping strategy under "disengagement" coping.	

For coping responses captured by multiple questionnaires, descriptors are provided adjacently for ease of comparison. Coping responses are classified as primary, secondary, and tertiary factors based on factor analytic results from the scale development studies. A-COPE, adolescent coping orientation for problem experiences; CSI, coping strategies inventory; CSQ, coping strategies questionnaire; CCSC, children's coping strategies checklist; CHAS, children's headache assessment scale; HICUPS, how I coped under pressure scale; PCQ, pain coping questionnaire; PPCI, pediatric pain coping inventory; PRI, pain response questionnaire; RSQ, response to stress questionnaire; SCSI, schoolagers coping strategies inventory.
[a]An ordered-categorical scale derived from factor analytic results from the scale development studies was used to classify coping responses. The terms "primary", "secondary", and "tertiary" correspond to first-, second-, and third-order factor levels, respectively [see Results section "Aim 3: Evaluations of Measurement and Conceptual Consistency (Questionnaires Only), subsection "Classifications"].

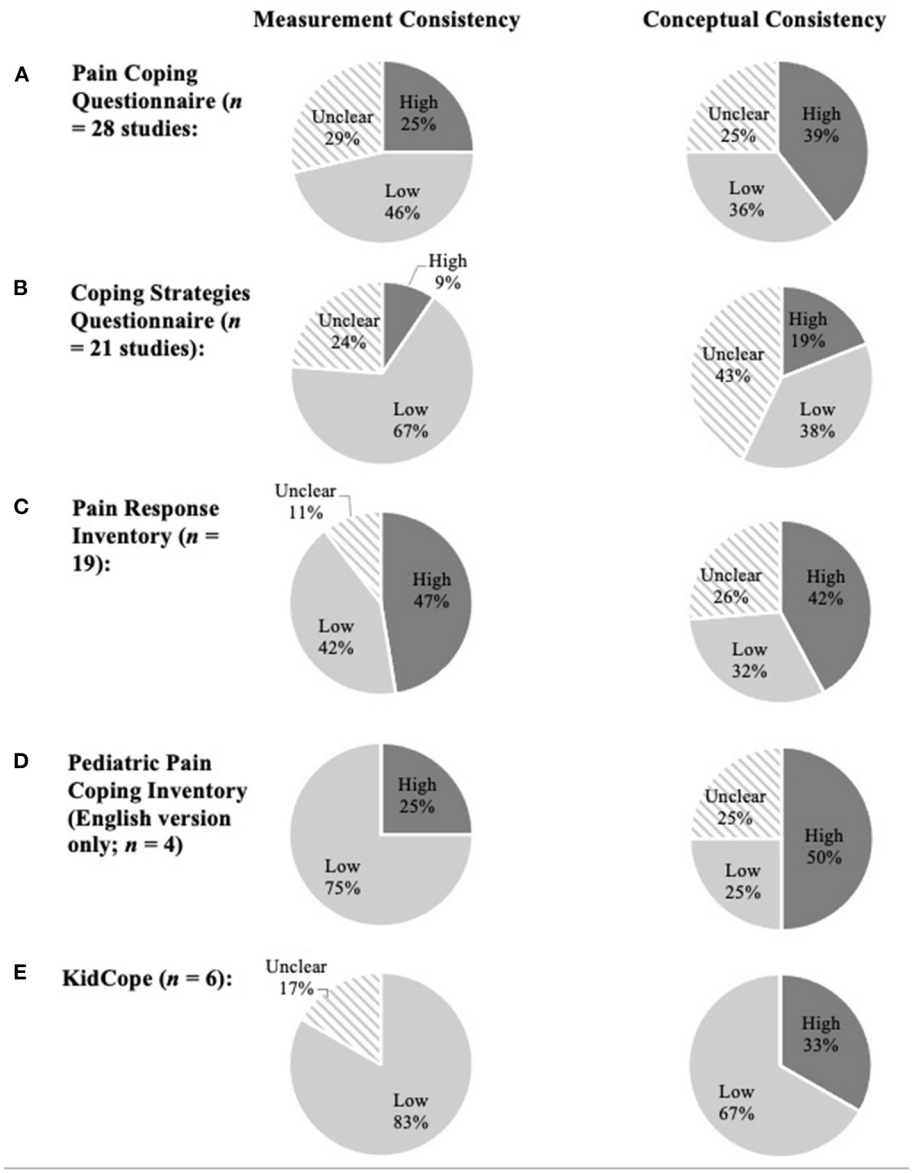

FIGURE 3 | For each measure, overall measurement and conceptual consistency ratings are presented as the percentage (%) of studies that were classified as "high" = no discrepancies, "low" = one or more discrepancies, and "unclear" = unable to evaluate (see full text, **Table 1**). Charts show the frequency distribution for both consistency ratings for the five most frequently cited questionnaires: **(A)** pain coping questionnaire; **(B)** coping strategies questionnaire; **(C)** pain response inventory; **(D)** pediatric pain-coping inventory (English version); and **(E)** KidCope. "Low" ratings indicate the presence of discrepancies that may impact our ability to directly interpret, consolidate, and/or compare research findings. These ratings do not reflect the quality of the research.

Types of Discrepancies

A wide range of discrepancies was identified in the measurement and conceptualization of coping responses by questionnaires across studies. Ratings of measurement and conceptual consistency (**Figure 3**), and the types of discrepancies (**Supplementary Table 5**) are summarized for the top five most frequently used questionnaires: PCQ (Reid et al., 1998), CSQ (Gil et al., 1991), PRI (Walker et al., 1997), PPCI (Varni et al., 1996), and KidCope (Spirito et al., 1988, 1991). These five questionnaires were used in 82.7% of studies which employed a questionnaire with known psychometric properties and 64.8% of all included studies. As such, the coping responses assessed by these measures were the focus. Given that the remaining questionnaires were included in few studies, our ability to draw conclusions about their conceptual and measurement consistency is limited and therefore, will not be explored in detail.

Pain Coping Questionnaire

Consistent with previous reviews (Blount et al., 2008), the PCQ was the most widely used measure of coping in the context of pediatric chronic pain (ages 7–17; $n = 28$). The

PCQ has been used in English (Reid et al., 1998) (24 studies), Dutch (Bandell-Hoekstra et al., 2002) (two studies), and Danish (Thastum et al., 1999) (two studies) samples. Twenty studies provided sufficient information about the PCQ to evaluate measurement consistency, of which 65% ($n = 13$) were found to have at least one discrepancy. Types of discrepancies included using samples outside of the recommended age range of 7–17 years ($n = 12$); alternate composite scores/subscales ($n = 6$); missing one or more subscales ($n = 2$); using different coding for response options ($n = 2$); and using the sum of item-level responses instead of the mean ($n = 1$). Low conceptual consistency was found for 47.6% ($n = 10$) of studies using the PCQ that provided descriptors of the coping responses assessed ($n = 21$). A common discrepancy included the inconsistent categorization of positive self-statements as a type of "problem-focused avoidance" instead of "approach" coping ($n = 3$). In addition, several studies used different labels for coping constructs, such as "distraction" instead of "problem-focused avoidance" ($n = 4$) and "positive approach" in place of "approach coping" ($n = 1$).

Coping Strategies Questionnaire for Sickle Cell Disease

Of the studies that provided sufficient information about the measurement characteristics of the coping strategies questionnaire for sickle cell disease (CSQ-SCD) ($n = 16$), 87.5% ($n = 14$) had at least one discrepancy. The most common discrepancy was the inclusion of participants beyond the age range of the scale development study (i.e., 7–17 years old; $n = 13$). Other discrepancies included using participants with a pain condition other than SCD ($n = 2$); alternate composite scores (e.g., total coping attempts, rational thinking; $n = 3$); missing subscales ($n = 1$); using a six- instead of seven-point Likert scale ($n = 2$); using a 44-item version of the CSQ-SCD ($n = 2$); and/or computing subscales by taking the sum of the item-level responses instead of the mean ($n = 2$). Conceptual consistency was evaluated for 12 of the included studies that used the CSQ-SCD. Of these studies, 66.7% ($n = 8$) reported a discrepancy in their conceptualization of particular coping responses. The primary reason for low ratings of conceptual consistency was the use of alternate terminology for constructs operationalized by the CSQ-SCD ($n = 4$). For example, the terms "illness-focused strategy" and "adherence" have been used in place of "passive adherence".

Pain Response Inventory

Upon evaluating measurement consistency ($n = 17$), 47.1% ($n = 8$) of studies had more than one discrepancy. The reasons for low ratings of measurement consistency included the following: the inclusion of participants outside of the age range of the PRI validation sample (i.e., 7–17 years old; $n = 3$); used in a sample of youth with pain conditions other than abdominal pain ($n = 2$); computing subscales by taking the sum of the item-level responses instead of the mean ($n = 1$); missing subscales ($n = 4$); and/or alternate categorizations of subscales ($n = 1$). Conceptual consistency was evaluated for 14 studies using the PRI. The original scale development study of the PRI did not provide definitions of the coping constructs assessed, and therefore, conceptual consistency was evaluated by the inclusion of specific examples of coping strategies that fall within each of the higher-order factors. As such, a common discrepancy was the exclusion or incorrect categorization of lower-order subscales in the conceptualization of higher-order ones ($n = 5$). For example, several studies excluded "stoicism" in the conceptualization of "accommodative coping" ($n = 3$). In addition, inconsistent labeling of coping responses was demonstrated by one study that used the term "activity restriction" in place of "behavior disengagement".

Pediatric Pain Coping Inventory

Measurement and conceptual consistency could not be evaluated for studies that used the German version of the Pediatric Pain Coping Inventory (PPCI) because the scale development study was not available in English. As a result, four studies were used to evaluate measurement and conceptual consistency for the PPCI. In terms of measurement consistency, 75% of studies that used the PPCI ($n = 3$) were rated "low". Types of discrepancies included the inclusion of participants beyond the intended age range (i.e., 5–16 years old; $n = 2$) and pain type ($n = 1$) validated for the PPCI. Moreover, conceptual consistency was rated "low" for one study (25%) that used the PPCI. This study reported using the *a priori* coping responses ("distraction", "problem-solving", and "helplessness") as opposed to those supported by factor analysis and psychometric properties of the PPCI assessed in children with chronic pain ("strive for rest and be alone", "cognitive refocusing", and "problem-solving self-efficacy").

KidCope

The majority of the six studies (83.3%) that employed the KidCope were rated low for measurement consistency. The most common discrepancy was the inclusion of participants aged two or more years outside of the age range validated for the KidCope (i.e., child version = 9–13; adolescent version = 12–18) ($n = 5$). While the KidCope was developed as a unidimensional scale, two studies computed alternate higher-order scores using the coping strategies measured by the KidCope. In addition, one study used a four-point Likert scale for the adolescent version rather than the original "yes/no" response items. With regard to conceptual consistency, 66.7% of studies that used the KidCope were rated as "low". Two studies proposed using the coping responses assessed by the KidCope to conceptualize "active" and "passive" coping; and, in contrast, one other study conceptualized "negative/avoidance" and "positive/approach". Neither of these higher-order conceptualizations of coping responses was proposed in the development of the KidCope. In another study, discrepancies included mislabeling coping responses (e.g., using the term "withdrawal" in place of "social withdrawal") or adding coping responses that are not typically conceptualized by the KidCope (i.e., remain positive, blame self instead of self-criticism, express emotions, relax, become helpless, different feelings, focus on future, taking medication/laying down).

DISCUSSION

This scoping review is the first to examine the sample and methodological characteristics, theoretical frameworks, and measurement tools used to measure and conceptualize coping in pediatric chronic pain. Overall, a lack of theory, conceptual clarity, conceptual consistency, and measurement consistency emerged in included studies. In the discussion, these results and their implications are explored in relation to four major gaps and are drawn upon to offer recommendations for future research.

Gap 1: Lack of Theory, Conceptual Clarity, and Conceptual Consistency

It is important for researchers to ground their research questions and methods in theory to ensure an appropriate use of measures and concepts. Also, explicit use of theory helps readers to make sense of and interpret findings. Less than 13% of included studies provided a clear explanation of a theoretical framework for the coping responses assessed. Even fewer studies (7%) used theory to justify the research methods employed. Current theoretical frameworks focus on the conceptualization of higher-order coping responses (i.e., secondary factors such as emotion-focused vs. problem-focused coping, tertiary factors such as engagement vs. disengagement) and organize lower-order coping responses within them (i.e., primary factors). The overreliance on quantitative and exploratory methods has contributed to an excessive number of lower-order conceptualizations (86 primary coping responses) that makes it challenging to synthesize, interpret, and apply this literature in future research and clinical contexts. Thus, theoretical frameworks for primary coping responses are needed to clearly and consistently operationalize overt thoughts and behaviors used for coping. Building and incorporating strong theoretical frameworks could help to establish a more parsimonious literature by avoiding redundant and synonymous conceptualizations (e.g., "ventilating feelings" and "expression of emotion").

The use of the terms "coping styles" to refer to higher-order categories was not as prominent as expected, and 75% of studies that focused on conceptualizing higher-order categories did not use any particular coping terminology to describe them. As such, a more explicit use of theory can also bring clarity and consistency to the conceptualization of higher- vs. lower-order categories of coping responses. For example, some researchers might use *dispositional/stylistic* categories, such as "coping styles", if the recommendation is to view coping responses as being stable over time and situation (Moos and Holahan, 2003). In contrast, researchers might avoid specific or highly restrictive terminology when using a *contextual* perspective, wherein coping responses are used differently in response to developmental (e.g., maturity) and environmental factors (e.g., access to resources) (Roberts et al., 2001; Moos and Holahan, 2003; Kim-Cohen et al., 2004). An *integrative* perspective might view some coping responses as stable and others as flexible over time and situations. As a field, it is important to further explore whether a particular theory or an integrative approach could drive a more clear and consistent understanding of the terminology used to refer to hierarchical classifications of coping responses.

Almost half of the included studies did not explicitly state descriptors of the coping responses assessed. Also, a third of the studies that used questionnaires described coping responses in a way that was inconsistent with the original scale development study, such as using a different definition(s), categorizations of lower-order into higher-order coping responses, or alternate terminologies for referring to factors. One issue contributing to low conceptual clarity and consistency is the tendency for authors to describe coping responses based on their relationship with coping *outcomes* rather than the *nature* or operationalization of the thoughts or behaviors underlying the coping response. For example, some authors have described both "passive coping" and "passive adherence" solely by their associations with maladaptive outcomes in youth with chronic pain (e.g., Thompson et al., 1994; Logan et al., 2012). This description is problematic for two main reasons. First, coping responses have inconsistent relationships with coping outcomes, and the factors contributing to these inconsistencies are not well-understood (Skinner et al., 2003; Zimmer-Gembeck and Skinner, 2016). Therefore, while "passive coping" and "passive adherence" are generally associated with negative outcomes, this might not be consistent across studies, individuals, or contexts. Second, relying on this description alone does not provide any information about what it looks like for a youth to use either of these coping responses and can lead to confusion between these two terms that are conceptually distinct (for an example, see **Figure 1**). While the adaptive and maladaptive qualities of coping responses are relevant to their conceptualization, researchers should prioritize using definitions and examples in their papers that help readers to reproduce these concepts in research and discuss them in clinical practice.

Gap 2: Lack of Diverse Research Methods and Measurement Consistency

Most of our knowledge about coping in the context of pediatric chronic pain is based on quantitative methods (85%) and cross-sectional designs (67%) with an emphasis on parent- and/or self-report questionnaires to assess coping responses in youth (86%). The five most common questionnaires identified were four pain-specific coping questionnaires (i.e., PCQ, PRI, PPCI, and CSQ-SCD) and one general coping questionnaire (i.e., KidCope). The use of questionnaires to assess coping is, generally, a cost-effective and convenient method for data collection. Some questionnaires (e.g., PCQ, KidCope) are available in different versions (e.g., language, age, and pain condition), enabling more tailored selection. Moreover, questionnaires can be used to promote more consistent conceptualizations of coping in the literature and allow for the consolidation of research findings across studies if used frequently and consistently by study authors. Unfortunately, regarding measurement consistency, just over half of studies had one or more discrepancies, and one in five was missing more than two relevant scale characteristics. However, as we employed a strict criterion for measurement consistency, a "discrepancy" does not necessarily mean that the study was flawed or inaccurate but rather highlights an inconsistency between studies that may impact our ability to directly consolidate and/or compare research findings. In addition, the high proportion of studies

missing information about the scale in their methods highlights a lack of transparency in the literature, which in turn, limits the reproducibility of studies and the ability to confidently consolidate findings across studies. The less we can consolidate findings, the less we can advance the field both in terms of theory building and testing as well as interventions.

Another potential limitation of questionnaires is the possibility for psychometric inadequacies, including an unstable or unsubstantiated factor structure (e.g., over-factoring, poor reproducibility), inadequate or non-existent construct validity, and no reports of test-retest reliability (Parker and Endler, 1992; Blount et al., 2008). Additionally, the use of exploratory factor analytic procedures contributes to an abundance of complex and difficult-to-interpret constructs that lack clinical utility and relevance (Parker and Endler, 1992; Blount et al., 2008). While overarching categories of coping responses (i.e., factors) can be useful for understanding and predicting coping outcomes, these constructs can be abstract and difficult to operationalize in the development of interventions (Blount et al., 2008). As such, Blount and colleagues (Blount et al., 2008) highlighted the importance of assessing and reporting on discrete, trainable mental actions or behaviors. One approach to assessing specific coping behaviors is by analyzing and interpreting item-level responses on questionnaires (Blount et al., 2008; Schwartz, 2016). For example, using the PCQ, researchers and clinicians might examine group or individual differences in responses to "talk to a family member about how I feel" and "talk to a friend about how I feel" instead of the subscale score for "seeking social support". This information helps to understand the specific ways that social support is used by youth to cope with pain. Although item-level responses are less psychometrically sound and provide less information about the latent variable being measured, they can be relevant to intervention studies where outcomes are used to inform intervention training and clinical recommendations. Alternatively, behavioral assessment tools are useful for assessing discrete, overt coping behavior (e.g., observable body movements, sounds, and words), which can be advantageous for identifying and monitoring specific behaviors, overcoming barriers to self-report (e.g., social desirability), and working with youth who have complex developmental or intellectual disabilities. To date, in contrast to acute pain, there are no well-established behavioral coping measures for pediatric *chronic* pain (Blount et al., 2008; Chorney and McMurtry, 2014), and therefore, developing such tools is an important direction for future research.

Gap 3: Poor Understanding of Coping Responses in Diverse Patient Populations

Although participant characteristics varied across studies in terms of age (3–20 years) and pain conditions, the lack of research in specific patient populations limits the validity of current measurement tools and conceptualizations of coping responses in certain populations. There is evidence to suggest that the use of coping responses may be influenced by age (Curry and Russ, 1985; Compas, 1998; Dubow and Rubinlicht, 2011). For example, young children may rely more on parental support for coping, whereas adolescents have a greater capacity for using more cognitive-oriented coping responses (e.g., cognitive restructuring) and for seeking a broader array of informational, emotional, and tangible supports beyond the family (Dubow and Rubinlicht, 2011). However, very few studies focused exclusively on children under 12 years with chronic pain (8%), and most studies included both child and adolescent participants (68%). Of note, no studies on coping in infants and toddlers with chronic pain were identified nor measures with known psychometric properties for children under 5 years. It is unclear whether this lack of research stems from actual low prevalence of chronic pain in infants (estimated at 1–3%) (Perquin et al., 2000; King et al., 2011) or challenges with detecting and adequately describing chronic pain in infancy (Pillai Riddell et al., 2009; DiLorenzo et al., 2016). Also, current models of conceptualizing coping responses as voluntary/intentional thoughts or actions may be inappropriate to understand infant coping as they have limited cognitive and language capacity as well as learned experiences to independently implement coping responses or describe their ways of coping. More nuanced research is needed to better understand how coping responses may change across development.

Studies were primarily conducted in the United States or Europe (88%) and included predominately female participants (65%). Apart from studies focused on youth with sickle cell disease [which primarily affects individuals of African or Caribbean descents (Hassell, 2010)], the majority of studies that reported on race/ethnicity included white participants (93%). This highlights a lack of measurement and conceptualization of coping responses in diverse cultural (i.e., cross-national, cross-ethnic, and cross-racial cultures) and/or sex/gender groups of youth with chronic pain. A systematic review of the adult chronic pain literature suggests that mean scores on measures of pain-related coping responses vary significantly between people of different countries and languages (Sharma et al., 2020). In addition, cultural and contextual theories of coping highlight that while stress and coping, in general, are universal experiences, members of different cultures might not only experience different/additional stressors but also consider and respond to stressors differently with respect to coping goals, responses, and outcomes (Kuo, 2011). For example, members of individualistic cultures tend to prioritize "externally targeted control" (i.e., changing the environment/stressor), whereas members of collectivistic culture tend to use "internally targeted control" (i.e., changing oneself) (Kuo, 2011). This highlights the importance of understanding which coping responses are relevant to assess and recommend for diverse youth.

Although there is evidence for sex-specific engagement in coping responses among adolescents with chronic pain (e.g., females reported greater use of social support networks than males) (Keogh and Eccleston, 2006), this research has not explored the biological and sociocultural mechanisms for these group differences. A poor understanding of the underlying mechanisms of sex differences and the interchangeable use of the terms "gender" and "sex" in this research poses risks for sex-based stereotypes regarding the effectiveness of certain coping responses that may not be consistent with individual preferences

and/or gender identity (Boerner et al., 2018; Samulowitz et al., 2018). Important gaps to be addressed in the literature include considering the role of gender identity/expression in coping as well as to make clear in research papers when birth-assigned sex vs. self-identified gender is being reported.

Gap 4: Lack of Measurement Tools and Conceptualizations for Proactive Coping

Few studies (<6%) assessed proactive coping responses using either interview or daily diary-recorded measures. Although research on proactive coping is limited in youth (Schwarzer and Luszczynska, 2008), studies in adults demonstrate that proactive coping is associated with various positive psychological (e.g., higher life satisfaction) and physical outcomes (e.g., rehabilitative functioning) (Katter and Greenglass, 2013; Miao et al., 2017; Bhattacharyya et al., 2018). Moreover, the concept of proactive coping is consistent with the push for long-term behavioral changes in the management of chronic pain (Landry et al., 2015; Miró et al., 2017). Taken together, proactive coping is deemed a promising approach to coping for youth with chronic pain.

To advance our ability to assess proactive coping, it is important to first clarify how to conceptualize proactive coping responses *via* theory. Thus, more qualitative research in this area would be useful for identifying different proactive coping responses. Based on this information, the next step would be to derive appropriate assessment tools that capture these responses. To date, the proactive coping inventory (PCI) (Greenglass et al., 1999) for adults is the only known questionnaire for assessing proactive coping responses. Perhaps, the adult-focused literature on proactive coping stems from the need for individuals to be able to anticipate pain-related stressors and independently implement lifestyle changes. As such, self-report assessments may be useful for older youth who may have accumulated experience with chronic pain and can take an active role in implementing lifestyle changes. In contrast, researchers may consider the role of parents in assessments for young children. A greater capacity for understanding and assessing proactive coping in the youth of all ages holds promise for preventing the impact of chronic pain on overall health and psychosocial well-being.

Recommendations for Future Research

The following sections of this review outline a proposed three-step process (**Figure 4**) for establishing more clarity and consistency in the literature.

Step One: Theory Testing/Validation and Revision

There is a critical need for more explicit, well-developed theories of coping in the pediatric chronic pain literature that can then provide the knowledge base for construct validation (Strauss and Smith, 2009). Theoretical frameworks serve as a rationale for *why* we measure particular coping responses by clarifying their meaning and conceptual relationships. Theories need to be culture specific and patient centered (Kuo, 2011). In addition, advancements in theory should clarify how we think about and relate higher- and lower-order coping responses (e.g., dispositional vs. conceptual frameworks).

Current theories of coping used in pediatric chronic pain research were derived from adult or non-chronic pain populations and were rarely cited in the research literature. As such, two pertinent steps need to be taken. First, considering that theory testing/validation is an iterative process (Strauss and Smith, 2009), there is a need for research aimed at either re-evaluating and revising current theoretical models of pain-related coping responses to be applicable to pediatric chronic pain or beginning anew. In these efforts, ways of conceptualizing proactive coping responses should be explored. Assimilating qualitative and/or observational methodologies may prove particularly helpful to test theoretical mechanisms and better capture the lived experiences of youth with chronic pain (Tutelman and Webster, 2020).

Second, it is important to recognize that theories and data are independent of each other (Strauss and Smith, 2009); therefore, researchers are responsible for drawing connections between their findings and theory to improve the clarity and consistency of literature. This would include using more explicit statements of theory and definitions in the introduction or the discussion section of a paper that would: (i) clarify the relevance of particular coping responses to the population of interest; (ii) justify the use of a given measure; (iii) promote a mutual understanding of research findings; and (iv) inform future efforts toward theory validation (Parker, 2020). The underuse or superficial use of theories and definitions in papers may relate, in part, to manuscript length restrictions put in place by publishers. In these situations, authors should consider the use of supplementary materials or open access repositories to share additional conceptual information needed.

Step Two: Measure Development, Evaluation, and Modification

Clear and well-defined theories should be used to guide *how* we measure coping responses. Although the development of new theories may dictate the need for new measures, researchers should, first, carefully consider whether existing measures can be adapted to fit new/modified theories. Adapting existing measures would be beneficial for reassessing the validity of former coping responses, avoiding the creation of redundant or interchangeable coping responses, and comparing new and extant research. More parsimonious literature can also be established by cross-validating or performing content analyses between different coping measures (Crombez et al., 2020). Measures of coping should be tested and periodically re-evaluated in samples of youth with chronic pain, which, in turn, may serve to validate and/or revise the theoretical frameworks used. As such, the process of developing/modifying and evaluating measures (Step 2) should be iterative and interactive with theory construction (Step 1).

This review highlights the need for research aimed at evaluating the reliability, validity, and clinical utility of measures and conceptualizations of coping responses in different age, gender, and cultural groups. In other fields, the use of item content analyses, focus groups, or cognitive interviews has been useful for capturing group differences and patient experiences using particular measures (Beatty and Willis, 2007; Amtmann

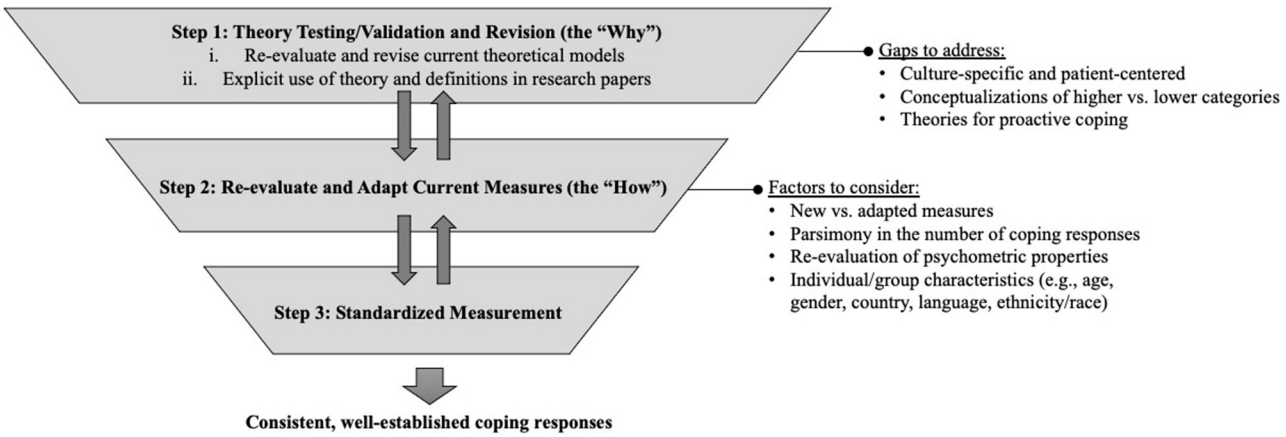

FIGURE 4 | Proposed three steps for future research aimed at establishing more consistent, well-defined coping responses. Double arrows indicate the iterative and interactional relationship between steps.

et al., 2018; Crombez et al., 2020). Researchers have also used culture-specific and patient-centered approaches for recruitment to increase the diversity of youth in research (Zamora et al., 2016; Winter et al., 2018). Alternatively, the recruitment of sufficient sample sizes can allow for age, gender, and ethnicity-specific analyses (Winter et al., 2018).

Step Three: Standardized Measurement

Once the field develops fewer, more well-established measures with clearly defined coping responses grounded in theory, researchers may consider the benefits of using a publicly available registry. This uniform approach has been adopted by the patient-reported outcomes measurement information system (PROMIS®) to measure pain sensations (i.e., intensity and quality), interference, and behaviors in the context of chronic pain (Jacobson et al., 2015; Askew et al., 2016; Witter, 2016; Singh et al., 2019). Registered coping measures specific to youth with chronic pain may help to establish a unified understanding of coping, meaningful and relevant measurement tools, and comparability across studies.

Recommendations for Clinical Practice

Although considerable work is needed to improve the clarity and clinical utility of the pediatric pain-coping literature, this review can serve as a resource for clinicians. For instance, clinicians can use this review to: (1) access a list of specific coping responses; (2) locate relevant research; and (3) identify and select measures for assessing the use of pain-coping responses in youth. However, there is currently, no best way to measure and conceptualize coping responses in this population. Instead, clinicians are encouraged to select measurement tools that have strong psychometric properties when used with their target population (i.e., age, pain condition) and based on their content (e.g., specific questions). For example, the PRI was developed for use in youth with abdominal pain, and therefore, a clinician working in a gastroenterology service may implement the PRI in his or her clinical practice. Alternatively, by comparing the item-level content for "seeking social support" on the PRI and PCQ, for example, it is apparent that the PCQ is better able to distinguish between the types of companions (friends vs. family) than the PRI. Therefore, the clinician may select the measure based on the goals of his or her assessment. Regardless, clinicians are encouraged to be intentional about their measure choice. In addition, clinicians can supplement their assessments with open-ended questions that create opportunities for patients to share information about more culturally relevant and person-specific approaches to coping. Furthermore, clinicians can play an important role in building consistency to the field by using evidence-based terminologies and conceptualizations of coping responses with their patients in a way that aligns with how they are intended to be used.

STRENGTHS AND LIMITATIONS

This review is the first to evaluate the clarity and consistency of the measurement and conceptualization of coping responses in youth with chronic pain. Strengths include the comprehensive search strategy and rigorous methodology used to identify and summarize published research. The findings of this review will allow researchers to design studies to address research gaps and inform more consistent and targeted assessments and conceptualizations of coping responses.

There are also limitations. One inherent limitation of a scoping review approach is that it does not formally evaluate the quality of evidence or allow for conducting a comprehensive synthesis of research findings (Pham et al., 2014; Munn et al., 2018). Therefore, concrete guidance on how best to measure or reconceptualize coping responses in the literature is beyond the scope of this review. In addition, this review did not consider the order and mode of administration (e.g., online, paper, and verbal), which may impact the types of coping responses reported by youth (Bowling, 2005). Also, we did not include a comparison between measures; however, it is critical to consider how assessments of frequency vs. duration vs. intensity may

influence how youth report on the same coping response (Stone et al., 1991).

Another limitation is the potential magnification of the extent of discrepancies. A set of strict criteria was used due to the lack of clarity as to which discrepancies hinder the reliability and validity of the results. For example, the most common discrepancy for measurement consistency was using a sample outside the age range validated by the scale. The extent to which a particular measure can be used in a sample of participants even 1 year outside of the age range is unclear.

Furthermore, there are factors that may have limited the breadth of this review. For instance, this review did not take into account studies that were not available in English, which may have limited the inclusion of research on geographically and ethnically diverse patient populations. In addition, coping is a nebulous concept; the distinction between thoughts and behaviors that would constitute coping as opposed to adaptation or self-management is not always obvious (Audulv et al., 2016). Likewise, coping responses are often confused with interventions for chronic pain (e.g., mindfulness training, medications, and exercise therapy) or self-care routines (e.g., getting enough sleep). Given that this review attempted to focus exclusively on studies that measured coping responses, the lack of clarity of how to differentiate coping responses from similar concepts may have led to the exclusion of relevant studies.

CONCLUSION

This review was a necessary first step toward providing concrete guidance on how best to measure or reconceptualize coping responses in the literature on pediatric chronic pain. The results demonstrate the complexity of the literature and highlight gaps and inconsistencies across studies. These gaps are underscored by the lack of theories and definitions/examples of coping responses across studies, which makes it challenging to interpret and apply research findings. Additionally, the wide range of measurement tools and inconsistencies in their use further contributes to the confusing state of this literature. It is recommended that future research prioritize the development and testing of theories and measures of coping responses for pediatric chronic pain. These efforts should be an iterative and interactive process and include a wider range of participants and cultures. Ultimately, the implementation of standardized measures grounded in theory with clear definitions of coping responses is critical to establish clearer and more consistent conceptualizations of coping in the field.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author/s.

AUTHOR CONTRIBUTIONS

This work was conducted as the master's thesis of ANN under the supervision of CMM. BAM is an advisory committee member. ANN, RMT, and CMM conceived the project with feedback from BAM. ANN and RMT conducted the searches. ANN analyzed results and wrote the manuscript. All authors read, edited, and approved the final manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2021.680277/full#supplementary-material

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Targeting Coping to Improve Surgical Outcomes in Pediatric Patients With Median Arcuate Ligament Syndrome: Feasibility Study

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Background: Median arcuate ligament syndrome (MALS) is a vascular compression syndrome leading to postprandial epigastric pain, nausea, and weight loss; it can be treated surgically. While most patients report improved quality of life following surgical intervention, 30% continue to experience chronic abdominal pain. Pre-surgical diagnoses of depression and/or anxiety have been found to significantly predict post-surgical: quality of life, highest experience of pain, anxiety, and parent- and self-reported coping strategies. As such, increasing the coping strategies of pediatric patients with MALS may impact their post-surgical outcomes. The purpose of the current study was to: (1) implement a pre-operative cognitive behavioral therapy protocol with a focus on psychoeducation and coping strategies; and (2) determine feasibility of a pre-surgical intervention for this population.

Method: Children (<18 years of age) with a diagnosis of MALS who were eligible for surgical intervention were invited to participate in a 7-week in-person or video-based pre-surgical cognitive behavioral therapy intervention. Psychiatric comorbidities were assessed at baseline and post-surgery; patient-reported distress, pain interference and intensity, health-related quality of life, and health status were assessed at four time points (baseline, week 4, week 7, and post-surgery). Descriptive analyses were used to characterize the sample, assess feasibility outcomes (i.e., attrition rates), and explore symptom-based outcomes across time.

Results: Twelve pediatric patients (*M* age = 15.2 ± 1.7; 91.7% female) and their parents (91.7% mothers) participated. Feasibility metrics based on protocol completion were exceeded for engagement at the stages of consent (68.4% vs. goal of ≥50%), treatment initiation (92.3% vs. 85%), and treatment completion (84.6% vs. 75%). Out of the 12 participants, nine (75%) met criteria for at least one comorbid psychiatric

diagnosis at baseline and nine (75%) elected to undergo MALS surgery after completing the intervention.

Conclusion: The intervention implementation was feasible, despite chronic pain symptoms experienced by the sample, a high prevalence of psychiatric diagnoses, and an international pandemic, suggesting that it would be beneficial to further evaluate the efficacy of the intervention. Future research should include stakeholder input in the design, deployment, and evaluation of a pilot efficacy trial of pre-surgical cognitive behavioral therapy for pediatric patients with MALS.

Keywords: median arcuate ligament syndrome, chronic abdominal pain, pediatric, cognitive behavioral therapy, coping

INTRODUCTION

Chronic abdominal pain (CAP) is a common medical concern among children and adolescents, with 8–20% of youth reporting pain symptomatology so severe that daily functioning is impeded (Perquin et al., 2003). CAP is associated with many negative psychosocial outcomes, including increased risk of depression and anxiety (Campo et al., 2004), decreased health-related quality of life (QOL; Warschburger et al., 2014), diminished perceived social competence (Scharff, 1997; Eccleston et al., 2008), frequent school absences (Ramchandani et al., 2007), diminished school-related functioning (Greco et al., 2007), and higher levels of peer victimization (Greco et al., 2007). In those with frequent abdominal pain (Scharff, 1997), as well as across children with diverse, chronic pain diagnoses (e.g., cystic fibrosis, juvenile chronic arthritis, diabetes melitus, osteogenesis imperfecta; Meijer et al., 2000; Palermo et al., 2014; Jones et al., 2021), endorsement of pain is associated with withdrawal from social activities. Generally, young people with chronic pain report a sense of loneliness and feelings of difference from their peers due to their pain experiences (Jones et al., 2021). Moreover, children who seek treatment for abdominal pain are more likely to experience physical discomfort, mental health concerns, and associated psychosocial impairment that persists into adolescence and adulthood (Campo et al., 2001). As such, timely and accessible psychological interventions are critical for youth with CAP.

While CAP is often not attributable to any organic disease (Di Lorenzo et al., 2005), one possible cause of CAP for some pediatric patients is median arcuate ligament syndrome (MALS). MALS is a vascular compression syndrome which occurs when the celiac artery is compressed by the diaphragm, leading to symptoms such as postprandial epigastric pain, nausea, and weight loss (Mak et al., 2013). The condition is rarely diagnosed, potentially due to the complexity associated with MALS being a diagnosis of exclusion, the poor understanding of the pathophysiology of pain associated with MALS, unpredictable treatment response, and lack of knowledge about the syndrome and its treatment (Skelly et al., 2018). Notably, while radiographic features of celiac artery compression are noted in upward of 50% of patients (Szilagyi et al., 1972), a much smaller percentage of individuals report the aforementioned symptoms or clinical profile associated with this diagnosis (e.g., 3.5%; Koç et al., 2018) and many report multiple barriers to diagnosis and treatment (Stiles-Shields et al., 2021), making both estimates of prevalence of MALS and clinical diagnosis challenging.

To receive a MALS diagnosis, patients must undergo a multipart diagnostic process involving radiologic studies and an extensive gastrointestinal workup (e.g., duplex ultrasound, computed tomography angiogram; Mak et al., 2013). Following this process and diagnosis, the MALS treatment team might recommend surgical release of the celiac artery. Surgery is voluntary and is therefore approached as a discussion with patients and their families, as the intervention does not guarantee full symptom relief (Skelly et al., 2018). Indeed, while the majority of patients report symptom relief and increased QOL following surgical intervention, about one third continue to experience CAP (Mak et al., 2013; Pather et al., 2020). Additionally, about half of pediatric patients who undergo surgery for MALS meet criteria for at least one psychiatric disorder, including depression and/or anxiety; these psychiatric symptoms do not appear to improve with surgical intervention (Mak et al., 2016; Stiles-Shields et al., 2018). Further, pre-surgical psychiatric diagnoses have been found to significantly predict lower post-surgical QOL for both pediatric and adult samples with MALS (Skelly et al., 2018; Stiles-Shields et al., 2018).

Due to the complexities and comorbidities of MALS, some teams have adopted an interdisciplinary assessment and treatment approach, consisting of a general pediatric surgeon, vascular surgeon, pain specialist, and psychologist (Stiles-Shields et al., 2018). This model is not universal across treatment settings. However, an interdisciplinary approach that includes a pre-surgical psychological evaluation has initial patient support as a beneficial practice (Stiles-Shields et al., 2021) and increases the likelihood of the identification of psychiatric comorbidities. While a pre-surgical psychological intervention is not always required to move forward with a MALS surgery, even when comorbidities are identified, it is possible that increasing the coping strategies of treatment-seeking pediatric patients with MALS may impact their post-surgical outcomes (Stiles-Shields et al., 2018). However, to date, there have been no psychological interventions designed specifically for this population.

For those individuals whose chronic pain persists post-surgically, effective coping strategies developed prior to surgery may be imperative for postoperative daily functioning. While not previously evaluated in pediatric MALS, it should be noted

that in other pediatric populations undergoing surgery (e.g., osteosarcoma; Allen et al., 2020), openness to psychological interventions related to chronic post-surgical pain is associated with shorter length of pain treatment. Despite this, to date, the majority of interventions developed to prevent the development of pediatric post-surgical chronic pain have focused on hypnosis, which has proved "possibly efficacious" at best (Accardi and Milling, 2009). However, in adult populations, psychological interventions, including acceptance and commitment therapy (ACT; Weinrib et al., 2017) and cognitive behavioral therapy (CBT; Landry et al., 2020), are known to reduce use of opioid medication post-operatively, and diminish likelihood of onset of new pain. Such findings are promising, as they suggest that with developmentally appropriate adaptations, similar interventions which address pain-related cognitions may be useful in optimizing pain-related post-surgical outcomes in pediatric patients with MALS.

A likely intervention candidate for pediatric patients with MALS is CBT, which has been established as an evidence-based treatment for pain, including pediatric CAP, and has been found to simultaneously increase health-related QOL and decrease pain catastrophizing (Levy et al., 2010; Palermo et al., 2010; Thorn et al., 2011). In general, CBT for pain management consists of psychoeducation about the pain cycle, training in cognitive and behavioral strategies, and developing a long-term maintenance and relapse-prevention plan (Keefe, 1996). First, CBT for pain presents the cognitive triad, explaining that behavior and cognitions contribute to the experience of pain to emphasize the control the individual holds within their pain experience. Next, coping skills training occurs, which focuses on relaxation (e.g., progressive muscle relaxation, brief guided visualizations), cognitive restructuring (i.e., identifying and challenging catastrophic, pain-related cognitions), and activity management (i.e., activity pacing, pleasant activity scheduling). Finally, CBT for pain ensures the practice of implementing such strategies, as well as anticipating and problem solving for potential challenges and pain flares. To the best of our knowledge, no study has assessed the effectiveness of a psychological intervention in improving post-surgical outcomes for pediatric patients with MALS. Given that this population has similar psychosocial profiles to those with CAP (Sanders et al., 1994; Humphreys and Gevirtz, 2000; Robins et al., 2005; van der Veek et al., 2013), pediatric patients with MALS may also benefit from CBT.

Consideration of how CBT may be readily adapted and delivered for a disease population is crucial. Naturally, there may be treatment barriers when attempting to engage children with MALS in a pre-surgical psychological intervention. First, since MALS is a rare diagnosis, families often travel long distances to reach a MALS specialist (Stiles-Shields et al., 2021) additional and frequent travel for psychotherapy might be burdensome. Second, given the difficulties in reaching a diagnosis, patients and their parents may experience healthcare fatigue, which discourages them from further engaging with mental healthcare providers to receive psychotherapy. Third, patients with MALS experience chronic pain which impedes their daily functioning to varying degrees. Some patients find a task as simple as getting out of bed or taking a shower to be extremely difficult, whereas others might accomplish such tasks with limited interference (Stiles-Shields et al., 2021). Finally, motivation to engage with a psychological intervention may be limited based on patient- and parent-held beliefs about the nature of chronic pain. Given that the presenting pain is associated with a physiological anomaly (i.e., compression of the celiac artery), patients and their parents may ascribe to the biomedical model of pain. This emphasis on the biological aspects of disease may lead to a familial focus on signs and symptoms of the diagnosis, with attempts to alleviate distress primarily via correction of the underlying pathology (Craig and Weiss, 1990; Craig et al., 1996; Bendelow, 2013). As biomedical models favor pharmaceutical interventions, a view of MALS through this lens may discourage engagement with and beliefs about the utility of CBT as an effective component of treatment. However, as such models are known to neglect consideration of social determinants of pain, as well as the role of social learning in expression and experience of pain (Craig et al., 1996), interventions which provide psychoeducation surrounding the nature of pain and the pain cycle may serve to introduce patients to a more readily received transactional model of stress (Lazarus and Folkman, 1984; Craig et al., 1996), which teaches patients to better control their chronic pain by adapting their thoughts, judgments, and beliefs (Craig et al., 1996). In sum, a psychological intervention for pediatric patients with MALS would require a flexible approach, adapting to the barriers and needs of each patient. Offering remote delivery options for interventions (e.g., including telehealth or the use of digital platforms) may be one way to reduce the difficulty of accessing psychological services and overcome symptom-based (e.g., pain interference) and practical treatment barriers (e.g., distance to treatment site; Palermo et al., 2018). Further, an intervention would require psychoeducation and practical skills for the experience of daily pain.

Given the aforementioned barriers to treatment, as well as a lack of interventions specific to those with MALS, establishing the feasibility of a pre-operative CBT intervention for MALS is warranted. Determining feasibility for this pediatric disease population is crucial, given promising outcomes observed in the study of pre-operative psychological interventions in other populations with chronic pain. For example, children with chronic hip pain who received a pre-surgical, psychologically focused pain management intervention reported significantly lower postoperative pain intensity, as well as less pain distress and sleep disturbances (Berge et al., 2004). Additionally, other pre-surgical psychological interventions for elective surgical patients that included components of CBT for chronic pain (i.e., guided imagery) have demonstrated some support for improved psychological well-being and lower post-operative pain levels (Nelson et al., 2013). Such findings suggest that when CBT is adapted to address the needs and barriers of a disease population, pre-operative CBT may be an accessible, inexpensive and low-risk intervention through which psychosocial functioning may be scaffolded post-surgically. Given the dearth of literature on the feasibility of CBT in patients with MALS and the relative infrequency with which this diagnosis is made, a preliminary study which focuses on addressing said barriers and determining

feasibility in a small sample is a crucial first step in determining the utility of pre-surgical CBT for MALS. Such a study allows for consideration of barriers to treatment (e.g., accessibility), allowing for adaptations and improvements to the delivery of treatment prior to engagement of a larger pilot or randomized controlled trial.

Pediatric patients with MALS present with chronic pain, lowered QOL, and frequently occurring comorbid psychiatric diagnoses, which may persist post-surgically. Therefore, the purpose of the current study was to: (1) implement a pre-operative CBT protocol with a focus on psychoeducation and coping strategies aimed to improve pain management, decrease subjective pain, and potentially reduce the need for surgical intervention; and (2) determine feasibility of a pre-surgical intervention for pediatric patients with MALS, with particular attention paid to adherence and patient election of whether to have surgery following intervention. Previous trials of CBT for pediatric chronic conditions have encountered enrollment refusal rates ranging from 0 to 75%; initial follow-up attrition ranges of 0–54%; and 0–59% for extended follow-up attrition rates (Karlson and Rapoff, 2009). We therefore hypothesized that feasibility would be established for pediatric patients with MALS based on the following metrics: (1) at least 50% consent to treatment when offered; (2) at least 85% of those who consent initiate treatment (i.e., complete at least one session); and (3) at least 75% complete treatment. While beyond the scope of a feasibility trial to assess impacts to clinical outcomes, we also aimed to characterize patient-reported symptoms across treatment (i.e., QOL, distress, pain, functioning, psychological characteristics).

MATERIALS AND METHODS

Participants

Pediatric patients (<18 years of age) presenting to Comer Children's Hospital, diagnosed with MALS, and who were eligible for surgical intervention based on an interdisciplinary assessment (Mak et al., 2013; Stiles-Shields et al., 2018) were invited to participate in a 7-week in-person or video-based pre-surgical CBT trial. Participants were excluded if they: (1) had difficulty reading, speaking, or understanding English; (2) were considered too medically unstable for outpatient psychotherapy; (3) were diagnosed with a psychotic disorder; (4) endorsed active suicidal or homicidal ideation; and/or (5) had a recent or planned change in antidepressant medication. A minimum age criterion was not enacted, as pediatric patients with MALS presenting for pre-surgical evaluation tend to be adolescent, as opposed to younger, pediatric patients.

Procedure

In compliance with The University of Chicago Institutional Review Board, informed consent was obtained prior to the baseline assessment from the patient's parent or guardian. Assent was obtained from all pediatric patients; any pediatric patients who turned 18 years old during the study period were re-consented as adults. Psychiatric comorbidities were assessed at baseline and post-surgery using a validated and structured clinical interview. Patient and parent-reported distress, pain interference and intensity, health-related QOL, and health status were assessed at four time points: baseline, following 4th session of CBT (mid-treatment), following 7th and final session of CBT (end of treatment), and 1 month after surgery. If patients and their families did not elect to undergo surgery, their 1-month post-surgical assessment was administered as soon as the decision was made and communicated with the research staff. Participants were compensated via gift card for study assessment completion. Specifically, they earned $25 for the baseline assessment (questionnaires and interview), $10 for the end of treatment/week 7 of CBT assessments (questionnaires only), and $25 for the post-surgery assessment (questionnaires and interview).

Pre-surgical Cognitive Behavioral Intervention

All participants underwent a seven-session, pre-surgical CBT protocol (see **Table 1**). Patients and their families could elect to engage in the intervention via in-person or video visits (conducted via a HIPPA-compliant Zoom account). Cognitive targets included identifying and challenging automatic thoughts related to pain, nausea, and stress, labeling and challenging cognitive distortions, and identifying and managing internalized and externalized stressors. Behavioral targets included coping/relaxation skills, pleasant activity scheduling, and time-based pacing. These targets were addressed across seven sessions. The intervention duration of seven sessions was selected to balance the need to address the core tenets of a CBT intervention for chronic pain with minimizing burden on patients (including their wait time to surgery). The intervention was administered by a postdoctoral fellow or master's level psychology doctoral student, and was conducted with the pediatric patient alone, without a parent present. A licensed clinical psychologist supervised all treatment. All sessions were audiotaped and randomly selected for review by a clinical supervisor. Treatment fidelity was ensured utilizing the CBT scale for children and young people (CBTS-CYP; Stallard et al., 2014). Participation in the study was voluntary and did not affect the patient's MALS care in any way. Recruitment was temporarily suspended mid-study (March 2020–September 2020) due to logistical challenges caused by the COVID-19 pandemic, including a temporary pause in funding.

Surgical Intervention

At the end of the 7-week intervention period, patients underwent an updated history, and physical and informed consent for surgical intervention was obtained. If surgery was still indicated and the family elected to proceed, minimally invasive (laparoscopic) surgical release of the median arcuate ligament was performed (please see Mak et al., 2013 for more details about the surgical intervention).

Measures

All self-report assessments were administered and managed via the REDCap (Research Electronic Data Capture; Harris et al., 2009).

TABLE 1 | Session goals and strategies of pre-surgical CBT intervention.

Session	Goals	Strategies
Session 1: Psychoeducation about pain	Identify ways in which pain has impacted activities, thoughts, and feelings	Illustrate concepts using pain impact sheet; draw diagram showing cycle between pain, distress, and disability
Session 2: Progressive muscle relaxation and visual imagery	Introduce coping skills and relaxation techniques	Practice diaphragmatic breathing, PMR, and visual imagery in session
Session 3: Automatic thoughts and pain	Understand the relationship between thoughts, emotions, and pain	Introduce cognitive errors; Introduce ABC model
Session 4: Cognitive restructuring	Teach patient to challenge maladaptive thoughts about stress and pain	Use completed ABC worksheet to identify and challenge automatic thoughts related to pain; Use Thought Challenger worksheet
Session 5: Time-based pacing	Provide psychoeducation on importance of breaks and pacing techniques to prevent increased pain and later avoidance	Illustrate concept using Activity Pacing worksheet
Session 6: Pleasant activity scheduling	Understand the role of pain in activity withdrawal and low mood	Help patient identify and schedule activities he/she enjoys that are realistic and achievable
Session 7: Relapse prevention and flare-up planning	Normalize pain relapse; review progress	Discuss past pain relapses; collaboratively create plan for future relapses

CBT, cognitive behavioral therapy; ABC, Activating Event, Beliefs, Consequences Model; PMR, progressive muscle relaxation. All sessions were conducted with the pediatric patient alone.

Interviews

Mini International Neuropsychiatric Interview for Children and Adolescents

The Mini International Neuropsychiatric Interview for Children and Adolescents (MINI-Kid) is a brief, structured diagnostic interview. Diagnoses are based on the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5) and International Statistical Classification of Diseases and Related Health Problems, Tenth Revision (ICD-10) criteria (Sheehan et al., 2010). The MINI-Kid was administered to pediatric participants by master's level doctoral students and supervised by a licensed clinical psychologist.

Median Arcuate Ligament Syndrome Diagnostic Interview

The MALS Diagnostic Interview is a semi-structured interview designed for administration to pediatric patients and their families prior to and following surgical intervention for MALS (Anderson et al., 2011; Stiles-Shields et al., 2018). The MALS Diagnostic Interview was administered by master's level doctoral students and supervised by a licensed clinical psychologist. Clinical information from this interview was used to supplement data from the MINI-Kid (i.e., provide parental/multi-informant input to diagnostic questions) to inform mental health diagnoses.

Self-Report Questionnaires

Demographic Characteristics

All pediatric participants were asked to report age, gender, current medical diagnoses, race/ethnicity, current living situation (e.g., with parents or independently), current work/school attendance, and socioeconomic status (SES). Demographic characteristics were administered at each time point.

Distress

The Kessler Psychological Distress Scale (K10) is a 10-item self-report questionnaire designed and validated to measure distress in youth and adults (Kessler et al., 2002, 2003). Scores range from 10 to 50, with scores categorized as non-clinical (<20), mild (20–24), moderate (25–29), and severe (30+). The K10 has previously been administered to pediatric and young adult patients with MALS (Stiles-Shields et al., 2021) and reliability was acceptable for the current pediatric sample at baseline ($\alpha = 0.86$).

Quality of Life

The Child Health Questionnaire-87, child version (CHQ-CF87) is a self-report health status and QOL measure designed for children and adolescents, ages 5–18 (Landgraf and Abetz, 1997; HealthActCHQ, 2013). The CHQ-CF87 contains 12 concepts, including two single items: global health and change in health; and 10 multi-item scales: physical functioning; role/social limitations-emotional, behavioral, and physical; bodily pain/discomfort; behavior; global behavior; mental health; self-esteem; general health perceptions; family activities; and family cohesion. The CHQ-CF87 has previously been used with a small sample of pediatric patients with MALS (Joyce et al., 2014) and the measure has demonstrated acceptable to high reliability in school-based and clinical samples (attention-deficit/hyperactivity disorder; cystic fibrosis, end stage renal failure; HealthActCHQ, 2013). Authorization and a completed license to use the CHQ-CF87 was obtained for this study. Reliability for the CHQ-CF87 was acceptable at baseline for all concepts (αs = 0.76–92), with the exception of bodily pain/discomfort ($\alpha = 0.55$). However, we have opted to still include the reporting of this two-item subscale to provide the characterization of this construct.

The Pediatric Quality of Life, version 4.0, child and teen reports (PedsQL) measures health-related QOL (Varni et al., 2001). The PedsQL yields a total score and subscale scores for physical, emotional, social, and school/work functioning. Higher scores indicate higher QOL, with a range of scores from 0 to 100. The PedsQL has been previously administered to pediatric and

adult patients with MALS (Mak et al., 2013, 2016; Skelly et al., 2018; Stiles-Shields et al., 2018). The PedsQL child and parent versions were administered at all time points and demonstrated acceptable reliability at baseline ($\alpha = 0.88$).

Pain

The PROMIS Pediatric Pain Intensity Form measures self-reported pain intensity as a one-item question for pediatric patients (Broderick et al., 2013). The PROMIS Pediatric Pain Interference Form measures the impact of pain on social, cognitive, emotional, physical, and recreational activities (Amtmann et al., 2010). The PROMIS Pediatric Pain Interference Form demonstrated acceptable reliability at baseline ($\alpha = 0.81$). For both measures, higher scores indicate greater pain intensity or hindrance, respectively.

Data Analysis

Descriptive analyses were used to characterize the sample, assess feasibility outcomes (i.e., attrition rates), and explore symptom-based outcomes across time (e.g., QOL).

RESULTS

Participants

The flow of participants through the feasibility trial is depicted in **Figure 1** and sample characteristics are presented in **Table 2**. The sample for this study included 12 pediatric patients diagnosed with MALS, ages 13–17 ($\boldsymbol{M} = 15.2 \pm 1.7$). The majority of the sample was female (91.7%), non-**H**ispanic/Latinx Caucasian (91.7%), and participated in treatment with their mothers (91.7%). The intervention trial began in August 2019, pausing due to the pandemic from March 2020 to September 2020. Two participants elected to complete the intervention face-to-face (16.7%), with the remainder opting for remote delivery.

Feasibility

Sixty-one patients presented to the interdisciplinary MALS team at Comer Children's Hospital and The University of Chicago Medicine for a pre-surgical evaluation during the feasibility trial recruitment phases. Of these, 19 (31.1%) were under the age of 18 and met the criteria for a diagnosis of MALS. Six of these 19 pediatric patients and their families declined participation, expressing: (1) immediate disinterest in participation ($n = 1$; 16.7%); (2) delayed disinterest and/or lost to follow-up contact ($n = 3$; 50%); (3) disinterest due to being too busy ($n = 1$; 16.7%); or (4) disinterest due to seeking surgical intervention at a different location ($n = 1$; 16.7%). The other thirteen pediatric patients and their families consented to the study (68.4%). Nearly all consented participants initiated treatment ($n = 12$; 92.3%). Of these 12 participants, 11 completed the seven-session treatment, resulting in a retention rate of 91.7%. Only one participant dropped from treatment (8.3%), with the dropout occurring prior to the mid-treatment assessment. In terms of surgery, nine out of the 11 participants who completed treatment elected to undergo surgery (81.8%). Out of the two participants who did not have surgery, one is currently waiting for their surgery to be scheduled, while the other elected to not undergo surgical intervention; when the family communicated this decision, their post-surgery assessment was conducted.

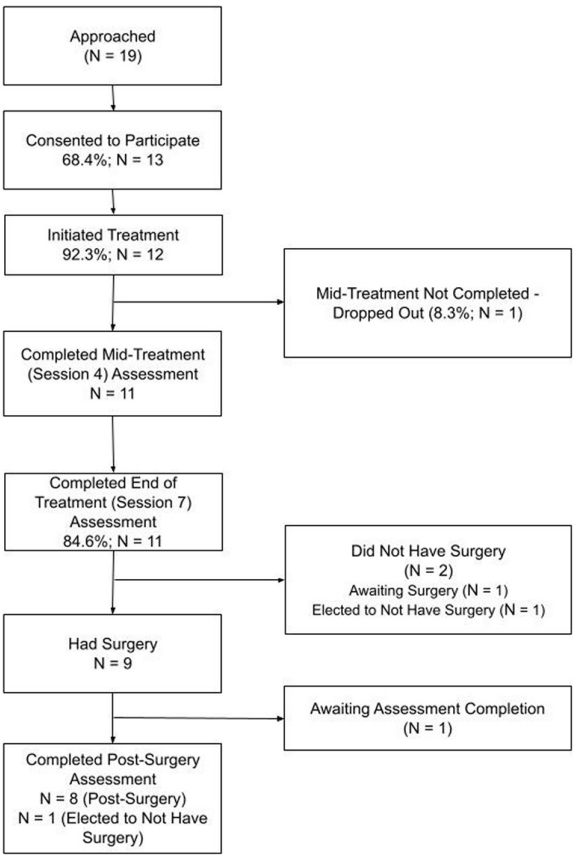

FIGURE 1 | Consort chart for feasibility trial, assessment completion defined by parent and/or child participation.

Patient Mental Health and Psychosocial Characteristics

Mental Health

Three participants did not meet criteria for any psychiatric diagnoses at the baseline interview. Nine participants met criteria for: depressive disorders [major depressive disorder (4), dysthymia (2)]; anxiety disorders [generalized anxiety disorder (3), separation anxiety disorder (2), social anxiety (2), obsessive compulsive disorder (1), agoraphobia (1), specific phobia (1), unspecified (1)]; adjustment disorders [with depressed mood (1)]; and attention-deficit/hyperactivity disorder [combined type (2)]. Average responses on the K10 across assessment time points indicated moderate (i.e., 25–30) to severe psychological distress (i.e., 30+).

Quality of Life and Pain

Table 2 displays the self-reported psychosocial functioning and pain scores across time. Health status and quality of life, as measured by the CHQ-CF87 and PedsQL, were similar at baseline

TABLE 2 | Sample characteristics, M (SD).

	Baseline (*n* = 13)	Mid-treatment (*n* = 12)	End of treatment (*n* = 12)	Post-surgery follow-up (*n* = 10)
Age	15.2 (1.72)	15.9 (1.46)	15.9 (1.60)	16 (1.4)
Gender, *n* (%)				
Female	12 (92.3%)	11 (91.7%)	11 (91.7%)	10 (100%)
Male	0	0	0	0
Non-binary	1 (7.7%)	1 (8.3%)	1 (11.1%)	0
Racial, ethnic identity, *n* (%)				
Non-Hispanic/Latinx Caucasian	12 (92.3%)	12 (100%)	12 (100%)	10 (100%)
Other (not specified)	1 (7.7%)	0	0	0
Highest level of education (grade)	9.1 (1.31)	–	–	–
Household income (US$)	$91,888 ($36,115.94)	–	–	–
Parent participation, *n* (%)				
Mother	11 (91.7%)	–	–	–
Father	1 (8.3%)	–	–	–
CHQ-CF87				
Global health	42.08 (26.24)	44.5 (24.55)	42.78 (29.49)	51.88 (25.63)
Physical functioning	69.75 (19.65)	65.19 (28.91)	55.97 (25.15)	61.57 (30.92)
Role/social limitations-emotional	68.52 (21.10)	58.89 (24.60)	58.02 (26.51)	76.39 (28.13)
Role/social limitations-behavioral	86.11 (19.03)	75.56 (34.27)	82.72 (24.91)	91.67 (16.53)
Role/social limitations-physical	74.07 (17.30)	61.11 (22.98)	59.26 (16.67)	72.22 (29.10)
Bodily pain/discomfort	30.83 (14.43)	21.0 (11.97)	22.22 (8.33)	40.0 (15.12)
Behavior	73.01 (18.21)	73.06 (11.98)	73.66 (14.10)	79.67 (13.64)
Global behavior	76.67 (20.26)	82.0 (16.53)	82.78 (14.39)	82.5 (15.35)
Mental health	57.29 (10.54)	52.03 (12.33)	52.43 (13.40)	63.09 (21.81)
Self esteem	63.54 (18.28)	64.29 (13.52)	62.30 (13.96)	70.54 (21.41)
General health perceptions	38.75 (17.90)	39.33 (17.02)	37.59 (15.25)	44.69 (19.13)
Change in health	2.25 (0.75)	2.2 (0.92)	2.22 (1.09)	3.75 (1.16)
Family activities	58.68 (20.91)	54.17 (20.31)	61.57 (16.37)	61.46 (19.38)
Family cohesion	3.92 (1.12)	3.66 (1.14)	4.16 (1.07)	4.05 (0.73)
PedsQL				
Physical	47.66 (14.0)	51.70 (20.56)	49.38 (15.92)	56.60 (19.73)
Emotional	51.57 (13.37)	49.09 (12.61)	49.5 (14.99)	60.0 (23.85)
Social	75.0 (24.03)	78.64 (21.34)	73.0 (21.11)	81.11 (18.16)
School	47.5 (12.34)	42.73 (16.49)	50.0 (16.67)	62.78 (30.43)
Total	54.44 (12.34)	55.04 (15.53)	54.67 (13.87)	64.0 (21.0)
K10	24.9 (6.02)	27.3 (6.63)	26.5 (6.08)	20.6 (8.03)
PROMIS pain intensity (0–10 scale)	6 (1.41)	5.8 (1.47)	6 (0.81)	4 (2.56)
PROMIS pain interference T-score	60.13 (5.41)	60.77 (4.93)	59.36 (6.41)	51.26 (9.88)

to previous samples of pediatric patients with MALS (Joyce et al., 2014; Stiles-Shields et al., 2018). At baseline, participants rated their pain on average at a "6" (SD = 1.41) on a 0–10 scale on the PROMIS Pediatric Pain Intensity Form, while T-scores on the PROMIS Pediatric Pain Interference Form indicated primarily normative pain hindrance in engaging with social, cognitive, physical, and recreational activities.

COVID-19 Related Considerations

Study therapists informally queried patients during the final session regarding impacts of the COVID-19 pandemic on the efficacy of the intervention, session structure, and patient concerns and related symptom presentation. Patients endorsed increased concerns about health and safety, the need to adapt sessions related to time-based pacing and pleasant activity scheduling (due to changes in available resources and need for social distancing), diminished pain intrusion due to having fewer functional demands, and diminished daily structure.

DISCUSSION

The current study aimed to implement a pre-surgical CBT protocol emphasizing psychoeducation and pain coping strategies, determine feasibility of engaging pediatric patients with MALS in this intervention, and characterize the sample that engaged. Implementation began prior to the COVID-19 pandemic and included a remote delivery option, which promoted the ability to resume the intervention following a pandemic-related delay. Feasibility metrics were exceeded for engagement at the stages of consent (68.4% consented to

treatment vs. goal of at least 50%), treatment initiation (92.3% completed at least one session vs. goal of at least 85%), and treatment completion (84.6% completed all sessions vs. goal of 75%). Additionally, nine participants (75%) elected to undergo MALS surgery following the CBT intervention. Of the patients and families who opted to participate in the intervention, pain experiences, QOL, and functioning were comparable to previous samples of pediatric patients with MALS (Joyce et al., 2014; Stiles-Shields et al., 2018). Further, mental health comorbidity was high, with 75% of the sample meeting criteria for at least one psychiatric diagnosis at baseline.

It was anticipated that multiple adaptations would be needed to effectively engage pediatric patients with MALS in a pre-surgical CBT intervention. Namely, a flexible approach in delivery options (face-to-face, video sessions) and emphases on pain psychoeducation and skill building around daily pain experiences and interferences. With the onset of the COVID-19 pandemic, remote methodologies became widely adopted and encouraged for clinical research efforts with pediatric populations (Stiles-Shields et al., 2020). However, at the onset of this feasibility trial, which predated the pandemic, additional efforts were required to justify to the Institutional Review Board (IRB) the need for a flexible delivery approach for pediatric patients experiencing CAP. We suspect that moving forward, many IRBs and institutions more broadly, will be open to remote delivery options. During treatment, multiple individual adaptations were made in response to patient-driven questions: (1) addressing symptoms related to comorbid conditions [e.g., Postural Orthostatic Tachycardia Syndrome (POTS)]; (2) means to improve medication adherence; and (3) how to plan for post-surgical adjustment (e.g., "How do I learn to eat and not be scared of food post-op?" "Will my hunger/fullness cues come back?"). While many of the questions were able to be easily woven into the established treatment, the questions regarding transition to post-surgical functioning support the likely benefit of offering supplemental post-surgical sessions.

Not surprisingly, the COVID-19 pandemic also appeared to impact the patient experience during the study period. First, multiple patients reported impacts to the health and safety of themselves and/or their family members (e.g., COVID-19 positive diagnoses). The pandemic may be viewed from a medical trauma perspective (Kazak et al., 2021), and as such, future adaptations of this protocol should consider these impacts regardless of the pandemic status (e.g., following a "return to normal"). Second, to comply with social distancing recommendations, time-based pacing and pleasant activity scheduling (sessions five and six) were particularly impacted. Some patients who participated in treatment early in the pandemic expressed initial feelings of relief that school attendance or social activities could be avoided, particularly during high pain days. As the pandemic continued, some patients reported that pain symptoms were less disruptive to their daily functioning, as they were not alone in missing typical activities (e.g., school, social outings). However, consistent with other youth populations during the pandemic (Brooks et al., 2020; Patrick et al., 2020), diminished daily activity structure relating to social distancing practices appeared to generally impact patients' reported mood states and pain flare-ups. As such, study therapists became more creative in their collaborations with patients across sessions five and six to plan for in-home, near-by, and virtual activities that could provide structure, feelings of accomplishment/pleasure, and complied with social distancing recommendations. Considerations should be made following the pandemic to account for a potentially expansive view of positive activities that may be conducted virtually without interfering with the benefits of exposure to typical social activity experiences when in pain.

Pediatric samples with MALS commonly endorse comorbid psychiatric symptoms both pre- and post-surgically (Mak et al., 2016; Stiles-Shields et al., 2018). Further, depressive and anxious disorders are common in both pediatric CAP and chronic pain, more broadly (Youssef et al., 2008; Shelby et al., 2013; Fisher et al., 2018; Soltani et al., 2019). It was therefore anticipated that the sample would likely have a high prevalence of mental health diagnoses. Indeed, 67.7% met criteria for at least one DSM-V diagnosis at baseline. Establishing feasibility of engaging pediatric patients with MALS who are simultaneously experiencing comorbid psychiatric diagnoses is important, as the clinical team has reported a long-standing history of their recommendations for pre-surgical psychological interventions being met with resistance. Indeed, consistent with a focus on a biomedical model of pain (Craig and Weiss, 1990; Craig et al., 1996; Bendelow, 2013), the team reported that many pediatric patients with MALS and/or their parents would insist that surgical release of the celiac artery would improve the symptoms of MALS and subsequently, mental health would also improve. Similarly, others insisted that CBT would not be an effective intervention to address pain, as their pain was a result of an overt physiological cause and would therefore be unmodifiable through learning and applying coping skills. The current study therefore adds to a preliminary body of evidence that pediatric patients with MALS will engage in pre-surgical psychological interventions, and may report deriving long-term benefit from doing so (Stiles-Shields et al., 2021).

The current study had multiple strengths, including being the first implementation and evaluation of feasibility for a psychological intervention for pediatric patients with MALS. However, the findings should be interpreted in light of specific limitations. First, the sample was homogenous; it was comprised primarily of non-Hispanic/Latinx Caucasian females who presented with their mothers. While this is not unusual for samples of pediatric patients with MALS (Mak et al., 2013; Joyce et al., 2014; Stiles-Shields et al., 2018), consistency is not cause for complacency. Efforts are required to ensure better: (1) knowledge dissemination about pediatric MALS, (2) screening for MALS, particularly with marginalized populations with CAP, and (3) treatment access for pediatric patients with MALS, paying particular attention to systemic barriers which might be impeding the chance for underserved populations to receive a diagnosis and treatment given the resources needed to complete a diagnostic evaluation for MALS (Stiles-Shields et al., 2021). Second, to establish feasibility, only youth under the age of 18 were included. Consideration of developmental differences within the aforementioned age range is encouraged to readily

evaluate how changes in cognitive development over these crucial years impact response to this intervention. Additionally, it is unclear how the current findings extend to older adolescent and young adult patients with MALS, many of whom presented for evaluation during the recruitment periods and expressed interest in a pre-surgical CBT intervention specific to MALS. Third, additional implementation outcomes and patient satisfaction with the intervention were not formally assessed. Engagement with treatment via treatment and assessment completion markers does not provide a full evaluation of feasibility or likelihood of clinical impact. Eliciting feedback from pediatric patients with MALS and their families, and including their voices in the development of a larger trial (e.g., stakeholder advisory board) is likely to promote better engagement and treatment experience. Fourth, this study evaluated feasibility of an intervention and was therefore not powered to evaluate clinical efficacy. Further, it is unclear how the effects of the COVID-19 pandemic may have contributed to patient-reported assessments and interviews. Given the impact of CBT-based interventions for other pediatric populations with CAP (Palermo et al., 2010; Groß and Warschburger, 2013; van der Veek et al., 2013), it is unlikely that CBT would have no positive effect for pediatric patients with MALS. However, the current study is unable to provide evidence for clinical impacts. Fifth, it must be acknowledged that the present study was underpowered to evaluate treatment efficacy, and findings related to feasibility should not be conflated with a statement of treatment efficacy. Evaluation of treatment efficacy is warranted in a larger, representative sample of pediatric patients with MALS. Finally, and as previously noted, this study was conducted during an international pandemic. Changes in how the medical system responded to the stress of this public health crisis delayed the testing of many patients suspected to have MALS (which is of note, as MALS is a diagnosis of exclusion, requiring multiple diagnostic tests). It is impossible to pull apart the effects of this phenomenon on the patients' experience nor to determine whether the current sample differs from those who may have presented for evaluation under typical circumstances. Future research efforts should include: (1) evaluating the efficacy of pre-surgical CBT intervention on post-surgical outcomes with a larger sample and a longer follow-up (e.g., 6 months post-surgery); (2) evaluating the intervention across developmentally appropriate age groups [i.e., late childhood/early adolescence (9–13), mid-adolescence (14–17), late adolescence/emerging adulthood (18–25)]; and (3) incorporating more assessments of implementation and the generalizability of the intervention to other treatment sites.

The current study demonstrated initial feasibility for engaging pediatric patients with MALS in a pre-surgical CBT-based intervention. Despite a high prevalence of comorbid psychiatric diagnoses, a pandemic, and chronic pain symptoms experienced by patients with MALS, the intervention was deemed feasible based on protocol completion, suggesting that it would be beneficial to further evaluate the efficacy of the intervention with this population. Implementation of the intervention called for some adaptations specific for this population, including the ability to offer sessions both in-person and virtually, a focus on coping and skill building and psychoeducation, and flexibility to manage concerns about comorbid conditions and larger barriers related to the COVID-19 pandemic. Future research will include stakeholder input to conduct a pilot trial evaluating the efficacy of a pre-surgical CBT-based intervention for pediatric patients with MALS.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by The University of Chicago Institutional Review Board. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

CS-S, CS, and TD conceptualized and designed the study. All authors contributed to the drafting of the manuscript and approved the final version.

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BRIEF RESEARCH REPORT
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Coping in Pediatric Burn Survivors and Its Relation to Social Functioning and Self-Concept

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Pediatric burn survivors experience increased risk for bullying, stigmatization, body image concerns, and problematic social functioning. Although coping behaviors are associated with engagement in social supports and positive self-concept in multiple pediatric illness populations, their relation has not been examined in pediatric burns. This study examined coping in relation to social functioning and self-concept in 51 pediatric burn survivors aged 7–17 years ($M = 12.54$; $SD = 2.65$). Survivors and their caregivers completed the *Child Coping Strategies Checklist* (CCSC; youth report); the *Burn Injury Social Questionnaire* (BISQ; parent and youth report); and the *Piers-Harris Children's Self-Concept Scale-2* (PH-2; youth report). Associations between coping, social functioning, self-concept, demographic features, and burn injury characteristics were examined *via* bivariate correlations. Hierarchical linear regressions examined whether coping strategies predicted social functioning and youth self-concept beyond burn injury and demographic variables. Social functioning concerns were positively correlated with total body surface area (TBSA; $r = 0.63$ and 0.40, respectively). TBSA was the only significant predictor of parent-reported social concerns ($\beta = 0.65$, $p < 0.001$). Greater distraction coping predicted fewer youth-reported social concerns ($\beta = -0.39$, $p = 0.01$). Greater active coping ($B = 0.67$, $p = 0.002$) and lower avoidance coping ($B = -0.36$, $p = 0.03$) predicted better youth-reported self-concept. This study advances our understanding of coping as potentially protective for psychosocial adjustment. Clinicians working with child burn survivors should incorporate active coping interventions into treatment. Further research including larger and more diverse samples is needed to understand the role of coping approaches on psychological adjustment during burn healing.

Keywords: adaptive coping, pediatrics, burn survivors, social functioning, self-concept

INTRODUCTION

Burns are among the leading causes of injury and unintentional death in the United States (American Burn Association, 2018). Pediatric populations are particularly vulnerable to burn injuries, with nearly a quarter of burn incidents in the United States occurring in children under 15 years of age (American Burn Association, 2018). Although many burn injuries are not treated medically or can be treated in outpatient settings, severe injuries require hospitalization or transfer to a burn center for critical care (Krishnamoorthy et al., 2012). Treatment of burn injuries can include dressing changes, wound cleansing, medicated salve, pain management, skin grafting, physical and occupational therapies, and pressurized garments (Gill, 2010; Krishnamoorthy et al., 2012). Consequentially, burn care can be medically extensive, painful, and last months to years.

Along with their physical repercussions, burns are associated with impairments in psychosocial functioning that hinder healing, long-term adjustment, and overall well-being (Lavigne and Faier-Rouman, 1992; Pardo et al., 2008; Gill, 2010; Enlow et al., 2019). One domain of psychosocial functioning that can be specifically impacted by burns is social functioning. A systematic review of 75 articles focusing on the psychosocial outcomes of pediatric burn injuries revealed that survivors were at an increased risk for difficulties in social functioning several years post-burn (Bakker et al., 2013). Another study (Andersson et al., 2003) identified that parents of pediatric burn survivors perceived their children as having reduced social initiative, while teachers perceived students who survived burns as having a less prosocial orientation. Additionally, problems with peers were more common in children who had sustained burn injuries compared to the general population (Willebrand et al., 2011). Despite these findings, the extant literature on social adjustment in pediatric burns is not extensive, and this is an area in need of additional study.

Although not all burn survivors experience long-term psychosocial concerns (e.g., Koon, 1993; Bakker et al., 2013), the trauma of the injury itself, the pain of burn care, and potential physical outcomes (e.g., disfigurement, limited mobility) place youth at substantial risk for maladjustment (e.g., Gill, 2010; Kazis et al., 2017). It is critical for caregivers to support pediatric burn survivors such that they may engage in effective coping skills to ameliorate or prevent adverse psychological experiences (Meijer et al., 2002; Gill, 2010; Enlow et al., 2019). Compas et al. (2001) defines coping as "conscious, volitional efforts to regulate emotion, cognition, behavior, physiology, and the environment in response to stressful events or circumstances." Specific psychosocial stressors that may be targeted by coping interventions in pediatric burn survivors include changes to social functioning and self-concept.

The physical sequelae from burns (e.g., visible scarring) can affect the self-concept of burn survivors, particularly during childhood and early adolescence, which is a formative period for a child's sense of self (Koon, 1993). Youth who experience disfigurement from burn injuries have reported deficits in psychosocial functioning, including lower self-esteem, difficulties with self-concept and social skills, and higher levels of anxiety (Gill, 2010). A key factor that influences adjustment in pediatric burn survivors is the reaction of peers and those in public. Burn survivors are more likely to adjust poorly if peers react negatively toward their physical disfigurement (LeDoux et al., 1996), and survivors who experience debilitating physical symptoms may be limited in their social participation (Kazis et al., 2017). Conversely, psychosocial support from family and a trauma-informed healthcare team during the acute phase following a burn injury is important for promoting healthy adjustment (De Sousa, 2010; Bakker et al., 2013).

Coping behaviors are a significant predictor of self-esteem, social skills, social anxiety, and behavioral difficulties in chronically ill patients broadly (Meijer et al., 2002). Negative coping strategies, such as avoidance, have been associated with increased social difficulties in pediatric patients with neonatal brachial plexus injuries (Mentrikoski et al., 2015). Similarly, a cross-sectional study on youth with spinal cord injuries revealed an association between avoidant coping strategies, lower quality of life, and higher anxiety and depressive symptoms (Smith et al., 2013). In contrast, active coping strategies (i.e., seeking social support, problem solving, and openly talking about stressors) are most effective for promoting the psychosocial adjustment of chronically ill pediatric patients (Meijer et al., 2002) and a key to family functioning and support in pediatric oncology patients (Trask et al., 2003; Martin et al., 2012). In the context of pediatric burns, adaptive coping strategies are negatively associated with post-traumatic stress symptoms (Enlow et al., 2019), whereas internalizing (e.g., rumination, fixating on anxieties) and externalizing coping behaviors (e.g., screaming, breaking things) have been related to greater panic and general anxiety disorder symptoms (Rimmer et al., 2015).

Although there are several studies on the psychosocial impacts of pediatric burns, the relations among coping behaviors, social functioning, and self-concept are not well understood in this population. The current study expands upon extant research by addressing the following aims: (1) examine associations among coping behaviors, social functioning, self-concept, and burn injury-related variables; (2) investigate the extent to which coping strategies predict social functioning; and (3) assess the extent to which coping strategies predict self-concept in pediatric burn survivors.

MATERIALS AND METHODS

Participants

Fifty-one pediatric burn survivors (M age = 12.54; SD = 2.65) and their primary caregivers participated. Families were recruited as part of a larger multi-project (e.g., Enlow et al., 2019) study examining the psychosocial outcomes of pediatric burn survivors. Eligible survivors were 7–17 years of age and had sustained a burn requiring medical treatment at least 1 month before recruitment. Survivors and caregivers were English speaking (measures validated only in English). Exclusion criteria included the child or caregiver having significant cognitive impairment,

as reported by the primary caregiver or medical team that would preclude completion of study measures.

Procedure

Researchers obtained hospital and university Institutional Review Board approval before project initiation. Participants were recruited during outpatient clinic appointments at two hospitals in Pennsylvania ($n = 14$) and Ohio ($n = 4$) and during registration for a summer camp in Pennsylvania for pediatric burn survivors ($n = 22$). A recruitment letter and informed consent forms were mailed to burn survivors in the Pennsylvania hospital's burn registry database who met inclusion criteria but were not approached in clinic or at camp (e.g., no longer required follow-up care), which yielded additional participants ($n = 11$).

Caregivers and survivors recruited from clinic and camp each independently completed a questionnaire packet. Caregiver packets included the *Family Information Form* and *Burn Injury Social Questionnaire* (BISQ), while survivor packets included the *Child Coping Strategies Checklist* (CCSC), *BISQ*, and the *Piers-Harris Children's Self-Concept Scale-2* (PH-2). Burn survivors recruited by letter from the hospital database were asked to contact research staff if interested in participating. They completed measures sent by mail, which were addressed individually for parent and child so that their responses could remain confidential from one another. Research staff also obtained data through medical records and the burn registry database.

Measures

Family Information Form

The *Family Information Form* was designed for this study to obtain demographic information (i.e., gender, age, race, education, and socioeconomic status) from caregivers.

Chart Review Form

Research staff documented relevant medical history (i.e., burn characteristics, total body surface area of the burn; TBSA, associated surgeries and hospital stays, and scarring and disfigurement) for patients on the *Chart Review Form*.

Child Coping Strategies Checklist

The CCSC (Ayers et al., 1996) is a 52-item self-report questionnaire that measures coping behaviors over the past month when trying to solve problems. Items in this measure are rated on a 4-point Likert scale ranging from 1 (never) to 4 (most of the time), yielding four subscales: active coping, distraction strategies, avoidance strategies, and support seeking strategies. In our sample, internal consistency was high for the active coping ($\alpha = 0.93$) and support seeking subscales ($\alpha = 0.88$), good for the avoidance strategies subscale ($\alpha = 0.75$), and somewhat questionable for the distraction subscale ($\alpha = 0.67$).

Burn Injury Social Questionnaire

The BISQ is a brief, 10-item self-report screening questionnaire that was devised for this study to assess social functioning after a burn injury (e.g., "Kids tease me about the way I look"; "It is hard for me to have friends or dates because of my burn injury"). Items in this measure are rated on a four-point Likert scale ranging from 1 (not at all true) to 4 (very true). Parent- and child-report parallel versions are available; both versions have a Flesch-Kincaid grade level of below 1.0, suggesting that they are very easy to read and understand. Although this measure has not yet been validated, the items are face valid. Cronbach's alpha for the current sample suggests good internal consistency for the BISQ-P ($\alpha = 0.79$) and BISQ-Y ($\alpha = 0.72$). A total score is calculated for analyses, with higher scores representing more problematic social concerns.

Piers-Harris Children's Self-Concept Scale-2

The PH-2 (Piers and Herzberg, 2002) is a common, validated measure evaluating self-concept in youth. This 60-item questionnaire is rated using a yes/no format and is divided into six subscales: behavioral adjustment, intellectual and school status, physical appearance and attributes, freedom from anxiety, popularity, and happiness and satisfaction. The total standardized *t*-score for the PH-2 was used in analyses ($\alpha = 0.89$ for our sample); higher scores represent higher overall self-concept.

Statistical Analysis

Bivariate correlations, independent samples *t*-tests, and ANOVA were used to examine the associations among coping behaviors, social functioning, self-concept, and demographic and burn injury-related variables (Aim 1). Hierarchical linear regression was used to assess whether coping strategies predicted social functioning (Aim 2) and self-concept (Aim 3). Demographic and burn injury-related variables significantly associated with self-concept and social functioning in bivariate analyses were included as covariates in the respective regression models.

RESULTS

Demographics/Descriptive Statistics

Frequencies (**Table 1**) and descriptive statistics (**Table 2**) for demographic, clinical, and variables are reported for our sample. Due to the non-normal distribution of the family income data, participants were divided into quartiles (1st quartile = ≤$29.9 k, 2nd quartile = $30 k–$49.9 k, 3rd quartile = $50 k–$89, and 4th quartile = ≥$90 k) for analyses. The median income was $40–$49.9 k and the interquartile range was $60–$69.9 k. The most common cause of burn was thermal (40.4%), followed by scald (32.7%). The average TBSA among participants was moderate (i.e., 9.19%), with approximately one-third having received skin graft surgery. Though the sample varied greatly with respect to time since injury (range = 1–141.5 months), participants completed study measures about 33 months, on average, after their injury. Examination of PH-2 total scores revealed that approximately 4% ($n = 2$) of participants had borderline low self-concept T-scores (≥1 SD but <2 SD below

TABLE 1 | Participant ($N = 52$) demographics.

Variable	Value	*n* (%)
Gender	Male	33 (63.5%)
	Female	19 (36.5%)
Race	White	46 (88.5%)
	Non-White	6 (11.5%)
Yearly family income	<$10k	3 (5.8%)
	$10k–$19.9k	5 (9.6%)
	$20k–$29.9k	6 (11.5%)
	$30k–$39.9k	5 (9.6%)
	$40k–$49.9k	7 (13.5%)
	$50k–$59.9k	4 (7.7%)
	$60k–$69.9k	3 (5.8%)
	$70k–$79.9k	4 (7.7%)
	$80k–$89.9k	6 (11.5%)
	$90k–$99.9k	3 (5.8%)
	>$100k	5 (9.6%)
Cause of burn	Thermal	21 (40.4%)
	Scald	17 (32.7%)
	Contact	8 (15.4%)
	Electrical	1 (1.9%)
	Friction	1 (1.9%)
	Other	1 (1.9%)
Received skin graft	Yes	35 (32.7%)
Visible burn injury	Yes	46 (92.0%)

Thermal = burn caused by external heat source (e.g., flame); Scald = burn caused by contact with heated liquids (e.g., coffee); Contact = burn caused by contact with heated objects (e.g., hot iron); Electrical = burn caused by contact with an electric current (e.g., exposed wire); and Friction = burn caused by rubbing against another surface (e.g., treadmill).

TABLE 2 | Descriptive statistics.

	n	Mean	SD	Min	Max
Clinical variables					
Time since burn (in months)	52	33.38	44.83	1.02	141.53
Total TBSA[a]	47	9.19	11.33	1.00	58.50
Psychosocial variables					
BISQ[b] total (parent)	47	4.96	5.24	0.00	25.00
BISQ total (youth)	45	5.18	4.74	0.00	18.00
CCSC[c] – active	46	2.65	0.65	1.25	3.96
CCSC – distraction	46	2.50	0.55	1.44	3.89
CCSC – avoidance	46	2.44	0.57	1.42	3.75
CCSC – support	46	2.31	0.78	1.00	3.56
PH-2[d] total self-concept	48	47.77	9.79	11.00	60.00

[a]*Total body surface area.*
[b]*Burn injury social questionnaire (BISQ).*
[c]*Child coping strategies checklist (CCSC).*
[d]*Piers-harris children's self-concept scale – 2nd edition (PH-2).*

the mean) and around 2% ($n = 1$) had significantly low self-concept T-scores (≥2 SD below the mean).

Bivariate Analyses

Results from bivariate correlations are presented in **Table 3**. To explore potential covariates ($p < 0.05$) for regression models, we examined bivariate correlations of age, TBSA, and time since burn with each of our primary outcomes (BISQ and PH-2 total scores). TBSA was positively associated with parent report ($r = 0.63$, $p < 0.001$) and youth report on the BISQ ($r = 0.40$, $p = 0.008$). Youth report on the PH-2 was negatively associated with age ($r = -0.34$, $p = 0.04$).

Correlations among primary study variables revealed that youth report on the PH-2 was negatively associated with youth reports on the BISQ ($r = -0.42$, $p = 0.005$) and positively associated with youth use of active coping strategies ($r = 0.41$, $p = 0.007$). The use of active coping strategies was positively correlated with the use of distraction ($r = 0.35$, $p = 0.017$), avoidance ($r = 0.61$, $p < 0.001$), and social support coping strategies ($r = 0.66$, $p < 0.001$). The use of social support coping was also positively correlated with avoidance coping ($r = 0.45$, $p = 0.002$). As expected, BISQ-parent and BISQ-youth scores were positively correlated ($r = 0.47$, $p = 0.001$).

Between-Group Comparisons

Independent-samples *t*-tests suggested that youth who had skin graft surgery reported more burn-related social problems on the BISQ ($M = 6.23$, $SD = 5.28$) than youth without a skin graft [$M = 2.86$, $SD = 1.75$; $t(40.78) = -3.19$, $p = 0.003$; $d = 0.86$]. There were no significant differences in youth PH-2 scores or parent BISQ scores between youth who did and did not have a skin graft. Male and female participants did not differ significantly in self-concept, or youth and parent report of burn-related social problems. Results from one-way ANOVA's suggested BISQ and youth PH-2 scores were not statistically significant between family income quartiles (p's > 0.05).

Regression Analyses

Hierarchical linear regression was used to examine whether coping strategies predicted (1) youth and (2) parent reports on the BISQ, as well as (3) youth report on the PH-2. Covariates (TBSA, presence of a skin graft) were entered in step one of the regression analysis, while all coping subscales from the CCSC were entered in step two. Results from three hierarchical linear regression analyses are reported in **Table 4**.

The first model examined whether coping variables predicted youth reports on the BISQ. The full model was statistically significant [$F(6, 36) = 4.01$, $p = 0.004$] and accounted for 30% of the variance in youth reports of social problems. In the full model, greater use of distraction coping predicted fewer youth-reported burn-specific social problems ($\beta = -0.39$, $p = 0.01$). Active, avoidance, and social support coping were not statistically significant predictors of youth reports on the BISQ. The presence of a skin graft ($\beta = 0.34$, $p = 0.02$) and TBSA ($\beta = 0.31$, $p = 0.03$) also significantly predicted more youth-reported burn-specific social problems on the BISQ.

The second model examined whether coping variables predicted parent reports on the BISQ. The full model was statistically significant [$F(5, 37) = 5.79$, $p < 0.001$] and accounted for 36% of the variance in parent reports of burn-specific social problems. In the full model, a greater TBSA predicted more parent reports of burn-specific social problems ($\beta = 0.65$, $p < 0.001$). None of the coping strategies were statistically significant predictors of parent reports on the BISQ.

The final model examined whether coping variables predicted youth self-concept as measured by the PH-2. The full model

TABLE 3 | Bivariate correlations.

	Age	Active coping	Distraction coping	Avoidance coping	Social support coping	BISQ – parent	BISQ – youth	PH2 – total	TBSA
Active coping	−0.10	---							
Distraction coping	0.04	0.35*	---						
Avoidance coping	−0.16	0.61***	0.27	---					
Social support coping	−0.14	0.66***	0.21	0.45**	---				
BISQ[a] – parent	−0.20	0.03	−0.08	0.09	−0.01	---			
BISQ[a] – youth	−0.01	−0.10	0.28	0.07	0.07	0.47**	---		
PH2[b] – total	−0.34*	0.37*	0.24	−0.03	0.15	−0.17	−0.46**	---	
TBSA[c]	−0.07	0.01	0.10	0.04	0.04	0.63***	0.40**	0.08	---
Time since burn	0.17	0.06	0.17	0.08	0.10	0.08	−0.27	−0.02	0.09

*$p<0.05$; **$p<0.01$; ***$p<0.001$.
[a]*Burn injury social questionnaire.*
[b]*Piers-harris children's self-concept scale – 2nd edition.*
[c]*Total body surface area.*

TABLE 4 | Hierarchical linear regressions.

Predictor variables	Outcome measure					
	BISQ youth total		BISQ parent total		PH-2 total self-concept	
	β	Adj. R^2	β	Adj. R^2	β	Adj. R^2
Step 1						
		0.17*		0.39***		0.04
Total TBSA	0.32*		0.63***		---	
Presence of graft	0.24		---		---	
Youth age	---		---		−0.25	
Step 2						
		0.30*		0.36		0.30**
Total TBSA	0.31*		0.65***		---	
Presence of graft	0.34*		---		---	
Youth age	---		---		−0.32*	
CCSC active	−0.25		0.09		0.67**	
CCSC distraction	−0.39**		−0.17		0.23	
CCSC avoidance	0.11		0.09		−0.36*	
CCSC support	0.28		−0.09		−0.18	
F(*df*)[a]	4.01 (6, 36)**		5.79 (5, 37)***		4.43 (5, 36)**	

[a]*F-ratio for full model.*
*$p<0.05$; **$p<0.01$; ***$p<0.001$.

was statistically significant [$F(5, 36) = 4.43$, $p = 0.003$] and accounted for 30% of the variance in youth self-concept. In the full model, greater use of active coping strategies predicted better youth self-concept ($\beta = 0.67$, $p = 0.002$), while greater use of avoidance coping predicted worse youth self-concept ($\beta = -0.36$, $p = 0.03$). Older youth also reported worse self-concept ($\beta = -0.32$, $p = 0.02$). None of the other coping strategies were statistically significant predictors of youth reports on the PH-2.

DISCUSSION

Burn injuries in children can lead to substantial psychosocial concerns stemming from the trauma of the injury itself, its painful treatment, and its physical sequelae. Thus, pediatric burn survivors are at an increased risk of negative psychosocial outcomes such as anxiety, depression, acute stress disorder, PTSD, and deficits in self-concept and social functioning (Landolt et al., 2009; Bakker et al., 2013; Kazis et al., 2017). To date, there is limited research on how coping relates to self-concept and social functioning in these youth (LeDoux et al., 1996; Gill, 2010). This study investigated associations between coping behaviors, social functioning, self-concept, and individual characteristics in pediatric burn survivors, with a particular focus on whether specific types of coping predicted social functioning and self-concept.

Our first hypothesis that there would be associations between coping, social functioning, self-concept, and burn injury-related variables was partially supported. Lower youth self-concept was associated with more youth reports of burn-specific social problems and older survivor age. Higher self-concept was

associated with greater use of active coping strategies. Youth and parent reports of more burn-specific social problems were also associated with higher TBSA.

Significant inter-correlations between coping subscales in our sample suggest that pediatric burn survivors rely on multiple forms of coping, rather than one specific or predominant coping strategy. This is similar to the findings of Camisasca et al. (2017), who found that youth who displayed secure attachment employed multiple coping strategies (i.e., active, social support, distraction, and avoidance) when faced with stressors. Likewise, Heffer and Willoughby (2017) alluded to the importance of coping flexibility with different coping strategies for different situations, which could explain why we obtained significant inter-correlations among coping strategies. The pattern of correlations was inconsistent, however, as not all coping subscales were correlated significantly with each other. It may be that inter-correlations were identified between active coping and social support, distraction, and avoidance coping because the use of active coping strategies involves attempts to reduce negative feelings and outcomes, which could consist of behaviors that resemble social support, distraction, and avoidance. Future research should continue to explore inter-correlations between use of different coping strategies to advance our understanding of the types of coping used in pediatric burn survivors and which combinations are most beneficial.

Our remaining hypotheses regarding the role of coping strategies as predictors of social functioning and self-concept were also partially supported. After controlling for burn injury variables and use of other coping strategies, greater use of distraction coping predicted fewer youth reports of burn-specific social problems. Similarly, less use of avoidance coping strategies and greater use of active coping strategies predicted lower youth self-concept after controlling for youth age and other coping strategies. Other coping strategies were not significant predictors of social functioning or self-concept in our sample. These findings align with prior research that suggests that the use of adaptive coping (e.g., active, social support, and distraction) is most effective in influencing positive youth psychosocial outcomes in youth with chronic illnesses (Meijer et al., 2002). Simultaneously, these results build upon the previous literature that found adaptive coping to be associated with positive parental mental health outcomes (Enlow et al., 2019). Adaptive coping is likely most effective because it requires youth to confront setbacks through problem solving and being cognizant of their thoughts and feelings toward the situation. Avoidance coping involves cognitive or behavioral strategies to avoid, deny, or minimize stressors and is associated with negative psychosocial outcomes (Smith et al., 2013; Mentrikoski et al., 2015). Consistent with past research, greater use of avoidance coping strategies predicted lower self-concept in pediatric burn survivors. However, use of avoidance coping was not significantly associated with burn-specific social problems in the current study. It may be that avoiding social problems with peers is less problematic and potentially adaptive in certain situations (e.g., staying away from bullies). Alternatively, this null finding may be due to the low statistical power of this study stemming from the small sample size.

Although the current study was conducted in the context of pediatric burns, our findings highlight the importance of adaptive coping strategies across illness populations. Empirically supported coping skills interventions can be delivered to help promote the use of adaptive coping strategies (e.g., open communication, conflict resolution, and problem solving; Jefferson et al., 2011). These interventions have been advantageous in other pediatric populations with chronic conditions, such as Type 1 diabetes (Hilliard and Hood, 2011). Taken together with the findings of the current study, it is likely that interventions which aim to increase the use of adaptive coping strategies (e.g., active, social support, and distraction), while decreasing the use of coping strategies associated with adverse psychosocial outcomes (e.g., avoidance coping) may be beneficial if tailored to pediatric burn survivors. In particular, a tailored intervention might focus on many of the same topics common to coping skills training (e.g., problem-solving, communication, and dealing with stress), but also target specific coping skills within burn-related scenarios (e.g., receiving wound care, responding to staring and bullying).

Consistent with prior research on burn-related psychosocial outcomes (Gill, 2010; Kazis et al., 2017), youth who had a skin graft stemming from a deeper burn and likely more scarring, reported more burn-related social problems. The association between burn-related variables and psychosocial outcomes has been mixed in the previous research. Some studies note that TBSA and scarring are associated with worse psychosocial outcomes (Karaçetin et al., 2014); yet, a meta-analysis concluded that clinical variables are not associated with psychosocial outcomes (e.g., anxiety, depression, and psychological maladjustment) in pediatric burn survivors (Noronha and Faust, 2007). Results from the current study suggest that burn-related variables may indeed be related to psychosocial outcomes and highlight a need for coping interventions to support survivors who experience more severe burn injuries. Active coping may underlie interventions that are often recommended for managing social reintegration after a severe burn (e.g., rehearsing responses in social situations, establishing contact with peer survivors; Phoenix Society, 2021). Although additional research is needed to fully understand the psychosocial impact of burn severity and skin grafts or other clinical variables in burn care, mental health interventions that emphasize coping in stressful social situations may be particularly beneficial for those survivors sustaining larger burn injuries, receiving a skin graft, or having visible scars or disfigurement.

A novel finding in the current study was the negative correlation between youth reports of burn-specific social problems and youth self-concept. It may be that youth who experience more social problems bear more negative feelings toward themselves. The previous research suggests that adolescents with negative self-concept are at a higher risk for social problems such as withdrawn behavior and internalizing problems (Ybrandt, 2008). With a larger sample, it would be possible to run analyses that examine the individual subscales of the PH-2 to have a clearer understanding of particular concerns in self-concept for pediatric burn survivors and how coping strategies foster or ameliorate these concerns. To this end, further evaluation of how specific coping approaches are related to specific aspects of self-concept may inform targeted mental health interventions.

Limitations

There were several factors that may have limited the results of the current study. The relatively small sample size resulted in reduced statistical power in detecting small and medium effects. Moderate associations were identified between some specific coping strategies and youth report of burn-related social problems and youth self-concept; therefore, the reduced power may have contributed to the lack of statistically significant findings. Future research should build upon this study by recruiting larger samples to achieve better statistical power and accuracy of estimates. A wide range of time elapsed since burn injury also was observed in our sample, limiting the extent to which these results can be generalized to specific populations of burn survivors. Additionally, our sample was primarily White. This unintentional lack of racial and ethnic diversity in our sample further limits the generalizability of our findings. A more diverse sample would also help highlight cultural considerations. It should also be noted that for participants selected through the registry, respondents may not have been representative of the pool of participants to which those letters were sent. The authors were unable to draw conclusions about the generalizability of these findings to other registry members as this data was unavailable for analysis. Further, this study is cross-sectional and did not allow for an evaluation of psychosocial outcomes over time. Future research would be strengthened by a longitudinal design, which would allow for evaluations of trajectory changes in psychosocial adjustment across time. Additionally, future research may wish to examine how the association between coping and social functioning changes as a function of time since the burn injury.

Strengths

The current study also demonstrated important strengths. For example, our sample was quite varied in terms of time since injury, which also was noted as a limitation. Our research design did not allow for examination of longitudinal trends within individuals but did permit a glimpse into how time since injury might related to psychosocial outcomes. An additional strength of this study was the use of both youth and parent reports of social functioning. This type of dual-report produced a more well-rounded understanding of each survivor's social functioning and reduced shared-method variance that may have increased the odds of a Type I error. Given some advantages (i.e., readability, brevity) of the BISQ, it may be useful to adapt and validate this tool with other pediatric illness populations, especially those involving appearance-related concerns. Other tools, such as the Pediatric Quality of Life Inventory (Varni et al., 1999) and the Children's Dermatology Life Quality Index (Parrish et al., 2020), include domains that measure interpersonal, social, and/or physical appearance concerns, which may be useful for validating the BISQ score. The inclusion of multiple coping strategies in relation to social functioning and self-concept was also a strength of this study, which is one of the first to measure the association between these particular psychosocial outcomes in pediatric burn survivors. As our understanding of coping develops, it would be beneficial to evaluate whether generic categories of coping that are studied across illness populations accurately reflect coping approaches used specifically in pediatric burns.

Observed associations between burn-injury variables, coping behaviors, social functioning and self-concept in this study suggest a need for psychosocial-based interventions that promote active coping and encourage further research of these specific outcomes in pediatric burn populations. Based on findings from previous studies (Holaday and McPhearson, 1997; Quezada et al., 2016), psychosocial interventions should focus on fostering positive social connections with family and peers as well as communication, social competence, problem-solving skills, self-esteem, and identification of self-worth in pediatric burn survivors. These factors are related to the development and growth of resilience – a characteristic that is essential for post-burn adjustment and well-being.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Allegheny Health Network Institutional Review Board and West Virginia University Institutional Review Board. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

CD, PE, and MS conceived and devised study. PE contributed to data collection. SY, PE, CA-N, and MS were involved in statistical analysis. MS, SY, and PE contributed to writing. CD, CA-N, and AA were involved in manuscript editing. All authors contributed to the article and approved the submitted version.

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